Guidance on Cancer Services

Improving Outcomes in Children and Young People with Cancer

The Evidence Review

NICE Stakeholder Consultation version

Contents

1. Introduction

This document contains a summary of the evidence reviewed for the production of the recommendations in *Guidance for Commissioning Cancer Services – Improving Outcomes in Children and Young People with Cancer - The Manual.* As with previous documents in this series, the topic areas are dealt with in the same order as in the Manual to facilitate cross referencing.

The purpose of the review is to determine the current evidence on interventions and models of care to guide and improve service provision for children and young people with cancer. The association between such evidence and patient outcomes is frequently lacking and in many instances it has been necessary to assume that improvement of health care service delivery and practice should enhance patient outcomes.

METHODOLOGY

- **Searching for evidence**

There are 3 stages to the identification and retrieval of evidence:-

(i) **Clinical question development:**

The members of the Guideline Development Group (GDG) were asked to consider the issues covered in the project scope and to submit clinical questions covering these issues. A total of 180 questions were submitted to the National Collaborating Centre for Cancer (NCC-C). It was clearly not possible to carry out full literature searches on

each question, due to time limitations. The clinical questions were therefore prioritised by the Director/Researcher/Chair/Clinical Lead for full searching (Appendix A) or 'high level' searching (Appendix B) and subsequent critical appraisal. The questions are presented in the evidence tables in the initial free form structure. These questions were converted to more structured questions for searching using the Population Intervention Comparison Outcome (PICO) format.

(ii) **Literature searching:**

Systematic. A systematic search strategy to identify published evidence for each clinical question was developed by the Information Specialists (Appendix A). The search period ended on the 6th December 2004.

High level searching: The wide range of the topic areas for consideration for children and young people with cancer necessitated the use of a pragmatic approach to searching for evidence in order to achieve production of the guidance within the timescales for delivery. It is clear that there had to be a balance between timeliness and rigour. Such an approach was also necessary to try and identify the type of literature relevant to service delivery. It is well known that the classical databases for medical literature, such as Medline, do not adequately index such literature. The Researcher used validated methods that involved the use of meta-search engines and other databases for 'high level' searching to quickly identify relevant evidence (Appendix B)

Identified titles and abstracts were initially screened for relevance to the clinical question by the Information Specialist and the Researcher. Definite inclusion/exclusion criteria were not employed for articles, because of the nature and variability of the literature on service delivery. In some instances help from a member of the GDG was enlisted to verify the relevance of selected articles and as a supplementary check on the completeness of the search. In general no formal contact was made with the authors for each paper identified, but occasionally communication was made for clarification of specific points.

(iii) Critical appraisal

The full papers were critically appraised using the methodology from the *NICE Guideline Development Methods* manual and the data relevant to the question was entered into an evidence table. Owing to practical limitations the final selection, critical appraisal and data extraction were undertaken by a single researcher. All tables were circulated to the GDG members for comments. References were also supplied by the GDG members and some stakeholder evidence was used. Both sources were always appraised for quality.

- **Synthesising evidence**

There were very few randomised controlled trials (RCTs) relevant to the majority of the clinical questions. This is a widely acknowledged problem with health service research and every effort was made to maximise the retrieval of

relevant high quality literature. Where available, evidence from good quality systematic reviews was appraised and included in the evidence tables; not all studies in the reviews were individually appraised.

Evidence for each topic was extracted into tables and summarised in the form of a considered judgement form (modified from the Scottish Intercollegiate Guideline Network methodology). The tables recommended for use in the NICE methodology manual were modified to accept the type of studies identified for service guidance. The quality of evidence was graded using the NICE hierarchy of evidence and the quality checklists. Evidence was usually rejected if graded as poor quality, apart from where it had been cited in the expert position papers and/or was of Level 1 type and was highly relevant to the question (Appendix C).

- **Expert position papers**

The GDG identified areas where there was a requirement for expert input. These areas were addressed by the production of a position paper by a recognised expert. Such experts were identified by contacting the relevant registered stakeholder and asking for a suitable nomination to deal with a particular topic area. A 'high level' search was performed to supplement these position papers, but there was usually no formal assessment of the papers cited within. These papers were presented at the GDG meetings for discussion. The papers that made a substantial contribution to the evidence are included in Appendices F-K.

Key strategic documents pertinent to paediatric oncology and/or child and young people's health were also identified as sources of evidence (Appendix A. The Manual). Relevant national and international guidelines were referred to during the guidance development process (Appendix A, The Manual). Where feasible the guidelines were appraised for quality using the Appraisal of Guidelines Research and Evaluation tool (AGREE).

- **Health economic evidence**

Economic evidence was extracted from the evidence tables, where it existed and was supplemented with searches performed by the Centre for the Economics of Health, University of Wales, Bangor.

- **Complementary research**

One complementary piece of research was commissioned to elicit children and young people's views about cancer service provision. The National Children's Bureau performed this study, the full results of which are given in Appendix D.

The results of a survey of teenagers (age range 14 -23 years) views on the provision of cancer services from a conference organised by the Teenage Cancer Trust in 2004 were also used to provide information on the specific requirements of this age group. (Appendix E).

- **Drafting recommendations**

The GDG members were allocated specific topic areas and asked to review the evidence tables pertaining to the topic and draft recommendations for the service guidance.

At the 10th GDG meeting of the 12 during the development phase, the GDG members participated in an event that involved external facilitation. This resulted in a list of three types of recommendations that were classified as essential, desirable and potential. The resulting recommendations were then examined by the Chair and Clinical Lead prior to the writing of the first draft of the guidance.

- **Agreeing recommendations**

Once an early draft of the guidance was produced, the GDG members were asked to review the draft document and consider whether:-

a) there appeared to be any major gaps in the synthesised evidence.
b) the recommendations were justified from the evidence presented and whether they were sufficiently practical and precise so that health service commissioners and the relevant front line health care professionals could implement them.

During the development of this guidance no formal consensus methods were used. Consensus was achieved by informal means during GDG meetings and correspondence outside the meetings.

The absence of high quality evidence for the majority of the clinical questions/topic areas made the grading of the recommendations impractical.

- **Writing of the guidance**

  The first formal draft version of the guidance was coordinated by the Chair and Clinical Lead of the GDG in accordance with the decisions of the GDG. The draft guidance was circulated for consultation according to the formal NICE stakeholder consultation and validation process prior to publication.

PRESENTATION, REFERRAL & DIAGNOSIS

The Question:

What is the evidence for delays in presentation, referral and diagnosis in children and young people with cancer?

Nature of the evidence

During the guidance development period the NICE clinical guidelines for general practitioners on *Referral for Suspected Cancer* [13] were released for consultation and some of the evidence contained in them was used, after critically appraising the articles.

Data was extracted from:-

10 historical case series, one of good quality, five of fair quality and four of fair to poor quality
1 retrospective comparative study with historical control of fair to poor quality
1 qualitative study of fair to poor quality
2 surveys, one of good quality and one fair to poor quality
1 audit of fair to poor quality

Summary of the supporting evidence for the recommendations

- There was consensus from the GDG members that implementation of the NICE GP referral guidelines for cancer should improve delays in referrals, but that training and resources would be required [13].
- The evidence from one historical case series indicated that 32% of patients with brain tumours are diagnosed within 30 days. The delay is contributed to by parental and physician delays and are caused by failure to recognise signs and symptoms [4].
- One survey of incidence rates of childhood cancer indicated that it was rare and that because of the rarity, guidelines for GPs were required [8]
- One historical case series demonstrated that the delay for brain stem tumours was greater than for other brain tumours. No effect of age or sex could be shown [12].
- Evidence from one historical case series demonstrated that age was significantly correlated with lag time. There was no correlation between lag time and outcome [16].

- Parental delays were shown to be shorter in one case series for children with acute lymphatic leukaemia (ALL) compared with brain tumours, doctor delays were the same [17].

- Evidence from one historical case series demonstrated that age, parental education level, lack of social security assistance affected time to diagnosis. Delays were greatest for Hodgkin's disease, retinoblastoma & unspecified neoplasms and shortest for leukaemia [7].
- The risk of local tumour invasion was increased with diagnostic delay in one historical case series. Primary healthcare professionals require education about the importance of ocular symptoms, especially squint, in paediatric patients [10].
- A review of qualitative studies concluded that there is a need for training in communication skills [1].
- Evidence from one historical case series of patients with retinoblastoma (RB) indicated that delays in diagnosis did not affect outcomes. There was a trend towards eye loss in bilateral RB [2.]
- There was evidence from one historical case series that younger children (0-2 yrs) are diagnosed more quickly than older children [5].
- In one comparative study the results showed that children < 5 yrs were diagnosed more quickly. The delay was greatest for brain tumours compared with acute lymphoblastic leukaemia and Wilm's tumours [9].
- Evidence from 1 historical case series suggests that the greatest delay in diagnosis is the failure of the family to recognise symptoms in patients with retinoblastoma [11].
- There was evidence of age correlation with lag time from one large historical case series for all solid tumour types except Hodgkin's disease[15].
- The preliminary results of an audit indicated that the 2 week referral method is not appropriate for childhood cancer [14].
- The results of the survey performed in 2004 by the Teenage Cancer Trust indicated that there are particular problems with delays in referral for older children and young people. (Appendix E)

There was a scarcity of papers that evaluated the reasons behind diagnostic delays. Furthermore the studies did not always distinguish between primary and secondary care related delays. Diagnostic delays do however appear to be correlated with age and the older the child, the longer the delay between presentation and diagnosis. For some cancers there is a lack of awareness by parents of the warning signs and symptoms. Delays are also contributed towards by difficulties that general practitioners have in recognising symptoms that may be vague and occur in other less serious illnesses.

EVIDENCE FOR DELAYS IN DIAGNOSIS, PRESENTATION AND REFERRAL IN CHILDREN AND YOUNG PEOPLE WITH CANCER

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
1. Arksey H,.Sloper P (1999) Disputed diagnoses: the cases of RSI and childhood cancer. *Social Science and Medicine* 49:483-97.	Adults with RSI Children (mean age 9.7 yrs, range 9/12 – 18 yrs) with cancer (leukaemia, lymphoma, solid tumours and CNS tumours) UK	Review of evidence for factors involved in diagnostic delays	• How much lay views count • Exercising choice • Referral pathways • Withdrawal of trust from medical practitioners	Interviews were performed with members of 98 (133 were identified as eligible; 74% response rate) families of children with cancer. 278 adults with RSI were interviewed. The evidence suggests:- • that the parents of children with cancer and adults with RSI felt that their experiences and knowledge were disregarded by doctors during the process of diagnosis. • There is a need for additional training in communication skills and occupational health problems.	*Reviews studies by Sloper 1996. Insufficient details of qualitative analysis methods used.*	Review of selected qualitative studies	3 +/-
2. Butros LJ, Abramson DH, Dunkel IJ (2002) Delayed diagnosis of retinoblastoma: analysis of degree, cause, and potential consequences. *Pediatrics* 109:1-5.	57 patients with retinoblastoma diagnosed between November 1993 – January 1998. US	Assessment of degree, cause and consequences of delays in diagnosis	Adverse effects of delayed diagnosis, such as eye loss.	The median time from presenting signs to diagnosis was 1.5 months (unilateral disease) and 2.25 months (bilateral disease). 77% of patients delayed seeking treatment. Primary care physicians delayed referral in 30% of cases. Diagnostic delays did not appear to have adverse effects on outcomes	*Recall bias possible. Small sample size. No p values stated. No discussion of reasons for patient attrition.*	Historical case series.	3 +/-

[1] See Appendix ? for explanation of evidence levels and quality grading

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
				although there was a trend towards eye loss being associated with longer delays in patients with bilateral retinoblastoma.			
3. Dixon-Woods M, Findlay M, Young B et al. (2001) Parents' accounts of obtaining a diagnosis of childhood cancer. *Lancet* 357:670-4.	20 parents whose children (aged 4-18 yrs) had a confirmed diagnosis of cancer (leukaemia) or brain or solid tumours UK.	Semi-structured interviews	The feelings of parents about the diagnosis process. Whether the narratives had implications for early diagnosis and referral.	Response rate 95%. There was good consistency between parent's accounts and the medical records. Data were analysed by the constant comparison method. The signs and symptoms of younger children were first noticed by parents. Parents of older children and adolescents often had to be told of problem. Early symptoms often vague. There were disputes in 7/20 families with the GP.	*The study is limited to 1 paediatric oncology unit. There were few examples of the types of tumour that can be prone to delays in diagnosis. Communication or information issues not addressed.*	Qualitative	3 +/-
4. Dobrovoljac M, Hengartner H, Boltshauser E et al. (2002) Delay in the diagnosis of paediatric brain tumours. *European Journal of Paediatrics* 161:663-7.	252 children with primary brain tumours diagnosed between 1980 – December 1999 Switzerland	Identification of reasons for delay in diagnosis.	Pre-diagnostic symptomatic interval (PSI) defined as interval between sign/symptom onset and the time of diagnosis by imaging.	The median pre-diagnostic symptomatic interval [PSI] (defined as the interval between onset of signs/symptoms and the time of diagnosis by imaging) was 60 days (range 0-8.2 years) with a parental delay of 14 days (range 0-6.3 yrs) and a doctor's delay of 30 days) range 0-8.2 yrs). 81 (32%) of the tumours were diagnosed within 30 days after symptom onset. Patients with raised intracranial pressure (ICP) had a statistically shorter PSI (median 60 versus 152 days; *P* = 0.007, Mann-Whitney test) and shorter doctor's	*Well described study with appropriate use of statistics. No distinction between delays in primary care and secondary care*	Historical case series	3 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
				delays (median 20 versus 60 days; *P* = 0.02, Mann-Whitney test) when compared with the children without increased ICP. However the parental delays for these two groups of patients were similar. Gender did not correlate with PSI, parental delay or doctor's delay. In 75 (45%), the doctor's delay was more than 30 days indicating misinterpretation of signs and/or symptoms. Common diagnostic difficulties included the correct interpretation of headache, nausea/vomiting, seizures, behavioural changes and squint/diplopia.			
5. Edgeworth J, Bullock P, Bailey AM et al. (1996) Why are brain tumours still being missed? *Archives of Disease in Childhood* 74:148-51.	74 children (0-16 yrs) with primary brain tumours admitted during 1990-1994 to a neurosurgical unit. UK	Examination of the duration and characteristics of symptoms and signs and the nature of consultations before diagnosis.		68% of children had not been correctly diagnosed at 1 month after symptom onset; at 6 months 28% were still not diagnosed. Children 0-2 years were diagnosed more quickly than older children (there was no difference present in histopathology grade, tumour location or number of consultations before diagnosis across the age groups). The mean duration of clinical history between the initial consultation and clinical diagnosis was 16.0 weeks (range 0-130 weeks). 62% of children were seen on f4 or more occasions before a diagnosis was made. There	*No examination of causal relationship. Some methodological problems with analysis of the qualitative interview data*.	Historical case series Questionnaire survey	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
				was no relationship between tumour site or duration of clinical history and incidence of psychological difficulty for any age group.			
6. Eiser C, Parkyn T, Havermans T et al. (1994) Parents' recall on the diagnosis of cancer in their child. *Psycho-oncology* 3:197-203.	30 families with a child diagnosed with cancer (ALL, lymphomas, solid tumours and brain tumours.	Determination of information parents' recall being given on diagnosis and assessment of information they would have liked.		In 20 cases mothers were told by the GP or local hospital before they received fuller information at the oncology unit or regional centre. 2/20 mothers reported that this initial explanation was incomplete. No real criticism of the way information was given at the oncology unit or regional centre. Policy in both centres was that children> 8 were told of diagnosis.	*Some relevance to question. Insufficient details given for appraisal*	Qualitative	3 ––
7. Fajardo-Gutierrez A, Sandoval-Mex AM, Mejia-Arangure JM et al. (2002) Clinical and social factors that affect time to diagnosis of Mexican children with cancer. *Medical and Pediatric Oncology* 39:25-31.	4940 children with cancer referred to secondary care Mexico	Estimation of delays in diagnosis and factors involved.		The time to diagnosis for all types of cancer ranged from 1 to 5 months. The shortest was for leukaemia (median = one month) and the longest for Hodgkin's disease, retinoblastoma and unspecified malignant neoplasms (median = five months). When grouped by age in years as < 1 (the reference age), 1-4, 5-9, and 10-14; the risk of a delayed time to diagnosis increased with age (x^2 = 29.12; P = 0.0001), the highest being for the 10-14 group (OR= 1.8; 95% CI = 1.4-2.3). Gender did not significantly affect time to diagnosis (OR	*Health service different in Mexico compared with UK.*	Historical case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
				= 1.1; 95% CI = 1.0-1.3). Parental educational level also influenced time to delay, and there was risk of delayed time to diagnosis in the lower compared to the higher educational level group (OR = 1.4; 95% CI = 1.1-1.8 for fathers, and OR = 1.5; 95% CI = 1.2-2.1 for mothers). The population without National Social Security had greater risk of delayed time to diagnosis (OR = 1.3; 95% CI = 1.1-1.4). The risk of delayed time to diagnosis varied among the different cancer types, but in general, age at diagnosis was the variable with greatest influence. .			
8. Feltbower RG, Lewis IJ, Picton S et al. (2004) Diagnosing childhood cancer in primary care – a realistic expectation? *British Journal of Cancer* 90:1882-1884.	1215 children < 15 yrs. ,diagnosed with cancer 1999-2001	Calculation of incidence rates across 2 strategic health authorities in Yorkshire (25 PCTs)	Rates of cancer for each PCT. Standardised morbidity ratios (SMRs).	The demographic and socioeconomic profiles of the PCTs in Yorkshire were highly representative of England and Wales: the median childhood population counts were 26,700 in Yorkshire compared with 27, 400 in the rest of E & W. No significant heterogeneity in SMRs across PCTs (p=0.09). The PCTs could expect 3-5 resident children to be newly diagnosed with cancer/yr. Based on the number of registered practitioners (defined as unrestricted principals & equivalents) [n=2050], a single GP will see a child	*Useful data in view of comparability with rest of E & W,*	Survey	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
				diagnosed with cancer once every 3 years on average. The authors conclude that childhood cancer is rare and therefore referral guidelines are required. There is also a need for availability of GP paediatricians i.e. GPs with a special interest.			
9. Flores LE, Williams DL, Bell BA et al. (1986) Delay in the diagnosis of paediatric brain tumours. *American Journal of Disease in Children* 140:684-6.	79 children (< 20 yrs) with primary brain tumours, diagnosed between 1976-1984. 45 patients with Wilm's tumours and 123 patients with ALL. US	Comparison of the interval from symptom onset to diagnosis in children with primary brain tumours with children with Wilm's tumours and ALL.		The mean diagnostic delay in patients with brain tumours was 26 weeks, with a median of six weeks. Patients less than 5 years of age who had infratentorial tumours and patients with more severe grades of signs and symptoms were diagnosed earlier. For patients with ALL the mean time to diagnosis was 4.5 weeks. The mean duration of symptoms for patients with Wilm's tumour was 2.8 weeks. When the three types of malignant neoplasms were considered, the primary brain tumour had a significant delay in diagnosis (p=<0.0001).	*Inadequate description of statistics. Small sample with inadequate power. Does not consider whether diagnostic delays are a function of the doctor or parent*	Retrospective comparative study	3 +/-
10. Goddard AG, Kingston JE, Hungerford JL (1999) Delay in diagnosis of retinoblastoma: risk factors and treatment outcome. *British Journal of Ophthalmology*	Parents of 100 children (with retinoblastoma treated between 1993-1996. 34 patients had bilateral disease and 66 unilateral.	Determination of extent of diagnostic delay and associated factors and the effect on treatment outcome	Parents were asked to recall the sequence of events from the time they first noted "something wrong" with their child's eye(s) to the diagnosis of	Leucocoria was the initial symptom in 52/100 patients. Squint, was the first symptom noted in 29 patients The parents of 10 patients noted change in the appearance of their child's eye(s). In nine patients the first	*Corroboration of the history obtained by parental interview by examination of patient records was achieved for 90/100 cases. Lag times could not be compared with visual outcome. Follow up*	Historical case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
83:1320-1323.	UK		retinoblastoma. Lag 1 = the time interval between the date the first symptom was noted and the date of first consultation with a primary healthcare professional (PHP) (parental delay) Lag 2 = the time interval between the date of the first consultation with a PHP and first consultation with a local ophthalmologist (health professional delay) Overall lag =the time from first symptom to referral for therapy in this institution.	symptom noted related to decreased visual acuity The median age at first symptom of patients with bilateral tumours was 5.0 (0-33) months. Patients with unilateral tumours were significantly older (p <0.001) with a median age of 18.0 (1-95) months at first symptom Although 49% of patients were referred to an ophthalmologist within 1 week of first 23% waited more than 8 weeks. There was a significantly increased risk of diagnostic delay in younger patients, those presenting with squint rather than leucocoria, and those first presenting to a health visitor rather than to a general practitioner. The risk of local tumour invasion was significantly increased by diagnostic delay. Treatment with primary enucleation was not increased by diagnostic delay. There were no deaths during the study period. The authors conclude that primary healthcare professionals require education about the importance of ocular symptoms, especially squint, in paediatric patients.	*ranged from 9 – 60 months. Correlation of diagnostic delay and outcome such as survival was **not** examined*		
11. Haik BG, Siedlecki A, Ellsworth RM et al. (1985)	250 cases of retinoblastoma referred to	Investigation of whether delays occur in diagnosis and	• Time from birth to first symptoms	28 patients (11%) had a family history of retinoblastoma. The median	*Poor description of statistics.*	Historical case series	3

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
Documented delays in the diagnosis of retinoblastoma. *Annals of Ophthalmology* 17:731-2.	ophthalmic oncology centre between 1974 and 1983.	referral	• Time from first symptom to examination in primary care • Time to subsequent referral to ophthalmologist	age at diagnosis was 6 months for such patients compared with 19 months for those with no family history. The longest interval was median time elapsed to first discernable symptom (4 months with positive family history [range 1-18 months], and 15 months without [range 1-115 months]). The next longest interval was median time elapsed from the primary care physician to referral to an ophthalmologist (five [range 1-32 weeks] and nine weeks [range 1-128 weeks], respectively). Significant percentages of primary care physicians (47% for children with no positive family history, and 25% for children with positive family history) delayed referral for a significant period of time (19 weeks for both groups). The mean time from first symptom to seeking the opinion of a primary care physician was 2 weeks (range 1-8 weeks) for children with a positive family history, and 5 weeks (range 1-100 weeks) for children with a negative family history. The authors conclude that the greatest delay in diagnosis is the failure of the patient's family to appreciate the significance of first symptoms.			+/-

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12. Mehta V, Chapman A, McNeely PD et al. (2002) Latency between symptom onset and diagnosis of paediatric brain tumours: an eastern Canadian geographic study. *Neurosurgery* 51:365-72.	104 patients (< 17 years) diagnosed with brain tumours between 1995 and 2000 Canada	Investigation of time required for diagnosis and factors involved in this diagnosis	Median time from symptom onset to diagnosis	The mean time from the onset of symptoms to diagnosis was 7.3 months (95% CI, 4.99-9.67 months) and only 41% of cases were correctly diagnosed within 3 visits to various doctors; 30% of children required > 7 visits. Sex or age did not affect time to diagnosis. Delays in diagnosis were significantly greater for brainstem tumours compared with those located elsewhere (mean = 11.76 months [95% CI, 3.13-20.39 months] versus 6.57 months [95% CI, 4.20-8.95 months], p = 0.014). Patients with medulloblastoma exhibited significantly shorter diagnostic times, compared with other pathological subtypes (mean = 3.78 months [95% CI, 1.97-5.59 months] versus 8.35 months [95% CI, 5.40-11.3 months], p = 0.006).	*Recall bias. Well described and designed study. The authors examined medical records and performed structured interviews. Appropriate use of statistics.*	Historical case series	
13. National Institute for Clinical Excellence (2004) *Referral guidelines for suspected cancer. Draft consultation 2004*. London: National Institute for Clinical Excellence 782p.	All cancer patients	Production of guidelines for referral for primary care		Reviews available evidence for factors producing delays in presentation and referral in primary care.		Guidelines	3/4 ++
14. Poirier V, Foot A, Walsh J et al. (2004)	Children diagnosed with a malignancy	Proforma sent to all GPs of children having	Route of presentation for	Proforma were available for 81/112 patients (72%).	*Small numbers. Only 64 responses for*	Audit	3/4 +/-

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Paediatric cancer – defining the pathway for children in the South West. SWCIS unpublished.		been diagnosed with a malignancy in 2003 in the South West Clinical Network	childhood cancer.	36/64 cases were referred within a week of their first visit to the GP. Fatigue, soft tissue mass and headache were the most frequent symptoms reported by GPs. Only 4 cases were referred using a faxed 2 week wait proforma. Giving a letter directly to the patient was the preferred mode of transmission for referral. The authors suggest that the 2 week referral method is not appropriate for childhood cancer.	*referral method. A similar study has been performed in secondary care, results available at end of 2004. The most common symptoms are in agreement with those recommended in the DoH referral guidelines.*		
15. Pollock BH, Krischer JP, Vietti TJ (1991) Interval between symptom onset and diagnosis of paediatric solid tumours. *Journal of Paediatrics* 119:725-32.	2665 children with solid tumours diagnosed between 1982-1988. (children were entered into POG therapeutic protocols) US	Identification of patient characteristics associated with longer lag times.	Lag times were calculated as the number of days between the onset of first symptoms and the date of diagnosis.	Median lag time ranged from 21 days for children with neuroblastoma to 72 days for those with Ewings sarcoma. (p <0.001). Age was positively and significantly correlated with lag time (p <0.001) for all tumour types except Hodgkin's disease (p = 0.58); Gender was significantly associated with lag time for non-Hodgkin's lymphoma (p = 0.02), for which girls had longer lag times. Race was significantly associated with lag time for osteosarcoma (p = 0.002), for which white children had longer lag times. Multivariate regression analysis was performed separately for each diagnostic group. With the exception of the Hodgkin's disease group, age remained	*Recall bias possible. The regression analyses for each diagnostic group only explained ~ 16% of the variance of lag time. In the non-Hodgkin lymphoma group the differences observed in the sexes could be accounted for in the distribution of histological subtypes of tumour Does not distinguish between physician related delay and parent/patient related delay.*	Historical case series	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
				a significant independent predictor of lag time for all diagnostic groups (p <0.05). Consistent with the univariate analysis, gender remained significantly associated with lag time for non-Hodgkin's lymphoma (p = 0.02). The multivariate analysis also revealed a significant association between gender and lag time for Ewing's sarcoma (p = 0.02). The association differed in these two tumour groups; girls had longer lag times in the non-Hodgkin lymphoma group but shorter lag times in the Ewing's sarcoma group. Also consistent with the univariate analysis, race maintained a statistically significant association with lag time only for osteosarcoma (p = 0.02). Signs and symptoms were compared for shorter (not more than the median) lag time and longer (greater than the median) lag time groups within each diagnostic category. Patients with shorter lag time for brain tumour had a 67% frequency of gait abnormalities and ataxia, compared with 59% for those with a longer lag time (p = 0.13), but were similar with respect to other common symptoms of brain tumour. For neuroblastoma, abdominal masses were			

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				more common in patients with shorter lag times (315 vs 19%; *p* = 0.037). Patients with shorter lag time for non-Hodgkin lymphoma had a higher frequency of abdominal masses (13% vs 5%; *p* = 0.06) and of breathing difficulty and coughing (32% vs 15%, *p* = 0.007).			
16. Saha V, Love S, Eden T et al. (1993) Determinants of symptom interval in childhood cancer. *Archives of Disease in Childhood* 68:771-4.	236 children (0-15 years) diagnosed with cancer from 1982-1990 UK	Identification of the determinants of lag time within multiple diagnostic groups and with age, sex and extent of disease. The study was designed to examine first the relation of lag time within multiple cancer diagnostic groups and with age, gender, and extent of disease in a cohort from the UK, and then compare the results with those of previous studies.	A child was considered to be symptomatic from the day that unrelieved symptoms that could be directly attributed to a malignancy were first recorded. The lag time was calculated from the date of onset of symptoms until the date of diagnosis to the nearest week, its relationship with patient demographic and clinical data was then examined.	The mean lag time varied from 2.8 weeks for nephroblastoma to 13.3 weeks in brain tumour. One way analysis of variance showed diagnostic group to be significant for length of lag time, F(6,161) = 5.5, *P* <0.001. There was no significant difference in the lag time between males and females. Age was a significant predictor for lag time, F(1,182) = 24.1, *P* <0.001, with older children having a longer lag time Both age, F(1,160) = 16.96, *P* <0.001 and diagnostic group, F(6,160) = 4.41, *P* <0.001 remained individually significant after multivariate analysis. The difference in lag time for children with acute leukaemia was not significantly related to a presenting white cell count of ≥ 50×1000³/l compared to those presenting	*Small numbers in each diagnostic category. Study does not assess directly the role of physician or parent. Recall bias possible..*	Historical case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
				with a lesser count. The difference in lag time between the stages in all diagnostic cancer groups was not significant either. The authors failed to find a positive correlation between of lag time and outcome.			
17. Thulesius H, Pola J, Hakanson A (2000) Diagnostic delay in pediatric malignancies – a population based study. *Acta Oncologica* 39:873-876.	64 children, mean age 7.8 yrs (0-16 yrs) with cancer (leukaemia & brain tumours)diagnosed between 1984-1995.	Investigation of the diagnostic process of childhood malignancies from the viewpoint of the GP, with the focus on the time course from initial symptoms until diagnosis and start of treatment.	Parental delay was defined as the interval from first symptoms to first consultation with a physician, and a doctor's delay as the time from first consultation to diagnosis. Treatment delay was the period from diagnosis to start of treatment. Lag time was the time from first symptoms to diagnosis.	Parent's delay was shorter than 4 weeks in 22 of 25 children with leukaemia, compared with 9 of 20 children with brain tumours (x^2 = 9.59, P = 0.002). For two children with leukaemia, parental delay was 3 months or more. The doctor's delay was <2 weeks for 17 of 25 children with leukaemia, compared with 7 of 21 children with a brain tumour (x^2 = 5.50, P = 0.019). Lag time was 4 weeks or less for 19 of 25 children with leukaemia, compared with 6 of 20 children with a brain tumour (x^2 = 9.52, P = 0.002). Median lag time also was 3 weeks (range 0-15) for children with leukaemia, and 9 weeks (range 1-199) for children with brain tumours (mean lag time was 3.8 [SD = 3.8] and 19.8 weeks [SD = 43.0], respectively). The mean number of visits to a GP in the year prior to tumour diagnosis was 2.3 for the children with leukaemia and 1.5 for the children with	*Insufficient details of statistics. Swedish medical system different from UK in that the GPs do not have a gatekeeper role, patients can see a specialist without a referral.* *Authors discuss the potential for bias in their study.*	Historical case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE[1] LEVEL/ QUALITY
				brain tumour (visits leading to diagnosis were included), and 0.2 and 0.6, respectively, the year after diagnosis. In the control group, the mean number of visits to a GP was 1.0 in both years. Diagnostic delays are longer for children with brain tumours compared with children with leukaemia.			

CHEMOTHERAPY

The Questions:

1. Does the place of administration and management of chemotherapy (CT) affect outcome?
2. What evidence is there that community delivered chemotherapy is delivered more safely and effectively by nursing staff than by parents?
3. Are there reliable methods to monitor chemotherapy treatment compliance?
4. Are protocol compliance and effectiveness greater when treatment is performed by a shared care centre compared with a tertiary care centre?
5. What evidence is there for non-compliance with cancer therapy in children and young people?

Nature of the evidence

Q.1

2 randomised trials of fair to poor quality
4 systematic reviews, 3 of good quality, 1 fair to poor quality

Q.2

2 systematic reviews of good quality
2 historical case series one of fair quality, one fair to poor quality

Q.3 & 4

3 case series, 1 of fair quality, 2 of fair to poor quality
1 review of good quality

Q.5

1 systematic review of RCTs of good quality
1 non randomised controlled trial of fair quality
2 qualitative studies of poor quality*
1 historical case series of fair to poor quality
1 guideline of fair quality
1 literature review of fair to poor quality
1 review of fair quality
1 expert opinion of poor quality*

* Included as forms part of the evidence in expert position paper

1 expert position paper (Appendix F)

Summary of the supporting evidence for the recommendations

Q.1

- The results of the two randomised controlled trials should be interpreted with caution because of methodological problems. One study [3] found no difference in patient satisfaction or quality of life in patients treated at home. The other trial findings suggest that home treatment may be feasible under some circumstances [4].
- The results of 1 systematic review (all patients with cancer) conclude that there is insufficient evidence on the clinical effectiveness of home versus non-home settings [5]. There is some evidence to show that home treatment delivery is safe, although patient selection and training are pivotal . 1 systematic review concluded that there is no evidence for patient acceptability or preferences [1].
- 1 systematic review indicated the requisite criteria for successful delivery of home CT [2].
- No conclusions on the clinical effectiveness of home CT could be made because of the variability of studies in 1 systematic review [5].
- A review of the effect of home treatment on quality of life concluded that the evidence was inconclusive [6].

Q. 2

- There was no evidence from 1 systematic review on the superiority of nurses vs parents [1].
- No conclusions could be drawn from 1 systematic review [4].
- The results from one small case series indicated that parents were enthusiastic about home CT treatment [2].
- 1 small historical case series demonstrated that parents could be trained to administer iv chemotherapy; no CT related adverse events [3].

N.B. There were no studies directly comparing nurses vs parents**.**

Q.3 & 4

- The evidence from 1 historical case series demonstrated that hamaglobin and weight changes were poor parameters for measuring compliance to prednisone [6].
- The evidence for the assessment of compliance to prednisone and penicillin by metabolite assays was poor [2].

- Assays of 6 mercaptopurine metabolites were used in 1 very small historical case series to measure compliance [3].
- The conclusions from 1 review were that the usefulness of urinary steroid assays was limited. Assays of 6 MP and its metabolites only provide information on short term compliance. Evidence from 1 study indicates that assays of RBC 6-TGNs and MeMPs allow early identification of non-compliance [1].

There was no evidence for shared care versus tertiary care with respect to superiority for compliance.

Q. 5

- The evidence from 1 systematic review indicated that there were no RCTs that fulfilled the inclusion criteria and the evidence on compliance was poor [4].
- The evidence from 1 non RCT for the effectiveness of 1 type of patient information leaflet was inconclusive [3].
- The evidence from 2 poor quality qualitative studies had some implications for communication between HC professionals and patients and the beliefs of patients and how they affect compliance [5].
- 1 historical case series demonstrated that younger patients preferred a more participatory role [2].
- 1 review (6 studies of relevance to C & A) indicated that non compliance in children was between 2-50%; adolescents were the least compliant [6].
- 1 guideline from SIOP gives the major reasons for non compliance in children and adolescents [8].
- 1 literature review indicated that the evidence on compliance was poor [1].
- 1 expert opinion suggested that patient information leaflets do not provide adequate help to patients and carers [7].

There was insufficient evidence on place of delivery of chemotherapy and its effect on outcomes and on the feasibility of home delivery of chemotherapy, although there was some evidence to indicate that home delivery produces improved quality of life for patients and carers.

The importance of suitable facilities and the presence of appropriately trained staff were confirmed by some Level 3 evidence.

There was evidence indicating that compliance is a particular problem in teenagers and young people. Electronic transfer of prescriptions (ETP) does appear to reduce prescribing errors, but there was no evidence specific for children and young people. Data is lacking for the effect of ETP on compliance in children and young people with cancer.

Q1. DOES THE PLACE OF ADMINISTRATION AND MANAGEMENT OF CHEMOTHERAPY AFFECT OUTCOME?

HOSPITAL vs COMMUNITY

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Agence d'Evaluation des Technologies et des Modes d'Intervention en Sante (AETMIS) (2001) *Home chemotherapy - systematic review (project)*. Agence d'Evaluation des Technologies et des Modes d'Intervention en Sante (AETMIS).	Adults and children with cancer	Review of literature on home chemotherapy for cancer patients. Qualitative interviews with service providers in Quebec and Ontario	Clinical effectiveness, QOL, implications for health care system, ethical, legal & social implications, alternatives	The authors conclude that there is insufficient evidence on the clinical effectiveness of home chemotherapy compared with non-home settings. There is more evidence to show that home treatment can be delivered safely, although patient selection & training is paramount. Evidence was poor for improvements in patient QOL with home CT. No direct evidence for patient acceptability and preference for home CT. A total of 17 interviews were performed between May 2001 and March 2002. These interviews revealed variable delivery of home CT and the need for well integrated collaborative teams of health care professionals.	*Well designed and comprehensive review of literature on home chemotherapy. Clear description of clinical questions and review methods. Valid conclusions drawn from available evidence. Highly relevant to CT questions. The authors make recommendations for conditions which are a perquisite for home CT.*	Systematic review	2++ ++
2. L'Agence Nationale d'Accréditation et d'Évaluation en Santé (2003) *Critères d'èligibilite des patients*	Cancer patients, receiving chemotherapy	Development of criteria for selecting patients who may be eligible to receive CT at home		The authors conclude after a review of the literature that the following conditions must be satisfied for CT to be delivered at home: • Written therapeutic	*Good review of literature and presentation of evidence based statements.*	Systematic review	2^{++} ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
à une chimiotherapie anticancereuse a domicile. Consensus formalise de professionnels.L'Agence Nationale d'Accréditation et d'Évaluation en Santé 27p.				protocol • Patient preference and consent to home treatment • Good adjustment (no psychological problems) to disease • Acceptance by the primary care physician to supervise the home treatment • Acceptance by the community nursing infrastructure to participate in the home care • Acceptance by other health care professionals involved in domiciliary care e.g. pharmacists, psychologists etc. • Adequate quality of home conditions • Adequate means of communication methods, particularly for medical emergencies.			
3. King MT,Hall J, Caleo S et al. (2000) Home or hospital? An evaluation of the costs, preferences, and outcomes of domiciliary chemotherapy. *International Journal of Health Services*	46 patients with breast cancer, 27 with colorectal cancer and 1 with head neck cancer	Comparison of home CT with OP or day care administration of CT	Questionnaire determination of preference, satisfaction, unmet need & QOL.	40/74 patients, completed the study. Home based care more expensive that IP treatment due to extra nurse time. No difference was found between patient satisfaction or QOL in the 2 settings.	*Good validation of questionnaire but study methodology poor No details of how randomisation was performed. Patient selection bias. No intention to treat analyses. In Australia most CT*	*Prospective randomised controlled trial*	1⁻ +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
30:557-579.					*is provided on an OP basis or day care. Not relevant to question*		
4. Mor V, Stalker MZ, Gralla R et al. (1988) Day hospital as an alternative to inpatient care for cancer patients; a random assignment trial. *Journal of Clinical Epidemiology* 41:771-785.	To compare adult day hospital care with usual in-patient care for cancer patients. 2 year single centre study. Total no of patients: 442 Adult Day Hospital (ADH): n=229; Inpatient n=213 Eligible patients required: 4-8 hr treatment plan, including chemotherapy, and other long-term intravenous treatments; stable cardiovascular status; mental competence; no skilled overnight nursing; helper to assist with home care. Patients ineligible if standard outpatient treatment possible.	Adult Day Hospital: 12-bed pilot unit included nursing core, treatment room; 2 follow-up rooms, education centre, satellite pharmacy, waiting lounge & administrative offices. Designed to create relaxing & comfortable environment facilitating communication among patients, families and staff. Physician director responsible for clinical management of unit. Patients dealt with by own physician. No house staff signed to unit. Telephone access to Nurse 24hrs/day, 7 days/week. No overnight accommodation; patients unable to return home for medical reasons admitted to hospital. Inpatient care: standard inpatient care, no details given.	Clinical, psychosocial and cost outcomes evaluated over 60day period.	No statistically significant ($p<0.05$) differences found between ADH and inpatient care in medical or psychosocial outcomes over 60-day study period. During study period 28 (6.3%) patients died (13 (5.7%) ADH patients; 15 (7%) Inpatients). Treatment stratas differed as did survival rates in treatment stratas. Patients interviewed for psychosocial status of patients and family: (ADH 198/229 (87%); Inpatient 188/213 (88%)). Patient Evaluation of ADH (ADH vs Inpatient) Scale of 1= worst; 7= best: Rated nurses self-care instruction (5.9 vs 4.5; $p<0.001$); helpfulness of staff (6.6 vs 5.3;$p<0.001$); access to follow-up care and attractiveness of the environment significantly (6.7 vs 6.1; $p\leq0.001$) higher than inpatients. No difference found in number of family provided direct hours of care,	*Few patients, within C & A age range. Authors conclude study demonstrates that day hospital care of medical oncology patients is clinically equivalent to Inpatient care, causes no negative psychosocial effects and costs less than Inpatient care. Findings support trend toward dehospitalisation of medical treatment.* *Analyses of cost, clinical & psychosocial outcomes performed on different sub samples of 442 randomised cases. Cost analyses included patients who died or removed from study. Other analyses seemed to "lose" patients, not include all data etc.*	RCT Randomisation stratified by treatment.	1- -

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
	Country: USA			subjectively assessed family disruption, or reported time lost from work. No observed differences in time lost from work between employed patients in ADH and Inpatient groups. Major difference was in medical costs – approximately one-third lower for ADH patients ($p<0.001$) than for Inpatient group. Initial concerns among physicians about ADH setting. By end of 2 yrs study period 76% of active medical oncologists in Hospital admitted patients to ADH.	*Study conducted some time ago (before 1988) and results may not apply to treatment protocols used today.*		
5. Parker G, Bhakta P, Lovett CA et al.(2002) A systematic review of the costs and effectiveness of different models of paediatric home care. *Health Technology Assessment* 6:1-118.	Children with all diseases receiving home treatment	Review of the literature on costs and effectiveness of home care	Costs	The variability in and quality of the studies reporting costs of home CT were so great that no conclusions could be drawn about the relative costs of home and hospital-based CT. No conclusions could be made on clinical effectiveness	*Well designed and reported review of world literature.*	Systematic review	2^{++} ++
6. Smeenk FW, van Haastregt JC, de Witte LP et al. (1996) Effectiveness of home care programmes for patients with incurable cancer on their quality of life and time spent in hospital: systematic review. *British Medical Journal* 316:1939-	All patients with incurable cancer, receiving home care programmes.	To investigate the literature on the superiority, in terms of QOL and reduction in readmission, of home care programmes compared with standard care	QOL; readmission to hospital.	9/358 prospective randomised controlled trials of moderate quality were identified that fulfilled the inclusion criteria. The evidence was inconclusive for the effectiveness of home care programmes.	*Not possible to determine patient characteristics.* *Well designed and reported study with clear description of inclusion criteria and search strategies.* *Not of relevance to question*	Systematic review	2^{++} –

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1944.							

Q2. WHAT EVIDENCE IS THERE THAT COMMUNITY DELIVERED CHEMOTHERAPY IS DELIVERED MORE SAFELY AND EFFECTIVELY BY NURSING STAFF THAN BY PARENTS?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Agence d'Evaluation des Technologies et des Modes d'Intervention en Sante (AETMIS) (2001) *Home chemotherapy - systematic review (project)*. Agence d'Evaluation des Technologies et des Modes d'Intervention en Sante (AETMIS).				**SEE EVIDENCE TABLE Q 1**			
2. Hooker L (1999) Safety, efficacy, and acceptability of home intravenous therapy administered by parents of pediatric oncology patients. *Medical & Pediatric Oncology* 32:421-426.	35 paediatric oncology patients in 1 UKCCSG centre.	Parents views on acceptability of home (iv antibiotics) treatment of FNP. Two groups of children received home antibiotic therapy: an early discharge group following hospital admission for treatment of infection	Admission to hospital	During study period there were 83 patient episodes of infection or FNP requiring admission. In 36 episodes, the course of antibiotics was completed at home. 16 episodes were managed at home. In the early discharge group 4 patients subsequently readmitted. Of the non-admitted patients 1	*Small numbers. Useful for written care protocol for parents for administration of iv therapy.*	Case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
		and those not admitted to hospital.		required subsequent admission. Parents felt that home treatment helped them to cope (72%) and learned more about their child's illness and treatment (82%)			
3. Jayabose S, van Haastregt JC, de Witte LP et al (1992) Home chemotherapy for children with cancer. *Cancer* 69:574-579.	Parents of 20 children with cancer receiving CT (most frequently cytosine arabinoside) at home US	Establishment of a home training programme for home delivery of intravenous CT.	Adverse events	The only criteria used for patient selection was • Frequent visits for CT to clinic 3 parents of poor socio-economic status were excluded. No CT related adverse events were noted. The authors conclude that home based CT is a safe and cost effective alternative to hospital or clinic based CT.	*Small study. Difficult to determine if patients were truly unselected.*	Historical case series	3 +/-
4. Parker G, Bhakta P, Lovett CA et al. (2002) A systematic review of the costs and effectiveness of different models of paediatric home care. *Health Technology Assessment* 6:issue 35.	Children with all diseases receiving home treatment	Review of the literature on costs and effectiveness of home care	Costs	The variability in and quality of the studies reporting costs of home CT were so great that no conclusions could be drawn about the relative costs of home and hospital-based CT. No conclusions could be made on clinical effectiveness	*Well designed and reported review of world literature.*	Systematic review	2^{++} ++

Q3. ARE THERE RELIABLE METHODS TO MONITOR CHEMOTHERAPY TREATMENT COMPLIANCE?

Q4. ARE PROTOCOL COMPLIANCE AND EFFECTIVENESS GREATER WHEN TREATMENT IS PERFORMED BY A SHARED CARE CENTRE COMPARED WITH A TERTIARY CARE CENTRE?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Davies HA, Lilleyman JS (1995) Compliance with oral chemotherapy in childhood lymphoblastic leukaemia. *Cancer Treatment.Reviews* 21:93-103.	Children with lymphoblastic leukaemia	Oral chemotherapy	Compliance with therapy	Overview of studies indicate that • Usefulness of urinary steroid assays as an assessment of compliance is limited • Assays of 6-MP and its metabolites only provide information about short term compliance. Assay of RBC 6-TGNs and MeMPs allow early identification of non-compliance *1 study only)*.	*Reviews major studies on compliance with ALL therapy and methods for detecting compliance.*	Review	3/4 ++
2. Festa RS, Tamaroff MH, Chasalow F (1992) Therapeutic adherence to oral medication regimens by adolescents with cancer. I. Laboratory assessment. *Journal of Pediatrics* 120:807-811.	21 patients (15.6 yrs ± 2.2yrs)15 with ALL and 6 with HD who were taking prednisone. 29 patients (19.1 yrs ± 4.1 yrs) with HD whose CT been stopped and taking penicillin	Laboratory assessment of outpatient adherence to CT (prednisone) and penicillin therapy.	Non adherence to therapy assessed by assays of metabolites.	11 patients (52%) nonadherent to prednisone treatment. 14 (48%) nonadherent to penicillin treatment.	*Only deals with prednisone and penicillin adherence. No discussion on validity of assay methods. No discussion of controls*	Case series	3 –
3. Lancaster D, Lennard L, Lilleyman JS (1997) Profile of non-compliance in lymphoblastic leukaemia. *Archives of Disease in Childhood* 76:365-366.	496 children with acute lymphoblastic leukaemia prescribed 6-mercaptopurine	Assays of 6-mercaptopurine to indicate compliance with therapy.	Levels of 6 MP. 2^{o} outcomes = remission	9 children (2%) had undetectable 6 MP metabolites. 5/9 were adolescents. 7/9 continue to be in remission.	*Small study. Follow up period not defined.*	Historical case series	3 –
4. Lilleyman JS, Lennard L (1996) Non-	Childhood ALL	-	-	-	-	Commentary	4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
compliance with oral chemotherapy in childhood leukaemia. *British Medical Journal* 313:1219-1220.							–
5. Partridge AH, Avorn J, Wang PS et al (2002) Adherence to therapy with oral antineoplastic agents. *Journal of the National Cancer Institute* 94:652-661.				**SEE EVIDENCE TABLE QUESTION 5**			
6. Smith SD, Rosen D, Trueworthy RC et al (1979) A reliable method for evaluating drug compliance in children with cancer. *Cancer* 43:169-173.	52 children, age range 8 mnths – 17 years. 43 with ALL, 5 with AML; 4 Non HL.	Evaluation of prednisone compliance by measuring Hb, weight changes & 17-ketogenic steroids.	Evaluation of compliance	33% children and 59% of adolescents were not compliant. Hb and weight change were poor parameters for measuring compliance.	*Numerous confounders. No description of validation of methods. Relevant to question*	Case series	3 +

Q5. NON-COMPLIANCE WITH CANCER THERAPY IN CHILDREN AND YOUNG PEOPLE

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Carter S, Taylor D, Levenson R (2003) *A question of choice - compliance in medicine taking* London: Medicines Partnership 87p.	Cancer patients			The authors conclude that the evidence on medication compliance is limited because most treatment is administered under the direct supervision of health professionals. Most research on compliance has been performed in the context of clinical trials and uses drop out rates as the measure of compliance. The paper by *Partridge et al. (2002)* revealed poor compliance in the paediatric oncology population (see below). The paper by Spinetta et al. (2002) was also identified (see below).	*Good quality search but articles not appraised for quality.*	Literature review	4 +/-
2. Cassileth BR, Zupkis RV, Sutton-Smith K et al. (1980) Information and participation preferences among cancer patients. *Annals of Internal Medicine* 92:832-6.	256 cancer patients. (type unspecified), all ages	Use of Information Styles Questionnaire and the Beck Hopelessness Scale to examine the degree to which patients, prefer to become informed about and to participate in their medical care.	Patents expressed views	The younger the patients the more closely they conformed to the well informed participant standard of patient behaviour. The older the patients the more likely they were to prefer the older nonparticipatory patient role.	*Insufficient details about patients,*	Historical case series	3 +/-
3. Dickinson D, Raynor DK, Duman P (2001) Patient information leaflets for medicines: using	Two matched groups of 20 consumers given either the European Commission leaflet	Comparison of consumers' ability to use 2 different patient information leaflets.	The groups were required to find and understand 15 pieces of information in the	The target that each question should be answered correctly by 16/20 consumers was achieved for 3/15 points in the EC leaflet compared with	*Indicates the importance of consumer testing. Not specific for cancer.*	Non randomised controlled trial	2⁻

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
consumer testing to determine the most effective design. *Patient Education and Counselling* 43:147-159.	(based on 'prescriptive' model for leaflets)or the Mark II leaflet (based on best practice in information design)		leaflets	8 in the Mark II leaflet.. Open questioning confirmed the problems with the EC leaflet.			+
4. Haynes RB, McDonald H, Garg AX et al. (2002) Interventions for helping patients to follow prescriptions for medications *The Cochrane Database of Systematic Reviews* Issue 2.	Patients with any medical condition requiring drug prescription.	Evaluation of the results of published RCTs on interventions to improve compliance with medications	Original data concerning medication adherence. One or > measures of treatment outcome. At least 6 month follow up from time of patient entry	No RCTs concerning cancer patients fulfilled the inclusion criteria. For short term treatments 1/3 interventions reported in 3 RCTs showed an effect on both adherence and outcome. 18/36 interventions reported in 30 RCTs were associated with improvement in adherence but only 16 interventions led to improved treatment outcomes. The effective interventions were complex; improvements were not large.	*Not directly relevant to C & A cancer. Good review with adequate description of methodology and limitations of included studies such as lack of concealment allocation, presence of confounding factors*	Systematic review of RCTs	1^{+} ++
5. Horne R, Weinman J (1999) Patients' beliefs about prescribed medicines and their role in adherence to treatment in chronic physical illness. *Journal of Psychosomatic Research* 47:555-567.	Patients with asthma, cardiac conditions, chronic renal failure and cancer patients	Investigation of whether patients beliefs and perceptions of their illness and treatment affected compliance	Factors influencing compliance with therapy	Specific beliefs about medicines were a strong predictor of compliance (19% of observed variance). Demographic variables were less significant. The authors conclude that it is important to address patients' beliefs when considering compliance.	*No patients within C & A range. Limited relevance. Small numbers*	Qualitative; interviews	3 –
6. Partridge AH, Avorn J, Wang PS et al. (2002) Adherence to therapy with oral antineoplastic agents. *Journal of the National Cancer Institute* 94:652:661.	All patients with cancer	Review of published studies	Non-compliance with anti-neoplastic agents.	Six studies were identified of children and adolescents with leukaemia or non Hodgkin's lymphoma, acute lymphoblastic leukaemia or Hodgkin's disease. The studies reviewed indicated that there was poor compliance in children with	*All observational historical studies with associated biases but some identified C & A studies of adequate quality.*	Review	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				cancer. Non compliance ranged from 2 – 50%. Measures and definitions of compliance varied between studies. Adolescents were the least compliant. Those most at risk had poorer understanding of their illness and raised levels of denial compared with those who were compliant. The relationship between the involvement of parents and compliance were important.			
7. Raynor DK, Savage I, Knapp PR et al. (2004) We are the experts: people with asthma talk about their medicine information needs. *Patient Education and Counselling* 53:167-174.	Patients with asthma	Effect of the provision of patient information leaflets.		The results indicate that patient information leaflets do not provide adequate help to patients and carers. This is a particular issue with paediatric prescriptions, where the drugs are often prescribed outside their licensed indications.	*Not directly relevant to C & A cancer.*	Expert opinion	4 –
8. Spinetta JJ, Masera G, Eden T et al. (2002) Refusal, non-compliance, and abandonment of treatment in children and adolescents with cancer: A report of the SIOP Working Committee on psychosocial issues in pediatric oncology. *Medical and Pediatric Oncology* 38:114-117.	Children and adolescents with cancer	Consideration of:- • the causes of refusal, non-compliance and abandonment of treatment. • The prevention of above • Judicial intervention, when essential		The major reasons for refusal, non-compliance and abandonment of oncology therapy in children and adolescents include • poor communication of diagnosis and treatment regimens • fear of side effects • poor understanding of the seriousness of the illness • physical discomfort • frustration with length of treatment • lack of knowledge about benefits of therapy	*Report of SIOP Psychosocial Working Group. Literature reviewed but no details given on how recommendations are formed.*	Guidelines	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				• increased availability of alternative medicine • availability of social support services			

SURGERY

The Question:

Does specialist (surgical) care improve outcomes for children and young people with cancer?

Nature of the evidence

2 systematic reviews 1 of good quality, 1 of fair quality
3 guidelines of good quality
2 reviews of good quality
4 expert opinions 2 of good quality, fair quality

Summary of the supporting evidence for the recommendations

- The paediatric studies did not meet the inclusion criteria in one systematic review and in the second review the evidence from the paediatric studies reviewed was inconclusive for the benefits of specialisation [8] [9].

- UK guidelines recommended specialist surgeons and anaesthetists for the treatment of children and young people with cancer(no evidence given) [3] [4] [11].

- The literature reviews provided evidence that survival in paediatric cancer was improved with specialist care, but the studies reviewed were generally of poor quality [7] [10].

- The expert opinions concluded that there was evidence for improved outcomes with specialist surgeons and anaesthetists [1] [2] [5] [12].

There is general consensus that specialisation is associated with improved patient outcomes, but there is a lack of good evidence to support this. The requirements to provide optimum surgical treatment are specified in a number of UK guidelines and strategic documents (Appendix 1 – the Manual). There is some observational evidence that specialisation is required in anaesthetic and pathology service provision (see Evidence table below).

DOES SPECIALISATION IN THE CARE OF CHILDREN AND YOUNG PEOPLE WITH CANCER IMPROVE OUTCOMES?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Arul GS, Spicer RD (1998) Where should paediatric surgery be performed? *Archives of Disease in Childhood* 79:65-72.	Paediatric surgery	Discussion and review of evidence for:- • Role of specialist paediatric surgery centre • Provision of non-specialist paediatric. surgery In district general hospitals (DGHs)		The authors conclude;- • There are arguments for and against large regional specialist paediatric centres. If specialist paediatric emergency transport is available the benefits of centralisation outweigh the adverse effects of the need to transport children to such a regional centre. • There is clear evidence that all neonatal surgery and anaesthesia should be conducted by specialists. • There is debate about the critical mass necessary to maintain specialist expertise in surgery. • There is lack of data from DGHs on the set up of paediatric surgical. services. Debate continues about the benefits of properly trained paediatric surgeons taking over surgery at a DGH.	*Good review of evidence for organisation of paediatric surgical services*	Expert opinion	4 ++
2. Atwell JD, Spargo PM (1992) The provision of safe surgery for children. *Archives of Disease Children* 67:345-349.	Paediatric surgery	Discussion and review of 1989 NCEPOD report and its implications for paediatric surgeons and anaesthetists		The authors conclude:- • there are no data comparing paediatric anaesthetic morbidity in a DGH with that in a regional paediatric centre. There are some surveys of perioperative complications post anaesthesia in children	*Limited review of evidence and dated*	Expert opinion	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				from UK, France & Canada, the results of which suggest the requirement for centralisation of paediatric anaesthesia (and intensive care) services. • The NCEPOD report highlighted the dangers of insufficient critical mass for paediatric surgery, but did not define the limits. • One solution may be the referral of all children < 3 yrs old to a specialist centre			
3. British Association Paediatric Surgeons (2001) *Response to the Kennedy Report. "Learning from Bristol"*. London: British Association of Paediatric Surgeons. Available from: www.baps.org.uk/Admin/Documents/Publications/BAPS%20Response%20to%20Bristol.htm		Response to the Kennedy report		The BAPS state:- • They strongly agree that where a small number of centres offer a specialist service, the requirements of quality and safety should prevail over ease of access • The designation of supra-regional units must be based on performance and outcome measures and not their geographical situation. • Centres must be constantly monitored by the designating authority to ensure that their continuing status as a supra-regional unit is justified. • Children's acute hospital services ideally should be located in a children's hospital close to an acute general hospital. • The Report does not distinguish between specialist paediatric surgery provided in specialist centres and general	*Important to take note of in any paediatric service issues*	Expert opinion	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				paediatric surgery provided by appropriately trained surgeons at DGHs • The provision of specialist paediatric surgery services on split sites is not acceptable • The recommendation that all surgeons who operate on children must obtain a recognised professional qualification requires clarification			
4. British Association of Paediatric Surgeons (1995) *A guide for purchasers and providers of paediatric surgical services.* Edinburgh: The Royal College of Surgeons of Edinburgh.	Paediatric surgical services	Recommendations for purchasers		The authors consider:- • The definitions of the spectrum of paediatric surgery • The provision of paediatric surgical services. The best clinical outcomes are achieved when the number of patients being treated in a unit is sufficient for a high level of surgical & nursing expertise to be maintained. • Specialist paediatric surgery must be provided in a specialist paediatric surgery. Unit. The **requirements** are:- • Trained and accredited paediatric surgeons and paediatric anaesthetists. • A full range of specialist services for children including paediatrics, neonatology, paediatric intensive care, radiology, neurosurgery, nephrology, cardiology,		Guidelines	4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				oncology & pathology • Nursing staff trained in paediatric nursing and paediatric critical care nursing Support services catering for the specific needs of children, including dieticians, social workers, play leaders and teachers • Facilities designed for children, including the accident and emergency department, outpatient department, wards, operating theatres, day case unit, radiology suite and laboratory services. • Accommodation for parents, who should have unrestricted access to their children **Population Base:-** There should be 1 specialist surgeon /500,000 population. It is unrealistic to plan a department with < 4 paed. surgeons and 1 paed. urologist. Thus 2.5 million is the minimum population required to ensure a sufficient critical mass. Strict adherence to this figure would increase the burden of travel in some rural areas and specialist paed surgery. should be provided at a regional unit.			
5. British Association of Paediatric Surgeons (2002) *Paediatric*	Paediatric surgical patients	Standards of care for paediatric surgery			*Detailed description of standards and manpower*	Expert opinion	4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
surgery; standards of care. London: British Association of Paediatric Surgeons 23p. Available from: www.baps.org.uk/documents/standards%20of%20care%20finaldoc.doc					*requirements and service arrangements.*		
6. British Association of Paediatric Surgeons (2003) *Reconfiguration in paediatric surgery*. London: British Association of Paediatric Surgeons 4p. Available from: www.baps.org.uk/reconfigpaed.doc				The authors conclude:- • At present the service is delivered in the British Isles by 120 consultant paediatric surgeons. This equates to 1 consultant/0.5 million population; in Australia, North America and most of Europe it is 1/250/000/300,000 population. This ratio is necessary in the British isles • Reconfiguration can best be achieved by an expansion of consultant numbers to 200.	*Latest figures for workforce*	Expert opinion	4 ++
7. Grilli R, Minozzi S, Tinazzi A et al. (1998) Do specialists do it better? The impact of specialization on the processes and outcomes of care for cancer patients. *Annals of Oncology* 9:365-374.	Patients with cancer receiving specialist carer	Assess the impact of specialisation on processes & outcomes of care for cancer patients.	Mortality, morbidity. Process outcomes e.g. specialisation of treating clinician, numbers of patients treated.	47/189 potential studies met the inclusion criteria. 12/24 (50%) studies provided information on process and 17/32 (53%) information on outcomes. Overall results were in favour of specialised clinicians/centres and were generally statistically significant. The study quality was however low	*Well described and designed study. Note is taken of the need to adjust in comparisons for case mix. The authors discuss the possibility of publication bias, influence of methodological flaws, use of observational studies causing an over estimate of effect size. The aims and inclusion*	Review	4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
					criteria were well defined. Care is required in concluding that there is good evidence for the apparent superiority of specialist versus non-specialist care		
8. Harding M, Lord J, Littlejohns P et al. (2002) *A systematic review of the evidence relating process of care or outcome to treatment in specialist and non-specialist hospital settings*. London: St George's Hospital Medical School 208p.	Patients with cancer	Assessment of difference in outcome between treatment in specialist and non specialist centres	Survival	The authors conclude that there was insufficient high quality evidence to indicate that specialist care affected outcomes in cancer patients	*High quality study No studies in paediatric cancer met the inclusion criteria. Publication bias significant.*	Systematic review	1^{-} ++
9. Hillner BE, Smith TJ, Desch CE (2000) Hospital and physician volume or specialization and outcomes in cancer treatment: importance in quality of cancer care. *Journal of Clinical Oncology* 18:2327-2340.	All types of cancer care	Evidence to support that hospital or physician volume or specialty affects outcome of cancer care.		A consistent literature was identified that support a volume-outcome relationship for cancers treated with technically complex surgical procedures. These studies identified 30-day mortality and used the hospital as the unit of analysis. For cancer treated with low-risk surgery there were fewer studies and there was only an association for colorectal and breast cancer.	*Search limited to Medline 1988-1999. Indirect relevance to question.*	Systematic review	2^{++} +
10. Parkes SE, Muir KR, Cameron AH et al. (1997) The need for specialist review of	Histopathology of 2104 biopsies of paediatric solid tumours	Assessment of variability in diagnosis of childhood cancer by paediatric and general	Analysis of conformity by percentage agreement, kappa	Birch Marsden classification was used. 348 (16.5%) of the 2104 original diagnoses were amended by the review	*Reasonable evidence to support sub specialisation of pathologists.*	Historical case series	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
pathology in paediatric cancer. *British Journal of Cancer* 75:1156-1159.		pathologists. Confirmation of diagnosis was made independently by 3 specialist pathologists	statistic & weighted kappa	panel. 23 cases originally diagnosed as malignant were reclassified as non-malignant The panel confirmed the original diagnosis of paediatric pathologists in 89% of cases (kappa = 0.76; w kappa = 0.78) compared with 78% 9 kappa= 0.59; w kappa 0.54) for general pathologists.			
11. Pheby DFH, Bray FI (1998) *Review of studies designed to explain variations in cancer disease outcome, particularly in relation to variations in patterns of practice.* Bristol: University of the West of England 161p. Available from: www.uwe.ac.uk/fas/uae/cancer3.pdf	Patients with ICD9 diagnosis 140-208 of cancer, of any age	Review of studies on variations in cancer outcomes in relation to variations in patterns of practice.	Survival	4 papers dealing with childhood cancer fulfilled inclusion criteria. There were data problems, but overall the studies indicated that survival was improved with treatment at specialist centres.	*Comprehensive literature review and discussion of the literature and factors affecting cancer outcomes.*	Review	4 ++
12. Royal College of Anaesthetists (2001) Guidance on the provision of paediatric anaesthetic services. *Royal college Anaesthetists Bulletin* 8:355-359.	Paediatric anaesthetic services			The authors conclude ;- • Anaesthesia for children requires specially trained medical and nursing staff & special facilities. • The service should be led at all times by consultants who anaesthetise children regularly • Adequate assistance to the anaesthetist by staff with paediatric training & skills must be available • Paediatric anaesthetic	*Detailed guidance for provision of paediatric anaesthetic services. Covers staffing, education organisation & administration*	Guidelines	4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				equipment must be available where children are treated and staff must receive regular retraining in paediatric life support • There should be properly funded acute pain services.			
13. Society of British Neurological Surgeons (2001) *Safe paediatric neurosurgery* London: Society of British Neurological Surgeons 7p. Available from: www.sbns.org.uk/docs/BOOKLET_SAFE_PAEDIATRIC_NEUROSURGEY_2001.doc	Children requiring neurosurgery	Standards	-	Results of task force set up in 1997 to set out minimum requirement of safe paediatric neurosurgery in the UK. • in neurosurgery the techniques for head injury, haemorrhage, hydrocephalus and some brain tumours do not differ radically between children and adults and adult neurosurgeons may provide an appropriate degree of care and level of skill. certain paediatric neurosurgical conditions are rare and it is generally accepted that they would be best managed by neurosurgeons with the appropriate paediatric specialist training and expertise. • Neurosurgical units providing specialist paediatric neurosurgical services should have sufficient facilities and resources to allow immediate transfer, urgent same day admission or admission within 48 hours as necessary. Specialist paediatric services must have appropriate support facilities including access to Paediatric Intensive		Expert opinion	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				and High Dependency Care. • Neurosurgeons providing specialist paediatric neurosurgical expertise should have a regular defined commitment to paediatric neurosurgery, including the necessary theatre and outpatient clinic time within their weekly timetable. • the neurosurgical training programme will give every trainee neurosurgeon exposure to paediatric neurosurgery and specific training in the management of paediatric neurosurgical emergencies sufficient to enable them to manage an emergency when on call. • In certain neurosurgical units which provide paediatric services some further subspecialty expertise may develop. • the management of children with childhood brain and spinal tumours is best accomplished by those with a subspecialty interest and expertise using a multi-disciplinary approach. Accordingly neurosurgical units which undertake the management of paediatric brain and spinal tumours must have access to paediatric oncologists who are members of or affiliated to the UK Children's Cancer Study Group.			

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				• Neurosurgical units which undertake paediatric work should be responsible for the development and dissemination of agreed guidelines for patient management and for their regular up-dating and perform regular audit.			

NEUROSURGERY

The Questions:

Do specialist paediatric neuroncology surgeons produce improved outcomes for children and young people with cancer?

Nature of the evidence

1 historical case series of fair quality
4 guidance/guidelines/policy documents 2 of good quality, 2 fair quality
2 commentaries/expert opinions of fair quality
1 overview of fair quality

Summary of the supporting evidence for the recommendations

- The evidence from 1 historical case series indicated that tumour resection was maximal with specialist paediatric neurosurgeons. Complication rates were less with accredited neurosurgeons [1].
- One policy document provides recommendations for safe neurosurgery [2].
- One guidance predating the above policy document makes recommendations for safe neurosurgery [4].
- Clear recommendations for paediatric neurosurgery are given in one guideline [5]
- Specialist neurosurgical units for paediatric brain tumours are recommended in one guidance [7].
- One commentary concludes that there is good evidence to support specialisation in paediatric neurosurgery [6].
- No definite recommendations for specialisation, specifically in the care of patients with gliomas, are made in one commentary [8].
- One overview makes no recommendations on specialisation [3].

There is evidence from expert opinion and formal consensus that care of children and young people with brain tumours should be delivered in the context of multidisciplinary teams (MDTs).

DO SPECIALIST PAEDIATRIC NEUROONCOLOGY SURGEONS PRODUCE IMPROVED OUTCOMES IN CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Albright AL, Sposto R, Holmes E et al. (2000) Correlation of neurosurgical subspecialisation with outcomes in children with malignant brain tumors. *Neurosurgery* 47:879-885.	732 children, 485 with medulloblastoma/primitive neuroectodermal tumours and 247 with malignant gliomas. US	Evaluation of association between the type of neurosurgeon (general or paediatric) and outcomes	Extent of tumour removal. Neurological complications.	Operations were performed by 269 neurosurgeons (213 general NS, 29 designated paediatric NS and 27 ASPN members. The mean number of operations /surgeon was 1.8, 4.9 and 7.6 for general, paediatric and ASPN respectively. There was a significant relationship between the extent of tumour resection and the type of neurosurgeon. Designated paediatric NSs and ASPN members were more likely to remove> 90% of the tumour than were general NSs ($p<0.05$). The probability of extensive tumour removal also correlated with the number of operations the neurosurgeon performed ($p<0.01$). Neurological complications occurred in 23% of cases operated upon by a general NS; 32% designated paediatric neurosurgeons and ASPN 18%	*Relevance of US practice to UK?*	Historical case series	3 +
2. Chumas P, Hardy D, Hockley A et al. (2002) Safe paediatric neurosurgery 2001. *British Journal of*	Paediatric neurosurgical	Update to 1998 policy document from SBNS.	-	The authors conclude:- • The management of children with brain and spinal tumours is best accomplished by those with	*No supporting evidence*	Policy document	4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Neurosurgery 16:208-210.				subspecialty interest and using a multidisciplinary approach. • NS units managing paediatric brain and spinal tumours must have access to paediatric oncologists who are members or affiliated to the UKCCSG. • Cross referral from other NS to these specialised services to be encouraged. • Specialised NS units will need to develop clinical networks. • Guidelines should be developed and audit of activity performed			+
3. Gerrard GE, Prestwich RJ, Franks KN et al (2003) Neuro-oncology practice in the UK. *Clinical Oncology* 15:478-484.	All cancer centres in UK	Questionnaire survey to determine current practice(July 2000)by UK clinical oncologists specialising in neuro-oncology. Workshops in 2000 and 2002		41/54 (76%) response rate. There were marked variations in practice. Results were obtained for controversial areas of management such as adults with high grade glioma, adults with grade !! glioma, ependymomas, meningiomas, primary adenomas & multiple brain metastases. The authors conclude that there are many controversial areas in which there is lack of trial evidence upon which to formulate a consensus for best practice. The development of a national database would be a useful tool.	*Good document to highlight controversial areas of neurosurgical oncological treatment*	Overview	4 +
4. Society of British	Paediatric	Recommendations		**The recommendations**	*Contains*	Guidance	4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Neurological Surgeons (1998) *Safe paediatric neurosurgery* London: Society of British Neurological Surgeons.	neurosurgery			**are:-** • High quality children's care should be delivered by appropriate staff and facilities. • Paediatric neurosurgery should offer at the very minimum, the same quality, degree of care and level of expertise regarded as a norm for adult NS practice. • Neurosurgical units offering a paediatric service must be capable of delivering a full comprehensive 24 hour service. This will require a minimum of 2 consultant WTEs and middle grade cover consisting of both NS and paediatric. staff The minimal requirement of a comprehensive 24hr service is paediatric beds, PICU, paediatric neuro-anaesthesia, paediatric nurses and on-site Ct scanning. The need for specialist services in neuro-radiology and neuropathology is recognised. Paediatric neurologists and general paediatric support is essential. • Audit is essential • In some regions there needs to be an assessment of the delivery of paediatric NS with some degree of rationalisation and co-	*specifications for workforce, training facilities etc. The 2001 update does not provide amendments to these data.*		+

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				ordination between departments			
5. Society of British Neurological Surgeons (2001) *Safe paediatric neurosurgery* London: Society of British Neurological Surgeons 7p. Available from: www.sbns.org.uk/docs/BOOKLET_SAFE_PAEDIATRIC_NEUROSURGEY_2001.doc	Paediatric neurosurgery	Standards		• All neurosurgical units providing care for neurosurgical emergencies should have clinicians with the necessary experience and training to undertake the immediate care of neurosurgical emergencies occurring in children. If separate facilities for children are not available then children should not be housed in adult facilities for longer than is required for their safe neurosurgical management and the child should be transferred to appropriate paediatric facilities as soon as is practicable. • Units undertaking the emergency care of children with neurosurgical problems must have access to CT and MR scanning. • Neurosurgical units providing specialist paediatric neurosurgical services should have sufficient facilities and resources to allow immediate transfer, urgent same day admission or admission within 48 hours as necessary. • Specialist paediatric services must have appropriate support		Guidelines	4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				facilities including access to Paediatric Intensive and High Dependency Care. Such facilities should be supported by specialist neuroradiologists, neuropathologists and Anaesthetists with the necessary expertise. Access to CT and MR imaging is essential. Paediatric neurologists should also be available. The Consultant medical team must be supported by properly trained and qualified nurses including theatre staff and professionals in allied disciplines. The specialist paediatric neurosurgical unit should have a paediatric environment able to support the social requirements of children and family, e.g. play area, schooling and family accommodation. • In certain neurosurgical units which provide paediatric services some further subspecialty expertise may develop. The development of such ultra-specialised paediatric neurosurgical expertise will normally require a further period of specialist training and experience Accordingly neurosurgical units which undertake the management of paediatric			

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				brain and spinal tumours must have access to paediatric oncologists who are members of or affiliated to the UK Children's Cancer Study Group.			
6. Stevens MC, Hockley AD, Spooner D et al (1995) Treatment for children with brain tumours. *British Medical Journal* 311:1213-1214.	Paediatric brain tumours	Recommendations for treatment		Author concludes:- • Most progress in the treatment of childhood cancer been due to improved survival from entry into trials • Treatment abroad is not supported by evidence for clinical benefit. Innovation in treatment is necessary Arguments for specialist referral are compelling. Only 62% of all children with tumours of the CNS diagnosed in the UK between 1991-1993 were referred to designated paediatric oncology centres in contrast with 90% of children with leukaemia and 79% with other forms of malignant disease.		Commentary	4 +
7. United Kingdom Children's Cancer Study Group & Society of British Neurological Surgeons (1997) *Guidance for services for children and young people with brain and spinal tumours*. Leicester: United Kingdom Children's Cancer Study Group 32p.	Children & young people with brain tumours			The authors conclude:- • Existing referral patterns in the UK not been clearly defined • In line with Calman/Hine report it is proposed that a network of UKCCSG centres be established which specialise in treatment of children and young people with brain tumours. All children and young people with suspected brain or spinal tumour should be treated in a	*Key document, 1997 no update. No supporting evidence.*	Guidance.	4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				unit where clinical services meet standards in DoH guidance documents about services for children and young people • It is hoped that concentration of expertise will lead to improved standards of care Quality of care will be further enhanced through audit & research			
8. Walker DA, Punt JAG, Sokal M (1999) Clinical management of brain stem glioma. *Archives of Disease of Childhood* 80:558-564.		Paediatric brain stem glioma		Author concludes:- • Gliomas are the most difficult of all paediatric tumours to treat because of problems with diagnosis, lack of effective treatments and consensus between specialists. • Modern neuroimaging techniques now facilitate diagnosis. • Lack of appreciation of presenting symptoms lead to delays in diagnosis • Surgery and/or radiotherapy are treatment options. • There is not standard chemotherapy regime. There should be specific recommendations developed for supportive care.	*Key review of management of gliomas. Some recommendations dated (1999)*	Commentary	4 +

RADIOTHERAPY

The Questions:

Do delays in radiotherapy (RT) and quality of radiotherapy affect patient outcomes in children and young people with cancer?

What evidence is there for the provision of specialist radiotherapy facilities producing improved outcomes?

Nature of the evidence

3 randomised controlled trials, two of fair quality, 1 fair to poor
2 retrospective cohort studies of good quality
1 systematic review of good quality
4 historical case series, 1 of fair quality, 3 of fair to poor quality
1 literature review of fair to poor quality

Summary of the supporting evidence for the recommendations

- One RCT of children with medulloblastoma concluded that there was no significant difference in overall survival or event free survival (EFS) starting radiotherapy within 42 days of surgery [9].
- The evidence from 1 RCT of patients with Wilm's tumours indicated that delays in RT were associated with tumour control [10].
- The evidence from 1 systematic review indicated that delaying RT in patients with high grade gliomas may affect outcomes [8].
- There is evidence from 2 retrospective cohort studies that demonstrate that delays in RT affect tumour control in patients with sarcoma [4 6].
- One historical case series demonstrated that in patients (> 3yrs) with medulloblastoma that delays in RT did not affect outcome [1].
- Multivariate analyses from 1 historical case series (Grade III or IV gliomas) demonstrated that increased time from presentation to the RT department was associated with reduced survival [2].
- The evidence from 1 historical case series showed that tumour recurrence rates of Wilm's tumours were not affected by delays in RT [3].
- The evidence from 1 historical case series of patients with Ewing's sarcoma did not indicate a significant effect of RT delays on outcomes [5].
- A recent literature review provides evidence for the effect of RT delays on high grade gliomas [7].

There appears to be a lack of consistent evidence for the effect of delays of radiotherapy on outcomes.

The recommendations for the provision of specialist RT facilities are in agreement with a move to sub specialisation in clinical oncology as outlined in the Calman Hine report and the publications from the royal colleges. The resource requirements are also specified in guidance from the UKCCSG and there is emphasis on the need to provide age appropriate facilities in line with the general recommendation in the children's National Service Framework.

DO DELAYS IN RADIOTHERAPY AFFECT PATIENT OUTCOMES IN CHILD AND YOUNG PEOPLE CANCER PATIENTS?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
1. DelCharco JO, Bolek TW, McCollough WM et al. (1998) Medulloblastoma: Time- dose relationship based on a 30-year review. *International Journal of Radiation Oncology, Biology, Physics* 42:147-154.	53 patients with medulloblastoma. Age range 3 to 77 years (7 children < 3 years were excluded from analysis). Country: USA	Postoperative craniospinal radiotherapy given with curative intent between 1963 and 1993 at one centre. Dose ranged from 23.3 to 45.6 Gy. Different fractionation regimes were used. 7 patients(13%) had biopsy alone; 28 patients (53%) had subtotal excision; 18 patients (34%) had gross total excision. 11 patients had adjuvant chemotherapy	Relationship between the following factors and survival (5 and 10 years), freedom from relapse, and disease control in the posterior fossa were examined: radiotherapy (dose to craniospinal axis and posterior fossa), fractionation, time from surgery to radiotherapy and duration of radiotherapy.	Absolute survival rates were 68% at 5 years and 64% at 10 years. Freedom from relapse rates were 61% at 5 years and 52% at 10 years. Rates of disease control in the posterior fossa were 79% at 5 years and 68% at 10 years. Only results pertaining to time between surgery and radiotherapy are reported below:- The time between surgery and the start of radiotherapy was not significantly associated with posterior fossa disease (P = 0.115); freedom from relapse (P = 0.311)or overall survival (P not reported). The authors conclude that radiotherapy treatment for more than 45 days increased freedom from relapse and improved posterior fossa disease control. Time from surgery to radiotherapy was not associated with outcome.	*Small sample size. Range of time between surgery and start of radiotherapy was not reported. Not restricted to children and adolescents. Results from 30 year period during which radiotherapy treatment regimes changed.*	Historical case series	3 +/-
2. Do V, Gebski V, Barton MB (2000) The effect of	182 patients with grade III/IV gliomas	Radiotherapy delays	Survival	3/182 survived at the end of the study. Median	*Authors acknowledge the known limitations*	Historical case series	3

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
waiting for radiotherapy for grade III/IV gliomas. *Radiotherapy & Oncology* 57:131-136.	with a follow up 10.5 months. Median age at presentation was 57 yrs. (range 16-84 yrs).			survival of the whole group = 8.5 months. Multivariate analysis indicated that reduced survival associated with older age, reduced dose and prolonged PWT. The hazard of death increased by 2% per day of PWT (p=0.03). Two waiting time variables were examined:- • time from biopsy to start of radiotherapy (BWT) • time from presentation to RT department (PWT) Poor prognostic variables such as age, surgery, dose, ECOG were not associated with longer PWTs.	*of their study – selection bias, uncontrolled patient and tumour factors and treatment bias. Median age 57 years, few patients in age range for C & A guidance*		+/-
3. Kalapurakal JA, Li SM, Breslow NE et al. (2003) Influence of radiation therapy delay on abdominal tumor recurrence in patients with favorable histology Wilms'' tumor treated on NWTS-3 and NWTS-4: a report from the National Wilms'' Tumor Study Group. *International Journal of Radiation Oncology, Biology, Physics* 57:495-499.	Total 1226 patients with Stage II, III or IV Favourable Histology (FH) Wilms'' Tumour who received flank or abdominal Radiation Therapy (RT) study guidelines. Male (567); Female (659). Randomised: eligible if between 1 & 15yrs inclusive & not received treatment for nephrectomy for unilateral disease.	Study NWTS-3: No. patients: 657; Regimen: DD1000 (n=116); DD2000 (n=168); K1000 (n=61); K20000 (n=137); DD-RT (n=107); J (n=68); Study NWTS-4: No. patients = 569 Regimen DD (n=313); DD-4A (n=234); DD4 (n=22) Details of	RT delay: Category 1: 0-9 days; Category 2 ≥10 days. Flank recurrence: recurrent disease in operative bed. Abdominal recurrence: infradiaphragmatic tumour recurrences, including flank recurrences.	Mean RT delay was 10.9 days; median delay was 9 days (range: 1-227 days). RT delay was in the range of 8 to 12 days after nephrectomy in 59% patients. RT delay did not vary substantially with patient's gender, race, age, tumour stage or treatment regimen. Univariate & multivariate analysis did not reveal RT delay of ≥10 days to significantly influence flank	*No patient characteristics given. Part of a larger study. Methods section mentions "randomised"; "switched" & "followed" as categories but no explanation given in study of if/how these applied.*	Historical case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
	Switched: if assigned regimen changed later in study. Followed: included those not randomised but treated according to one of the arms of study. Exclusions: received preoperative chemotherapy, bilateral (Stage V) tumours, tumours arising from fused kidney, extrarenal Wilms', or adult Wilms' (age ≥16). Country: USA	chemotherapy regimens not supplied in this report (referred to previous reports)	Excluded: Tumour recurrence in liver Tumour recurrence in opposite kidney.	and abdominal tumour recurrence rates in NWTS-3 or NWTS-4. Flank recurrence: N=18/1226 (1.5%); Abdominal tumour recurrence: 59/1226 (4.8%). Median follow-up for 1,070 was 12 yrs (range 0.1-22yrs). Median time to death for 156 deceased patients was 1.75yrs from diagnosis. 8yr flank tumour recurrence risk of 0-9 days was 1.9% and ≥10 days was 1.2% (5 recurrences vs 7.2 recurrences expected)). (p=0.3). 8yr abdominal tumour recurrence rates for 0-9days were 4.8% and ≥10 days 5.3% (25 recurrences vs 23.6 recurrences expected) (p=0.7). Study reports Median RT delay for NWTS 1 to 4: 1: 4; 2: 8; 3: 9; 4: 9. Authors concluded that RT delay of ≥10 days did not significantly influence flank or abdominal tumour recurrence rates among children with FH tumours			

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
				treated on NWTS-3 and NWTS-4. However unable to test for meaningful difference as concentration of RT delay close to 10 days.			
4. Raney RB Jr, Tefft M, Newton WA et al. (1987) Improved prognosis with intensive treatment of children with cranial soft tissue sarcomas arising in nonorbital parameningeal sites. A report from the Intergroup Rhabdomyosarcoma Study. *Cancer* 59:147-155.	95 patients (median age 6 yrs) with nonorbital cranial parameningeal sarcoma comprised the preintensive treatment group (RT began 6 weeks after start of CT) and .68 patients(median age 5 yrs) the intensive treatment group (RT started at day 0).	Comparison of CT and nonintensive RT with intensive RT	Remission rate. Tumour free survival	The remission rate in the preintensive group was 65/95 patients (68%) and 52/68 (76%) in the intensive group (p <0. 25). The authors attribute the better results to the early institution of wide-field RT for those patients with risk of meningeal extension	. *Well designed and described study with appropriate analyses*	Retrospective cohort study	2 +
5. Schuck A, Rube C, Konemann S et al. (2003) Postoperative radiotherapy in the treatment of Ewing tumours: influence of the interval between surgery and radiotherapy. *Strahlentherapie und Onkologie* 178:25-31.	153 patients with Ewing tumours treated between 1985 and 1998. Age Range : 3 to 33) Male (94); Female (59); Patients were part of other ongoing trials reported elsewhere from Country: Germany	Surgery, chemotherapy, radiotherapy. Patients received different regimens of chemotherapy. Median follow-up 70 mths (range 7 to 169 mths). Postoperative radiotherapy applied either in conventional fractionation or in hyperfractionated accelerated split course. Dose dependent on intraoperative resection margins and	Local and combined relapse as first event; Local or combined relapse as second event. First events: local relapse, systemic relapse, death for any reason and secondary neoplasm.	Median interval between surgery & irradiation was 79 days. **Group 1:**46 patients: postoperative radiation started ≤60 days after surgery; (9 patients ≤30 days) Combined and local systemic relapse as first event: 1/46 (2%) Local or combined relapse as second event: 3/46 (6.5%) Freedom of local and combined local and systemic relapses after 5 yrs for >30days ≤60 was	*Results not specifically for children/adolescents. Results include patients from other trials – unclear as to which patients on what chemotherapy regimens etc.*	Historical case series	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
		on response to initial chemotherapy. Patients with wide resection, poor response to chemotherapy or with marginal resection and good response to chemotherapy received 45 Gy; Patients with intralesional resection or marginal resection, poor response to chemotherapy received 54 Gy. After completion of local therapy systemic therapy continued for patients.		98%. 9 Patients ≤30 days was 100%. Event free survival for both subgroups was identical. p=0.7085. Group 2: 107 patients ≥60 days after surgery. (51 patients ≥90 days) Local relapse as first event: 5/105 (5%) Combined Relapse as first event: 4/107 (5%) Local or combined relapse as second event: 1/107 (0.9%) Freedom of local and combined local and systemic relapses after 5 yrs was 92%. For subgroups ≤90 days & ≥90 days no difference in local control & event free survival, p0.7447 No substantial difference between groups concerning risk factors for local failure and survival. No statistically significant difference in event free survival between groups: 64% after 5 yrs. Authors conclude patients with early onset of postoperative irradiation			

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
				show a trend for improved local control compared to patients with later onset; difference is statistically not significant. This trend has no influence on survival.			
6. Schwartz DL, Einck J, Hunt K et al. (2002) The effect of delayed postoperative irradiation on local control of soft tissue sarcomas of the extremity and torso. *International Journal of Radiation Oncology, Biology, Physics* 52:1352-1359.	102 patients, median age 50 yrs (14-76) with soft tissue sarcomas of the extremity and torso	Radiotherapy timing. . The group was dichotomised according to time interval from definitive resection to the start of adjuvant radiation. 26 patients delay of ≤ 4 months and 32 patients long delay of ≥ 4 months.	Local relapse free survival	Univariate analysis indicated that 5 year local relapse free survival was significantly improved in the short delay group (88% vs 62% for the long delay group, p= 0.048 by log rank). Overall distant relapse free survival, disease free survival or overall survival were not statistically different. Authors conclude that postoperative RT delays of 4 months or greater were associated with inferior local disease control for intermediate and high grade soft tissue sarcomas of the extremities and torso	*Authors consider bias. Only 2 factors differed between the group – age and study period. Both factors had no effect on local control and event free survival*.	Retrospective cohort study	2⁻ +
7. Seel M, Foroudi F (2002) Waiting for radiation therapy: does it matter? *Australasian Radiology* 46:275-279.	All cancers	Radiation therapy	Multiple	Great majority of the research concentrates on radical RT. Breast – multiple studies High grade gliomas – 1 study Head & neck – many studies Oesophagus – 1 study. Lung – 4 studies Prostate – 0 articles Cervix -0 articles Evidence exists for head	*Where there were papers, the evidence suggested that delays in RT did have an adverse effect on outcomes.* *Details of searching process inadequate – time period, inclusion & exclusion criteria not specified.*	Literature review.	4 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
				and neck cancer, small-cell lung cancer and high grade gliomas that tumour control might be adversely affected by delaying RT.			
8. The Swedish Council on Technology Assessment in Health Care (2003) *Radiotherapy for cancer. A systematic literature review. SBU Report 162/2.* Stockholm: The Swedish Council on Technology Assessment in Health Care 556p.	Patients all ages with cancers:- Head & neck Oesophageal Rectal Non small cell lung cancer Soft tissue sarcomas Breast Cervical Uterine Ovarian Prostate Bladder Brain Hodgkin's Non-Hodgkin's lymphoma Skeletal metastases	-	-	Soft tissue sarcomas= Evidence indicates that preoperative RT is most appropriate treatment. On the negative side is that preoperative RT is associated with higher wound complication rate. Post operative RT is more widely used that preoperative. Brain = Addition of RT to surgery for high grade malignant gliomas extends life by 3 to 4 months. The value of RT as palliative treatment in low grade glioma is unclear.	*Well performed review with good documentation of methods used. Evaluation of evidence based practice and costs are limited to Sweden.*	Systematic review	2^{++} ++
9. Taylor RE, Bailey CC, Robinson K et al. (2003) Results of a randomized study of preradiation chemotherapy versus radiotherapy alone for nonmetastatic medulloblastoma: The International Society of Paediatric Oncology/United Kingdom Children's cancer Study group PNET-3 Study. *Journal of Clinical Oncology* 21:1581-1591.	217 patients median age 7.67 yrs(range 3 – 16 years) with nonmetastatic medulloblastoma (Chang stage M0-1). Median follow up 5.4 years	Patients randomised to preradiation chemotherapy or radiotherapy.	Overall survival, event free survival.	179/217 patients were eligible for analysis (CT + RT, 90 patients; RT alone, 89 patients). It was recommended that RT should commence within 28 days of surgery. 12 patients (13.5%) achieved this. The interval between surgery and RT was 24-42 days for 46 patients (51.7%) and > 42 days for 28 patients. There was no significant difference in OS (p=0.2113) or event free survival (p=0.1263) for those patients, starting RT	*Adequate details of randomisation process. Problems with recruitment to trial resulted in reduction in power of study. Good multicentre study with adequate details of methods etc.*	Randomised controlled trial	1+ +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
				within 42 days of surgery compared with those starting > 42 days after surgery.			
10. Thomas PRM, Tefft M, Compaan PJ et al. (1991) Results of two radiation therapy randomizations in the third National Wilm's tumor Study. *Cancer* 68:1703-1707. Note: Also co-author of Kalapuraka study which also reports results from NWTS 3 & 4.	Children with Stage II and Stage III Favourable histologic type (FH) tumours. 268 patients Stage II FH Wilms'' tumour; 277 patients Stage III FH Wilms'' tumour . Country: US	To resolve some issues raised in NWTS 1 & 2 incorporated RT randomisations for patients with Stage II & Stage III (residual abdominal disease) FH Wilms'' tumours. Stage II: Surgery and then 2 groups: Group 1) no radiotherapy or 2000 cGy radiotherapy and chemotherapy of dactinomycin (AMD); Vincristine (VCR); and Doxorubicin (ADR) for 15mths; OR Group 2) no radiotherapy and intensive AMD + VCR for 15mths; Patients on radiotherapy arm of trial required to receive RT to operative bed (excised tumour plus kidney). Stage III Surgery and then 2 groups: 1) either 1000 cGY or	Survival and Intra-abdominal relapses.	10/15 patients who experienced an abdominal relapse had delays of > 10 days after surgery before initiation of RT. A comparison of the 10 relapses in 103 patients, with the 5 relapses in 174 patients whose RT started within 10 days is significant (continuity-corrected Pearson chi-squared test, 4.664; two-sided p=0.03). No other overall effect of RT delay was demonstrated. Authors conclude delay of start of RT seems to have been implicated in the development of abdominal relapse. Factors such as slower recovery from surgery in patients with more extensive tumours may contribute to this. NWTS-4 (next stage of study) mandates RT start <10 days of surgery.	*Insufficient details of patient characteristics, randomisation methods, statistical analyses, attrition. Without details of randomisation study could be a nested case control study. Controversy exists about use of continuity corrected Pearson chi tests.*	Randomised controlled trial?	1⁻ +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics = reviewers comments*	DESIGN	EVIDENCE LEVEL /QUALITY
		2000 cGY and AMD+VCR+ADR for 15mths or 1000 cGY or 2000 CGY plus Intensive AMD+VCR for 15mths. .					

FEBRILE NEUTROPENIA

The Questions:

1. Does the place of treatment of febrile neutropenia (FNP) episodes for children and young people with cancer affect outcome?

2 Are there safe and reliable methods for selecting and treating children and young people with FNP in an outpatient setting?

Nature of the evidence

Q. 1

3 RCTs of fair quality
1 guideline of good quality
1 literature review of fair quality

Q. 2

1 systematic review of good quality
1 prospective cohort study of fair quality
1 prospective cohort of fair quality
2 guidelines, 1 of good quality, 1 of fair quality
2 historical case series of fair quality

Summary of the supporting evidence for the recommendations

Q. 1

- The conclusions from one RCT are that oral antibiotics and early hospital discharge for patients who remain stable >24 hours of in-patient monitoring offers an alternative to conventional management of low risk FNP [4].
- The evidence from one RCT indicated that children can be managed as outpatients providing they meet certain criteria [5].
- The evidence from one RCT indicates that the safety of outpatient treatment requires further research [1].
- The conclusions from one guideline are that some selected patients may be treated as outpatients [3].
- The evidence from 1 literature review suggests that there is a sub-population of children who can be managed as outpatients [2].

Q. 2

- One systematic review in which it is concluded that FNP cancer patients can be considered low risk if they are clinically well with evidence of marrow recovery and no disqualifying comorbidities [4].
- The authors of one prospective cohort conclude that clinical and laboratory parameters can be used to select children in an outpatient setting but that formal evaluation is required [6].
- One prospective case series indicates which criteria can be used to select children [5].
- One historical case series provides indications for selection criteria for low risk children [2].
- One guideline provides partially evidence based indications for selection criteria [1].
- One guideline describes the Multinational Scoring System for Identification of low Risk FNP cancer patients [3].

The guidelines that exist are from the United States and there is consensus that there is an urgent need for UK guidelines on the management of FNP. As yet there is insufficient high quality evidence to determine whether it is safe to treat FNP in an outpatient setting

Q1. DOES THE PLACE OF TREATMENT OF FEBRILE NEUTROPENIA (FNP) EPISODES FOR CHILDREN/YOUNG PEOPLE WITH CANCER AFFECT OUTCOME?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Freifeld A (1999) A double-blind comparison of empirical oral and intravenous antibiotic therapy for low-risk febrile patients with neutropenia during cancer chemotherapy. *New England Journal of Medicine* 341:305-311.	232 episodes in 163 patients, aged between 5 to 74yrs.) of neutropenia with an estimated projection to last no more that 10 days and no coexisting medical conditions	Comparison oral amoxicillin-clavulanate plus ciprofloxacin with iv ceftazidine. Patients hospitalised.	Treatment failure	From May 1992-1997, 211 patients had a total of 284 episodes of fever & neutropenia. Fifty two episodes not evaluated. The authors conclude that oral antibiotics are as effective as iv treatment but the safety of outpatient treatment requires further research.	*The use of a double blind design with a dummy iv or oral treatment eliminated bias toward modifying therapy earlier in the oral therapy group. The size of the sample provided an 80% probability that the upper 95% CI for the true difference in the success rates for oral treatment and iv treatment was > than 15%. The observed difference was 3% in favour of oral medications. A confounding factor is that 200/232 episodes of fever & neutropenia were treated with CSFs. According to ASCO guidelines low risk patients do not need treatment with growth factors. Suggests that patients were not truly low risk. No QoL measures.*	Double blind, randomised controlled trial	1^+ +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
					Difficult to determine attrition bias.		
2. Holdsworth M, Hanrahan J, Albanese B et al. (2003) Outpatient management of febrile neutropenia in children with cancer. *Paediatric Drugs* 5:443-455.	Children with cancer	Using available evidence, make recommendations for the care of children with FNP.	Out patient management of FNP	The authors conclude that the available literature suggests that there is a sub-population of children with FNP who may be managed in the OP arena. Such candidates for this approach represent the minority of children with FNP. Social and environmental factors are important as well as clinical ones. Criteria to select low risk children with FNP should include absence of comorbidity, evidence of haematopoietic recovery & Lack of positive cultures. No clear evidence for whether children with leukaemia more likely to develop complications during OP management compared with children with solid tumours. Choice of antibiotics for OP management should conform to the IDSA guidelines. The benefits of continuation of adequate antibacterial therapy until neutrophil recovery appear to outweigh the risks.	*Search of PubMed 1993-2003. Insufficient details of exclusion criteria etc. but study gives very relevant overview of world literature with referenced recommendations. High relevance to questions. Recommendations follow literature review*	Literature review	4 +
3. Hughes WT, Armstrong D, Bodey GP et al. (2002) Guidelines for the use of antimicrobial agents in neutropenic patients	-	Update of 1997 guidelines.	-	Level of Risk for Oral Antibiotics and Outpatient Management = The guideline panel consider there is good evidence for the treatment of carefully selected patients with oral	*The guidelines are partially evidence based, but rely heavily on expert consensus opinion. Thee evidence from which the*	Guidelines	3/4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
with cancer. *Clinical Infectious Diseases* 43:730-751.				antibiotics alone. Some patients may receive their therapy as outpatients, although the majority of studies that have supported treatment with oral antibiotics have been performed in hospitals. Vigilant observation and prompt access to 24h per day/ 7 days a week medical care must be in place. The factors favouring low risk for severe infection in patients with neutropenia are :- • ANC ≥100 cells/mm^2 • AMC ≥100 cells/mm^2 • Normal findings on a chest X ray • Nearly normal results of hepatic & renal function tests • Duration of neutropenia < 7 dyas • Resolution of neutropenia expected in < 10 days • No intravenous catheter-site infection • Early evidence of bone marrow recovery • Malignancy in remission • Peak temperature of < 39.0^{o}C • No neurological or mental changes • No appearance of illness • No abdominal pain	*recommendations are made is graded. The up to date nature of the guidelines and their evidence base make them useful to the question of criteria for patient selection for OP/shared care treatment..*		

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				• No comorbidty complications As an alternative to initial OP treatment early discharge with continued OP therapy for selected patients may be considered after a brief IP admission during which iv therapy is initiated, a fulminant infection is excluded and the status of initial culture specimens is ascertained.			
4. Innes HE, Smith DB, O'Reilly SM et al. (2003) Oral antibiotics with early hospital discharge compared with in-patient intravenous antibiotics for low-risk febrile neutropenia in patients with cancer: a prospective randomised controlled single centre study. *British Journal of Cancer* 89:43-49.	102 Patients undergoing conventional dose cytotoxic chemotherapy representing 126 episodes of fever associated with neutropenia. Clinical symptoms at randomisation were mild to moderate. Age range: 18 to 78yrs Female 61.9% Majority episodes occurred in women reflecting underlying diagnoses of breast cancer & small-cell lung cancer. Patients could be entered in study more than once following subsequent episodes	Assessment of efficacy and safety of oral antibiotics in conjunction with early hospital discharge in comparison with standard in-patient intravenous antibiotics in patients with low-risk neutropenic fever. Compared oral and early hospital discharge with inpatient IV antibiotics. Oral arm: oral regimen of ciprofloxacin 750mg/12hrs plus amoxicillin 500mg+ clavulanate 175mg every 8hrs for total of 5 days Patients eligible for discharge following 24h hospitalisation if clinically stable and symptomatically improved and according	Primary outcomes: success and safety. "Success" defined as lysis of fever and resolution of symptoms and signs with no modification to initial antibiotic regimen and with no recurrence within 7days. Safety assessed by frequency of serious medical complications and deaths. Secondary outcomes: total duration of hospital admission, frequency of readmission, toxicity of treatment and resource utilisation.	**Oral arm:** 66 episodes (51 first episodes) **Intravenous Arm:** 60 episodes (51 first episodes). Total of 36.5% had no symptoms other than fever. **Efficacy & Safety:** Success rate to initial antibiotic therapy similar in both groups. **Intravenous:** Successful 90% of episodes (95%CI: 82.4 to 97.6%) **Oral:** 84.8%; P=0.55: absolute differences between groups 5.2%; 95%CI for difference minus 7 to 17.3% Success rates of 102 first episodes: Intravenous: 45 /51 (82%); Oral: 43/51 (84.3%); P=0.77. Failure: **Intravenous arm:**	*Small sample; Study states retrospectively scored patients' baseline characteristics at randomisation. Some retrospective analysis done due to publication of "risk scales" during study period.*	Prospective randomised controlled single centre study (retrospective scoring of baseline characteristics) Randomisation : consecutively drawn sealed envelopes.	1+ +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
	of febrile neutropenia Patients required to be haemodynamically stable with no signs or symptoms requiring intravenous fluid support. Adequate renal function and ability to maintain satisfactory oral intake required. Responsible adult to act as carer; Exclusion criteria: Patients with poor compliance history; Undergone autologous bone marrow; peripheral blood stem-cell transplantation; received antibacterial medication ≤7days of enrolment. Use of CSFs and cytokines not permitted; any co-existing medical condition requiring in-patient treatment or monitoring; clinically documented infection likely to require targeted or prolonged duration of antibiotic therapy; inability to tolerate oral medication; known allergy to study drugs. UK	to patient's wishes. Patients supplied with diary to record temp at 6h intervals and associated symptoms. Telephone contact maintained with clinical research team. Oral & written instructions and 24h contact no. of specialist centre, emphasising need for early reporting of symptomatic deterioration. After discharge patients reviewed 7-10 days later in Oncology Outpatient Dept. If not discharged after 24h reassessed daily. Intravenous arm: Intravenous regimen of gentamicin 80mg every 8hrs and dose adjusted according to therapeutic levels plus tazocin (piperacillin) 4g+tazobactam 500mg) every 8hrs until hospital discharge. Patients eligible for discharge when afebrile for 24h with a rising neutrophil count (irrespective of absolute value). Patients did not routinely receive antibiotics on discharge. Indication for changes in		death (1) not attributed to treatment; Persistence of fever with microbiological evidence of resistance (2); without microbiological evidence of resistance (3); **Oral arm:** Serious complication or clinical deterioration while in-patient (1); Intolerance of antibiotics due to vomiting (1); due to severe oesophagitis (2); Persistence of fever without microbiological evidence of resistance (6). All 9 failures converted to intravenous antibiotic regimens. 5 patient readmitted to hospital, 4 described above; 1 with pulmonary embolism. Toxicity: Both arms well tolerated. **Oral:** 1 episode (0.8%) severe toxicity, CTC grade 3. Other: mild-moderate gastrointestinal toxicity not requiring change to regimen: 14 (21%) patients CTC grade 1-2 diarrhoea; 5 (7.6%) grade 1-2 nausea/vomiting; **Intravenous**: no episodes of toxicity CTC grade >1; Median in-patient stay: Intravenous: 4 (range 2-8); Oral: 2 (range 1-16days); P<0.0005. Overall oral antibiotic policy resulted in reduction of 66 in-patient days (199 compared to			

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
		treatment regimen for both groups included persistent fever ≥72h, positive culture results with resistant organisms, or clinical deterioration. Study states no significant differences between groups at baseline.		265); **Resource utilisation:** Overall costs over £19,000 less in oral arm compared with intravenous arm. Authors conclude that oral antibiotics in conjunction with early hospital discharge for patients who remain stable after a 24hr period of in-patient monitoring offers a feasible and cost-effective alternative to conventional management of low-risk neutropenic fever. Authors urge caution when applying findings outside setting of a single specialist centre. Also suggest larger trials needed to further evaluate policy.			
5. Mullen CA, Petropoulos D, Roberts WM et al. (1999) Outpatient treatment of fever and neutropenia for low risk pediatric cancer patients. *Cancer* 86:126-34. Aim: to assess the safety of treating low risk paediatric patients with FNP as outpatients	73 episodes of fever and neutropenia in 41 children receiving chemotherapy for cancer. Aged 3 to 20 years. Children had to have reliable caretakers and be < 1 hour from hospital. Absolute neutrophil count < 500 cells/microL or < 1000 cells/microL and declining. Oral temp >	Children and parents attended for baseline clinical evaluation, blood samples. All give single dose ceftazidime (50 mg/kg) then randomised to intravenous ceftazidime (50 mg/kg/dose every 8 hrs via portable pump) or oral ciprofloxacin (12.5 mg/kg/dose every 12 hrs). Children returned to	Number of episodes treated entirely as outpatients. Duration of raised temperature and treatment. Proportion of episodes requiring change of initial antibiotic. Description of problems encountered for episodes requiring hospitalisation.	Overall, 63/73 (86%) of episodes were managed as outpatients. There was no statistically significant difference between oral and intravenous antibiotics in the proportion of episodes treated entirely as out patients (31/33 with iv ceftazidime v 32/40 with oral ciprofloxacin, P = 0.10) Mean duration of: raised temp was 2.7 days;	*Episodes rather than children were randomised and analysed. Small number of children. Children had to fulfil certain criteria such as < 1 hr from hospital reliable carer etc and attend clinical every day.*	RCT	1⁻ +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
This RCT compares oral and intravenous antibiotics in outpatients	38.5 C once or > 38C on three occasions over 6 hrs Country: USA	clinic for daily evaluation till afebrile for 48 hrs.	Deaths, ICU transfers, serious complications.	antibiotic treatment was 4.7 days. 77% episodes required no change of initial antibiotic. 10 children were hospitalised (4 had prolonged fever; 3 had emesis; 1 deteriorating condition; 1 parents non-compliance; 1 protocol violation). There were no deaths, ICU transfers or serious complications 60% of children presenting with febrile neutropenic episodes were not eligible for outpatient treatment. initially treated as outpatients. The authors concluded that carefully selected children with fever and neutropenia can be safely treated as outpatients provided they are evaluated every day.			

Q 2. ARE THERE SAFE AND RELIABLE METHODS FOR SELECTING AND TREATING CHILDREN AND YOUNG PEOPLE WITH FNP IN AN OUTPATIENT SETTING?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Hughes WT, Armstrong D, Bodey GP et al. (2002) Guidelines for the use of antimicrobial agents in neutropenic patients with cancer. *Clinical Infectious Diseases* 43:730-751.				**SEE EVIDENCE TABLE QUESTION 1**			
2. Lucas KG, Brown AE, Armstrong D et al. (1996) The identification of febrile, neutropenic children with neoplastic disease at low risk for bacteremia and complications of sepsis. *Cancer* 77:791-798.	161 children (mean age 9.2 years; range 1-18 years) with cancer, hospitalised for 509 episodes of FNP between January 1990 and June 1992. 63 patients, had leukaemia or lymphoma and 98 had solid tumours.	Identification of criteria for children at low risk for FNP	Hospital admission. Occurrence of episodes of FNP.	509 episodes of fever in 161 patients. 27% of the episodes had microbiologically documented infections with 16% being associated with positive blood cultures. 12% of patients without clear signs of sepsis at presentation had a positive blood culture, compared with 44% of patients who had 1 or more clinical signs of sepsis at presentation. There was a low incidence of complications during hospitalisation amongst low risk patients who also had resolution of fever and an ANC>100/mm^2 within 48 hours of admission. The authors concluded that this subset of patients may be candidates for early hospital discharge.	*Good quality study. Appropriate use of statistics.*	Historical case series	3 ++
3. National	All patients with cancer	Consensus guidelines		Flow charts for all steps in the	*Not specific for C &*	Guidelines	3/4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Comprehensive Cancer Network. Clinical Practice Guidelines in Oncology. Fever and Neutropenia. Version 1.2002		for the management of fever and neutropenia		treatment of FNP. Description of the Multinational Scoring System for Identifying Low-Risk Febrile Neutropenic Cancer Patients	*A ages range The NCCN guidelines do not score highly on assessment with the AGREE tool because of their lack of evidence*		+
4. Orudjev E, Lange B (2002) Evolving concepts of management of febrile neutropenia in children with cancer. *Medical & Pediatric Oncology* 2002:39:77-85. Systematic review Aim: to determine risk factors in identifying low-risk paediatric patients for outpatient treatment of fever and neutropenia, to assess alternative antibiotic regimens and create an algorithm for managing patients. The review included the studies by Sahu, Rackoff, Petrilli Shemesh and Santolaya.	27 prospective trials and 5 reviews of febrile neutropenia in paediatric cancer patients. Aged 0 to 18 years. Studies of adult patients were included in the assessment of different antibiotic strategies.	Various oral and intravenous antibiotic regimens. Treatments took place in hospital, home, outpatient departments. Some regimens involved early discharge.	The association between the following factors and treatment failure: comorbidities at presentation; absolute neutrophil count (ANC), absolute monocyte count (AMC) and fever; and therapeutic strategies (site of care, route of administration of antibiotics and the duration of antibiotic treatment).	1/3 to ½ of children with febrile neutropenia are at low risk of life threatening complications. Low risk patients can be identified by an experienced nurse or physician, physical examination and complete blood count. The review presents a list of specific comorbidities. Children who are well and have evidence of marrow recovery (rising ANC or AMC; children in relapse from leukaemia require an APC≥100 x 10^9) are low risk and are suitable for outpatient treatment or early discharge. Studies of antibiotic regimens generally used 1 or more doses of intravenous broad spectrum antibiotics followed by observation plus daily assessment. Continuation of treatment with oral and intravenous had similar treatment failure rates. Standard inpatient treatment is required for outpatients with bacterial infection, fever for > 4 days, clinical deterioration, and intolerance	*Search date: not stated. Primary sources: Medline, references. Validity of studies was not assessed. No details of methods used to conduct the review.*	Systematic review	2+ +=

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				of therapy or non compliance. Up to 25% may experience treatment failure. ***The authors concluded that febrile neutropenic paediatric cancer patients can be considered low risk if they are clinically well with evidence of marrow recovery and no disqualifying comorbidities.*** ***The authors report that one study stated that results reported in trials may not generalise to settings outside clinical trials.*** A treatment algorithm is presented.			
5. Rackoff WR, Gonin R, Robinson C et al. (1996) Predicting the risk of bacteremia in children with fever and neutropenia. *Journal of Clinical Oncology* 14:919-924. Study aim: To identify factors in children with cancer who present with fever and	115 episodes of fever and neutropenia in 72 children with cancer treated in hospital. Age 9 months to 18 years. Children had solid tumours or haematological malignancies Fever was defined as temperature of 38 C on 3 occasions over 24 hours or single temperature recording ≥38.5C. Neutropenia was	Treated in hospital. Study setting: Children's Medical Centre, Indiana, USA	The following predictors of bacteraemia were examined: diagnosis; disease status; type of central venous access device; admission clinical signs and symptoms; prophylactic antibiotics; use of G-CSF; admission blood count and picture; need for	Only the absolute monocyte count (AMoC) and temperature on admission were significant predictors of bacteraemia. Bacteraemia was significantly increased for high risk episodes compared with low risk episodes. OR 4.4 (95% CI: 1.6, 12.9). Risk classification was validated using data from 57 different episodes of fever and bacteraemia. The authors concluded that three levels of risk were		Prospective case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
neutropenia at admission that predict bacteraemia	defined as absolute neutrophil count (ANC) < 500/µL. Bacteraemia was defined as positive blood culture using BACTEC. Country: USA		iv resuscitation; and chest X-ray. Episodes of fever and neutropenia were classified as low risk (AMoC ≥100 mu/L). intermediate (AMoC < 100 m/l and temp < 39C) and high risk (AMoC <100mu/L and temp ≥39C) for bacteraemia	defined using the AMoC and temperature at admission. They suggest future studies could examine the safety of abbreviated antibiotic therapy in children at low or intermediate risk of bacteraemia. Authors note generalisability of study findings may be limited since: various regimes used in children with cancer; different underlying disease; year to year and site specific variation in rate and type of infection.			
6. Santolaya ME, Alvarez AM, Becker A et al. (2001) Prospective, multicenter evaluation of risk factors associated with invasive bacterial infection in children (IBI) with cancer, neutropenia, and fever. *Journal of Clinical Oncology* 19:3415-3421. Aim: to determine factors that predict the presence of invasive bacterial infection (IBI) in paediatric cancer patients with acute fever and	447episodes of febrile neutropenia in 257 children with cancer. Aged 6 months to 18 years. Most had acute lymphocytic leukaemia . All were receiving chemotherapy. Fever defined as ≥38.5C once or ≥ 38C twice. Neutropenia defined as absolute monocyte count (AMC) ≤500/mm^3 Country: Chile	Setting; 5 hospitals in Santiago, Chile. All children were hospitalised and received intravenous broad spectrum antibiotics. Monitored daily in hospital till fever settles and AMC > 500 mm^3	Investigation of the following potential predictors of invasive bacterial infection (IBI): demographic variables; cancer-related variables; febrile episode variables; admission clinical and laboratory variables. Demonstrable IBI defined as confirmed bacteraemia and positive bacterial	Five variables were independent risk factors for IBI: CRP ≥ 90 mg/L (RR 4.2); hypotension (RR 2.7); relapse of leukaemia (RR 1.8); platelet count ≤ 50,000/mm^3 (RR 1.7); ≤ 7 days since last chemotherapy (RR 1.3). 95% CIs were reported for the above RR. Results were similar for demonstrable IBI and probable IBI when analysed separately and in analysis using only the first episode per child. The authors concluded that clinical and laboratory admission parameters can help predict the risk of invasive bacterial infection.		Prospective cohort	2- +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
neutropenia			culture from usually sterile site. Probable IBI defined as no positive culture but clinical and lab signs suggestive of sepsis plus focal organ involve	The authors state that validation of this predictive model is required before it can be adopted for use. The authors state that they are currently conducting such a validation.			

CENTRAL VENOUS ACCESS

The Question:

What is the evidence for the optimum method of central venous catheter (CVC) insertion in children and young people with cancer?

Nature of the evidence

1 randomised controlled trial of fair quality
1 non randomised controlled study of poor quality
2 systematic reviews one of fair to poor quality and one poor quality
1 prospective cohort study of fair quality,
1 retrospective comparative study of poor quality
2 guidelines one of good quality, one fair quality
1 audit of fair to poor quality
1 non systematic literature review of poor quality

Summary of the supporting evidence for the recommendations

- There is evidence from both randomised trials and non randomised trials to support the view that trained clinical nurse specialists can provide high quality CVC care [2 4].
- The systematic and non systematic review evidence indicated that there was a lack of high quality evidence to indicate the optimum method for CVC insertion but that there are some simple measures that are effective in reducing complications [5 6 8].
- The cohort study aimed to determine risk factors for infection but the results were not separated for children and young people. The study provided indirect evidence that the insertion of CVCs should be performed in an operating theatre or clean special procedure room [8].
- Ultrasound locating devices can improve the insertion success rate and reduce complications – NICE technology appraisal [7].
- The audit study demonstrated that there was a wide variation in insertion techniques used in the UKCCSG centres. The authors concluded that such variations make any interpretation of data difficult [9].

No randomised evidence specific for child or adolescent cancer patients was identified. No clear evidence was found to indicate the best model of care for CVC insertion in children and young people with cancer.

WHAT IS THE EVIDENCE FOR THE OPTIMUM METHOD FOR CENTRAL VENOUS CATHETER INSERTION?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Alonso-Echanove J, Edwards J, Richards M et al. (2000) Risk factors for central line associated bloodstream infections: preliminary analysis of the detailed intensive care unit surveillance component study. *Infection Control and Hospital Epidemiology* 21:93.	*8 adult general medical and/or surgical ICUs.* Patients admitted to ICU >24 hrs	To identify risk factors for central line associated bloodstream infections Intervention: insertion of central line catheters (CL) Data collection: 24mths. In first 20mths 7,913 CL contributed 91,474 CL days. Catheters: Non-tunnelled CL = 59% :29% antibiotic impregnated; Swan-Ganz (25%): 24% antibiotic impregnated. Insertion sites: Jugular = 43% of CL Subclavian = 38% of CL	Central line-associated bloodstream infection (LA-BSI) rates. Data collection daily for 24 mths on potential intrinsic and extrinsic risk factors for blood stream infection	Multi-variate analysis controlling for CL days identified following risk factors for LA-BSI: Suggests intrinsic factors affecting LA-BSI rates represented by Transplant (TR), Mechanical Ventilation (MV)or Pulmonary Oedema (PE) . Independent Extrinsic risk factors : inserting a CL outside an operating room (SPR), using Total Parenteral nutrition (TPN) and keeping CL312 days. Line insertion outside a special procedure room or operating room (SPR) odds ratio (OR) =2.5; (95% CI: 1.6 to 4.0) Total parenteral nutrition (TPN) OR=2.3; (95%CI: 1.7 to 3.0) Duration of CL 312 days OR=1.8 (95%CI: 1.3 to2.6); Mechanical ventilation (MV) OR=2.5; (95%CI: 1.7 to 3.9) Pulmonary Oedema (PE) OR=2.0; 95%CI: 1.3 to 3.1)	*No results separately for children and adolescents with cancer* *Numbers of patients not given, nor any other patient characteristics.*	Prospective multicentre cohort surveillance study.	2^{+} +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				Transplant patient (TR) OR=2.6; (95%CI: 1.2 to 4.7)			
2. Boland A, Haycox A, Bagust A et al. (2003) A randomised controlled trial to evaluate the clinical and cost-effectiveness of Hickman line insertions in adult cancer patients by nurses. *Health Technology Assessment* 7:1-99.	470 adult (>18 years) cancer patients due to have a Hickman line insertion who were clinically and physically compliant with specified protocols. 80% patients of normal physical state; 75% had Karnofsky performance score from 80% to 100%; 54% had gastrointestinal cancer; 66% treated as in patients.	To compare the clinical and cost-effectiveness of image guided Hickman line insertion with blind Hickman line insertion undertaken by nurses in adult cancer patients. Setting: large acute cancer hospital in Manchester. Blind versus guided insertion of Hickman line by nurses (1 trainer and 2 trainees). Both treatments involved blind venipuncture of the subclavian vein. Lines inserted by 3 trained nurses 74% were single lumen Hickman lines Blind insertion of a Hickman line at the patients' bedside: Hickman line routinely inserted without the use of image guidance at any point in the procedure. Patients could be transferred immediately to the X-ray suite for image	Primary clinical outcome measure: catheter-tip misplacement. For comparison of skill level of the trainer and the trainees, pneumothorax was the primary clinical outcome. Secondary outcomes were: arterial puncture, haematoma; infection; failed insertion; and assistance from other healthcare professionals.	Catheter-tip misplacement was significantly higher with blind insertion (14%[32/235] v 1%[1/235], P < 0.001), This was the only significant difference. No significant difference between blind and image guided in: arterial puncture (6% v 5%), pneumothorax (3% v 1%), haematoma (1% v 2%), line infection (4% v 6%), tunnel infection (5% v 2%). Significantly higher proportion of lines were inserted without complications or help from other staff using image guided method (81% with image guided v 67% with blind, P < 0.001). Only 3 patients were transferred from the bedside to X-ray unit during insertion. Time Blind: mean 38 mins (95% CI: 36, 39 mins); Range 15 to 90 mins Image-guided: mean 40 mins (95% CI: 38, 42 mins); Range 20 to 150 mins. There was no significant difference between the two	*Few patients, of relevance to C & A group. RCT; treatment groups comparable at baseline; detailed report* *Lots of background information including types of CVCs; methods of insertion of Hickman lines; choice of access site; choice of operator; complications; analysis of empirical evidence (1980 to 2000); comparison of Hickman with other types of CVC; different settings, operators and techniques; review of health economics literature (reviews 8 papers); summary of*	RCT	1^+ +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
		guidance if required V Image-guided insertion in X-ray suite: the position of the guide wire was checked before the Hickman line was introduced and later the Hickman line was positioned with the use of X-ray fluoroscopy.		trainees combined and the trainer for pneumothorax, catheter tip misplacement; arterial puncture, haematoma; line infection, tunnel infection, successful insertion; nurse assistance; oncologist assistance; radiologist assistance). Trainees were significantly more likely to require the assistance of another nurse than the trainer (13% v 4%, P = 0.002). Authors concluded that: Nurses previously inexperienced in the procedure can be trained to insert Hickman lines successfully both at the bedside and under image guidance within a 3-month period	*published literature* Authors concluded that the insertion of Hickman lines is safe and effective for most adults with cancer		
3. British Committee for Standards in Haematology (1997) Guidelines on the insertion and management of central venous lines. *British Journal of Haematology* 98:1041-1047.	All patients with skin tunnelled catheters	Development of recommendations for insertion and management		Major recommendations (relevant to question):- • Single lumen catheters cause fewer problems. • Fully implantable catheters more suitable for children • Lines should be inserted in children by paediatric specialists • Imaging facilities must be available. • Line insertion should take place in an operating theatre or similar clean	*Publication date 1997 – still current, not updated*	Guidelines	3/4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				environment • Thrombosis and infection must be diagnosed promptly. Both complications may require line removal • Catheters should be removed only by experiences personnel. Catheter breakage requires radiological intervention. • Patients should receive clear and comprehensive information and be encouraged to look after their own lines. • Units should audit complication rates and use the data to develop preventative measures.			
4. Cardella JF, Cardella K, Bacci N*, et al.* (1996) Cumulative experience with 1,273 peripherally inserted central catheters at a single institution. *J Vasc Interv Radiol* ;**7**:5-13 **Phase 2 of study (Phase 1 previously reported elsewhere)** Aim: To compare bedside insertion of PICCs by Nurses with	Total of 869 Peripherally inserted central catheters (PICCs) inserted in 655 patients. Mean age 49.7 yrs (range 1 to 93yrs). No statistically significant difference in patient ages between groups.	**(Group A)** Nurses (N) performed 327 (37.6%) bedside insertions with palpatory, through-the-needle technique in 301 patients. (3 PICCs inserted by nurses under guidance of interventional radiologists in fluoroscopy suite for training purposes.) *(Group B) Radiologists*	Technical success; Service Interval Complications	Follow-up information available for 808 of 869 (93%) PICCs inserted; 50 of 61 PICCS lost to follow up did have identifiable removal date. Outcome of insertion attempts: Technical Success: Nurses: 327/396 (82.6%) Radiologists: 542/555 (98.2%) Failure:	*No results separately for children and adolescents with cancer.* *PICC : Each group used own preferred Vendor of PICC.* *Groups different at baseline.* *Radiologists were assigned*	Non randomised controlled study with non-comparative control group	2

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
insertion by Radiologists	**Sex:** 51% male (n=340); 48.1% female (n=315) **Group A (Nurses)** 51% male 41.9% female **Group B (Radiologists)** 48.2% male; 51.8% female. Statistically significant difference in sex (P=0.004) Indication for PICC insertion: Antibiotic therapy: Group A: n=198 (45.4%) Group B n=324 (52.3%) Difference NS Hyperalimentation: Nurses n=65 (14.9%) Radiologists n=190 (30.7%) P<0.001 Hydration: Nurses n=107 (24.5%) Radiologists n=39 (6.3%) P<0.001 Chemotherapy: Nurses n=13 (3.0) Radiologists n= 14 (2.3%) Difference NS	*performed 542 (62%) insertions with a venographic-fluroscopic direct puncture & sheath technique in 354 patients.* Radiologists required for difficult initial insertions, PICC salvage and PICC exchange. From 14 May 1992 to 31 December 1994		Inability to cannulate Nurses: 31/69 (44.9%) Radiologists: 10/10 (100%) Inability to thread: Nurses: 26/69 (37.7%) Radiologists: 0/10 (0%) Errant threading (to wrong site) Nurses: 12/69 (17.4%) Radiologists: 0/10 (0%) In all cases of failed attempts by nurses PICCS successfully inserted by radiologists. All failed attempts at PICC insertion in radiology dept occurred in patients (n=63) referred directly by nurses. Overall mean service interval for PICC insertions was: Nurses: 21.0 days (range 0 to 288 days) Radiologists: 32.2 days (range, 0-432 days) P=0.002 Size & Type of catheter: No statistically significant difference between Nurses group and Radiologists Group with regard to PICC type or size. Due to different insertion strategies there were significant differences in preferred insertion site	*more difficult patients. Not stated how patients were selected for reporting- not stated that consecutive patients were treated.*		-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
	Plain Medication: Nurses n=14 (3.2%) Radiologists n=6 (1.0%) P=0.003 Immunosuppressive Therapy: Nurses n=0 Radiologists n=16 (2.6%) P<0.001 Other: Nurses n=39 (8.9%) Radiologists n=30 (4.6%) P=0.001 20% cases had more than one indication			(P<0.001) and final tip position (P<0.001) between Groups A & B. Patient Status at end of study period: Difference in patient status between groups not statistically significant. 695/869 PICCS were alive; 85/869 PICCS (9.8%) died 28/869 PICCs (3.2%) still in use 61/869 PICCs (7%) lost to follow up. Reasons for removal of PICC: There were no statistically significant differences between Nurses and Radiologists groups with regard to reasons for PICC removal. Complications: <u>Death:</u> Nurses: 0 Radiologists: 0 <u>Thrombophlebitis:</u> Nurses: 13/301 patients (4.3%) Radiologists: 12/354 (3.4%) P=0.133 Infection: Nurses: 2/301 (0.7%) Radiologists: 11/354 (3.1%) P=0.147			
5. Harrison M	? People having	Appears to be	Not explicitly	**Dressing:** One study found	*Inclusion criteria*	Non systematic	4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
(1997) Central venous catheters: a review of the literature. *Nursing Standard* 11:43-5.	long term intravenous therapy	methods aimed at preventing infection	stated. Appears to be infection	that cleansing the end of a Hickman catheter with 70% alcohol, placing in sterile finger cot and sealing with tape covering at least 2 inches of the cap resulted in no further episodes of *S epidermidis* catheter infection. **Hygiene**: One study found that intensive training in strict hygiene and hand washing reduced infection rates in a children's' hospital from 40% to 8%. ***Antibiotics:*** *One study found that exit site infections respond to intravenous antibiotics and that removal of the catheter is not required. One study recommended catheter removal for tunnel infection with Pseudomonas.* **The exit site:** Three studies found that skin cleansing with chlorhexideine in spirit reduces infection compared with alcohol or povidone iodine. One study found that dry dressings reduce infection compared with Tegaderm or Opsite. Two studies found that dressings like IV3000 are better at prevent accumulation of moisture under the dressing than Tegaderm. **Securing the catheter:** No studies reported. Just comments.	*not stated in terms of population* *Inclusion criteria not stated in terms of interventions* *No results separately for children and adolescents with cancer.* *No defined inclusion criteria for review, no stated search strategy, no details of methods used to select studies, assess validity, extract data.* *Inadequate reporting of individual studies included in this report. Study design and number of patients/ catheters not mentioned.* *High risk of bias. Unable to assess the quality of the evidence.*	literature review	–

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				Pyrexia: No studies reported. Just comments. **Prevention of thrombosis:** One study reported that most hospitals heparinised catheters daily when in use. **Treatment of thrombosis:** Two studies recommended that catheters be flushed with Hepsal once weekly when in use. One study recommended more frequent flushing in children with small lumen catheters. One study found that suction of blood into the catheter tip may be prevented by clamping catheter while the last ml of heparin. One study found that Exoparin is effective in treating and preventing venous thrombosis in bone marrow transplant patients. One study suggested that low dose warfarin may prevent venous thrombosis in high risk patients. Treatment of thrombosis: One study found that clot could be removed from Hickman catheters if care were taken. Two studies found that the length of time urokinase has to remain in place is controversial. One study found that using an algorithm for the management of occluded catheters helps and can be used as a teaching tool.			

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				As a results of the review the following aspects of catheter care were changed: **Cleansing solution:** changed from saline to chlorhexide in spirit. **Hygiene:** bacterial cultures taken from nursing and medical staff to highlight need for hygiene. **Bungs:** change to bungs rather than membrane caps and use Bionectors **Reporting Audit:** reporting infection rates to surgeons. And monitoring infection rates. **Surveillance:** collection of information on catheters with annual audit. Audit sooner if increase in infection. Need to consider most effective tool for collecting information **Communication**: Monthly multidisciplinary clinical haematology audit meetings.			
6. Mermel LA (2000) Prevention of intravascular catheter-related infections. *Annals of Internal Medicine* 132:391-402.	Patients having catheters inserted into new sites.	To assess methods used to prevent intra-vascular catheter infection	Guidelines Catheter cultures using semi quantitative/ quantitative methods. Catheter related blood stream infections confirmed by microbial growth from percutaneously drawn blood	*Intravenous antimicrobial Prophylaxis* Prophylaxis with vancomycin or teicoplanin at insertion of a central venous catheter is not recommended on the basis of the available data [IIa]. Prevention of intravascular catheter-related infections should not involve vancomycin or other therapeutic agents [IV]. *Warfarin and Heparin Prophylaxis*	*Search date: 1999* *Primary sources: Medline; conference proceedings; reference lists; contact with primary authors. Included RCTs where these were available, if no RCTs, case control and cohort studies*	Systematic review.	1⁻ ⁻

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
			cultures that matched microbial growth from catheter. Grade 1: Evidence from a well-designed meta-analysis of randomised, controlled trials; Grade IIa, evidence from at least one randomised, controlled trial. Grade IIb: evidence from at least 1 RCT that allowed catheter exchange over guide wires into old sites. Grade III: evidence from at least one well-designed clinical trial without randomisation Grade IV: evidence from opinions of authorities in the field based on clinical experience, descriptive studies or expert committee reports.	Prophylaxis with very-low-dose warfarin should be strongly considered for patients with long-term, indwelling intravascular catheters [IIa]. Prophylactic heparin should be administered to patients with short-term central venous catheters [I]. <u>Site of Insertion</u> No randomised trials have assessed the risk for infection associated with catheter insertion into the subclavian, internal jugular, or femoral vein. Insertion into a subclavian vein is preferred to reduce the risk for infection [III]. (4 observational studies) Femoral venous catheterisation should be limited to circumstances that prevent the use of alternative sites.[III]. *Subcutaneously Tunnelled Catheters* Subcutaneous tunnelling of short-term internal jugular or femoral vein catheters is recommended if the catheters are not accessed for drawing blood. [IIa]. .**Cutaneous Antisepsis** Chlorhexidine containing antiseptics should be used, where approved, or skin preparation before catheter	*were used.* *No details of methods used to select studies, extract data, assess validity. Insufficient details of individual RCT to assess the evidence* *Catheters inserted into new sites were included. Catheters inserted into old sites over a guide wire were excluded.*		-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				insertion [IIa]. Tincture of iodine is superior to povidone-iodine as sites a cutaneous antiseptic and should be considered for preparation of intravascular sites [IV]. <u>Sterile Barrier Precautions</u> Full barrier precautions should be the standard of care during central venous catheter insertion [IIa] and should be considered during insertion of midline and artery catheters [IV]. <u>Catheter Dressing</u> On the basis of all available evidence, the choice of central venous catheter dressing may be a matter of thin preference and cost [IIb]; however, gauze dressings are preferred if blood is oozing from the catheter insertion site [lib]. <u>Ointments</u> Applying triple antibiotic ointment (polymyxin, bactracin, neomycin) to the catheter insertion is not recommended [IIa]. Mupirocin ointment should not be applied to catheter insertion sites [IV]. Applying povidone-iodine ointment to insertion sites of nontunnelled, long-term central venous catheters in			

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				immunocompromised patients with heavy S. aureus carriage (such as patients with AIDS, or cirrhosis) should be considered [IV]. **Contamination-Shielded Pulmonary Artery Catheters** A contamination shield should be used for all pulmonary artery catheters [IIa]. There were also additional recommendations for catheter maintenance. Authors concluded that simple interventions can reduce the risk for serious catheter-related infections. Adequately powered RCTs are required			
7. National Institute of Clinical Excellence (2002) Guidance on the use of ultrasound locating devices for placing central venous catheters. *NICE Technology Appraisal Guidance* No. 49. London: National Institute of Clinical Excellence. Available from: www.nice.org.uk	Patients requiring CVCs.	Two types of Real-time ultrasound guidance : two-dimensional (2-D) imaging ultrasound guidance and audio-guided Doppler ultrasound guidance. Evaluated against a venepuncture method known as "landmark method".	Major recommendations on use of ultrasound locating devices for placing CVCs	Two Dimensional (2-D) imaging ultrasound guidance recommended: 1) as preferred method for insertion of CVCs into internal jugular vein (IJV) in adults and children in elective situations. 2) should be considered in most clinical circumstances where CVC insertion necessary either electively or in emergency situations	*No specific patient details or numbers given.* *Very little data presented for children, particularly limited for infants weight <3kg. Evidence: 22 RCTs. 6 evaluated audio-guided Doppler ultrasound*	Based on systematic review of 22 RCTs. (full search strategy given, selection criteria, evaluation of studies & recommendations supported by evidence. See Comment by Carey CR,	1^{++} ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				3) that all those involved in placing CVCs using 2D imaging should undertake appropriate training to achieve competence. Audio-guided Doppler ultrasound guidance not recommended for CVC insertion.	*against landmark method; 13 evaluated 2-D ultrasound guidance against landmark method. 1 Evaluated both Doppler & 2D against landmark method. No trials compared ultrasound against surgical cut-down method. None addressed PICCs or ports.* *Procedure carried out by anaesthetists in 7 studies & other medical staff in 4 studies. None of the studies involved nurses.* *Only 3 trials for 2D ultrasound and one for Doppler ultrasound using internal jugular vein evaluated effect of guidance on infants.*	Stenz R, (2003) Paediatric central venous catheter insertions A*naesthesia,* **58**: 11, 1127-1128 Anaesthetists	
8. Randolph AG, Cook DJ, Gonzales CA et al. (1996) Ultrasound	493 patients requiring 513 placements of	To evaluate effect of real-time ultrasound guidance using regular	Rapidity of placement; Number of	<u>Rapidity of placement:</u> Results comparing ultrasound guidance vs landmark	*Experience of operators varied. Settings for CVC*	Systematic Review – Meta Analysis	1^{+}

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
guidance for placement of central venous catheters: a meta-analysis of the literature. *Critical Care Medicine* 24:2053-8.	Central Venous Catheters (CVCs). Only one study specified children included. No numbers or patient details provided.	or Doppler ultrasound technique for placement of CVCs compared with landmark placement. .	attempts before successful placement; Success of placement; rate of complications; rate of success after failure by landmark method.	technique were heterogenous (p<0.0001). Some showed took less time, some more. Mean difference was 9 seconds (95%CI: –80 to 62.2). <u>Number of attempts before successful placement:</u> Ultrasound guidance significantly decreases requirement of multiple placement attempts. Overall relative risk for ultrasound guidance 0.60 (95%CI: 0.45 to 0.79). <u>Success of placement:</u> Ultrasound guidance significantly decreases relative risk of catheter placement failure compared with landmark placement.. Overall relative risk of 0.32 (95% CI: 0.28 to 0.55). Catheters placed in internal jugular vein (IJV) (relative risk 0.38; 95%CI: 0.21 to 0.71); Subclavian vein (SCV) (relative risk 0.15; 95%CI: 0.04 to 0.53) 1 RCT for infants showed reduced number of attempts to success. <u>Rate of complications:</u> Frequency rate of complications during placement significantly decreased using ultrasound	*insertion varied. Only 1 Study detailed results on children and states due to their smaller vessel size may be beneficial for children but requires further investigation under controlled settings.* *All studies unblended : assessor bias.* *Variable definition of failed catheter placement.* *Provided search, details of included studies, evaluation of evidence*		+/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				guidance (relative risk 0.22; 95%CI: 0.10 to 0.45); IJV (relative risk 0.26; 95%CI: 0.11 to 0.58); SCV (relative risk 0.11; 95%CI: 0.02 to 0.56). Rate of success after failure by landmark method: Not all trials reported results. Success rates where reported ranged from 33% to 100% with ultrasound guidance following failure by landmark method.			
9. Tweddle DA, Windebank KP, Barrett AM et al. (1997) Central venous catheter use in UKCCSG oncology centres. *Archives of Diseases in Childhood* 77:58-59.	Children with cancer admitted to UKCCSG centres.	• *Characterisation of CVC use, insertion techniques & reinsertion rates.* • *Identification of variations in aftercare practice* • *Survey opinion of diagnosis of CVC sepsis among multiple centres belonging to a single cooperative group*		13/22 UKCCSG centres participated and returned 347 data forms. External catheters were inserted in 84% of cases & subcutaneous ports in 16%. There was a wide variation in surgical insertion technique. Most surgeons used the right internal jugular vein. There was considerable variation with respect to aftercare. Subcutaneous ports were flushed monthly in 88% of centres. Nurses were taught line care in 80% of centres compared with 33% for doctors. Criteria for diagnosing infection due to CVC colonisation indicated that positive blood cultures and flush associated rigor, fever were considered pathognomic. The authors conclude that the	*No age or diagnosis data given but authors state that these were representative of the prevalence of individual centres in the UK.*	Cross sectional audit	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				variations highlight the difficulties in interpreting the published data on CVC efficacy			

BLOOD PRODUCT SUPPORT

Nature of the evidence

3 national guidelines of fair quality
1 national surveillance report of fair quality
1 expert position paper (Appendix G)

Summary of the supporting evidence for the recommendations

- The three guidelines and the expert position paper recommended the use of agreed protocols although there was no supporting evidence specific for children and young people with cancer [1 2 4].
- The results of the national surveillance of adverse incidents indicated that medical and nursing and laboratory staff should be aware of the specific transfusion requirements of children [3].

The expert position paper was accepted by the GDG as providing advice on this topic and a detailed literature search was not performed.

BLOOD PRODUCT SUPPORT

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. British Committee for Standards in Haematology 2003. Transfusion guidelines for neonates and older children.	Newborn and older children	Development of guidelines for blood transfusion		Provides recommendations for blood transfusion in children		Guidelines	¾ +
2. British Committee for Standards in Haematology 2004. Guidelines for the compatibility procedures in blood transfusion laboratories.	-	Development of guidelines for blood transfusion		Standards to reduce the risk of transfusion of incompatible blood		Guidelines	3/4 +
3. Serious Hazards of Transfusion. Annual Report 2003.	-	Description of adverse incidents occurring during blood transfusion		The commonest error in 2003 was the failure to request irradiated blood appropriately		Guidelines	¾ +
4. United Kingdom Blood Transfusion Services 2002. Guidelines for the blood transfusion services in the UK	-	Development of guidelines for blood transfusion		Service guidelines		Guidelines	3/4 +

PAIN MANAGEMENT IN CHILDREN AND YOUNG PEOPLE WITH CANCER

The Question:

What are effective methods for pain management in children and young people with cancer?

Nature of the evidence

- 1 systematic review of fair quality
- 2 guidelines, 1 good quality, 1 fair quality
- 1 government policy of fair quality
- 1 expert opinion of fair quality

Summary of the supporting evidence for the recommendations

- There is evidence from a systematic review that relaxation and cognitive behavioural therapy (CBT) are effective in reducing effects of headache [2].
- There are guidelines on the management of pain control that has implications for service provision and emphasises the importance of protocols for the safe and effective use of analgesia [4].
- The Children's NSF states the importance of effective pain management and staff training [1].
- 1 expert opinion that concludes that effective pain management should be high priority in service provision and that there should be adequate numbers of paediatric oncology nurses [3].

It was clear from the evidence that multidisciplinary protocols should be in place for pain assessment and treatment and all children should have access to play specialists.

WHAT ARE EFFECTIVE METHODS FOR PAIN MANAGEMENT IN CHILDREN AND YOUNG PEOPLE WITH CANCER?[2]

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Jassal, SS (2002) *Basic symptom control in paediatric palliative care. The Rainbows Children's Hospice guidelines*. Association for Children with Life-threatening or Terminal Conditions and their Families. www.act.org.uk [accessed 14 December 2004]	Paediatric palliative care	Development of protocol for doctors and nursing staff in specialised units and in the community.		There is a detailed protocol for pain assessment and treatment	*More useful for guideline development. Contains little on service delivery implications. Not evidence based*	Guidelines/protocols	3/4 +
2. Hooke C, Hellsten MB, Stutzer C et al. (2001) *Pain management for the child with cancer in end of life care. APON Position Paper* Association of Pediatric Oncology Nurses. www.apon.org//files/public/Pain_Management.pdf [accessed 14 December 2004]	Children with terminal cancer.			• The APON conclude that effective management of pain can be achieved by :- • Ensuring that pain relief is made a high priority in service provision. • The use of appropriate pharmacological and non pharmacological interventions. • Adequate provision of specialist paediatric oncology nurses	*Presents the nursing perspective.*	Position paper/Expert opinion	4 +
3. Department of Health (2003) *Getting the right start: National Service Framework for Children, Young People and Maternity Services.*	Paediatrics	-	-	• Separate facilities for young children from those provided for adolescents • Designated play areas for young children and	*Not specific for child and adolescent cancer. States how important pain management is for*	Government policy	3/4

[2] Cross refer to palliative care evidence table

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Part 1: Standard for hospital services. London: Department of Health 56p.				privacy for adolescents • Specialist training for staff dealing with children • Play specialists who help children cope with the distress of being in hospital	*children in hospital*		+
4. Eccleston C, Yorke L, Morley S et al. (2003) Psychological therapies for the management of chronic and recurrent pain in children and adolescents *The Cochrane Database of Systematic Reviews* Issue 1.	Children and adolescents with chronic pain	Investigation of the effectiveness of psychological therapies	Relief of chronic or recurrent pain:- Pain experience Affect Cognitive coping and appraisal Pain behaviour Social role performance Biological and physical fitness measures Quality of life/satisfaction for recipients of care or carers	There is good evidence (18 RCTs included in analyses) that relaxation and CBT are effective in reducing severity and frequency of headache. There is no evidence for their effectiveness in other conditions.	*Considers chronic and recurrent pain. Not cancer specific No references on C & A cancer fulfilled inclusion criteria..*	Systematic review	1^{+} +
5. Scottish Intercollegiate Guidelines Network (2000) *Control of pain in patients with cancer. A national clinical guideline. No 44*. Edinburgh: Scottish Intercollegiate Guidelines Network 61p.	All patients with cancer	Development of recommendations for the assessment of pain, its management, choice of analgesia, interventional techniques for treatment, education on pain management and psychosocial issues.		**SOME OF THE RECOMMENDATIONS RELEVANT TO SERVICE GUIDANCE** Prior to treatment an accurate assessment should be performed to determine the type and severity of pain, and its effect on the patient The patient should be the prime assessor of his or her pain. All health care professionals involved in cancer care should be educated and	*Good quality guideline with high score on AGREE tool.*	Guideline	3/4 ++

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				trained in assessing pain as well as in the principles of its control. Patients should be given information and instruction about pain and pain management and be encouraged to take an active role in their pain management Analgesia for continuous pain should be prescribed on a regular basis not 'as required. Breakthrough analgesia should be administered at any time outwith regular analgesia if the patient is in pain. All staff using syringe drivers, including community based health care professionals, must be fully trained in their correct use. Safe systems for use and management of syringe drivers must be in place as detailed in guidance issued by the Scottish Executive Department of Health. All professionals looking after patients with pain from cancer should be aware of the range of neurosurgical and anaesthetic techniques available for the relief of pain. All professionals looking after patients with pain from cancer should have access to a specialist pain relief service, able to offer the			

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				techniques described above. If a patient's pain is not controlled by other measures, then the advice of a specialist in pain relief should be sought, with a view to performing one of the above procedures. Pre-registration curricula for health care professionals should place greater emphasis on pain management education. Continuing pain management education programmes should be available to all health care professionals caring for patients with cancer. All patients with cancer should have access to a health care professional appropriately qualified to offer advice and information, both verbal and written, regarding pain and effective pain management. Patients with cancer pain should be given an opportunity to be trained in some form of relaxation as an adjunct to pharmacological pain control. Family members should be offered information and education regarding the principles of pain and its management in order to address their lack of			

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				knowledge and concerns regarding analgesic administration, tolerance and addiction. A thorough assessment of the patient's psychological and social state should be carried out. This should include assessment of anxiety and, in particular, depression, as well as the patient's beliefs about pain. Patients with cancer pain should be given an opportunity to be trained in some form of relaxation as an adjunct to pharmacological pain control.			

MANAGEMENT OF NAUSEA AND VOMITING

The Question:

What is the evidence for the optimum management of nausea and vomiting?

Nature of the evidence

1 quasi randomised controlled trial of fair to poor quality
3 guidelines, two of good quality, one fair quality
1 expert opinion of fair quality

Summary of the supporting evidence for the recommendations

- The results of the quasi RCT on the use of evidence based guidelines on the symptoms of nausea and vomiting showed that such guidelines do improve control of nausea and vomiting [4].
- Data from one US and two UK guidelines give detailed information on protocol use to treat nausea, vomiting and diarrhoea. The evidence for the guidelines was considered however to be poor and in some instances the recommendations are formed from expert consensus [1 3 5].
- An expert opinion provides a review of the current evidence for management of nausea and vomiting in children with cancer [2].

EVIDENCE FOR THE OPTIMUM MANAGEMENT OF NAUSEA AND VOMITING

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Armon K; Stephenson T; MacFaul R et al. 2001 An evidence and consensus based guideline for acute diarrhoea management. Archives Disease Childhood 85:132-142	Children with acute diarrhoea presenting to hospital	Development of evidence based guideline	-	The authors recommend that all children with dehydration should be admitted to a paediatric facility.	*Not specific for children with cancer but treatment and service principles applicable*	Guideline	3/4 ++
2. Antonarakis E; Hain RDW. 2004. Nausea and vomiting associated with cancer chemotherapy: drug management in theory and in practice. Archives Disease Childhood; 89:877-880	Children with cancer	Review of the management of nausea and vomiting in children	-	In all children receiving cancer treatment, antiemetic prophylaxis should be given on each day that chemotherapy is given. It is very important to consider the most effective route	*Good review of current management of N & V in children with cancer. The conclusions have service implications*	Expert opinion/evidence review	3/4 +
3. Department of health 2002Prodigy Guidance – Palliative care – nausea/vomiting/malignant bowel obstruction	Patients > 16 years. UK	Development of guidance to aid management of nausea and vomiting during palliative care	-	Describes the incidence, causes and management of nausea and vomiting in cancer patients.	*Detailed clinical information on the treatment of nausea and vomiting.*	Guidance	3/4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
4. Kearney N; Miller M; Weir-Hughes D et al. 2004. Workflow Information systems for European Nursing Care. Wisecare +	Patients (> 18 years) with cancer undergoing chemotherapy. Pilot project of 11 patients aged between 13 and 20 years (mean age 16 yrs).	To assess the influence of the integration of patient symptom assessment and the promotion of evidence based guidelines on the symptoms of nausea and vomiting. Two groups one group received additional self care information. Both groups received evidence based guidelines. Both groups then received evidence based care. Data collection was over 20 months.		Results were available for 235 patients. All symptoms measured except fatigue improved following the introduction of evidence based intervention. The authors conclude that the introduction of structured patient assessment and evidence based guidelines significantly improves patients' symptoms during chemotherapy. 10/11 patients returned the questionnaire in the pilot project. The modifications of the questionnaire were considered by most teenagers to be adequate for symptom assessment	*No details of randomisation process. Exclusion criteria well described. Useful evidence..*	Quasi randomised controlled trial	1^- +/-
5. National Comprehensive Cancer Network 2004. Practice Guidelines in Oncology. Antiemesis. Version 1	All cancer patients US	Development of evidence based guidelines	-	The guidelines present flow diagrams with the different methods to treat emesis	*The evidence for the guidelines was considered to be of low quality. The statements were based on NCCN consensus based on this evidence.*	Guidelines	3/4 ++

NUTRITION

The Question:

What is the evidence for the optimum method of provision of nutritional support for children and young people with cancer?

Nature of the evidence

1 historical case series of fair quality
1 guideline of fair quality
2 expert opinions, one of fair quality, one fair to poor quality
1 expert position paper (Appendix H)

Summary of the supporting evidence for the recommendations

- The evidence from one historical case series indicates that children who were malnourished at diagnosis have poorer outcomes compared with well nourished children [3].
- The guideline provides information for optimum artificial nutritional support but is not specific to children with cancer [2].
- The expert opinions emphasise that it is necessary to understand the metabolic changes that occur in cancer patients and that nutritional support is vital in children with cancer [1 4].

The expert position paper was accepted by the GDG as providing advice on this topic and a detailed literature search was not performed

WHAT IS THE EVIDENCE FOR THE OPTIMUM METHOD OF PROVISION OF NUTRITIONAL SUPPORT FOR CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Andrassy RJ 1998 "Nutritional support of the paediatric oncology patient. Nutrition 14; 124-129.	Paediatric oncology patients	Description of nutritional support interventions	-	The author emphasises that it is necessary to understand the metabolic alterations occurring in cancer patients that cause nutritional depletion.		Expert opinion	4 +/-
2. British Society Gastroenterology 1996. Guidelines for artificial nutrition support	All hospitalised patients requiring nutritional support	Development of evidence based clinical practice guidelines	Improved nutritional status in previously malnourished patients	The guidelines give explicit instructions for nutritional support in patients with different diseases.	*Does not deal specifically with cancer patients.*	Guidelines	3/4 +
3. Donaldson SS 1981 "A study of the nutritional status of pediatric cancer patients" American .Journal Diseases. Childhood 135;1107.	455 paediatric cancer patients	Review of case notes to determine the effect of nutritional status on outcomes	Survival, weight loss, anorexia, fatigue.	Serum albumin levels were not correlated with nutritional status. Nutritional status was correlated with freedom from relapse in children with solid tumours. In children with localised disease nutritional status was correlated but this correlation was absent in those with advanced disease. Children who were malnourished at diagnosis have a significant poorer outcome compared with children who are well nourished at diagnosis. Nutritional status has prognostic implications.	*Dated study but provides information for an association of poor nutritional status with outcomes.*	Historical case series	3 +
4. Van Eys J 1998 "Benefits of nutritional intervention on nutritional status, quality of life and survival" Inter Journal of Cancer [supp] 11 66-8	Paediatric oncology patients	Reviews evidence for the benefits of nutritional intervention on nutritional status, quality of life and survival	-	The author concludes that nutritional support improves outcomes such as quality of life and survival. Nutritional support should be given with appropriate tumour directed therapy if curative intent is the goal f treatment.		Expert opinion	4 +

ORAL & DENTAL CARE

The Question:

What is the evidence for the optimum method of provision of oral and dental care for children and young people with cancer?

Nature of the evidence

1 systematic review of good quality
2 historical case series of fair quality
1 survey of fair quality
2 guidelines, 1 of fair quality, 1 fair to poor
1 expert opinion

Summary of the supporting evidence for the recommendations

- The results of one systematic review indicated that there was a lack of high quality evidence for effective treatment for oral infections and mucositis [7].
- It was demonstrated in one historical case series that root surface area of mandibular teeth is reduced in long term survivors of paediatric cancer [2].
- Untreated decay and problems accessing dental care was shown in one historical case series of children with cancer [1].
- One survey of all 22 UKCCSG centres revealed variation in service provision for oral and dental care [4].
- Two guidelines provided some recommendations for oral care [5] [6].
- The author of one expert opinion concluded that the development and implementation of evidence based guidelines could improve the oral and dental care of children and young people with cancer [3].

WHAT IS THE EVIDENCE FOR THE OPTIMUM METHOD OF PROVISION OF ORAL AND DENTAL CARE FOR CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Clarkson JE; Eden OB 1998. Dental health in children with cancer. *Archives Diseases Childhood*; 78:560-561	60 children with cancer 1-14 years (mean age 6.2 yrs)	Assessment of dental health 4-6 months post diagnosis	Presence of infection and dental problems	Untreated decay was diagnosed in 26 children and 20 children had visible plaque with gingivitis. 8 patients had problems gaining access to dental care. 25 children had received preventive dental advice. 21 children required urgent dental treatment. The authors conclude that the results highlight the need to improve the integration of dental services into the medical care structure.		Historical case series	3 +
2. Duggal MS 2003. Root surface areas in long-term survivors of childhood cancer. *Oral Oncology*; 39:178-183	69 long term survivors of paediatric cancer.	Quantification of root surface area of mandibular teeth in long term survivors of childhood cancer	Comparison of RST of cancer survivors with normal controls	RSA of mandibular teeth was significantly smaller in survivors of cancer patients than in normal controls. There was no relation between the RSA and the age at which cancer was diagnosed.	*Well designed study with appropriate use of statistics*	Historical case series	3 +
3. Gibson F2004. Best practice in oral care for children and young people being treated for cancer: can we achieve consensus? *Eur. J Cancer*;40:1109-1110	Children and young people with cancer	-	-	The author concludes that good quality evidence based guidelines are required for best practice in oral care.	*The UKCCSG; RCN through SIGN are in the process of developing clinical guidelines for oral care*	Expert opinion	4 +/-
4. Glenny AM; Gibson F; Auld E et al. (2004). A survey of current practice with regard to oral care for children	Paediatric oncology patients being treated at 22 UKCCSG centres	Establishment of current UK oral care practice by telephone survey of 22 UKCCSG centres	-	19/22 centres (86%) of the centres used protocols/guidelines for mouth care. There was wide variation in the use of		Survey	3/4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
being treated for cancer. *European Journal Cancer;* 40:1217-1224*:*				preventive oral care therapies. Only at 8/22 centres (36%) did children undergo dental check ups before the commencement of cancer treatment. There was little variation in advice given to parents. Patients on basic oral hygiene. The authors stress the need to establishing evidence based strategies for oral and dental care.			
5. Royal College Surgeons 1999. Clinical Guidelines. The oral management of oncology patients requiring radiotherapy: chemotherapy: bone marrow transplantation. RCS England	All cancer patients requiring RT,CT and BMT	The development of clinical guidelines for oral and dental care	–	Provides recommendations based on current evidence (1999) for management of oral and dental problems occurring during cancer treatment	*Partial evidence based guidelines, but slightly dated*	Guidelines	3/4 +
6. St James's University Hospital Leeds 2001. Guidelines for mouth care in paediatric/adolescent oncology patients.	Children and adolescents with cancer	Development of local clinical guidelines for mouth care	-	The authors provide evidence based recommendations for oral care.		Guidelines	3/4 +/-
7. Worthington HV;Clarkson JE; Eden OB. Interventions for treating oral mucositis for patients with cancer receiving treatment. Cochrane Review.	All cancer patients	Any intervention for the treatment of oral mucositis or its associated pain	Mucositis; days to heal; oral pain; scores; dysphagia; systemic infection incidence;analgesia;LOS; cost of oral care & QOL	Only 1 RCT met the inclusion criteria. The authors conclude that there is weak and unreliable evidence that allopurinol mouthwash, vitamin E, immunoglobulin or human placental extract improve or eradicate mucositis. There is no evidence that patient controlled analgesia is better than continuous infusion method for controlling pain.	*The reviewers also mention that there is no good evidence to support the use of antimicrobial agents for reducing oral mucositis.*	Systematic review	$1^{==}$ ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				Further well designed trials are required.			

REHABILITATION

The Question:

What is the most effective strategy to provide effective rehabilitation services for children and young people with cancer?

Nature of the evidence

1 systematic review of good quality
1 systematic review of fair quality (head injury in children)
1 literature review of fair/poor quality

Summary of the supporting evidence for the recommendations

- The NICE supportive and palliative care guidance contains evidence from a systematic review on general rehabilitation services for cancer patients [2].
- There is good evidence from the systematic review on the literature for traumatic brain injury in children on the effectiveness of rehabilitation of children and adolescents [1].
- The results of 1 literature review on the effectiveness of occupational therapy indicate that they have a positive role in providing psychosocial support, maximisation of function and family assistance [3].

There is a lack of good quality evidence for children and young people with cancer. Consensus opinion exists that adequate AHP input is vital and that timing of commencement of rehabilitation is important.

WHAT IS THE MOST EFFECTIVE STRATEGY TO PROVIDE REHABILITATION FOR CHILDREN AND YOUNG PEOPLE DURING TREATMENT AND FOLLOW UP?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Agency for Healthcare Research and Quality (1999) 2*S. Supplement. Rehabilitation for traumatic brain injury in children and adolescents*. Available from: http://www.ncbi.nlm.nih.gov/books/bv.fcgi?rid=hstat1.chapter.2633	Children and adolescents with head injury	Systematic review of the literature		There is no good evidence on the effectiveness of rehabilitation for C & A with head injury. Certain models for social skills training and cognitive rehabilitation have been shown to be ineffective in people who have similar disabilities, yet these models are still being used in children and adolescents with head injury. Interventions must be tested with experimental designs that incorporate concepts of child and adolescent development.	*The requirements of C & A with head injury have some similarities to those of C & A with cancer. Useful for type of questions and outcomes that should be formulated.* *Of relevance to C & A with neurological tumours.*	Systematic review	2^{++} +
2. National Institute for Clinical Excellence (2004) *Guidance on cancer services - improving supportive and palliative care for adults with cancer – the manual*. London: National Institute of Clinical Excellence. Available from: www.nice.org.uk	All patients, with cancer	Review of evidence on rehabilitation services and the effect of rehabilitation on patient outcomes		There is a growing body of evidence to support the effectiveness of various interventions. To inform service provision more research need to be done on the relative effectiveness of different interventions.	*Good review of evidence, but very little specific to C & A age range.*	Systematic review	2^{++} ++
3. Strong J (1987) Occupational therapy and cancer rehabilitation. *British Journal of Occupational Therapy* 50:4-6.	Cancer patients	Role of occupational therapy in rehabilitation		Review of the paediatric literature indicates that the occupational therapist's role has been advocated as largely one of psychosocial intervention and support, in addition to maximisation of		Review of literature	4 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				function, assistance with developmental tasks and provision of assistance to families on environmental matters. The psychosocial area is very important in paediatric oncology and play therapy is often use to help children with terminal cancer deal with their feelings.			

PSYCHOSOCIAL CARE

The Question:

What is the evidence for the best model of psychosocial care for children and young people with cancer?

Nature of the evidence

2 systematic reviews of fair quality
1 overview & survey of good quality
1 questionnaire study of fair quality
1 expert opinion of fair to poor quality
1 expert position paper (Appendix I)

Summary of the supporting evidence for the recommendations

- 1 systematic review concluded that for children and adolescents with cancer the evidence for the best model of psychosocial provision was poor [3].
- 1 systematic review for mixed cancer patients including children and adolescents concluded that the published evidence was poor but that there was some evidence to suggest that group therapy, education, counselling, cognitive behaviour therapy, relaxation therapy and guided imagery were of benefit [5].
- 1 questionnaire study examining unmet needs illustrated that for cancer patients (few patients, in C & A age range) that these needs were variable particularly with age and social class [4].
- 1 detailed overview and survey of UKCCSG centres gives current levels of service provision and makes suggestions for future developments [2].
- 1 expert opinion makes recommendations that are not evidence backed [1].

Whilst high quality evidence was lacking on the optimum psychosocial service provision, the NICE guidance on *Improving Outcomes in Palliative and Supportive Care in Adults with Cancer* recommended that cancer networks have an important role in coordinating service improvement to meet the demonstrated unmet need for psychosocial input.

WHAT IS THE EVIDENCE FOR THE BEST MODEL OF PSYCHOSOCIAL CARE FOR CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Children's Cancer Centre Royal Children's Hospital, Melbourne (2003) *Psychosocial care in paediatric oncology. Towards best practice at the Royal Children's Hospital*. Melbourne: Royal Children's Hospital	Australia. Service users of a paediatric oncology psychosocial care department.	Parent focus groups and survey Audit Staff consultation Benchmarking Consultation with community based cancer support organisations		The authors conclude that:- • There must be a commitment to and vision of psychosocial service development within the Centre. • Engagement of expertise within the hospital and where indicated, external agencies to assist in the development of services • Substantial funding for additional psychosocial services	*Recommendation for service improvement but specific to the Melbourne hospital. Can extrapolate to UK for ideal components of service provision. Useful*	Review/expert opinion	4 +/-
2. Clarke S, Mitchell W, Sloper P et al. (2003) *Current patterns of provision of psychosocial support and practical support services at NHS paediatric oncology treatment centres in the UK: an overview*. York: University of York .	21 UKCCSG treatment centres, including their associated Teenage Cancer Trust (TCT) units. The 2 TCTs not based at a UKCCSG centre were also surveyed.	Questionnaire survey to investigate the provision of psychosocial and practical support services.		All 21 centres completed the questionnaire and 2/3 separate TCTs replied. • The number of children and teenagers registered as new patients, per year varied from 250-15, with a mean of 97. • Data is available on age distribution and expertise at the centres. • 15/23 centres share care with other hospitals • 21/22 centres employed social workers, the majority of posts were funded by the voluntary sector • 11/20 centres employed psychologists. • 8/22 employed a psychiatrist • 1/21 centre employed a	*Excellent questionnaire survey with good questionnaire design and reporting of results.*	Overview/ Questionnaire survey	3 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				counsellor • 19/20 employed a play therapist • 21/22 employed at least 1 full time POONS • All 23 centres had family accommodation • Data was available from 22 centres on patient and family facilities, hospital transport and teenage facilities • Formal psychological assessments of patients are not routinely made. Patients, usually assesses by social worker, psychologist or nurse. • Support groups can be accessed from 21/23 centres • 17/23 centres have formal bereavement support • 12/23 centres offer some form of complementary therapy • 23 centres provide informal support • data available on cultural needs, information and transition support • 22/23 centres provide an outreach service for families within their local community. • 18/23 centres have procedures for transition of care from hospital to home. • 20/23 centres have a designated person responsible for assisting patients to return to school • there was a lack of			

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				consensus & information in relation to hand over of care from paediatric to adult services. • 11/23 centres provide psychosocial support for long-term survivors. • For palliative care all 23 centres offer a combination of care in the home, hospital and hospice The gaps in service provision most frequently reported were psychology support (11/23) , social work support (9/23), provision of age appropriate facilities, support for survivors/long term follow up, communication and duplication between statutory and voluntary sector			
3. Eiser C, Hill JJ, Vance YH (2000) Examining the psychological consequences of surviving childhood cancer: systematic review as a research method in pediatric psychology. *Journal of Pediatric Psychology* 25:449-460.	Children and adolescent survivors with cancer	Results of a systematic review of literature on psychological consequences of surviving childhood cancer	Psychological consequences	20 studies were identified, 17 from the US. Anxiety depression or low self esteem were not significantly different in child cancer survivors compared with population norms or matched controls. The studies were of poor quality and no definite conclusions can be drawn from the results.	*Well designed and described study with adequate description of inclusion and exclusion criteria.*	Systematic review	2^{++} +
4. McIllmurray MB, Thomas C, Francis B et al. (2001) The psychosocial needs of cancer patients: findings from an observational study. *European Journal*	1000 patients > 18 years, with breast, colorectal, lymphoma and lung cancer	Assess and identify the prevalence of psychosocial need	Unmet need for psychosocial care using a 48 point inventory.	Response rate 40%. Logistic regression analysis indicated that the statistically significant variables of need vary by both clinical and social characteristics. Results however do indicate the range of psychosocial needs	*Few patients, in age range of C & A. Clear patient selection bias*	Cross sectional questionnaire study	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Cancer Care 10:261-269.				required by cancer patients.			
5. Newell SA, Sanson-Fisher RW, Savolainen NJ (2002) Systematic review of psychological therapies for cancer patients: overview and recommendations for future research. *Journal of the National Cancer Institute* 94:558-584.	All cancers. Only 1 study in which population stated to be children. All other studies either adult or mixed ages.	Systematic review of literature on psychological therapies	Effectiveness of interventions targeting anxiety, depression, hospitality, stress, QOL, coping skills, domestic adjustment, social relationships, nausea, vomiting , pain and survival.	627 relevant papers were identified that reported on 329 intervention trials. Only 1 trial was rated as good for its methodology. Using effectiveness results from 34 trials with psychosocial outcomes, 28 trials with side effect outcomes, 10 trials with survival or immune outcomes, the authors suggest that the following have a positive effect on outcome:- • Group therapy, education, structured counselling & CBT. • Relaxation therapy and guided imagery for side effects	*Comprehensive review of the literature with good methodology. The authors stress the poor quality of published literature.*	Systematic review	2^{++} +

LONG TERM FOLLOW UP/SEQUELAE

The Questions:

1 What is the evidence for the most effective strategy to provide long term follow up (FU) for children and young people with cancer?
2 What is the evidence for the optimum type of late effects services for children and young people?
3. Should fertility (cryo) preservation strategies be routinely offered to all young people deemed at significant risk of infertility and competent to consent?

Nature of the evidence

Q.1

1 historical case series of fair quality
1 guideline of good quality
1 questionnaire survey of fair quality
1 expert opinion of fair quality
1 review of fair to poor quality
1 qualitative study of fair to poor quality
1 expert position paper (Appendix J)

Q.2

1 retrospective cohort study of fair to poor quality
1 cross sectional study of fair to poor quality
1 expert opinion of fair quality
1 expert position paper (Appendix J)

Q. 3

1 audit of fair quality
1 questionnaire survey of fair quality
1 survey of fair to poor quality
1 expert position paper (Appendix K)

Summary of the supporting evidence for the recommendations

Q.1

- The evidence from 1 large historical case series indicates the extent of chronic medical problems and that compliance with follow up was good [3].
- Three levels of follow up care are described in 1 guideline [2] and there are recommendations for GP and patient/carer information. Further issues, not dealt with specifically in the SIGN guideline are addressed in the position paper (Appendix J).

- The evidence from the questionnaire survey demonstrates the pattern of FU arrangements for UKCCSG patients [4].
- One expert opinion emphasised the important role for nurses in FU and that UK arrangements lack coordination and evaluation [1].
- One evidence based review summarises the clinical effects of childhood cancers and their treatments. The authors highlight the need for further research and the cost implications [5].
- One qualitative (US) study examined the barriers to FU [6].

Q.2

- The results of the US cohort study and historical case series demonstrated the problems with establishing a comprehensive late effects service and the authors recommend a national policy for adult survivors of childhood cancer [4 6].
- The US cross sectional study again indicated the problems with late effects services and noted the lack of outcomes based research to evaluate the components of follow up [5].
- The authors of 1 expert opinion conclude that follow up strategies are made empirically due to limited evidence and stress the importance of late sequelae [3].

Q.3

- The report of the audit of current provision of fertility services and the development of service guidance makes a series of recommendations for the development of comprehensive fertility services [1].
- The questionnaire survey provides information on the decision process surrounding sperm storage but makes no recommendations about whether fertility preservation strategies should routinely be offered to adolescents [2].
- The expert position paper provides recommendations and evidence for providing endocrine and fertility services for children and young people with cancer.

Q.1 WHAT IS THE MOST EFFECTIVE STRATEGY TO PROVIDE LONG TERM FOLLOW UP FOR CHILDREN AND YOUNG PEOPLE WITH CANCER?[3]

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Gibson F, Soanes L. Long term follow-up following Childhood Cancer: maximising the contribution from nursing. Eur J Cancer 2001: 37:1859-1868.	Children and young people with cancer. UK authors	Follow-up in paediatric oncology		Author's comments: - • Importance of physical, social and emotional adjustment • Role of nurse providing structured and continuing follow-up • Establishing re-entry into school • UK arrangements often informal, lack of co-ordination and evaluation • Can some patients be discharged from follow-up? For continuing care – by whom and in what setting?	*Important for UK practice. Nurses are interested in LTFU – to provide health education programmes and support in hospital based clinics and as nurse practitioners running nurse led clinics as occurs in USA.*	Expert opinion	4 +
2. Scottish Intercollegiate Guidelines Network. Long term follow up of survivors of childhood cancer. A national clinical guideline. Edinburgh; SIGN 2004; SIGN publication 76.	Young people who have survived cancer	Development of clinical guidelines		The recommendations from the guideline are:- • All survivors of childhood cancer should be actively followed up • At the end of a course of cancer therapy. Patients, their carers and GPs should be given a summary of the treatment and a list of signs of late effects to lookout for. • Each patient should have access to an appropriate designated key worker to coordinate care. • With appropriate	*Well designed guidelines with good AGREE score. Some issues not covered – see extra position paper from Levitt*	Guidelines	3/4 ++

[3] Cross refer to late effects question

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				training, specialist nurses can make a significant contribution to care. Describes 3 levels of follow up care			
3. Stevens MC, Mahler H, Parkes S. The Health status of adult survivors of Cancer in Childhood. Eur J Cancer 1998:34;694-698	Adult survivors of childhood cancers. Median age at the time of analysis was 22 years 10 months (18 yrs-35 yrs) Between 1968 and 1990, 1954 patients were registered with tumours.	Investigation of the pattern of morbidity in long term survivors of childhood cancer.	Chronic medical problems within 7 functional clinical areas:- Endocrine Fertility Sensory Neuropsychological Organ toxicity Mobility Cosmetic	600 patients (52%) had survived 5 years from first diagnosis but 67 (6%) died later. 290 patients attending the long term follow up clinic were available for investigation. 34 (12%) had survived treatment for relapse. Overall 169 (58%) had at least one chronic medical problem and 93 (32%) had two or more. Infertility problems (14%); nephrectomy (11%), thyroid hormone replacement therapy (9%) and visual handicap were the most common problems. Compliance with long term follow up was good and the audit of unselected sub group of all the survivors in the study showed that 84% had attended for surveillance. The results of the study confirm that the sequelae of cure are not trivial.	*Well described study with important results for service provision. Outcomes could not be related to specific modalities of treatment.*	Historical case series	3 +
4. Taylor A, Hawkins M, Griffiths A, et al.. Long term follow-up of survivors of Childhood Cancer in the UK. Paediatric Blood Cancer 2004;42(2); 161-168	All 22 UKCCSG centres.	Questionnaire to all clinicians at UKCCSG centres. British Childhood Cancer Survivor Study (BCCSS)	Questions to determine:- Discharge policy arrangements before and after 5 years from completion of treatment. Other clinics	Clinicians were divided into paediatric oncologists and other specialists. Completed questionnaires were received from 71 clinicians in 21/22 centres, a response rate of 77%. The unit of analysis was clinician not centre.		Questionnaire survey	3/4 +

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
			available to survivors and facilities available to 5 year survivors with, or at an increased risk of endocrine dysfunction. Involvement of nurses in long-term follow up clinics. Preliminary results of BCCSS, a population based cohort study established to study risks of adverse health outcomes in childhood cancer survivors	Subsequent to 5 years after the end of treatment 37/71 (52%) of UKCCSG clinicians follow up all survivors for life, whilst 32/71(4%%) discharge some patients. The majority of patients discharged had benign or stage 1 tumours or were treated with surgery alone without specification of disease. There were 5 clinicians who reported discharging all patients. Of the 32 clinicians discharging beyond 5 years, 97% of clinicians discharged to the GP. 14/32 clinicians reported that such patients were kept on alternative methods of follow up.			
5. Wallace WHB, Blacklay A, Eiser C, et al. Developing strategies for long-term follow-up of survivors of childhood Cancer. BMJ 2001;323, 271-274	Childhood cancer survivors in Britain	Development of long term follow up strategies	Long term complications considered: - • Second primary tumours • Cardiovascular disease • Fertility • Education, psychosocial and quality of life issues • Growth, bone mineral density and body composition	Evidence based on retrospective studies. The authors summarise the clinical effects of childhood cancers and their treatments. They highlight the need for research, such as the British Childhood Cancer Survivor Study, to provide an evidence base. The authors also highlight the financial implications of following all childhood cancer survivors for life		Review	4 +/-
6. Zebrack BJ, Eshelman DA, Hudson MM et al Health care of	20 young adult 9 median age 38; range 21-51)	Identification of barriers to the utilisation of follow up	Major barriers to health care	The barriers could be grouped in to 4 categories:- • Survivor related		Qualitative	4

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Childhood Cancer survivors. Cancer 2004 ;100;8433-850.	survivors of childhood cancer US	care using the Delphi technique.		barriers • Psychological barriers • Provider related barriers • Insurance or System related barriers. With regard to the preferred setting of care the chosen setting was a long term follow up clinic staffed by a physician experienced with late effects and a nurse practitioner and based at a teaching hospital or cancer centre but separate from the children's hospital/cancer centre.			+/-

Q.2 WHAT IS THE OPTIMUM TYPE OF LATE EFFECTS SERVICE FOR CHILDREN AND YOUNG PEOPLE WITH CANCER?

Q.3 SHOULD FERTILITY (CRYO) PRESERVATION STRATEGIES BE ROUTINELY OFFERED TO ALL YOUNG PEOPLE DEEMED AT SIGNIFICANT RISK OF INFERTILITY AND COMPETENT TO CONSENT?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. British Fertility Society Multidisciplinary Working Group A strategy for fertility services for survivors of childhood cancer. *Human* Fertility 2003;6(2): A1-A40BFS 2003	Children with cancer	Audit of services for the preservation of fertility before and during treatment. Production of strategic guidance.	Patent reproductive system	Important document for service provision with 45 recommendations	*Comprehensive series of recommendations to facilitate the development of a multidisciplinary service for children being treated for cancer and the adults they will become.*	Audit of activities and Strategic guidance	3/4 ++
2. Crawshaw M; Glaser A; Hale J et al. A study of the decision-making process surrounding sperm storage for adolescent minors within paediatric oncology. Department of Social Policy and Social Work, University of York. 2003	Staff in UK 1.Paediatric Oncology Centres 2.Assisted Conception Units	Questionnaire surveys of oncology centres and assisted conception units. Face to face interviews with • Professionals • Young men • Parents	**Paediatric oncology centres** -Storage services -Written guidelines -Service provision -Information provision and consent -Psychosocial support **Assisted conception units**	**Paediatric oncology centres** 40% have written guidelines regarding fertility preservation in adolescent males 55% have written information 55% Did not know which consent form to use 90% response rate from oncology centres. 19/20 centres would welcome the introduction of national guidelines for adolescent male fertility	*Well designed and reported study*	Questionnaire survey	3/4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
			-Storage services -Screening -Service provision -Consent -Psychosocial support	preservation. **Assisted conception units** 23 units currently offered storage facilities for sperm and/or testicular tissue. 61% screened for HIV 56% for Hepatitis B 60% for Hepatitis C 4% for Syphilis 4% of units had sperm stored for adolescent males 105 Assisted conception units selected for questionnaire from total of 160. Response rate 62% of those selected, 41% of total. 87% would welcome the introduction of national guidelines. Young men's issues -Choice -Information -Communication -Consent Young men understood the need for quick decision making about fertility preservation issues. Parent's issues -Role -Information -Communication -Coping with feelings			
3. Jenney MEM and	UK authors			Author's comments	*Useful for UK*	Expert	4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY

Levitt GA. Paediatric Update: The quality of survival after childhood cancer. *European Journal of Cancer,* 2002, 38: 1241-1250.				• Survival rates now so good, quality of life increasingly important
				• Need to maintain survival rates while minimising adverse sequelae
				• Evidence limited by changes in treatment schedules and supportive care guidelines
				• Increasing importance of late sequelae
				  • Cardiotoxicity
				  • Endocrine function
				  • Osteoporosis
				  • Fertility
				  • Psychosocial issues
				• Follow-up strategies made empirically due to limited evidence.
				• Need for objective measure of quality but also importance of child/adolescent and family perspective
					practice	Opinion	+

4. Oeffinger KC Longitudinal cancer-related health care for adult survivors of childhood cancer. Commissioned Report provided to the National Cancer Policy Board 2003.	USA 1. Childhood Cancer Survivor Study. A retrospective cohort study of 14 000 adult survivors aged 18 and older. Information available for 9434 adults. 2. The RWJ	Subset of the retrospective cohort study randomly selected and sent an 88 item questionnaire	Health care visit, in previous 2 years, related to cancer treatment	CCSS study 41% RWJ Barriers study 26% Decreased attendance associated with: - • Increasing age • Male gender • College education • Lack of medical insurance • Lack of concern about future health	*Only 67% of the original cohort represented in the CCSS study results.* *70% response rate to the RWJ Barriers study*	Retrospective cohort study.	2⁻ +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
	Barriers study was a subset of 1600 patients from the CCSS cohort			The author highlighted the growing population of childhood cancer 'survivors' who are vulnerable to the late effects of disease and therapies. Author's recommendations: - • National Health Care Policy for adult survivors of childhood cancer • Evidence based guidelines for screening and surveillance (research required) • Minimise barriers to care and maximise enablers	*questionnaire. The results reported are preliminary findings. Both studies use self-reported data.*		
5. Oeffinger KC ; Eshelman DA; Tomlinson GE et al. Programs for adult survivors of childhood cancer. *J Clin. Oncol.* 1998 16 (8):2864-2867. Ref ID: 27	USA Members of the Children's Cancer Group and the Paediatric Oncology Group 219 participants	16 item questionnaire	4 Categories of questions • Existence of a programme to follow-up young adults • Setting of the programme • Routine activities of the programme • Commonly encountered barriers to care	44% of institutions had a mechanism for following up adult survivors. 93% lead by paediatric oncologist 13% had assistance from an adult oncologist 8% had primary care physician input 70% had nurse clinician or nurse practitioner involvement Problems routinely followed-up included: - • Employment • Cardiac function • Educational attainment • Fertility • Insurability	*Response rate 83%*	Cross sectional study	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				• Other psychosocial measures • Quality of life Measures The authors noted the lack of outcomes based research to evaluate the value and components of follow-up. Patients were said to be uncertain about follow-up or unwilling to attend, particularly in the paediatric clinic setting. The authors suggest a need for purposeful, planned movement from child-centred to adult oriented health care.			
6. Oeffinger, K. C., Mertens, A. C., Hudson, M. M.et al.. Health care of young adult survivors of childhood cancer: a report from the Childhood Cancer Survivor Study. Annals of Family Medicine 2004 2[1], 61-70	9434 adult childhood cancer survivors, mean age 26.8 years (18-48 years).		General or non specific contact with healthcare provider . General physical examination Cancer related medical visit Medical visit to a cancer centre	87% reported general medical contact, 71.4% a general physical examination, 41.9% a cancer related visit and 19.2% a visit to a cancer centre. The authors conclude that primary care doctors provide health care for most of this increasing high risk population. Communication is vital between primary care and cancer centres. Univariate analyses were performed to assess the associations of demographic and cancer-related variables with the medical outcome measures.	*Well designed and described study. Authors discuss the limitations of the study design such as self reporting, survivors' perceptions of the reason for the medical visit. Selection bias, ethnic groups were under reported.*	Historical case series	3 ++

PALLIATIVE CARE

The Question:

For children & young people with cancer what is the evidence for the requirements for a comprehensive palliative care service?

Nature of the evidence

1 systematic review of good quality
1 guideline of good quality
4 questionnaire surveys, 1 of fair quality; 3 fair to poor
7 expert opinions, 2 of fair quality; 5 fair to poor quality
1 expert position paper (Appendix K)
1 strategic document of fair quality

Summary of the supporting evidence for the recommendations

- The evidence for child and adolescent cancer palliative care service requirements from one comprehensive systematic review was poor. The authors stress the difficulties in evaluating the palliative care team because of lack of measurable outcomes. Possible benefits of effective palliative care teams are reduced time in hospital, improved symptom control and increased carer satisfaction [10].
- The NICE guidance provides evidence based recommendations for the requirements for palliative care and supportive care services for adults with cancer; many of these can be extrapolated to children's services [13].
- The surveys provide information on audit of GP referral patterns and service requirements [7]; coordination of palliative care in shared care settings [9] and the role of the specialist palliative care clinical nurse in service provision [11]; symptoms and management of children with progressive malignant disease and practice within the 22 UKCCSG centres in 1997 [15]..
- The guidance/expert opinion from Addenbrooke's hospital provides details of their structures for community and shared care palliative care [1].
- The Association for Children with Life Threatening Conditions and their Families have recommendations for palliative care services specifically for young people aged 13-24 years and following a UK needs assessment recommendations for commissioners [3] [14].
- Two expert opinions stress the importance of specialist nurses in the provision of palliative care in the community [5] [6].
- The position statement provided estimates of the numbers of children requiring palliative care in the UK and recommendations for service provision [8].

- The Norfolk Children and Young People's Palliative Care Group performed a local needs assessment and produces a local strategy for palliative care service provision based on this assessment and in response to national guidance [12].
- The document from the Children's Hospital in Melbourne gives recommendations for best practice for children requiring palliative care [14].
- The strategic document (Wales) addresses palliative care for all ages but addresses the specific requirements of children [16].

There is a considerable amount of observational evidence on the requirements for effective palliative care service provision to children and young people with cancer. Outcome measurement is difficult and there is a need for well designed high quality studies to evaluate different models of service provision.

FOR CHILDREN & YOUNG PEOPLE WHAT ARE THE REQUIREMENTS FOR A COMPREHENSIVE PALLIATIVE CARE SERVICE?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Addenbrooke's Hospital (2004) *Addenbrooke's paediatric oncology symptom control & palliative care guidelines for community & shared care*. Cambridge: Addenbrooke's Hospital 18p.	Children receiving terminal care in the community linked to Addenbrookes Hospital			Description of current structures and services, contact details. Guidance for symptom control including prescribing advice	.	Expert opinion/guidelines	4 +/-
2. American Academy of Pediatrics (2000) Palliative care for children. *Pediatrics* 106:351-357.	USA. Children with cancer requiring palliative care.			The statement defines the purpose of palliative care to: - • Enhance quality of life • Relieve symptoms • Support families • Manage psychological symptoms The statement calls for development of clinical policies and minimum standards, training and research. Barriers to the development of the service were described: - • Infrequency of events in primary care • Finance (In USA) • Lack of paediatric expertise in hospices	*Statement from American Academy of Pediatrics. US practice but problems appear identical to those in UK*	Expert opinion	4 +/-
3. Thornes R (2001) *Palliative care for*	UK. Young people aged			Recommendations: - • Recognise young people	*Limited research literature.*	Expert opinion (consensus	4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
young people aged 13-24. Bristol: Association for Children with Life-threatening or Terminal Conditions and their Families 69p.	13-24 years with terminal conditions			as a distinct group • Involve young people in decision making • Undertake needs assessment • Flexibility of teams important but need a named key worker. • Joint planning health and social services • Plan for transition to adult care • Increase training. The numbers of young people receiving palliative care is not known as data is not collected routinely. There are increasing numbers of clients attending children's hospices. In late adolescence / early adulthood there is a higher proportion of individuals requiring palliative care. There is also and increasing number of survivors of childhood cancer reaching adulthood. Issues raised by young people: - • Involvement in decision making • Psychological needs • Transition from child to adult health services • Inexperience of adult health services Concerns about parents and siblings	*Wide ranging response to the consultation. Consensus document based upon limited research literature and written and oral submissions from: -* • *Professionals* • *Voluntary organisations* • *Young people's forum* • *Parents* • *Families*	document)	+
4. Association for	UK			Definition of palliative care as	*Guidance including*	Expert	3/4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Children with Life-threatening or Terminal Conditions and their Families, The Royal College of Paediatrics and Child Health (2003) *A guide to the development of children's palliative care services*. Bristol: Association for Children with Life-threatening or Terminal Conditions and their Families 53p.	Children with cancer requiring palliative care			including an active and total approach to care: - • Physical • Emotional • Social • Spiritual Need : - 8 per 50,000 children die annually 60-85 per 50,000 have life limiting conditions with some palliative care needs. Recommended model of care: - • Locally based multi-disciplinary team • Ready access to children's hospice and specialist palliative care advice • Most day to day care in the community • Key worker • Flexibility • Specific needs of adolescents and young adults Author's comments: - • High proportion of services provided and/or funded by voluntary sector. NHS and voluntary sector need to act quickly to resolve issues over funding. • Many illnesses requiring respite and palliative care are familial, raising	*needs assessment, review of existing services and recommendations for commissioners*	opinion/Guidance	+

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				the need for family support and guidance. • Good information available on cancers, particularly in children. Less information on other conditions. • Needs of minority, ethnic communities must be recognised. Locally based registers might be helpful.			
5. Beardsmore S (2002) Palliative care in paediatric oncology. *European Journal of Cancer* 38:1900-1907.	UK. Paediatric palliative care	Development of protocol for doctors and nursing staff in specialised units and in the community.		Authors conclude:- Transition to palliative care complicated by: - • Prognosis difficult to accept for patient, staff and family. • Desire for second opinion. • Participation in phase 1 trials Phase 1 trials can benefit by: - • Stabilising progression of disease • Alleviate symptoms • Psychological benefit • Potential gains for future patients • Adult trials not an adequate predictor Authors recommend: - • A flexible response • Paediatric outreach nurses and 24 hour access to specialist advice and information		Expert opinion.	4 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				WHO guidelines for cancer pain relief and palliative care are directly applicable in the paediatric setting			
6. Edwards J (2001) A model of palliative care for the adolescent with cancer. *International Journal of Palliative Nursing* 10:485-488.	UK Adolescents with cancer			Distinct needs of adolescents • Cancer care • Adolescent care • Specialist palliative care Problems due to small numbers 700 new cases in 13-19 year olds/year in the UK. 155 cancer deaths. Clinical issues: - • Transition from health to illness • Wide range of physical and emotional maturity • Different spectrum of cancers • Patient involvement- dependent upon level of maturity and understanding • Educational needs of professionals The model described uses collaboration between acute hospital, adult Specialist Care team, paediatric oncologists and community teams.	*Nurse Specialist description of a model of palliative care for adolescents with cancer*	Expert opinion;	4 +/-
7. Finlay I, Wilkinson C, Gibbs C (1992) Planning palliative care services. *Health Trends* 24:139-141.	UK. Palliative care services, all ages.	Audit of GPs referring to hospice, inpatient, outpatient and day care facilities(child and adult)		Response 137 GPs (60%) from 74 practices (85%). 91% used planned inpatient admission 4% used paediatric services (small numbers) 20% used palliative medicine		Questionnaire survey	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				clinic. 78% of GPs didn't know whether more or less services were needed. This reflects the rare occurrence and therefore limited experience for individual practitioners.			

8. Hain R (2004) *Paediatric palliative care, a position statement*. unpublished paper.	UK. Paediatric palliative care			An estimated 12 000 children in England and Wales require palliative care.
				120 likely to die from the condition. A proportion of these deaths will be due to malignancy, but not all.
				
				Recommendations: -
				• Sound community children's nursing infrastructure
				• Skilled medical support from general practitioners with an interest and some training in paediatric palliative care and from tertiary specialists
				• Expansion of consultant-led tertiary services
				• Expansion of teaching and research
				• Co-ordination and continuity of care
				  • Named key workers
				  • Multidisciplinary teams
				  • Parent or patient held records
					The document also includes longer-term goals such as increased respite provision and the development of adolescent and young people's palliative care services.	Expert opinion (based on a regional research project and published guidance).	4
							
							++

9. Harris N, Myers P (2003) *Palliative care*	UK . 8 Paediatric	Regional survey; Two part		8 out of nine units responded. Most palliative care for	*Authors commented that*	2.	3/4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
provision by paediatric oncology teams in the Southwest –a regional survey. San Antonio: Southwest Paediatric Oncology Palliative Care Network 6p.	oncology shared care units and the regional centre in Bristol.	questionnaire. 1. General section covering workload, team organisation and training over the previous three years. Confidential section examining aspects of personal involvement in palliative care.		children dying from cancer taking place in the community, but is co-ordinated and provided by hospital based paediatric oncology palliative care teams. Deaths: - 19% in hospital 78% at home 3% in hospice or other setting. Areas of concern: - • Pain control • Symptom control • Information for parents • Support for staff	*hospital staff underestimate the potential assistance to families from local primary care teams.*		+/-
10. Higginson IJ, Finlay IG (2003) Improving palliative care for cancer. *Lancet Oncology* 4:73-74.	UK. Palliative care, all ages	Hospital based palliative care teams	Process or outcomes of care for patients and families at the end of life. • Symptoms • Quality of life • Time in hospital • Total length of time in palliative care • Professional behaviour change (e.g. prescribing practice)	Results suggest possible benefits: - • Reduced time in hospital • Improved symptom control • Increased carer satisfaction • Influence on prescribing of opioids and non-steroidal anti-inflammatory analgesics	*Ten databases searched. Hospital setting, mainly UK and large teaching hospitals, though including studies from Sweden, Canada, Argentina, France and Italy. One randomised controlled trial. Only three studies included a control group. No adjustment for confounding. Effect sizes generally small. The authors comment on the "poor quality of studies". The effectiveness*	Systematic literature review; qualitative meta-synthesis and quantitative meta-analysis.	1^{+} +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
					of the palliative care team, working as unit, is difficult to measure. Standardised outcome measures would be valuable for practice and research.		
11. Jack B, Oldham J, Williams A (2003) A stakeholder evaluation of the impact of the palliative care clinical nurse specialist upon doctors and nurses, within an acute hospital setting. *Palliative Medicine* 17:283-288.	UK. Palliative care, all ages	A stakeholder evaluation of the role of the palliative care clinical nurse specialist in the acute hospital setting. 31 semi -structured interviews giving opinions from:- • Senior nurses • Consultants • Junior doctors Nurses of different Clinical nurse specialist appointments in palliative care		Emerging themes: - • Core components of the role include expert practice, education, consultation and research • Colleagues value support and advice • Education is particularly welcomed by senior nursing staff and doctors Clinical nurse specialists identify education as an important part of their role	*The study was based in a large UK NHS hospital (1300 beds) with 4 full time, hospital based, clinical nurse specialists. Results may not be generaliseable to smaller units.* *UK does not require graduate level entrants as defined by the American Nursing Association.*	Survey	3 +/-
12. Leeson J (2002) *Norfolk palliative care strategy for children and young people*. Norwich: Norfolk Children and Young People's Palliative Care Group 24p. Available from: www.cancernw.com/content/palliative_care/palliative_care_strategy_children.doc	UK authors. Children and young people (aged < 19 years) in Norfolk requiring palliative care.			Morbidity 10-12 children per 10 000 population. Mortality 1.1 per 10,000 child population. Children's palliative care different because:- • Small numbers requiring the service • Many rare diagnoses specific to paediatrics • Genetic illnesses- issues for family members • Families directly involved in care Issues raised by the needs assessment: -	*Local strategy and recommendations based on a local health needs assessment and in response to national guidance.*	Expert opinion	4 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				• Poor co-ordination of services • Lack of bereavement support / training • Patchy provision of respite care • Inequality of access to services • Lack of good practice guidelines • Lack of consistent key worker for families • Little support for siblings Recommendations • Locally based services • Named keyworker • Individual care plan for each family • Strengthen arrangements for tertiary services Education and information			
13. National Institute for Clinical Excellence (2004) *Guidance on cancer services - improving supportive and palliative care for adults with cancer – the manual*. London: National Institute of Clinical Excellence. Available from: www.nice.org.uk	All patients with cancer	Review of evidence on palliative care services. Recommendations for service provision	'Effective' service provision.	Numerous key recommendations for commissioners of palliative care services.	*Good review of evidence, but very little specific to C & A age range.*	Guidelines/ Systematic review	2^{++} ++
14. Royal Children's Hospital, Melbourne (2002) *Best practice in palliative care*. Royal Children's Hospital, Melbourne. 2002.	Australia. Paediatric palliative care			The site cites the Royal College of Paediatrics and Child Health (UK) description of best practice, with essential components: - • Assessment and care		Expert Opinion	4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Internet Communication.				plan • Key worker • Local clinicians and nurses skilled in paediatric palliative care • 24 hour support • Regular respite • Emotional support • Provision of medication and equipment • Financial assistance Barriers described: - • Rarity of conditions Only 40% child deaths due to malignant • conditions • Difficult to develop and maintain skills • Dual role for parents of care givers and decision makers Developmental factors in children affect understanding, ability to communicate and decision making			+/-
15. United Kingdom Children's Cancer Study Group, Paediatric Oncology Nursing Forum. *Survey of signs and symptoms. Symptoms and management of children with progressive malignant disease. Draft.* Leicester: United Kingdom Children's Cancer Study Group, Paediatric Oncology	Children and young people with cancer	Prospective survey of 22 UKCCSG centres May 1997-November 1997	Symptoms; current management and variation in service delivery	Study documents frequency of symptoms in children with CNS tumours; leukaemia; neuroblastoma; soft tissue sarcoma; nephroblastoma; osteosarcoma; lymphoma. The study showed symptomology differences between tumour types. The authors discuss the difficulties of evaluating palliative care services and suggest that by using symptoms that these are proxy measures of quality of	*Useful preliminary data for UKCCSG patients.*	Questionnaire survey	3/4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Nursing Forum 14p.				life			

16. Welsh Assembly Government (2003) *A strategic direction for palliative care services in Wales*. Cardiff: Welsh Assembly Government 26p.	Wales. Palliative care, all ages			The Strategy recognises that paediatric palliative care requires a specific needs assessment and strategy.
				
				The document covers issues relevant to palliative care at all ages: -
				• Pain control
				• Patient involvement
				• Out of hours services
				• Barriers to co-ordinated care
				• GP knowledge
				• Work force planning
				• Education and training
				  • Undergraduate
				  • Postgraduate
				• Voluntary sector issues
				• Carer support
				
				Recommendations are made for: -
				Generic services
				• Primary care
				• Secondary care
				• Tertiary care
				
				and Palliative care services.
					The document addresses palliative care services for all ages, though there is a short section on palliative care for children. Reference is made to other documents, such as The Calman Hine report, Cancer Services in Wales and Paediatric Palliative Care in Wales.	Strategy document	4
							
							+

BEREAVEMENT SERVICES

The Question:

What is the evidence for best practice in the provision of bereavement services for children and young people with cancer, their families and carers?

Nature of the evidence

1 systematic review of good quality
1 draft guideline of fair quality
1 questionnaire survey of good quality
1 questionnaire survey of fair quality

Summary of the supporting evidence for the recommendations

- A systematic review on palliative and supportive care gives general recommendations for bereavement care [2].
- 1 draft guideline from the newly set up National Child Bereavement group gives preliminary guidance on child bereavement services [1].
- The evidence from 1 questionnaire survey provides an up to date picture of current childhood bereavement services in the UK [4].
- The evidence from 1 questionnaire survey illustrates the problems of one health authority in providing bereavement services (not paediatric specific) [3].

There is a lack of evidence on what constitutes an effective bereavement service but there is consensus on the need for key worker support and that each treatment centre should provide bereavement support for a suitable period depending on the needs of individual families. Good communication skills and the provision of adequate information are vital in providing bereavement support.

WHAT IS THE EVIDENCE FOR BEST PRACTICE IN THE PROVISION OF BEREAVEMENT SERVICES FOR CHILDREN AND YOUNG PEOPLE WITH CANCER AND THEIR FAMILIES AND CARERS?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Child Bereavement Network (2003) *Service development and best practice guidelines for bereavement care for children*. London: National Children's Bureau. Available from: www.ncb.org.uk/cbn/projectdetail.asp?ProjectNo=275	Children with cancer	Project (ending 2006) to promote new service development in specific areas highlighted by mapping exercise in 2002 and to develop guidelines for best practice		The preliminary version of the guidelines is complete.	.	Draft guidelines	3/4 +/-
2. National Institute for Clinical Excellence (2004) *Guidance on cancer services - improving supportive and palliative care for adults with cancer – the manual*. London: National Institute of Clinical Excellence. Available from: www.nice.org.uk	All patients, with cancer	Review of evidence on bereavement care for families and carers		Differing forms of support are available for those experiencing bereavement, ranging from information, through befriending and self-help groups to more formalised psychological interventions such as counselling. There is inequitable distribution of bereavement services and the quality varies. Families and carers may never undergo screening to assess their level of vulnerability. Professionals are often not adept at assessing, predicting and responding to families' and carers' bereavement needs, both before and after	*Good review of evidence, but very little specific to C & A age range.*	Systematic review	2^{++} ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				death. Professionals need to pay greater attention to the needs of carers			
3. Northern & Yorkshire Cancer Registry & Information Service (2000) *The provision of bereavement support services. A pilot study*. Northern & Yorkshire Cancer Registry & Information Service	All subjects requiring bereavement support.	.Interviews with health professionals (primary, secondary and community care) who had experience of working with the bereaved.	Level of bereavement support provided by health professionals for a specific health authority in Yorkshire	The study indicated that bereavement services provided within the hospital setting were reactive not proactive. The use of an assessment tool was not reported and bereavement need was identified informally through contacts with the bereaved. There appeared to be insufficient bereavement policy statements. There were significant communication problems between secondary and primary care systems	*Illustrates problems within a typical HA for provision of a bereavement service.*	Questionnaire survey	3/4 +
4. Rolls L, Payne S (2003) Childhood bereavement services: a survey of UK provision. *Palliative Medicine* 17:423-32.	Bereaved children	Identification of current provision of bereavement services in the UK.		A questionnaire was sent to 127 services that were wither solely dedicated to childhood bereavement or offered a service within a host organisation. The response rate was 85% (108/127 services). The findings identified that 85% of bereavement services are located within the voluntary sector; 14% are dedicated childhood bereavement services. 44% of host organisations are hospices. 73% of services relied on both paid and unpaid staff ,with 11% relying entirely on paid staff and 14% of services entirely on unpaid staff.	*Good review of current UK service provision of bereavement services.*	Questionnaire survey	3/4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				Interventions were offered to children and young people (<18yrs) by 86% of the services. The authors conclude that the number of variables that need to be held constant between services makes the use of RCTs or other comparative research methods difficult as tools of service evaluation.			

MULTIDISCIPLINARY TEAMS/CARE

The Question:

What is the evidence for the role of the multidisciplinary team (MDT)on the outcomes of care of children and young people with cancer?

Nature of the evidence

1 good quality randomised controlled trial
2 case series of fair to poor quality
2 expert opinions of fair to poor quality
1 survey of fair to poor quality
1 guide/guidance of fair to poor quality
1 consensus of fair quality

Summary of the supporting evidence for the recommendations

- The advantages of multidisciplinary care for young people were demonstrated in one good quality RCT performed in Denmark. There were improvements in patients' attitudes to the healthcare system but quality of life scores were not significantly different [7].
- There was indirect evidence from two case series on multidisciplinary treatment of paediatric Hodgkin's disease and medulloblastoma and its beneficial effect on quality of life [3 4].
- The expert opinions conclude that multidisciplinary care offers the best opportunity for improved outcomes [6 8].
- The survey gives an estimate of the number of MDTs in a small sample of NHS trusts as less than 30% [1].
- The guide and consensus document provides standards and quality measures for cancer MDTs [2 5].

In children and young people with cancer there is a lack of high quality evidence that directly supports the positive effect of multidisciplinary care on survival. Observational evidence suggests that such care leads to improved quality of life for patients.

WHAT IS THE EVIDENCE FOR THE ROLE OF THE MULTIDISCIPLINARY TEAM ON THE OUTCOMES OF CARE OF CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Commission for Health Improvement, Audit Commission (2002) *National Service Framework assessments: No.1 – NHS cancer care in England and Wales. Supporting data: 5 Multidisciplinary team working*. London: Commission for Health Improvement and the Audit Commission 18p.	NHS Cancer Care in England and Wales	Survey MDT working in 22 NHS trusts (within 9 networks) in E & W		Less than 30% of trusts reported regular patient-planning MDTs for neurological/brain and CNS patients. Where an MDT was present the percentage membership was:- Lead physician/surgeon 100% Pathologist 83% Non-surgical oncologist 81% Other surgeon/physician specialising in same cancer 78% Nurse specialist 74% Radiologist 69% Palliative care nurse 34% Palliative care doctor 31% Medical trainees 23% Therapy radiographer 10% Information specialist 9% Service manager 9% Dietician 9% Ward nurses 7% Speech therapist 4% Physiotherapist 4% Social worker 4% Trials/audit 1% Pharmacist 1% OT 1%	*Small sample, difficult to draw conclusions*	Survey	3/4 +/-
2. Department of Health (2004) *Manual of cancer services. Topic 2a – The generic*	All patients with cancer UK	Standards for generic MDT		Detailed description of standards and measures of compliance.	*These generic MDT standards will be replaced with site specific ones*	Consensus	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
multidisciplinary team (MDT). London: Department of Health 22p.					*as such guidance becomes available.*		
3. Donaldson SS, Whitaker SJ, Plowman PN et al. (1990) Stage I-II pediatric Hodgkin's disease: long-term follow-up demonstrates equivalent survival rates following different management schemes. *Journal of Clinical Oncology* 8:1128-1137.	All patients were 15 years of age or younger with stage I - II Hodgkin's disease. Patients were treated between 1971 and 1985. The case series included 100 patients from the first centre (USA) and 71 from the second (UK).171 children (<15yrs) with stage I-II Hodgkin's disease from two institutions with differing approaches to management.	To compare the prognosis of children with stage I-II Hodgkin's disease	Actuarial survival and freedom from relapse (calculated using Kaplan -Meier technique). Prognostic factors were also analyzed using Cox regression.	The first centre (USA) used an aggressive approach: pathologic staging, extended-field radiation alone or involved-field radiation plus combination chemotherapy. The second centre (UK) used a less aggressive approach: clinical staging only and involved/regional-field radiotherapy. Combined modality therapy was used in both institutions in some cases. The 17 year survival of the entire group was 87% and 17 year freedom from relapse was 87%. Actuarial survival (91% at 10 years in both centres) and freedom from relapse (90% at 10 years USA, 83% UK) showed no significant difference between the 2 institutions. Authors' conclusions Treatment strategies should be directed toward the long-term goal of cure of disease with maximal quality of life. A multidisciplinary management philosophy undertaken at a centre with extensive experience in pediatric Hodgkin's disease is important to achieving this	*The many differences in management policy between the centres make a direct comparison difficult to interpret. Institution was not included as a prognostic factor in the Cox analysis. Minimum follow-up was 2 years. Median follow-up was 7 years 8 months (USA) and 6 years 1 month (UK).* *Comparison of the patient characteristics showed differences in Hodgkin's disease histology between the 2 centres. This may confound direct comparison.*	Case series / prognosis study	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				goal. Staging and therapy should be individualized taking age, extent of disease, and acute or long-term toxicity of therapy into account.			
4. Gerosa MA, di Stefano E, Olivi A et al. (1981) Multidisciplinary treatment of medulloblastoma: a 5-year experience with the SIOP trial. *Child's Brain* 8:107-118.	30 children (< 14 yrs) with medulloblastoma involved in a SIOP trial	Major surgical resection, extensive irradiation and combined chemotherapy (vincristine + CCNU).	Overall survival and actuarial survival. Performance status.	Results only refer to multidisciplinary treatment protocols not MDTs	*Paper dated, treatment now altered*	Historical case series	3 +/-
5. NHS Modernisation Agency, Cancer Services Collaborative Improvement Partnership *Multidisciplinary team resource guide*. www.ebc-indevelopment.co.uk/mdt/ [accessed 14 December 2004].	Cancer patients requiring multidisciplinary care.	Resource guide	-	Lists questions to help achieve the 12 standards for MDTs in the Manual of Cancer Standards. Gives recommendations for service improvements	*Useful information for generic MDTs*	Guide	4 +/-
6. Newman KD (1997) Hepatic tumors in children. *Seminars in pediatric surgery* 6:38-41.	Children with hepatic tumours	-	-	The author concludes that a multidisciplinary team approach to hepatic tumours in children offers the best opportunity for improved outcomes.	*Deals with multidisciplinary forms of treatment.*	Expert opinion	4 +/-
7. Nielsen JD, Palshof T, Mainz J et al. (2003) Randomised controlled trial of a shared care programme for newly	248 cancer patients, > 18 years	To determine the effect of a multidisciplinary shared care programme on the attitudes of newly	QOL & performance status. Patients attitudes to healthcare system and GPs.	Shared care is defined as:- when the responsibility for the health care of the patient is shared between individuals or teams who are part of separate organisations, or	*Well designed and described study. Non-blinded. Power of study discussed and adequate to detect*	Randomised controlled trial	1-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
referred cancer patients: bridging the gap between general practice and hospital. *Quality & Safety in Health Care* 12:263-272.		referred cancer patients. The shared care programme included transfer of knowledge from the oncologist to the GP, improved communication between the parties and active patient involvement.		where substantial organisational boundaries exist. It implies personal communication and organised transfer of knowledge from hospital doctors to GPs and patient involvement. 48 patients dropped out of study. (24 from each group); 17 had died. The shared care programme had a positive effect on patient evaluation of cooperation between primary & secondary care. The effect was particularly significant (p==0.003)in young (18-49) men. There were no differences in QOL.	*a difference. Appropriate statistical analyses. Good discussion about limitations of study – power, bias etc.. Only relevant to upper age group patients for question.*		++
8. Wittig J, Bickels J, Priebat D et al. (2002) Osteosarcoma: a multidisciplinary approach to diagnosis and treatment. *American Family Physician* 65:1123-1132.	All patients with osteosarcoma			The authors conclude that Improvement in survival is due to multimodality treatment	Deals with multidisciplinary treatment	Expert opinion	4 –

CONTINUITY OF CARE

The Questions:

1. How can the transition from paediatric to adult services best be managed to ensure quality services for teenagers and young people?

2. What is the evidence for the role of the key worker in the care of children and young people with cancer?

Nature of the evidence

Q.1

1 review of good quality
2 formal consensus papers of fair quality
2 expert opinion papers of fair quality

Q.2

1 RCT of fair quality
1 policy document
1 questionnaire survey of fair quality
1 literature review of fair to poor quality
1 guidance/resource pack

Summary of the supporting evidence for the recommendations

Q.1

- The conclusions of one literature review describing current UK provision of services for young adults were that there is a dearth of high quality evidence to indicate the best service model to ensure continuity of care [2].
- 1 formal consensus provides useful recommendations on service structure. A UK formal consensus concluded that further research is required to determine service provision and gives current recommendations [1 3].
- Two expert opinions, specific for young people with cancer provide overviews of issues and recommendations for successful transition of care [4 5].

Q.2

- The evidence from one randomised controlled trial did not provide evidence directly about the role of a key worker but indicated that a nurse coordinator acting in a key worker role significantly improved the coordination of palliative care for terminally ill patients (all ages) [5].
- A questionnaire survey to providers of care in the UK to disabled children indicated that there was considerable variation in the provision of key worker schemes. Thirty schemes were in operation in the UK in 2004 [2].
- Evidence from a review of the literature suggested that where key worker schemes were used in the treatment of disabled children that quality of life for families was improved [3] A further review produced a resource pack for key worker schemes for disabled children [4].
- An expert opinion concluded that there is no evidence on whether one profession is better than another in the key worker role [6.]

No evidence from high level research was identified to indicate the optimum model of service provision to ensure continuity of care for children and young adults with cancer; this also applied to disabled children.

The Children Act [1] states the importance of the key worker in coordinating the care of children. Observational evidence supports the role of the key worker in successful coordination in the transition of care.

Q.1 HOW CAN THE TRANSITION FROM PAEDIATRIC TO ADULT SERVICES BEST BE MANAGED TO ENSURE QUALITY SERVICES FOR TEENAGERS AND YOUNG PEOPLE?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. American Academy Pediatrics (2002) A consensus statement on health care transitions for young adults with special health care needs. *Pediatrics* 110:1304.	Young adults with special healthcare needs	Improving the transition of health care for young adults		The authors consider: • What is meant by healthcare transitions • Why planning for healthcare transitions are important They recommend: • All children have an identified healthcare professional • Core skills and knowledge of healthcare professionals are identified to develop healthcare transition services • Portable up-to-date medical summary for each child • Written healthcare plan for each child by age 14 • The same guidelines for primary & preventative care should be applied for all adolescents & young adults • Ensure all young people with healthcare needs have health insurance coverage which is affordable & continuous	*Not specific to cancer.*	Formal consensus	4 +
2. NHS Service Delivery and Organisation R&D Programme (2002) *The transition from*	Young adults with disabilities or chronic diseases	Improving the transition of health care for young adults		The authors review current practice and provide a review of literature (they note that little high-level research is available).	*Comprehensive literature review. Good critical appraisal and good questionnaire survey.*	Review of current practice	4 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
child to adult health and social care. London: NHS Service Delivery and Organisation R&D Programme 6p.				Four main models of transition services were identified	*Not specific to cancer.*		
3. Rosen DS, Blum RW, Britto M et al. (2003) Transition to adult health care for adolescents and young adults with chronic conditions. Position paper of the Society for Adolescent Medicine. *Journal of Adolescent Health* 33:309-311.	Young adults with special healthcare needs	Improving the transition of health care for young adults		Outlined principles of transition that authors believe have been nearly universally endorsed. Endorses the American Academy Pediatrics document: A consensus statement on health care transitions for young adults with special health care needs (2002). Additional recommendations from the Society for Adolescent Medicine: • Primary care provider should be responsible for coordinating appropriate services • Ongoing education for all concerned about the importance of appropriate transition • All adults should receive adult-orientated primary health care • Adult health care sector be encouraged to make adult-orientated services available to adolescents and young adults with chronic health conditions • Continued collaborative development of best practices for management of adults	*Not specific to cancer*	Formal consensus/ position paper	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				with diseases of childhood • Removal of restrictions/barriers preventing timely transition • Further research in the area of transition to adult health care			
4. Viner R (2003) Bridging the gaps: transition for young people with cancer. *European Journal of Cancer* 39:2684-2687.	Young people with cancer	Improving the transition of health care for young adults		Overview of issues occurring with transition of care. Elements of good transition practice are outlined: • A policy on timing of transfer • A preparation period and education programme • A co-ordinated transfer process • An interested and capable adult service • Administrative support • Primary care involvement Potential models are briefly discussed. The gold standard model for long term follow up is a seamless clinic which begins in childhood or adolescence and continues into adulthood with both paediatric and adult professionals providing ongoing life-long care as appropriate. Alternatively dedicated long-term follow up services may be set up within an adult setting, but without paediatric input. In this situation more detailed attention to transition		Expert opinion	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				planning is required. Where care for adolescents has started in an adolescent service staffed by adult oncologists and haematologists, life time follow up may be possible within that service. This follow up is best arranged as a formal Late-effects clinic. Life-long follow up in the paediatric setting runs the risk of lack of access to fertility issues and diseases of ageing. Least preferable model is transfer of young people to their local general adult cancer service with no dedicated Late-effects service			
5. Viner R (1999) Transition from paediatric to adult care. bridging the gaps or passing the buck? *Archives of Disease in Childhood* 81:271-275.	Young adults with special healthcare needs			Recommendations: • Transition preparation is essential • All paediatric general and specialty clinics should have a transition policy • Young adults should be taught skills to function in an adult setting • An identified person should be responsible for transition eg clinical nurse specialist • Management links developed between hospitals/adult & paediatric services • Large children's services develop a transition map	*Not specific to cancer.*	Expert opinion	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				for each specialty • Evaluation of transition should be conducted			

Q.2 WHAT IS THE EVIDENCE FOR THE ROLE OF THE KEY WORKER IN THE CARE OF CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. (1989) *Children Act 1989 Elizabeth II. Chapter 41*. London: HMSO.	All children	-	-	States about the coordination of care for children that there should be a named person i.e. key worker	-	Government policy document	4
2. Greco V, Sloper P (2004) Care co-ordination and key worker schemes for disabled children: results of a UK-wide survey. *Child Care, Health & Development* 30:13-20.	225 disabled children UK	Postal survey to determine the prevalence and nature of care coordination and key worker services		The response rate was 70% (159/225 questionnaires). Thirty five areas (22%) reported having a coordination scheme, 26 in England, 5 in Scotland and 4 in Wales and 0 in Northern Ireland. Thirty schemes provided key workers to families. The authors conclude that there was considerable variation in service models and little is known on how such variations affect outcomes for children and families.	*The authors address the problems with questionnaire surveys and emphasise that the survey gives a small picture of care coordination in the UK*	Questionnaire survey	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
3. Liabo K (2001) *A review of key worker systems for children with disabilities and development of information guides for parents, children and professionals*. Cardiff: Wales Office of Research & Development/Barnardos 41p.	Children with disabilities	Identification from the literature of major issues in managing key worker systems, the key worker's role and families needs and requirements for a successful service	Development of information guides for parents, children and professionals	Where a key worker system is present the QOL of families with disabled children is improved. The model is only available to a third of families in the UK. Key worker systems focus on parents' needs rather than the needs of services.	*Limited search of non medical databases. Some of the conclusions cannot be substantiated by the evidence presented*	Review of the literature	4 +/-
4. Mukherjee S, Sloper P, Beresford B et al. (2000) *A resource pack: developing a key worker service for families with a disabled child*. York: University of York, Social Policy Research Unit 67p.	Disabled children	Resource pack for implementing a key worker system for families with disabled children.		Review of the evidence for effectiveness of key worker systems indicates that:- ▪ Families with key workers have less unmet needs and improved relationships with professionals ▪ A key worker service needs to be located within a formal key worker service ▪ A key worker service should contain the following elements – proactive regular contact, supportive open relationship, family centred as opposed to a child centred approach, working across agencies, working with families' strengths and working for the family not the agency	*Care required in extrapolating to children and adolescent with cancer. Statements in evidence review not always backed by references.*	Guidance/resource pack	4 +
5. Raftery JP, Addington-Hall JM MacDonald, LD et al. (1996) A randomized controlled trial of the cost-effectiveness of a	167 terminally ill cancer patients with a prognosis of less than one year, in a single health authority.	To compare the cost effectiveness of a co-ordination service with standard services for terminally ill cancer patients with a	Survival at the end of the study, inpatient days, and cost per patient.	One group of patients was allocated to receive the services of a nurse coordinator who acted as a 'broker of services', the other received routine services.	*Stratified cluster randomization was used (the GP with which the patients were registered) but analysis was done on*	.Randomized controlled trial	1˙ +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
district co-ordinating service for terminally ill cancer patients. *Palliative Medicine* 10:151-161.	UK	prognosis of less than one year		There was no difference in patient survival between the groups. The co-ordination group used fewer inpatient days (mean 24 versus 40 inpatient days; t = 2.4, p = 0.002) and nurse home visits (mean 14.5 versus 37.5 visits; t = 0.3, p = 0.01). Mean cost per coordinated patient was lower than that of the control group patients 4774 pounds versus 8034 pounds, t = 2.8, p = 0.006). Authors' conclusions:- Assuming that the observed effects are real, improved co-ordination of palliative care offers the potential for considerable savings.	*an individual basis. Could spuriously overestimate the significance of differences.* *Unclear whether the investigators involved in data collection were blind to treatment allocation.* *Service use data was extracted from patients' case notes, and hospice records. 32% co-ordination group, and 22% control group were lost to follow-up. Service use data were collected in 53% of the co-ordination group, and 63% control group.* *Patients too ill to be interviewed were excluded.*		
6. Sloper P, Jones L, Triggs S et al. (2003) Multi-agency care co-ordination and key worker services for disabled children. *Journal of Integrated Care* 11:9-15.	Disabled children	Optimisation of coordination of care and key worker services. Review of evidence for effectiveness of key workers.	-	There is some observational evidence for the effectiveness of the key worker model for families of disabled children and also the staff working with them. There is no evidence on whether one profession is better than other in the key worker role. The authors report on a number of projects being		Expert opinion	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				performed in the UK			

PROTOCOL BASED CARE

The Question:

What is the evidence that protocol driven treatment improves outcomes for children and young people with cancer?

Nature of the evidence

1 systematic review of good quality
1 prospective cohort study of fair quality
2 historical case series, 1 of fair quality; 1 of fair to poor quality
1 literature review of fair quality

Summary of the supporting evidence for the recommendations

- There is evidence from one systematic review that there is some evidence for the positive effect of protocols on outcomes [1].
- One prospective cohort study provides some evidence, with methodological problems, for positive effect of protocol on outcomes for children with nephroblastoma [2].
- The evidence from 1 historical case series indicates that children can be successfully treated on a protocol at a non specialist cancer centre i.e. it is the protocol not the centre that is important [3].
- The evidence from 1 large historical case series indicated a positive protocol effect [7].
- One literature review concludes that there is no evidence for a positive protocol effect on survival [6].

WHAT IS THE EVIDENCE THAT PROTOCOL DRIVEN TREATMENT IMPROVES OUTCOMES FOR CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Edwards S, Lilford RJ, Braunholtz DA et al. (1998) Ethical issues in the design and conduct of randomised controlled trials. *Health Technology Assessment* 2:1-96.	Systematic review of literature on ethical issues and randomised controlled trials			636 articles containing ethical issues relating to RCTS were identified. 17 of these examined the effects of participating in clinical trials on patient outcomes, including the effect of protocol entry. 10 articles involved cancer therapy and examined the effect of trial entry on survival/disease free survival, of which 4 involved childhood cancer. There were few relevant studies and these were of variable quality. The results however support the belief that treatment protocols benefit patients. There are two options to redress the balance between trial and non-trial patients. The first is based on the so called 'treatment effect' and in this case all eligible patients are offered entry into trials. The second assumes that the 'trial' effect is really a 'protocol effect' and therefore all treatments whether trial or non trial should be performed under protocol regimes.	*Good quality review with adequate description of methodology. Extensive search, limited to English language.*	Systematic review	2^{++} ++
2. Lennox EL, Draper GJ, Sanders BM (1975) Retinoblastoma: a	UK. 313 children diagnosed with nephroblastoma in 1970.	98 children were entered into the MRC nephroblastoma study between October 1970	3 year survival	The 3 year survival rate was 58%. The rate in the children entered into the trial (77%) was significantly better than	*Trial participants and non trial controls were not matched for*	Prospective cohort study	2^{+}

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
study of natural history and prognosis of 268 cases. *British Medical Journal* 3:731-4.		– December 1973. 288/313 (92%) children had a nephrectomy, 248 (79%) received a course of RT and 267 (85%) were given at least 4 days CT. To identify a group of children who would have been eligible for inclusion in the MRC trial but were not entered and to compare the treatment and survival rates of this group with those of the trial patients.		that among children who were eligible for the trial but not included (58%) p<0.01. This result was more pronounced when allowance was made for the distribution of age and tumour stage (p<0.001). The authors conclude that all children with nephroblastoma should be treated according to well defined protocols	*prognosis variables but statistical adjustment was made. Recruitment rates not given. Appropriate use of statistics*		+
3. Meadows AT, Kramer S, Hopson R et al. (1983) Survival in childhood acute lymphocytic leukaemia: effect of protocol and placement. *Cancer Investigation* 1:49-55.	327 children < 15 yrs with ALL diagnosed between 1970-1975. US	Effect of treatment protocol and place of treatment.	Survival	The 4 year survival was 60% in the treatment protocol group compared with 19% for non protocol treated patients (p<0.001). Significantly higher survival rate at specialist paediatric cancer centres and for children treated on protocols, but little variation with type of centre within the protocol group.	*Indicates that it is protocol that is important not centre effect.*	Historical case series	3 +
4. NHS Modernisation Agency, National Institute of Clinical Excellence (2003) *What is protocol-based care?* London: NHS Modernisation Agency 7p. Available from: www.modern.nhs.uk/protocolbasedcare/whati	All patients receiving NHS treatment			Protocol based care is developed around NICE guidance or other recognised standards. Definition of protocol:- detailed descriptions of the steps taken to deliver care or treatment to a patient. **Definition equivalent to that of an integrated care pathway**	*This is really equivalent to integrated care pathway use and not 'protocol' as understood within the context of a clinical trial.*	Expert opinion	4 –

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
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5. Norum J, Nordoy T, Wist E (1995) Testicular cancer treated in a minor general oncology department. *European Journal of Cancer Part A: General Topics* 31:293-295.	98 patients, with testicular cancer. Norway	Evaluation of outcomes in patients treated at a district general hospital oncology department.	Remission; survival	All 98 patients obtained complete remission. The 5 year cancer corrected cumulative survivals according to Kaplan-Meier method were 0.9787 and 0.9804 in the seminoma and non-seminoma groups, respectively. These results were similar to those reported from major oncological centres. In Norway almost all treatment centres treat their patients, according to the same protocols. The use of protocols means that patients, can be successfully treated in non specialist oncology centres.	*Few patients in C & A age range, Insufficient details to assess quality.*	Historical case series	3 –
6. Shochat SJ, Fremgen AM, Murphy SB et al. (2001) Childhood cancer: patterns of protocol participation in a national survey. *CA: A Cancer Journal for Clinicians* 51:119-130.	200 cancer registries in US 2208 children and adolescents(< 21 years) DIAGNOSED IN 1987 AND 2293 DIAGNOSED IN 1992.	Assessment of patterns of protocol.	Protocol participation	53.8% of children were treated on protocols in paediatric centres compared with 25.1% treated at other institutions. In general the younger the patients, the more likely the chance of being treated in a protocol(paediatric centres 63.7%; others 42.0%) with very poor adolescent protocol participation (paediatric centres 34.8%; others 42.0%). The authors conclude that measures must be taken to increase participation in protocols.	*Results not directly applicable to UK. Not of direct relevance to question. Concentrates on inequality of protocol participation between children and adolescents and between specialist and no specialist centres.*	Survey	3/4 –
7. Stiller CA (1994) Centralised treatment,	All cancer patients	Review of published literature from 1984-	Survival	. Children entered into MRC trials had a significantly	*The papers are not critically appraised.*	Literature review	3/4

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
entry to trials and survival. *British Journal of Cancer* 70:352-362.		1993 (Medline, Embase) on patterns of care.		higher survival rate. Trial entry had little effect on survival at high volume centres. For children with A non LL entry to a trial and treatment at a teaching hospital were both associated with a higher survival rate. In 2 studies of children with retinoblastoma survival rate was highest at the national referral centre. For children with Wilms' tumour survival rates were highest for those included in MRC trials than those who were eligible but not included. Patients, who had surgery at a specialist centre had higher survival rates. The author concludes there is no evidence that referral or treatment according to protocols leads to improved survival rates. **CHECK**	*.The author discusses the possible sources of bias. Other possible outcome measures are discussed. Some of the studies reviewed predate the introduction of current treatment methods*		+
8. Youngson JHAM, Jones JM, Chang JG et al. (1995) Treatment and survival of lymphoid malignancy in the north-west of England: a population-based study. *British Journal of Cancer* 72:757-765.	1663 patients, entered into a specialist population based (cases 15 years) registry in North West England of patients, with lymphoid leukaemia and non Hodgkin's lymphoma. Cases diagnosed between Jan 1983-December 1986.	Estimation of treatment variations	Survival	**RESULTS OF RELEVANCE TO PROTOCOL:** 1009 patients were analysed. 159/1003 patients were entered into clinical trials of which 111 were managed at a specialist oncology centre. Patients were unlikely to have been entered into a trial unless they had been treated at a specialist oncology centre. Whether treatment had followed a recognised protocol was a significant		Historical case series	3 +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				factor affecting survival. The importance of the use of an appropriate protocol was marked for patients managed at 'other hospitals'. CLL patients had particularly poor survival when not treated on a protocol.			

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PLACE OF CARE

The Questions:

1. What evidence is there for the optimum place of treatment for children and young people with cancer?

2. What evidence is there for the effects of accessibility and centralisation of cancer services for children and young people?

3. Is there evidence for an association between the number of cases of cancer in child and adolescent cancer seen and outcome?

4. Is there evidence that shared care improves patient outcomes?

Nature of the evidence

Q1

1 non randomised controlled trial of poor quality[4].
1 systematic review of good quality.
1 retrospective cohort study of fair to poor quality
3 reviews, two of good quality and one of fair quality
6 historical case series, two of good quality, four of fair quality
1 guidance of fair quality
1 dissertation/evidence review of fair quality
1 expert opinion of fair quality

Q2

2 systematic reviews one of fair quality, one fair to poor quality
1 thesis/expert opinion of fair quality
1 literature review of fair quality
1 survey of fair quality

Q3

2 systematic reviews of fair quality, no specific paediatric papers
1 literature review of fair quality with specific paediatric papers
4 historical case series, 3 of good quality, 1 of fair quality

[4] Retained despite methodological problems - highest level of evidence available with relevance to the question

Q4

1 RCT of good quality
1 systematic review of fair quality
2 qualitative studies of fair to poor quality
1 review of fair quality
3 reports/guidance 1 of good quality; 2 of fair quality
1 expert opinion of fair quality

Summary of the supporting evidence for the recommendations

Q1

- No difference in outcomes was demonstrated in one non RCT with treatment in the community vs specialist centre. This trial had methodological problems [5].
- A systematic review indicated that there was insufficient evidence to recommend specialist treatment. The paediatric cancer papers reviewed did not meet the inclusion criteria [4]
- In adolescents with acute lymphoblastic leukaemia (ALL) one retrospective cohort study concluded that they should be treated in a paediatric setting [1].
- One review of the literature concluded that survival was improved in children with ALL, acute non lymphoblastic leukaemia, non Hodgkin's lymphoma (NHL), retinoblastoma and Wilm's tumours when treated in a specialist cancer centre/teaching hospital department. Two other reviews of all types of cancer patients were also were in favour of specialist treatment [12 8 3].
- Another historical case series demonstrated that survival for children with acute non lymphoblastic leukaemia NHL, Ewings sarcoma, rhambdomyosarcoma and osteosarcoma was improved at paediatric oncology centres compared with non UKCCSG centres [11].
- The evidence from one historical case series indicated that survival for children with ALL did not vary between paediatric oncology centres and other hospitals [13].
- For adolescents with ALL and acute myeloid leukaemia survival rates were similar at teaching and non-teaching hospitals in one historical case series [14].
- A difference in survival for patients with medulloblastoma and rhabdomyosarcoma treated in cancer centre vs a non cancer centre was seen in one historical case series; this difference was not seen in Wilms' tumour patients [6].
- The evidence from one historical case series indicated that for children with ALL survival in the non protocol group was improved at specialist paediatric cancer centres. For children treated on protocols there was little variation with type of centre [7].

- For children with Wilms' tumours evidence from one historical case series demonstrated that survival was reduced when patients were treated at non UKCCSG centres compared with paediatric oncology centres [9].
- The evidence from a dissertation/review of the evidence for the improvement in outcomes with dedicated adolescent units was equivocal [15].
- An expert opinion concluded that referral to a specialist centre was not always indicated [10].

Q2

- Evidence from 1 systematic review suggested that shared care was a safe option for cancer patients. The review was not specific for child and adolescent patients [1].
- The evidence from 1 systematic review was poor and no conclusions could be drawn about the relationship between distance and mortality and morbidity for cancer patients. The 2 paediatric studies did not meet the inclusion criteria [3].
- The conclusions from a thesis/expert opinion that studied paediatric cancer patients indicated that travel was not an issue for the decentralised group but that the parents using the more centralised service identified several problems with travel [4].
- Evidence from 1 literature review was contradictory for the burden of travel, but suggests that it is an inconvenience for patients, and may be a barrier for compliance. Not specific for C & A age range [5].
- The results of one survey showed that there was no significant correlation between travel times for treatment and overall radiotherapy uptake [2].

Q3

- The conclusions from one systematic review were that whilst the evidence from the published literature does support the volume/outcome association it is possible that different case mix and processes of care between high and low volume providers may partially explain the results. There were no specific child and adolescent cancer studies [1].
- One systematic review provided evidence for the volume/outcome (30 day mortality) association for cancers that require complex surgical interventions compared with those patients treated with low risk surgery. No specific child and adolescent cancer studies [2].
- Evidence for improved survival for children with ALL, acute non lymphoblastic leukaemia, retinoblastoma and Wilm's tumours treated at high volume hospitals was provided from one literature review [6].
- There was evidence from one historical case series that survival of children with acute nonlymphoblastic leukaemia, non Hodgkin's lymphoma, Ewing's sarcoma and osteosarcoma was better in high volume treatment centres [3].

- Survival of children with ALL and AML did not vary with case load in one historical case series [4].
- One historical case series provided evidence that survival was significantly higher for children with ALL treated at high volume hospitals and entered into clinical trials [3].

Q4

- Randomised controlled trial evidence indicated that shared care had a positive effect on patients' views of cooperation between primary and secondary care; there was no difference in quality of life. There were no patients less than 18 years old in the trial [6].
- The safety of shared outreach care was supported by evidence from one systematic review [2].
- Qualitative studies indicated that parents had problems with shared care in district general hospitals and that parents thought shared care promoted the security of the whole family [5 7].
- The problems with agreeing a standardise definition of shared care were discussed in one review [4].
- 3 reports/guidance provided information on requirements and standards for a UK shared care centre [1 8 9].
- The conclusions of an expert opinion were that centralised care can have negative outcomes for families and shared care requires efficient organisation [3].

There is limited good quality evidence to suggest the optimum place of treatment for children and young people with cancer. The choice of outcome measures is difficult and survival has most frequently been used, with no conclusive supporting evidence being found. Other measures such as quality of life and patient satisfaction are also important and several studies have addressed these outcomes. The evidence for shared care improving outcomes appears to depend on whether the care is well coordinated with good communication methods.

Q 1. WHAT EVIDENCE IS THERE FOR THE OPTIMUM PLACE OF TREATMENT FOR CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Boissel N, Auclerc M-F, Lhéritier V et al. (2003) Should adolescents with acute lymphoblastic leukaemia be treated as old children or young adults? Comparison of the French FRALLE-93 and LALA-94 trials. *Journal of Clinical Oncology* 21:774-780.	Adolescents with ALL aged 15 to 20 years. 77 treated in paediatric FRALLE-93 trial (1993 to 1999) 107 treated in adult LALA-94 trial (1994 to 2000), 100 with complete follow-up were analysed. Treatment groups were similar apart from median age (15.9 years for FRALLE versus 17.9 years for LALA)	To compare treatment of adolescents with acute lymphocytic leukaemia (ALL) using paediatric and adult protocols Adolescents diagnosed by paediatricians are treated in paediatric trials. Adolescents diagnosed by GPs or internists are treated in adult departments. Treatment regimens for both trials were reported. Higher doses of all major ALL drugs were given in FRALLE-93 within shorter time period compared with LALA-94. Country: France.	Complete remission (CR); Disease free survival (DFS); event free survival (EFS); relapse free survival (RFS). Predictors of EFS at 5 years.	Treatment with the paediatric protocol (FRALLE-93) significantly improved CR rates compared with the adult protocol (LALA-94) especially for patients with BCP-ALL; CR for all: 94% with FRALLE versus 83% with LALA, P = 0.04. CR for BCP-ALL: 98% with FRALLE versus 81% with LALA, P = 0.002. Treatment with the paediatric protocol (FRALLE-93) significantly improved EFS and DFS at 5 years compared with the adult protocol; EFS: 67% with FRALLE versus 41% with LALA, P < 0.0001. DFS: 72% with FRALLE versus 49% with LALA, P = 0.0004. The only prognostic factors for EFS were WBC (P < 0.0001) and the trials (P = 0.004).	The authors concluded that adolescents with ALL should be treated with intensive paediatric protocols. *Treatment groups were not randomly allocated to treatment. The authors did compared baseline characteristics between groups and found groups to be similar.*	Retrospective cohort with control group	2^- +/-
2. Foreman NK, Thorne RN, Mott MG	678 children < 15 yrs with cancer	Quality of primary and hospital based care.	Survival by diagnosis period &	At 5 years patients, with CNS tumours experienced a 58%	*Well designed study. Appropriate*	Historical case series	3

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
(1996) Variation in survival of children with cancer within a region of the United Kingdom. *Cancer* ;77:785-90.		Large vs small hospital size.	type of cancer, survival by county & cancer.	survival rate in large hospitals and a 41% survival rate in small hospitals (p=0.03). The rate of entrance into trials was similar for each of the hospital types. There were significant differences in survival between the 5 counties studied, particularly for CNS tumours.	*statistical analyses. Provides evidence that children treated at larger hospitals likely to have improved prognosis. Significantly improved survival rates at 'large' hospitals (≥6 patients per year) compared with 'small' hospitals (< 6 patients per year)*		+
3. Grilli R, Minozzi S, Tinazzi A et al (1998) Do specialists do it better? The impact of specialization on the processes and outcomes of care for cancer patients. *Annals of Oncology* 9:365-374.	Patients with cancer receiving specialist carer	Assess the impact of specialisation on processes & outcomes of care for cancer patients.	Mortality, morbidity. Process outcomes e.g. specialisation of treating clinician, numbers of patients treated.	47/189 potential studies met the inclusion criteria. 12/24 (50%) studies provided information on process and 17/32 (53%) information on outcomes. Overall results were in favour of specialised clinicians/centres and were generally statistically significant. The study quality was however low	*Well described and designed study. Note is taken of the need to adjust in comparisons for case mix.*	Review	4 ++
4. Harding M, Lord J, Littlejohns P et al. (2002) *A systematic review of the evidence relating process of care or outcome to treatment in specialist and non-specialist hospital settings.* London: St George's Hospital Medical School 208p.	Patients with cancer	Assessment of difference in outcome between treatment in specialist and non specialist centres	Survival	The authors conclude that there was insufficient high quality evidence to indicate that specialist care affected outcomes in cancer patients	*High quality study No studies in paediatric cancer met the inclusion criteria. Publication bias significant.*	Systematic review	1⁻ ++
5. Kisker CT,	24 children	Evaluation of medical	Febrile episodes,	Results presented on 46/82	*Study power not*	Non	2⁻

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Strayer F, Wong K et al. (1980) Health outcomes of a community-based therapy program for children with cancer – a shared management approach. *Pediatrics* 66:900-906	receiving shared care and 22 who received specialist care. Both centres used the same treatment protocols.	outcomes when care is provided by a shared care system.	infections, drug toxicities, neutropenia, thrombocytopenia. Physician performance (protocol compliance)	patients. No significant differences were reported in febrile episodes & infections, drug toxicity, blood dyscrasias or protocol compliance.	*reported, but likely to be low. (small number of patients). Patient characteristics & severity of disease at diagnosis not described. Incomplete adjustment for confounding factors. High likelihood of bias*	randomised controlled trial	–
6. Kramer S, Meadows AT, Pastore G et al. (1984) Influence of place of treatment on diagnosis, treatment and survival in three paediatric solid tumours. *Journal of Clinical Oncology* 2:917-923.	147 patients with Wilms' tumours, 87 with rhabdomyosarcoma and 76 with medulloblastoma.	Determination of effect of place of treatment between cancer centres and non-cancer centres	Disease free survival (DFS)	Differences in 3yr DFS between CC and NCC were noted for medulloblastoma (52% v 24%) and rhabdomyosarcoma (48% v 10%, but not for Wilms' tumours (79%v68%). The principle management contrast found in rhabdomyosarcoma was that multiagent CT was used less often in NCC. Wilms' tumour patients were evaluated and treated similarly in the CC and NCC, except for surgical approach and FU.	*No case mix adjustment. Patterns of care, US orientated. Insufficient details of statistical analyses.*	Historical case series	3 +
7. Meadows AT, Kramer S, Hopson R et al. (1983) Survival in childhood acute lymphocytic leukaemia: effect of protocol and placement. *Cancer Investigation* 1:49-55.	327 children < 15 yrs with ALL diagnosed between 1970-1975. US	Effect of treatment protocol and place of treatment.	Survival	The 4 year survival was 60% in the treatment protocol group compared with 19% for non protocol treated patients ($p<0.001$). Significantly higher survival rate at specialist paediatric cancer centres and for children treated on protocols, but little variation with type of centre within the protocol group.	*Indicates that it is protocol that is important not centre effect.*	Historical case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
8. Pheby, DFH, Bray FI (1998) *Review of studies designed to explain variations in cancer disease outcome, particularly in relation to variations in patterns of practice*. Bristol: University of the West of England 161p. Available from: www.uwe.ac.uk/fas/uae/cancer3.pdf	Patients with ICD9 diagnosis 140-208 of cancer of any age	Review of studies on variations in cancer outcomes in relation to variations in patterns of practice.		4 papers dealing with childhood cancer fulfilled inclusion criteria. There were data problems but overall the studies indicated that survival was improved with treatment at specialist centres.	*Comprehensive literature review and discussion of the literature and factors affecting cancer outcomes.*	Review	4 ++
9. Pritchard J, Stiller CA, Lennox EL (1989) Over treatment of children with Wilm's tumour outside paediatric oncology centres. *British Medical Journal* 299:835-836.	30 Wilms'' tumour Patients diagnosed 1980-1982 *not* treated in centres in the UK Childhood Cancer Surveillance Group (UKCCSG)	What is the treatment regimen for children with Wilms'' tumour outside of paediatric oncology centres?	Disease type by treatment given, 3- year survival rate by centre type	10 of 20 children studied at these centres received more treatment than the authors would recommend. The three- -year survival rate was significantly lower in non-UKCCSG centres compared with that of paediatric oncology centres. Authors recommend that patients should be included in multicentre studies after and after establishment of prognosis, care should be shared with a paediatric oncology centre.	*Log-rank Test. Dealing with very small numbers (20-30) but authors give some evidence that treatment may be substandard where treatment does not occur at a UKCCSG centre.*	Historical case series	3 +
10. Selby P, Gillis C, Haward R (1996) Benefits from specialised cancer care. *Lancet* 348:313-318.	All cancers	-	-	The author concludes that there is some evidence to support the view that referral is not always necessary to a cancer centre.	*Low relevance to question.*	Commentary	4 +
11. Stiller CA, Draper GJ (1989) Treatment	Children with acute non-lymphoblastic	Comparison of survival rates between	Survival	Children with acute non-lymphoblastic leukaemia, non	*Relevant to question*	Historical case series	3

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
centre size, entry to trials and survival in acute lymphoblastic leukaemia. *Archives of Disease in Childhood* 64:657-61.	leukaemia, Hodgkin's disease, non HL, neuroblastoma, Wilms' tumour, osteosarcoma, Ewing's tumour & rhabdomyosarcoma .	UKCCSG patients and non-UKCCSG patients		HL, Ewing's tumour, rhabdomyosarcoma and osteosarcoma treated at paediatric oncology centres had significantly (p= < 0.05) higher survival rates than those treated elsewhere.			+
12. Stiller CA (1994) Centralised treatment, entry to trials and survival. *British Journal of Cancer* 70:352-362.	All cancer patients	Review of published literature from 1984-1993 (Medline, Embase) on patterns of care.	Survival	For children with ALL there was a significant trend towards higher survival rates in children being treated at high volume centres. Children entered into MRC trials had a significantly higher survival rate. Trial entry had little effect on survival at high volume centres. For children with A non LL entry to a trial and treatment at a teaching hospital were both associated with a higher survival rate. In 2 studies of children with retinoblastoma survival rate was highest at the national referral centre. For children with Wilms' tumour survival rates were highest for those included in MRC trials than those who were eligible but not included. Patients, who had surgery at a specialist centre had higher survival rates.	*The papers are not critically appraised. .The author discusses the possible sources of bias. Other possible outcome measures are discussed*	Literature review	3/4 +
13. Stiller CA, Eatock EM (1999) Patterns of care and survival for	4998 children aged between 0 and 14 years	Effect of patterns of care	Survival. Hospitals were classified as:-	5 year survival improved from 67% in 1980-84 to 81% in 1990-94.	*Large well designed multicentre study.*	Historical case series	3

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
children with acute lymphoblastic leukaemia diagnosed between 1980 and 1994. *Archives of Disease In Childhood* 81:202-208.			• Mean annual number of new patients aged 15-29 with ALL & AML • As teaching or non-teaching hospitals	The authors conclude that survival did not vary systematically with hospital case load or between paediatric oncology centres and other hospitals. Trial entry had an effect on survival.			++
14. Stiller CAS, Benjamin RA, Cartwright JV et al. (1999) Patterns of care and survival for adolescents and young adults with acute leukaemia--a population-based study. *British Journal of Cancer* 79:658-665.	879 patients, aged 15-29 yrs. with acute leukaemia during 1984-1994.	Effect of patterns of care	Survival. Hospitals were classified as:- • Mean annual number of new patients aged 15-29 with ALL & AML As teaching or non-teaching hospitals	For ALL actuarial survival rates were 43% at 5 years after diagnosis and 37% at 10 years. Survival improved significantly between 1984-88 and 1989-94 for those aged 15-19 at diagnosis. Entry into trials had no effect on survival. Survival rates were similar at teaching and non-teaching hospitals & at hospitals treating different numbers of study patients per year. For AML survival rates 42% at 5 yrs after diagnosis & 395 at 10 years. Survival did not vary with category of hospital. Trial effect was equivocal.	Survival. Hospitals were classified as:- • Mean annual number of new patients aged 15-29 with ALL & AML As teaching or non-teaching hospitals	Historical case series	3 ++
15. Wilkinson JR (2001) *Will the creation of an adolescent centre lead to improved outcomes in Yorkshire?* Edinburgh: University of Edinburgh. Unpublished thesis.	Patients aged between 10 to 24 years of age with cancer.	Evaluation of the proposal for creation of a teenage and young person's cancer unit in Leeds.	Needs assessment of young people with cancer in Yorkshire. Qualitative work examining the needs of young people with cancer.	The evidence is equivocal that place of treatment improves survival. There is insufficient evidence to state that the quality of care which can be offered by specialist teenage units is superior to that offered by smaller local hospitals.	*Discrepancy between overall conclusion and data presented.*	Thesis	4 +

Q.2 WHAT EVIDENCE IS THERE FOR THE EFFECTS OF ACCESSIBILITY AND CENTRALISATION OF CANCER SERVICES FOR CHILDREN AND YOUNG PEOPLE?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Campbell NC, Ritchie LD, Cassidy J et al. (1999) Systematic review of cancer treatment programmes in remote and rural areas. *British Journal of Cancer* 80:1275-1280.	All cancers	Identification of problems and effectiveness of oncology service provision in remote and rural areas	Patient and physician satisfaction. Survival	The authors concluded that there was some evidence to support the safety of shared outreach care. Such care could make specialist care more accessible to outlying patients.	*. All studies were small and had methodological problems. Little evidence of relevance to the question*	Systematic review	2^{++} +
2. Cosford P, Garrett C, Turner K (1997) Travel times and radiotherapy uptake in two English counties. *Public Health* 111:47-50.	Registered cancer patients who received radiotherapy from 14 local authority districts in UK in 1991. Residents recorded at a single cancer centre as receiving palliative or radical radiotherapy at that centre in 1991.	To examine whether longer travel times for radiotherapy are associated with reduced overall uptake of radiotherapy treatment, or with reduced uptake of palliative as opposed to radical radiotherapy.	Uptake of radiotherapy.	There was no significant correlation between travel times for treatment and overall radiotherapy uptake (correlation coefficient r=0.40, 95%CI -0.19 to 0.78, P=0.18). The non-significant trend towards increasing uptake with increasing travel time disappeared with the exclusion of the four districts where treatment after six months was included in the data (r=0.08, -0.61 to 0.70, P=0.84). There was no significant change in the ratio of palliative to radical radiotherapy at one cancer centre with increasing travel time to that centre (r=-0.29, -0.72 to 0.31, P=0.34).	*Considerable variability was observed between local authority districts for both measures of uptake. Longest travel times were about one hour. Total study population not stated.*	Survey	3/4 +
3. Ferguson B, Place M, Posnett J (1996) *Accessibility and*	All cancer services			3000 articles were identified. and approximately 300 were screened against inclusion	*93% of studies were cross sectional and thus*	Systematic review	2^{++}

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
centralisation in cancer services. A report by the Yorkshire Collaborating Centre for Health Services Research. Leeds: Nuffield Institute for Health 57p.				criteria of relevance, outcome and design. 243/300 papers were rejected. The quality of the evidence was generally poor with a lack of properly controlled trials. Direct evidence of the relationship between distance and mortality or morbidity was rare, although 2 studies of cancer patients indicated that outcomes are not affected by distance. Data for other outcome measures such as QOL was lacking The author concludes that there is not widespread agreement about what constitutes specialist care. Specialist care should not be equated with centralised care.	*vulnerable to confounding. There were no RCTs. Useful review of all literature to 1995.*		++
4. Kearney P (2003) *The burden of travel in paediatric oncology*. Cork: University College, Cork 131p. Unpublished thesis.	22 parents (5 fathers and 17 mothers) of children with cancer	Investigation of the experience of travelling to paediatric oncology centres and to identify the burdens of travel. Analyses of transcripts using a grounded theory approach		There were 2 paediatric units associated with centres that had different policies of decentralising care. Four focus group interviews performed by an experienced sociologist were recorded. The experience of having a child with cancer was very intense for both groups. Travel was not an issue for the decentralised group. The parents using a more centralised service identified several burdens of travel:- • Exhaustion with uncertainty of travel. arrangements &		Thesis	3/4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				• huge round trips • Experience of travelling with sick child was frightening. • Serious financial problems • Family disruption was more severe for the centralised service • Sibling neglect The author concludes that the recommendation of 1 paediatric oncology centre /5 million population remains suitable in densely populated areas. Maximum devolution of care reduces the burden of travel on families. There is a need for appropriate standards of staffing and accommodation for units with maximal devolution of care			
5. Payne S, Jarrett N, Jeffs D (2000) The impact of travel on cancer patients' experiences of treatment: a literature review. *European Journal of Cancer Care* 9:197-203.	All cancer patients	Identification of evidence on impact of travel on cancer patients' experiences of treatment.	Patients views on the burden of travel for cancer treatment.	296 papers were identified. 11 papers, from 6 countries, fulfilled the inclusion criteria. Most studies had methodological flaws. The evidence that travel distance and difficulty increases psychological distress and reduces compliance with treatment and take up of treatment is inconclusive. The author concludes that the literature is contradictory but travel for cancer treatment appears an inconvenience for	*Well designed and reported review with adequate description of methodology. No specific & A studies*	Literature review	3/4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				patients and may be perceived as a barrier to compliance.			

Q.3 IS THERE EVIDENCE FOR AN ASSOCIATION BETWEEN THE NUMBER OF CASES OF CHILD AND YOUNG PEOPLE WITH CANCER SEEN AND OUTCOME?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/QUALITY
1. Halm EA, Lee C, Chassin MR (2002) Is volume related to outcome in health care? A systematic review and methodologic critique of the literature. *Annals of Internal Medicine* 137:511-520.	All types of health care	Organisation. To review systematically the research evidence linking volume and outcome in health care	Health outcomes e.g. death, stroke or clinical complication	135 studies met the inclusion criteria**.** No C & A cancer studies. Overall 71% of all studies of hospital volume and 69% of studies of physician volume reported statistically significant associations between higher volumes and better outcomes. Differences in case mix and processes of care between high and low volume providers may explain part of the observed relationship between volume and outcome. The authors discuss the methodological problems with some of the primary studies and emphasise about making policy decisions based on the evidence. .	*Studies were too heterogenous to combine for metaanalysis but were combined according to procedure. Appropriate methods used for analysis of volume/outcome via pooling were used. Possibility of publication bias not formally tested*	Systematic review	2^{++} +
2. Hillner BE, Smith TJ, Desch CE (2000) Hospital and physician volume or specialization and outcomes in cancer treatment: importance in quality of cancer care. *Journal of Clinical Oncology* 18:2327-2340.	All types of cancer care	Evidence to support that hospital or physician volume or specialty affects outcome of cancer care.		A consistent literature was identified that support a volume-outcome relationship for cancers treated with technically complex surgical procedures. These studies identified 30 day mortality. And used the hospital as the unit of analysis. For cancer treated with low-risk surgery there were fewer studies and	*Search limited to Medline 1988-1999*	Systematic review	2^{++} +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/QUALITY
				there was only an association for colorectal and breast cancer.			
3. Stiller CA, Draper GJ (1989) Treatment centre size, entry to trials, and survival in acute lymphoblastic leukaemia. *Archives of Disease in Childhood* 64:657-661.				**SEE TABLE 1**			
4. Stiller CA, Eatock, EM (1999) Patterns of care and survival for children with acute lymphoblastic leukaemia diagnosed between 1980 and 1994. *Archives of Disease in Childhood* 81:202-208.				**SEE TABLE 1**			
5. Stiller CA, Benjamin S, Cartwright RA et al. (1999) Patterns of care and survival for adolescents and young adults with acute leukaemia - a population-based study. *British Journal of Cancer* 79:658-665.				**SEE TABLE 1**			
6. Stiller CA (1994) Centralised treatment, entry to trials and survival. *British Journal of Cancer* 70:352-362.				**SEE TABLE 1**			

Q.4 IS THERE EVIDENCE THAT SHARED CARE IMPROVES PATIENT OUTCOMES?

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Birmingham Children's Hospital NHS Trust and its Catchment Population (2002) *Standards for the care of children and young people with cancer.* Birmingham: Birmingham Children's Hospital NHS Trust and its Catchment Population 48p.	Children and young people with cancer	Agreement on standards of care for an acute trust and its designated shared care units	-	The standards are based on the national cancer standards and developed by a sub-group of the Supra-network paediatric Tumour Advisory Group	*The standards draw heavily from those developed for the London. South Eastern and South Western Regions. Not really evidence for place of treatment setting, but useful background document .for auditable standards*	Report	4 +
2. Campbell NC, Ritchie LD, Cassidy J et al. (1999) Systematic review of cancer treatment programmes in remote and rural areas. *British Journal of Cancer* 80:1275-1280.	All cancers	Identification of problems and effectiveness of oncology service provision in remote and rural areas	Patient and physician satisfaction. Survival	The authors concluded that there was some evidence to support the safety of shared outreach care. Such care could make specialist care more accessible to outlying patients.	*. All studies were small and had methodological problems. Little evidence of relevance to the question*	Systematic review	2^{++} +
3. Edwards J, Hooker L Caring for the child with cancer within a model of shared care unpublished	Children with cancer	Description of shared care		The authors conclude that; • Centralised care has improved outcomes, but can have negative consequences for families	*Useful review chapter*	Expert opinion	4 +

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				and is expensive for service providers • Successful shared care requires good organisation by all parties involved. • There is a need for high quality research to evaluate outcomes			
4. Genefke J, Holm T, Melchiorsen H (2003) *Working Paper 2003 – 9: Multipartner collaboration with many partners. Shared care: management fad – or tool for the few*. Aarhus: University of Aarhus 8p.	All patients, receiving shared care.	Evaluation of shared care programmes	Clinical effectiveness	Only 2 oncology studies included in the review. The authors distinguish between 3 types of treatment:- • Mono-care, where 1 medical entity covers all the patients medical need with respect to the disease • Distributed care where more that one medical entity is needed but where each only performs and bears responsibility for part of the treatment • Shared care, where 2 parties each take part in the treatment and bear common responsibility	*Good paper on theory and definition of shared care with review of published studies. The paper discusses the potential problems of shared care and how to address them. Makes the point that a lot of what is called shared care is in fact distributed care.*	Report	4 +
5. Jones CEM (1998) *Shared care for children with acute lymphoblastic leukaemia: the parent's perspective*. London: Roehampton Institute. Thesis	9 parents of children with ALL in 2 focus groups	Analysis using grounded theory of parents views on shared care		The themes which arose form the interviews were convenience, variability of shared care services and the role of the parent. Shared care promoted the security of the whole family	*Small sample size; selection bias. Researcher bias (employed by regional centre)*	Qualitative	3/4 +/-
6. Nielsen JD, Palshof T, Mainz J et al. (2003) Randomised controlled trial of a shared care programme for newly referred cancer patients: bridging the gap between general	248 cancer patients, > 18 years	To determine the effect of a shared care programme on the attitudes of newly referred cancer patients. The shared care programme included transfer of	QOL & performance status. Patients attitudes to healthcare system and GPs.	Shared care is defined as:- when the responsibility for the health care of the patient is shared between individuals or teams who are part of separate organisations, or where substantial organisational boundaries	*Well designed and described study. Non-blinded. Power of study discussed and adequate to detect a difference. Appropriate*	Randomised controlled trial	1^- ++

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
practice and hospital. *Quality & Safety in Health Care* 12:263-272.		knowledge from the oncologist to the GP, improved communication between the parties and active patient involvement.		exist. It implies personal communication and organised transfer of knowledge from hospital doctors to GPs and patient involvement. 48 patients dropped out of study. (24 from each group); 17 had died. The shared care programme had a positive effect on patient evaluation of cooperation between primary 7 secondary care. The effect was particularly significant (p==0.003)in young (18-49) men. There were no differences in QOL.	*statistical analyses. Good discussion about limitations of study – power, bias etc.. Only relevant to upper age group patients for question..*		
7. Sepion B (2004) Shared care. In: Gibson F, Soanes L, Sepion B, editors. *Perspectives in paediatric oncology nursing.* London: Whurr Publishers, p176-191.	6 parents of children with cancer	Assessment using phenomenological method of parents feelings about shared care		The major issues identified were:- • Poorer facilities in the DGH compared with cancer centre • Parents felt isolated both physically and emotionally • Conflict between the knowledge base of some parents and the healthcare professionals caring for them in the DGH, • Communication was identified as an important problem	*Phenomenology is often criticised as a technique but is often used in nursing to explore issues that are different to every human being.* *Small study of limited relevance to question*	Qualitative study	3/4 +/-
8. United Kingdom Children's Cancer Study Group. Standard Operating Procedure (2004) *Shared care*. Leicester: United	All children with cancer	Guidance for shared care arrangements		Describes objectives, procedures with the levels of treatment		Guidance	4 +

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Kingdom Children's Cancer Study Group 3p.							
9. West Midlands Cancer Peer Review (2003) *Shared care of children and young people with cancer: overview report*. Birmingham: West Midlands Cancer Peer Review Team 24p.	Children and young people with cancer	Peer review visits to units that share care with Birmingham Children's Hospital NHS Trust.	Adherence to standards.	Adherence to standards was generally over 50% for all units apart from 1 (~47%). The authors found that there • did not appear to be clear understanding of the type of care that a SCU is able to offer. • there was not a robust commissioning framework for care of children with cancer The authors propose the following principles for the future organisation of care for children and young people with cancer • Equity of access to high quality services. • A 'critical mass' of staff so that the quality of service does not vary unreasonably when staff are away. • Care as near to home as possible. • Support from Birmingham Children's Hospital for care delivered in other hospitals and in the community. • Access to specialist treatment and care from Birmingham Children's Hospital when this is needed. • Commissioning arrangements that support the agreed model of care. Elements that make up service for care of children & young people with cancer	*Useful document for possible service configuration guidance.*	Report	4 ++

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				Community-based care • Blood counts and blood product support • Central venous line care • Cytarabine administration • Nasogastric tube feeding and support **Hospital-based care** • Diagnosis and initiation of treatment for 'low risk' acute lymphoblastic leukaemia • Care for patients on an agreed list of regimes • In patient chemotherapy for an agreed list of regimes • Out-patient chemotherapy for an agreed list of regimes • Treatment of febrile neutropenia **Specialist care** • Diagnosis and initiation of treatment (except for 'low risk' acute lymphoblastic leukaemia) • Care for patients with the rarest cancers (including chemotherapy) • Care for complex cases (including chemotherapy) • Provision of training, including for shared care unit staff • Advice and support to shared care units, including a consultant clinic in each shared care unit and BCH Macmillan			

STUDY	POPULATION	INTERVENTION/ AIMS	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				Nurse attendance at some shared care unit multi-disciplinary teams.			

COMMUNICATION / INFORMATION

The Questions:

What is the evidence for effective means of communication?

What is the evidence for effective means of information giving?

Nature of the evidence

1 RCT of fair to poor quality
4 systematic reviews, 3 of good quality, 1 fair to poor quality
2 qualitative studies of fair to poor quality
1 guideline of fair quality
1 policy/expert opinion of fair quality
2 surveys

Summary of the supporting evidence for the recommendations

- The results of one RCT indicate that training courses to improve communication are effective [3].
- The evidence from one systematic review indicated that communication skills training programmes are effective [4].
- The evidence from one systematic review indicated that communication skills training programmes are effective and that tailored information is the most effective [7].
- There was good quality evidence from one systematic review on the best use of communication methods and information exchange in cancer [9].
- One systematic review of RCTs indicated that the use of effective mechanisms such as audio visual aids improve patient outcomes [6].
- One systematic review of randomised and non randomised trials indicated that the evidence was poor on interventions to enhance communication involving child and adolescent patients. Some evidence for tailored information [10].
- One qualitative study indicated that patients recall about the information they were given by the GP was poor [2].
- One qualitative study indicated that parents feel that the executive role they have to adopt *vis a vis* their children is a problem for effective communication [11].
- One guideline specifically for paediatric cancer patients provides good recommendations for communication of diagnosis [5].
- One policy document details basic principles for communication in cancer services [8].

- There was evidence from a survey of teenagers that approximately 50% felt that information was not suitable for their age group. (Appendix E)
- A survey of very young children showed that they are able to express their feelings when staff skilled in communicating with them are employed. (Appendix D)

There is very little high quality evidence to indicate the optimum service provision for children and young people with cancer who have very specific information requirements.

WHAT IS THE EVIDENCE FOR EFFECTIVE MEANS OF COMMUNICATION AND INFORMATION GIVING?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Dixon-Woods M, Findlay M, Young B et al. (2001) Parents' accounts of obtaining a diagnosis of childhood cancer. *Lancet* 357:670-4.	20 parents whose children (aged 4-18 yrs) had a confirmed diagnosis of cancer (leukaemia) or brain or solid tumours.	Semi-structured interviews	The feelings of parents about the diagnosis process. Whether the narratives had implications for early diagnosis and referral.	Response rate 95%. There was good consistency between parent's accounts and the medical records. Data were analysed by the constant comparison method. The signs and symptoms of younger children were first noticed by parents. Parents of older children and adolescents often had to be told of problem. Early symptoms often vague. There were disputes in 7/20 families with the GP.	*The study is limited to 1 paediatric oncology unit. There were few examples of the types of tumour that can be prone to delays in diagnosis. Communication or information issues not addressed.*	Qualitative	3 +/-
2. Eiser C, Parkyn T, Havermans T et al. (1994) Parents' recall on the diagnosis of cancer in their child. *Psycho-oncology* 3:197-203.	30 families with a child diagnosed with cancer (ALL, lymphomas, solid tumours and brain tumours.	Determination of information parents recall being given on diagnosis and assessment of information they would have liked.		In 20 cases mothers were told by the GP or local hospital before they received fuller information at the oncology unit or regional centre. 2/20 mothers reported that this initial explanation was incomplete. No real criticism of the way information was given at the oncology unit or regional centre. Policy in both centres was that children> 8 were told of diagnosis.	*Some relevance to question. Insufficient details given for appraisal*	Qualitative	3 +/-
3. Fallowfield L, Jenkins V, Farewell V et al. (2002) Efficacy of a Cancer Research UK communication skills training model for oncologists: a randomised controlled	160 oncologists from 34 cancer centres in the UK, 72% male and of consultant grade (61%) 39% SpR. 640 patients with cancer (61%	Videotaped consultations of consultation sin cancer patients	Behavioural changes	5/160 doctors withdrew from study and were replaced. Data was presented for 640 patients. The authors conclude that:- Communication problems of senior doctors working in oncology are not resolved by	*Method of randomisation or allocation concealment not given. Complete blinding was not always possible. Outcomes not*	Randomised controlled trial.	!- +/-

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
trial. *Lancet* 359:650-656.	female; 48% aged between 51-70 years). 5% < 30 years and 22% > 70 years. 43% in palliative stage of disease.			time and clinical experience. Training courses significantly improve key communication skills.	*properly defined*		
4. Fellowes D, Wilkinson S, Moore P (2004) Communication skills training for health care professionals working with cancer patients, their families and/or carers. *The Cochrane Database of Systematic Reviews* Issue 2.	Qualified health professionals within all hospital, hospice and ambulatory care settings, working in cancer care.	Studies in which the intervention group has communication training	Changes in behaviour skills	3/2824 references were included. Two/3 trials concerning communication skills training programmes did have a positive effect on the communications behaviour of experienced nurses and doctors working in cancer care. The authors conclude that further research into the long-term efficacy of communication skills training is needed	*Good quality review with good description of methodology. There is some evidence to suggest labour intensive communication skills training can have a beneficial effect on behaviour change in professionals working with cancer patients.*	Systematic review	2^{++} ++
5. Masera G, Chesler MA, Jankovic M et al. (1997) SIOP Working Committee on psychosocial issues in paediatric oncology: guidelines for communication of the diagnosis. *Medical and Paediatric Oncology* 28:382-5.	Paediatric oncology	Development of guidelines		**Summary of principles for communicating the diagnosis** 1. Establish a protocol for communication. 2. Communicate immediately at diagnosis and follow up later. 3. Communicate in a private and comfortable space. 4. Communicate with both parents, and other family members if desired. 5. Hold a separate session with the child. 6. Solicit questions from the parents and child. 7. Communicate in ways that	*Not evidence based. Based on consensus.*	Guidelines	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				are sensitive to cultural differences. 8. Share information about the diagnosis and the plan for cure. 9. Share information on lifestyle and psychosocial issues. 10. Encourage the entire family to talk together.			
6. McPherson CJ, Higginson IJ, Hearn J (2001) Effective methods of giving information in cancer: a systematic literature review of randomized controlled trials. *Journal of Public Health Medicine* 23:227-234.	Cancer patients, their families and carers. Majority of studies included newly diagnosed patients.	Review of published literature	Knowledge, recall, symptom management, satisfaction, health care utilisation and affective states.	10/1120 studies met the inclusion criteria. Interventions ranged from written information to audiotapes, audiovisual aids and interactive medium. The evidence indicated that the interventions had a positive effect on patient outcomes.	*Few patients within the C & A age range. Some relevance to question*	Systematic review	2^{++} +
7. NHS Centre for Reviews and Dissemination (2000) Informing, communicating and sharing decisions with people who have cancer. *Effective Health Care* 6:1-8.	All patients with cancer	Review of published literature	Effective methods for informing and communicating and sharing decisions with people who have cancer	• Limited trial data suggest that training programmes in communication for healthcare staff are beneficial for patients, with cancer • Informing patients- there is evidence (44 articles, data quality poor) to indicate that tailored information best meets the need s of cancer patients.	*No studies specific for C & A cancer. Some relevance to question*	Systematic review	2^{++} +/-
8. NHS Modernisation Agency, Cancer Services Collaborative Improvement	All cancers	-	-	Document setting out basic principles in communication in cancer services.	*Useful for general principles of communication across all sectors. Not referenced.*	Policy	4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
Partnership (2004) *Improving communication in cancer care*. London: NHS Modernisation Agency 22p.					*Fax pack, safe haven policy.*		
9. National Institute for Clinical Excellence (2004) *Guidance on cancer services - improving supportive and palliative care for adults with cancer – the manual*. London: National Institute of Clinical Excellence. Available from: www.nice.org.uk	All cancers	Review of literature on communication and information giving.		There is good quality evidence available on the effective use of communication and information exchange in cancer.	*Good review of evidence, but very little specific to C & A age range.*	Systematic review	2^{++} ++
10. Scott JT, Harmsen M, Prictor MJ et al. (2003) Interventions for improving communication with children and adolescents about their cancer. *The Cochrane Database of Systematic Reviews* Issue 3.	Children and adolescents with cancer	Randomised and non-randomised controlled trials and before and after studies that evaluated the effects of interventions to improve communication with children and/or adolescents about their cancer, its treatment and their implications	C & A's knowledge about cancer and its treatment. Psychological, social & behavioural outcomes and social activities Physical health outcomes	The reviewers conclude that interventions to enhance communication involving C & A with cancer have not been widely or rigorously assessed. The weak evidence that exists suggests that C & A with cancer may derive some benefit from specific information giving programmes & from interventions that aim to facilitate their reintegration into school and socially. Nine studies met the criteria for inclusion. They were diverse in terms of interventions, study designs and outcomes. • One study of a computer assisted education programme reported	*Adequate description of inclusion and exclusion criteria for studies and outcomes. Search terms and databases searched comprehensive. Updated search in January 2003.*	Systematic review	1^{++} ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				improvements in knowledge & understanding about blood counts and cancer symptoms • One study of a CD-ROM about leukaemia reported an improvement in children's feelings of control over their health • One study of art therapy during painful procedures reported an increase in collaborative behaviour in children • 2/2 studies of school reintegration programmes reported improvements in some aspects of psychosocial well being			
11. Young B, Dixon-Woods M, Windridge K et al. (2003) Managing communication with young people who have a potentially life threatening chronic illness: qualitative study of patients and parents. *British Medical Journal* 326:305-308.	Young people with cancer and their parents. 13 families comprising 19 parents and 13 patients aged 8-17 yrs. 1 paediatric oncology unit, UK.	Semi structured interviews analysed using constant comparison method.	-	13/20 families agreed to be interviewed. Most parents described acting in an executive-like capacity managing what their children were told about their illness. The diagnosis was usually told to the parent first without the child being present. This executive role both facilitates and constrains communication with young people. Some young people feel marginalised in consultations	*Well designed and reported study*	Qualitative study	3 +/-

RESEARCH

The Questions:

1. Do children and young people with cancer have equal access to entry into clinical trials?

2. Does inclusion in a clinical trial improve outcomes for children and young people with cancer?

Nature of the evidence

Q.1

2 retrospective analyses, one of fair quality, 1 fair to poor quality
2 expert opinions one of good quality and one fair to poor quality

Q.2

1 prospective cohort study of fair quality
1 literature review of good quality
1 literature review of selected cancer trials (8 trials in C & A age range), of fair quality
2 historical case series of good quality
1 expert opinion of fair to poor quality

Summary of the supporting evidence for the recommendations

Q.1

- One large retrospective analysis indicates that adolescents do not have equal access to clinical trials [6].
- The evidence from one retrospective analysis indicates that adolescents are more likely to enter a clinical trial if treated at a paediatric centre [8].
- It is concluded in one expert opinion that recruitment to clinical trials is low in children with some types of tumour e.g. brain and in adolescents [1]. Another expert opinion emphasises that adolescents do not have equal access to clinical trials [7].

Q.2

- The evidence from one prospective cohort study indicates that 3 year survival is improved in children with nephroblastoma entered into a clinical trial [5].
- There was no difference in survival in adolescents with ALL entered into a clinical trial in one historical case series [11], but in children with ALL survival was improved in those treated within a clinical trial [10].
- A statistically non significant increase in survival was noted in one historical case series of children with ALL entered into clinical trials [14].
- One review of the literature indicated that for children with ALL clinical trial entry resulted in a significantly higher survival but there was no effect at high volume centres. Trial effect however, was demonstrated in children with Wilm's tumours [13].
- An investigative literature review compared trial subjects with non-trial subjects to investigate the possible confounding effect. Eight paediatric oncology trials were included and the results indicated that trial entry produced improvement in outcomes in 1/8 paediatric cancer trials after correction for confounding factors [9].

It is accepted that, while there is currently insufficient high quality evidence to definitely conclude that entry into a clinical trial improves outcomes in children and young people with cancer, patients should be encouraged to enrol in trials. There is observational evidence to indicate that adolescents and young people do not have as good access to clinical trials as children.

Q1. DO CHILDREN AND YOUNG PEOPLE HAVE EQUAL ACCESS TO ENTRY INTO CLINICAL TRIALS?

Q2. DOES INCLUSION IN A CLINICAL TRIAL IMPROVE OUTCOMES FOR CHILDREN AND YOUNG PEOPLE WITH CANCER?

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
1. Ablett S, Pinkerton CR (2003) Recruiting children into cancer trials - role of the United Kingdom Children's Cancer Study Group (UKCCSG). *British Journal of Cancer* 88:1661-1665.	Children with cancer ≤ age 15 years			Author concludes:- • Referral into specialist centres for children with cancer & recruitment into trials is very high and exceeds the targets currently being set for the NCRN for adult cancer trials in the UK. • Recruitment is low in children with e.g. brain tumours and adolescents. • There is also geographical variation in centre facilities which may lead to differences in recruitment. • Issues remain about randomisation rates to certain studies compared with European centres.		Expert opinion	4 ++
2. Benjamin S, Kroll ME, Cartwright RA et al. (2000) Haematologists approaches to the management of adolescents and young adults with acute leukaemia. *British Journal of Haematology* 111:1045-50.	Adolescents and young adults (15-29 years) with acute leukaemia. UK	Questionnaire survey of haematologists from 121 hospitals, entering patients into clinical trials	Types of hospital treating the patients; haematologists perceived practice for trial entry and reasons for non entry.	There was a 79% response rate (96 hospitals) 82% of haematologists stated that they entered patients 'always' or 'whenever possible' for AML and 76% for ALL but actual entry rates were 46% of 239 AMI patients and 36% of 182 ALL patients. The respondents gave 3 main types of reason for the failure to enter patients into national clinical trials	*The data obtained were linked to the MRC trials data to determine the actual proportion of patients actually treated in MRC leukaemia trials in the 5 years prior to the questionnaire. Adequate response rate; well described study. No discussion of study*	Questionnaire survey	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				• Preference for non trial protocols • Administrative reasons – ethical approval, cost, extra workload • Participation in local trials or non MRC trials	*limitations.*		
3. Boissel N, Auclerc MF, Lheritier V et al. (2003) Should adolescents with acute lymphoblastic leukaemia be treated as old children or young adults? Comparison of the French FRALLE-93 and LALA-94 trials. *Journal of Clinical Oncol*ogy 21:774-80.	Adolescents with ALL aged 15 to 20 years. 77 treated in paediatric FRALLE-93 trial (1993 to 1999) 107 treated in adult LALA-94 trial (1994 to 2000), 100 with complete follow-up were analysed. Treatment groups were similar apart from median age (15.9 years for FRALLE versus 17.9 years for LALA)	To compare treatment of adolescents with acute lymphocytic leukaemia (ALL) using paediatric and adult protocols. Adolescents diagnosed by paediatricians are treated in paediatric trials. Adolescents diagnosed by GPs or internists are treated in adult departments. Treatment regimens for both trials were reported. Higher doses of all major ALL drugs were given in FRALLE-93 within shorter time period compared with LALA-94. Country: France.	Complete remission (CR); Disease free survival (DFS); event free survival (EFS); relapse free survival (RFS). Predictors of EFS at 5 years	. Treatment with the paediatric protocol (FRALLE-93) significantly improved CR rates compared with the adult protocol (LALA-94) especially for patients with BCP-ALL; CR for all: 94% with FRALLE versus 83% with LALA, P = 0.04. CR for BCP-ALL: 98% with FRALLE versus 81% with LALA, P = 0.002. Treatment with the paediatric protocol (FRALLE-93) significantly improved EFS and DFS at 5 years compared with the adult protocol; EFS: 67% with FRALLE versus 41% with LALA, P < 0.0001. DFS: 72% with FRALLE versus 49% with LALA, P= 0.0004. The only prognostic factors for EFS were WBC (P < 0.0001) and the trials (P = 0.004). The authors concluded that	*Treatment groups were not randomly allocated to treatment. The authors did compared baseline characteristics between groups and found groups to be similar*	Retrospective cohort with control group.	2⁻ +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				adolescents with ALL should be treated with intensive paediatric protocols.			
4. Estlin EJ, Ablett S (2001) Paediatric update: practicalities and ethics of running clinical trials in paediatric oncology - the UK experience. *European Journal of Cancer* 37:1394-1401.	UK authors	Clinical trials		The authors conclude:- • Small patient numbers • Increasing subdivision by clinical and biological prognostic factors • Increasing complexity of trials • Collection of large amounts of patient data often justified due to rarity of disease and lack of knowledge about long term effects of the tumour and its treatment. • Need for explicit consent, age specific information sheets and good clinical practice guidelines. Need realistic expectations for toxicity and benefit (e.g. 7.9-10% objective response rate, 0.6-0.7% drug related toxicity		Expert review	4 +/-
5. Lennox EL, Stiller CA, Jones PH et al. (1979) Nephroblastoma : treatment during 1970-73 and the effect on survival of inclusion in the first MRC trail. *British Medical Journal* 2:567-69.	UK. 313 children diagnosed with nephroblastoma in 1970.	98 children were entered into the MRC nephroblastoma study between October 1970 – December 1973. 288/313 (92%) children had a nephrectomy, 248 (79%) received a course of RT and 267 (85%) were given at least 4 days CT.	3 year survival	The 3 year survival rate was 58%. The rate in the children entered into the trial (77%) was significantly better than that among children who were eligible for the trial but not included (58%) p<0.01. This result was more pronounced when allowance was made for the distribution of age and tumour stage (p<0.001).	*Trial participants and non trial controls were not matched for prognosis variables but statistical adjustment was made. Recruitment rates not given. Appropriate use of statistics*	Prospective cohort study	2^{++} +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
6. Liu L, Krailo M, Reaman GH et al (2003) Childhood cancer patients' access to cooperative group cancer programs: a population-based study. *Cancer* 97:1339-1345.	10,108 children <20yrs old with cancer, identified by the 11 SEER registries between 1992-1997.	Analyses of Children's Oncology Group (COG) to determine whether it would serve as a resource for identifying children with cancer		Not all children are registered by the cooperative groups. The annual age-adjusted registration rate (AARR) was 71% for children <15yrs, 24% for adolescents 15-19 years, and 57% for children <20 years. Registration rates varied by geographic region and were higher among children with advanced disease. Registration rates were highest for children (<15yrs) with leukaemia (84%), hepatic tumours (82%)& renal tumours (80%) and were lowest for carcinoma (26%) and retinoblastoma (30%)	*US. Confirms UK studies of low registration for older children and adolescents and differences with tumour type.*	Retrospective analysis	3/4 +/-
7. McTiernan A (2003) Issues surrounding the participation of adolescents with cancer in clinical trials in the UK. *European Journal of Cancer Care* 12:233-239.	Adolescents (15 - 19?) with cancer	Consideration of issues about participation of adolescents in trials		Author concludes:- • Clinical trials imperative to improve treatment and prognosis. • Adolescents do not have equal access to trials due to fragmentation of care between paediatric and adult settings • Compliance is less in adolescents and needs research.	*Age definition of adolescents not given*	Expert opinion/overview	4 +/-
8. Mitchell AE, Scarcella DL, Rigutto GL et al. (2004) Cancer in adolescents and young adults treatment and outcome in Victoria. *Medical Journal of Australia* 180:59-62.	All adolescents & young adults, aged 10-24 yrs. Diagnosed with cancer (leukaemia, lymphoma, germ cell tumours, brain tumours and bone tumours) between 1992-1996.	Questionnaire survey of referring physician.	Treatment regimen. 5 year survival. Compliance with protocol	Questionnaires completed for 576/665 eligible subjects (87%). Recruitment into trials decreased with increasing age. Adolescents aged 10-19 yrs. were more likely to be recruited into a trial if treated at a paediatric hospital (38% and 3% respectively; P<0.005; 95% CI for difference 25%-41%). Only 4% of young adults aged 20-24 years were treated within clinical trials. There was no significant difference in overall 5 year survival between the three	*Australian study of relevance to UK situation. Well designed and described study. Appropriate use of statistics. Authors discuss limitations of small numbers in each age group to*	Retrospective analysis	3/4 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
				age groups (10-15, 16-19 & 20-24 yrs). Brain tumours had the lowest trial entry. 1% of patients did not complete treatment	*make conclusions about differences in survival. Examination of figures for 5 year survival suggest differences in survival between age groups for brain and bone tumours.*		
9. Peppercorn JM, Weeks JC, Cook EF (2004) Comparison of outcomes in cancer patients treated within and outside clinical trials: conceptual framework and structured review. *Lancet* 363:263.	Cancer patients enrolled and not enrolled in clinical trials.	Literature review of studies comparing trial with non trial subjects. Reviewed 8 major paediatric oncology trials.		21 comparisons used retrospective cohort designs. 14 comparisons provided some evidence that trial patients have improved outcomes. A third of the studies were restricted to children. Only 8 comparisons restricted non-trial patients to those meeting trial eligibility criteria. Of these three noted better outcomes in trial patients than in non-trial patients; one of these (Lennox et al – nephroblastoma) was in a paediatric population. Strategies to control for confounding were frequently inadequate. Positive studies were more likely than negative studies to involve children, patients treated before 1986.	*Authors discuss limitations of search strategy and standardisation of quality assessment standards*	Literature review	3/4 +
10. Stiller CA, Eatock EM (1999) Patterns of care and survival for children with acute lymphoblastic leukaemia diagnosed	4998 children aged between 0 and 14 years	Effect of patterns of care	Survival. Hospitals were classified as:- • Mean annual number of new patients aged 15-	5 year survival improved from 67% in 1980-84 to 81% in 1990-94. The authors conclude that survival did not vary systematically with hospital	*Large well designed multicentre study.*	Historical case series	3 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
between 1980 and 1994. *Archives of Disease in Childhood* 81:202-208.			29 with ALL & AML • As teaching or non-teaching hospitals	case load or between paediatric oncology centres and other hospitals. Trial entry had an effect on survival.			
11. Stiller CA, Draper GJ (1989) Treatment centre size, entry to trials and survival in acute lymphoblastic leukaemia. *Archives of Disease in Childhood* 64:657-61.	Children with acute non-lymphoblastic leukaemia, Hodgkin's disease, non HL, neuroblastoma, Wilms' tumour, osteosarcoma, Ewing's tumour & rhabdomyosarcoma.	Comparison of survival rates between UKCCSG patients and non-UKCCSG patients	Survival	Children with acute non-lymphoblastic leukaemia, non HL, Ewing's tumour, rhabdomyosarcoma and osteosarcoma treated at paediatric oncology centres had significantly (p= < 0.05) higher survival rates than those treated elsewhere.	*Relevant to question*	Historical case series	3 +
12. Stiller CAS, Benjamin RA, Cartwright JV et al. (1999) Patterns of care and survival for adolescents and young adults with acute leukaemia--a population-based study. *British Journal of Cancer* 79:658-665.	879 patients, aged 15-29 yrs. with acute leukaemia during 1984-1994.	Effect of patterns of care	Survival. Hospitals were classified as:- • Mean annual number of new patients aged 15-29 with ALL & AML As teaching or non-teaching hospitals	For ALL actuarial survival rates were 43% at 5 years after diagnosis and 37% at 10 years. Survival improved significantly between 1984-88 and 1989-94 for those aged 15-19 at diagnosis. Entry into trials had no effect on survival. Survival rates were similar at teaching and non-teaching hospitals & at hospitals treating different numbers of study patients per year. For AML survival rates 42% at 5 yrs after diagnosis & 395 at 10 years. Survival did not vary with category of hospital. Trial effect was equivocal.	Survival. Hospitals were classified as:- • Mean annual number of new patients aged 15-29 with ALL & AML As teaching or non-teaching hospitals	Historical case series	3 ++

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
13. Stiller CA (1994) Centralised treatment, entry to trials and survival. *British Journal of Cancer* 70:352-62.	All cancer patients	Review of published literature from 1984-1993 (Medline, Embase) on patterns of care.	Survival	For children with ALL there was a significant trend towards higher survival rates in children being treated at high volume centres. Children entered into MRC trials had a significantly higher survival rate. Trial entry had little effect on survival at high volume centres. For children with A non LL entry to a trial and treatment at a teaching hospital were both associated with a higher survival rate. In 2 studies of children with retinoblastoma survival rate was highest at the national referral centre. For children with Wilms' tumour survival rates were highest for those included in MRC trials than those who were eligible but not included. Patients, who had surgery at a specialist centre had higher survival rates.	*The papers are not critically appraised. .The author discusses the possible sources of bias. Other possible outcome measures are discussed*	Literature review	3/4 +
14. Stupnicki A, von der Weid N, Imbach P et al (1995) Incidence of childhood acute lymphoblastic leukemia (ALL) and population-based treatment results in Switzerland: experiences with 507 study and 149 nonstudy patients. 15. *Medical &*	656 children < 15 years with acute lymphoblastic leukaemia entered into protocols (1980-1983; 1984-1987; 1988-1991)		Number entered into trials	507/656 were entered into trials and 149 were not. The authors conclude that the true incidence of ALL in Switzerland in children < 15 is higher than that reported. The rate of survival at 4 years for both trial and non trial patients increased but the increase was greater in trial included patients, although this difference was not	*Confounding not discussed adequately. Study period 11 years.*	Historical case series	3 +

STUDY	POPULATION	INTERVENTION	OUTCOMES	RESULTS	COMMENTS *Italics= reviewers comments*	DESIGN	EVIDENCE LEVEL/ QUALITY
*Pediatric Oncol*ogy 25:79-83.				statistically significant			

APPENDIX A

Main Child and Young People with Cancer Search Strategy

Relevant studies were identified using the search strategy for Medline shown below:-

Medline, Embase (a modified search strategy with different index terms was used for Embase) and the Cochrane Library were searched as core databases - Cinahl, British Nursing Index, Psychinfo and Amed were searched, if relevant to the subject of the search.

1. exp Adolescent/ or exp Child/ or exp Child, preschool/ or exp Infant/ or exp Infant, newborn/ or exp Minors/ or exp Pediatrics/
2. (child$ or paediatric$ or pediatric$ or perinat$ or neonat$ or newborn$ or infan$ or baby or babies or toddler$ or boy$ or girl$ or kid$1 or schoolage$ or juvenil$ or young).mp.
3. (underage$ or teen$ or youth$ or pubescen$ or adolescen$).tw. or (infan$ or child$ or pediatric$ or paediatric$ or adolescen$).jw.
4. or/1-3
5. exp neoplasms/
6. (cancer$ or neoplas$ or tumo?r$ or oncol$).tw.
7. leuk?emi$.tw.
8. lymphoma$.tw.
9. (Hodgkin$ or non Hodgkin$).tw.
10. reticulosarcoma$.tw.
11. lymphosarcoma$.tw.
12. granuloma$.tw.
13. astrocytoma$.tw.
14. glioma$.tw.
15. glioblastoma$.tw.
16. medulloblastoma$.tw.

17. ependymoma$.tw.
18. craniopharyngioma$.tw.
19. neuroblastoma$.tw.
20. ganglioneuroblastoma$.tw.
21. meningioma$.tw.
22. neuroepithelioma$.tw.
23. neurilemmoma$.tw.
24. neuroma$.tw.
25. oligodendroglioma$.tw.
26. pineoblastoma$.tw.
27. primitive neuroectodermal tumo?r$.tw.
28. pnet.tw.
29. retinoblastoma$.tw.
30. (wilm$ or nephroblastoma$ or nephroma$).tw.
31. (hepatoblastoma$ or hepatoma$).tw.
32. (renal adj (carcinoma$ or tumo?r$)).tw.
33. sarcoma$.tw.
34. angiosarcoma$.tw.
35. dermatofibrosarcoma$.tw.
36. ewing$.tw.
37. (askin$1 adj1 tumo?r$).tw.
38. osteosarcoma$.tw.
39. (haemangiopericytoma$ or hemangiopericytoma$).tw.
40. (haemangiosarcoma$ or hemangiosarcoma$).tw.
41. (haemangioendothelioma$ or hemangioendothelioma$).tw.
42. oligodendroglioma$.tw.
43. histiocytoma$.tw.
44. rhabdomyosarcoma$.tw.
45. rhabdosarcoma$.tw.
46. fibrosarcoma$.tw.
47. desmoid$.tw.

48. kaposi$.tw.
49. leiomyosarcoma$.tw.
50. liposarcoma$.tw.
51. myosarcoma$.tw.
52. angiosarcoma$.tw.
53. mesenchymoma$.tw.
54. neurofibroma$.tw.
55. neurofibrosarcoma$.tw.
56. schwannoma$.tw.
57. chondrosarcoma$.tw.
58. choriocarcinoma$.tw.
59. dysgerminoma$.tw.
60. (germ cell or germimoma$).tw.
61. teratoma$.tw.
62. seminoma$.tw.
63. carcinoma$.tw.
64. exp adrenal gland neoplasms/
65. adenocarcinoma$.tw.
66. exp thyroid neoplasm/
67. phaechromocytoma$.tw.
68. exp nasopharyngeal neoplasms/
69. melanoma$.tw.
70. or/5-69
71. 4 and 70

APPENDIX B

High Level Search Strategy

The following sites were searched:-

Automated Childhood Cancer Information System
Agency for Healthcare Research and Quality (AHRQ)
Appraisal of Guidelines for Research & Evaluation (AGREE) Collaboration
AltaVista
Audit Commission
Agency for Quality in Medicine (AZQ)
Cancer and Public Health Unit
Cancer and Public Health Unit, London School Hygeine & Tropical Medicine
Cancer Care Ontario Practice Guidelines Initiative
Cancer links - Cancer guidelines and standards
Cancer Management Guidelines British Columbia Cancer Agency
Cancer Research UK - Science and Research
Cancer Research UK Home
Cancer Services Collaborative Group
Cancer.gov - Cancer Information
Cancer.gov - Cancer Literature in PubMed
CancerBACUP
Canadian Coordinating Office for Health Technology Assessment (CCOHTA)
Centre for Evidence Based Medicine
Centre for Evidence-Based Child Health
Centre for Health Services Research - Population and Health Sciences - University of Newcastle
Centre for Reviews Dissemination
Childhood Cancer Research Group
Children's Cancer Centres and Units
College of Health
Commission for Health Improvement
Department of Health
Department of Health - Cancer
Department of Health National Specialist Commissioning Advisory Group (NSCAG)
Eastern Cooperative Oncology Group (ECOG)
Effective Professional Practice Initiative
Evidence Network - The UK Centre for Evidence Based Policy
Evidence-Based Medicine
Finnish Medical Society Evidence-Based Medicine Guidelines for primary care

French Cancer Resources Directory - CancerIndex
Guidelines International Network
Global ChildNet Bibliographical Database
Google
Guide to Internet Resources for Cancer - CancerIndex
Health Care Policy Research Development Unit
Health Development Agency
Health Evidence Bulletins
Health Management Information Consortium
Health of Wales Information Service
Health Technology Assessment.Programme
http—www.anaes.fr-ANAES-anaesparametrage.nsf
International Confederation of Childhood cancer Parent Organization (ICCCPO)
International Agency for Research on Cancer
International Network of Agencies for Health Technology Assessment
International Society of Paediatric Oncology (SIOP)
Kings Fund
Leitlinien.de
Macmillan Cancer Relief Fund
National Assembly for Wales
National Cancer Research Network
National Comprehensive Cancer Network
National Electronic Library for Health (NeLH) - Cancers
National Guideline Clearinghouse
National Electronic Library for Public Health
National Horizon Scanning Centre
National Institute for Clinical Excellence
National Public Health Service for Wales
NeLH Guidelines Finder -
New Zealand Guidelines Group
NHS Centre for Reviews and Dissemination
NHS Modernisation Agency
The National Research Register
OncoLink
Oncology Tools
Organising Medical Networked Information
Public Health Information
Public Health Institute of Scotland
Public Health Knowledge
Royal College Paediatrics & Child Health
Scottish Intercollegiate Guidelines Network (SIGN)
Société Française du Cancer (SFC)
SUMSearch
Swiss Network on Health Technology Assessment

Trent Research Information Access Gateway
Turning Research Into Practice (TRIP) Database
UK Cancer Links
United Kingdom Childrens'Cancer Study Group UKCCSG
UpToDate
World Health Organisation
Young Adults & Cancer WebSite

Appendix C

EVIDENCE LEVELS AND QUALITY GRADING (modified from NICE Methodology Manual)

Level of evidence	Type of evidence
1^{++}	High-quality meta-analyses, systematic reviews of RCTs, or RCTs with a very low risk of bias
1^{+}	Well-conducted meta-analyses, systematic reviews of RCTs, or RCTs with a low risk of bias
1^{-}	Meta-analyses, systematic reviews of RCTs, or RCTs with a high risk of bias*
2^{++}	High-quality systematic reviews of case–control or cohort studies High-quality case–control or cohort studies with a very low risk of confounding, bias, or chance and a high probability that the relationship is causal
2^{+}	Well-conducted case–control or cohort studies with a low risk of confounding, bias, or chance and a moderate probability that the relationship is causal
2^{-}	Case–control or cohort studies with a high risk of confounding bias, or chance and a significant risk that the relationship is not causal*
3	Non-analytic studies (for example, case reports, case series)
4	Expert opinion, formal consensus

Quality grading

++ = good quality
+ = fair
+/- = fair to poor
- = poor

APPENDIX D

Consultation with children with cancer, their siblings and parents for the NICE child and adolescent cancer service guidance

Commissioned by the National Collaborating Centre for Cancer

Jessica Datta, Claire Lanyon, Lucy Read, Emma Sawyer, Janine Shaw, Ben Street

July 2004

Contents

Acknowledgments

We would like to thank the children and parents who participated in the four consultations and who shared their experiences, views and aspirations for cancer services. We hope this report accurately reflects these.

Thanks are due to Dr Andrew Champion at the National Collaborating Centre for Cancer who commissioned the National Children's Bureau to facilitate the consultation days and to Rachel Hollis and other members of the Child and Adolescent Cancer Guidance Development Group sub-group for their support for this work. We would also like to thank staff at the regional cancer centres who were so helpful in organising and co-facilitating the days and whose expertise and professionalism were invaluable. We would particularly like to thank Jeanette Hawkins, Louise Soanes, Rachel Hollis and Judith Armstrong for their support.

A special vote of thanks goes to catering staff at Birmingham Children's Hospital who provided a fabulous lunch. Thanks also to the two clowns from Theodora Children's Trust who provided entertainment at Great Ormond Street Hospital. We would also like to thank staff at Wolf+Water Arts Company (www.wolfandwater.org) for their advice in developing some of the consultation activities.

We would like to thank Julie McLarnon who facilitated the sessions with very young children and Young NCB's Hannah Gibney, Seun Fajolu and Graham Duffy who co-facilitated the siblings groups.

1. Summary of report

Background to the consultation

The National Institute for Clinical Excellence (NICE) commissioned the National Collaborating Centre for Cancer (NCC-C) to develop child and adolescent cancer service guidance early in 2003. In spring 2004 the NCC-C commissioned the National Children's Bureau (NCB) to facilitate this consultation. It was agreed that the consultation would consist of four one day events in four cities in England held in May and June 2004.

Aims of the consultation

The broad aims of the consultation were to examine the perspectives and to elicit the views of children with cancer, their siblings and parents in relation to the relevant criteria within the scope of the NICE guidance (see http://www.nice.org.uk/pdf/Child_Adolescent_Final_Scope.pdf), in order to inform its development.

The participants

In all 114 people took part in the four consultation days. These included 49 parents, 39 children with cancer and 26 siblings. Children's ages ranged from two to 14 years. Participants at each consultation event were assigned to a group of either children with cancer (differentiated by age), siblings or parents.

Programme of activities

A programme of activities and areas for discussion was developed for each group. These included games and creative activities as well as ideas for discussion. Although participants were given the opportunity to set the agenda to some extent, topics for discussion were relevant to the development of the guidance and were agreed in advance by staff at NCC-C.

Key points

The broad areas covered by the consultation were the diagnosis of cancer and how families are told, access to health services, hospital treatment and care, families' information needs, communication, community and home care and family support. Participants shared their experiences and opinions about health services and the key points for improvements in services raised by both children and parents are listed below. The consultation's findings are given in more detail in the following chapters.

Diagnosis of cancer

- Primary care professionals should be well informed about childhood cancers and take parental concern seriously.
- Diagnostic tests should be undertaken quickly.
- Families should be warned if a diagnosis of cancer is suspected.
- Families should be told about a diagnosis of cancer in a comfortable, private room by one or two professionals with plenty of time available for discussion.
- There is no uniform way of telling family members about a diagnosis – children and their parents should be treated as individuals.
- The diagnosis and its consequences should be explained to children in simple, direct language.
- Children should be given the opportunity to contact another child or children of a similar age and with the same condition.
- Families should have opportunities to ask questions more than once and be given access to support by telephone.
- Parents are likely to search the internet for information and would like consistent advice on how best to do this and how to interpret their findings.
- Both parents and children suggested a 'buddy' system which would give families the opportunity to support each other after diagnosis.

Access to health services

- Services should be located near home.
- Hospitals should be near other facilities which families could visit.
- Free or affordable parking should be available.
- Access to more responsive and better organised hospital transport service.
- Appointment systems should be designed to meet individuals' needs.
- Shorter waiting times at outpatient appointments.
- Children with cancer should have access to beds on specialist wards.
- Speedy, consistent procedures for referral to specialist wards.
- Consistent, universal access to home care and social work support.
- Effective pharmacy services.

Hospital treatment and care

- ***Hospital environment and facilities***
  - Wards should be colourfully decorated and comfortable with opportunities for privacy.
  - Space for older children separate from babies and toddlers.
  - Separate visiting and resource rooms were suggested.
  - Play facilities outside the wards including access to outdoor play space.
  - Facilities (such as showers) for family members.
  - All groups mentioned entertainment wanting more age appropriate toys, games, art materials, television and DVD.
  - Easy access to free or affordable telephones.

- ***Food***
  - Hospital food should reflect what children like to eat while not being consistently unhealthy. Younger children wanted pizza, burgers and chips while older ones sometimes preferred lighter meals.
  - Food should be well prepared.
  - Opportunities to eat outside normal mealtimes.

  - Opportunities to eat with family members.
  - Affordable food available for family members.
  - The provision of a ward snack trolley.
  - Easy access to cold drinking water.

- ***Education***
  - Teaching in hospital to be appropriate for age and ability.
  - Effective hospital/school liaison.

- ***Relationships with staff***
  - Staff should talk honestly to children and not just to their parents using accessible language.
  - Children should be given opportunities to be involved in their care whenever possible.
  - Children appreciated reward systems after treatment.
  - Staff in shared care centres should be trained in specialist cancer care and treatment.

- ***Treatment and care***
  - Consistent quality of care across services.
  - Consistent treatment protocols across services.
  - Parents' involvement in care to be valued and training offered to parents when appropriate.

Providing and sharing information

- Information about all aspects of cancer and cancer treatments should be made available to children, parents and other family members in a range of formats and appropriate for their age and circumstances.
- Information should be made available about rare conditions.
- Information about medication and its effects.
- Regular updates on a child's progress which parents can understand.

- Clear lines of communication between professionals.
- Information provided to GPs about a child's progress.
- Regular consultation with patients and parents about services.
- Wider understanding of childhood cancer and its effect on families.
- A sensitive approach to sharing information about the death of a child.

Community and home care

- Consistency in the availability and quality of home care services.

Family support

- Consistent, easily accessible advice about welfare benefits.
- Support for siblings of children with cancer.
- Access to psychological and family support when needed.

2. Introduction

The National Institute for Clinical Excellence (NICE) commissioned the National Collaborating Centre for Cancer (NCC-C) to develop child and adolescent cancer service guidance early in 2003.

To help inform the development of the guidance, NCC-C was asked by the child and adolescent cancer guidance development group (GDG) to commission a consultation with children and young people with cancer and their parents, carers and siblings. The GDG considered input of patient experience into the guidance a high priority. In spring 2004 NCC-C commissioned the National Children's Bureau (NCB) to facilitate such a consultation. It was agreed that it would consist of four one day consultation events in four cities in England held in May and June 2004. Young people aged 15 and over were consulted separately in collaboration with the Teenage Cancer Trust.

NCC-C was responsible for the administration of the days which included booking the venues, recruiting participants, transport, catering and providing rewards for participants. NCB's Project Team was responsible for planning and facilitating activities. This included developing a programme of activities and games, preparing discussion topics, facilitating activities and discussion, collecting and analysing information and preparing a report for the GDG. The topics covered in the consultations were agreed between NCC-C and NCB.

3. The National Children's Bureau

The National Children's Bureau is a national voluntary organisation. It promotes the voice, interests and well-being of all children and young people across every aspect of their lives. It advocates the participation of all children and young people in all matters affecting them. It challenges disadvantage in childhood. Young NCB is a membership organisation for young people with its own magazine and website.

NCB achieves its mission by:

- ensuring the views of children and young people are listened to and taken into account at all times
- playing an active role in policy development and advocacy
- undertaking high quality research and work from an evidence based perspective
- promoting multidisciplinary, cross-agency partnerships
- identifying, developing and promoting good practice
- disseminating information to professionals, policy makers, parents and children and young people

NCB has adopted and works within the UN Convention on the Rights of the Child.

The consultation team included staff from NCB's Participation Unit, Research Department and Family Support Unit. An early years consultant was employed to undertake the work with children aged two to four. The team also included three young facilitators (aged 16-18) who are members of Young NCB and who have trained as group facilitators.

4. Aims of the consultation

The broad aims of the consultation were to examine the perspectives and to elicit the views of children with cancer, their siblings and parents/carers in relation to the relevant criteria within the scope of the guidance (http://www.nice.org.uk/pdf/Child_Adolescent_Final_Scope.pdf), in order to inform its development. These criteria included access to health care, experiences of health care and treatment and how information needs are met.

The consultation focused on the following aspects of cancer care:

- Diagnosis of cancer
- Hospital treatment and care
- Meeting information needs
- Community and home care
- Family support

The report covers these subjects and key points based on the views of participants are included after each section.

5. Methodology

The participants

The sampling frame for participants was children aged two to 14 years, their siblings and parents or carers. The children who were recruited were or had been patients at participating regional cancer centres and had malignant disease, including leukaemia and related conditions (as defined by the International Classification of Childhood Cancer) or benign cancer. The GDG stipulated that there should be a mix of boys and girls in each age group and that recruitment from black and minority ethnic groups should be included at each event. It was agreed that a maximum of 48 children would take part in the consultation.

The four consultation events were held in different cities in England (Birmingham, London, Leeds, and Bristol) in collaboration with the regional cancer centres. NCB produced a recruitment flyer (see Appendix A*) and staff at the regional centres made contact with a wide range of potential participants from individuals, families and local groups already known to them. Details of each local centre's recruitment strategy are not known.

Participating children with cancer were grouped by age and it was planned that approximately 20 families would participate in each event. In order to cover the wide age range identified by the GDG, it was agreed that there should be two consultations for each age group. Age groups included children aged two to four, five to eight, nine to 11 and 12 to 14 (see table 1).

Table 1: Age groups planned for the consultation

Hospitals	**Groups consulted**
Birmingham Children's Hospital	Parents, siblings and children with cancer aged 5-8 years and 9-11 years
Great Ormond Street Hospital, London	Parents, siblings and children with cancer aged 2-4 years and 12-14

	years
St James's Hospital, Leeds	Parents, siblings and children with cancer aged 2-4 years and 12-14 years
Bristol Children's Hospital	Parents, siblings and children with cancer aged 5-8 years and 9-11 years

Altogether 114 individuals (including children with cancer, siblings and parents) took part in the consultation days. There were 39 children with cancer of whom 22 were boys and 17 girls. There were 26 siblings and 49 parents of whom 31 were mothers and 18 fathers (see table 2). Although it was planned that a group aged 12 to 14 would be included in the London consultation, only one child of that age group attended and so that group did not take place.

Table 2: Numbers and sex of participants

Group	**No. of participants**	**Male**	**Female**
Aged 2 to 4	15	8	7
Aged 5 to 7	7	5	2
Aged 8 to 11	10	6	4
Aged 12 to 14	7	3	4
Siblings	26	13	13
Parents	49	18	31

The large majority of participants described themselves as 'White British'. Only 7% of all participants were from a black or ethnic minority background.

Participants came from East Anglia, the Midlands, the south-east, south-west, north, north-west of England and from south Wales. Some participants had met

each other or some of the facilitators before during hospital visits while others knew nobody else.

Consulting with children and young people

Over recent years there has been a growing acceptance that children and young people should be given opportunities to be involved in decisions that affect their lives. This has been driven by a number of ideas which include the increased involvement of the consumer in the development and modification of both private and public goods and services, the children's rights agenda, which includes the right to participate in decision making, and a relatively new understanding of children – including very young children - as a competent social actors who have a role to play in shaping their own lives and those of others. In accordance with these ideas, children and young people have increasingly been asked to take part in consultation exercises and decision making in diverse areas of their lives. These include health services, the family court, community initiatives and education. The current consultation is an example of how service providers and policy makers take account of the views of children and young people. A separate report of the consultation and its findings has been written for young participants (see Appendix H*).

A consultation of this kind cannot represent the views of all family members in England and Wales who are affected by childhood cancer. However, it does provide the opportunity for a number of children and parents to share their experiences of and views about living with cancer and cancer care. The issues raised at the separate consultation days were broadly similar which points to the relevance of the information gathered and key points made.

Consultation techniques and activities

The programme for consultation days was divided into sessions with age appropriate activities organised around a theme (see Appendices for details of

the programme). Children's activities included opportunities to make pictures and play games as well as to discuss issues while the sessions with parents were more like traditional focus groups where issues were discussed and recorded in writing. Although activities and discussion were based broadly on the issues agreed with the GDG, participants were able to widen the remit and talk about other aspects of cancer services that they felt were important. Consultations with very young children used play and medical equipment to encourage interaction and discussion.

The consultation tools were carefully prepared with consideration given to age appropriate activities, the health of the children, keeping children engaged and having fun. Separate programmes were devised for children aged two to four years and five to eight years which focused on creative play techniques (see Appendices B and C*). One programme was developed for the sessions with nine to 11 years and 12 to14 year olds (see Appendix D*) with minor adaptations according to the age group. In this programme a series of activities was designed to engage the children, to be fun, to be participative and to ensure that relevant topics were raised and discussed. The activities included group discussions around a topic using post-it notes to record experiences and views, a reporter exercise in which children interviewed each other, and making collages to express views and suggestions. All the activities were designed to promote discussion and enable children to give their views in fun ways. The programme for siblings was similar to the programme for nine to 14 year olds but included slightly different exercises (see Appendix E*). The programme for parents was much more discussion-based in order to allow participants as much opportunity as possible to talk about their individual experiences and express their views in addition to creating a group consensus about issues of importance (see Appendix F*). All participants were asked to stick 'leaves' on a wish tree at the end of each day on which they had written a 'wish' for improvements in cancer services.

In order to establish a friendly, informal atmosphere at the consultations, each group session (including parents' sessions) began with a brief fun game which gave participants the opportunity to familiarise themselves with each other and with facilitators. Ground rules were introduced and agreed by participants. These included an agreement that personal experiences were confidential and should not be repeated outside the sessions and that children should not leave a session without letting facilitators know. Children were able to leave a session and go to their parents at any time.

Collecting and analysing information

Members of the Project Team with the invaluable help of local facilitators made copious notes of what participants said on flipchart paper and notepads. These notes, with the post-it notes, drawings, etc., produced at the consultation days, were written up into reports of each session. The resulting reports were collected together for each group of participants (i.e. for each age group and for parents and siblings) and themes relating to the main issues covered by the guidance scope were drawn and compiled together to produce this report. As noted above, another report was produced for young participants.

6. The views of children affected by cancer

This section presents the experiences and views of children with cancer who were consulted about their health care and treatment. It is divided into three sections which detail the responses from each age group. These were children aged two to four, five to seven, and eight to 14. Each section describes the activities, consultation findings and key points.

Younger children affected by cancer aged 2-4 years

The consultations with children aged two to four took place in hospitals in London and Leeds. An early years consultant facilitated the sessions with this group of children.

Methods

An outline of the day's activities was planned and intended to be used as a guide for the kinds of age appropriate activities that could take place on the day rather than as a rigid schedule (see Appendix B*). To ensure that the process was both child-led and child-focused the children dictated the pace of the consultation and how it developed. The different activities were integrated into an ordinary play session so that the children could choose what they wished to play with, how they played and could join in activities only if they wanted to. Flexibility was essential in order to work with the children's normal routines. This meant that sessions did not always go according to plan – for example, at one hospital children were too tired to participate in all the sessions. Discussions with co-facilitators highlighted the many different techniques used to support these children through the different stages and circumstances of their treatment. Examples include at initial diagnosis, during treatment, when in remission or isolation, terminally ill children and those at the transition stage between illness and becoming well. Because children of this age can be wary of being separated from parents or other familiar adults, the facilitator felt that meaningful consultation with this age group would be more effective on an ongoing (rather than one-off) basis.

The topics for consultation were simplified so that they were meaningful to those taking part. Written observations were made on the day. The topics became:

- **I know I am not well** – drawing around each child's body and prompting discussion about how they felt;

- **Making me better** – a sensory walk around the hospital setting, the children being prompted to talk about what they saw and smelt and what happens in particular areas of the hospital;
- **The very worried doctor** - an interactive story and rhyme session about a newly qualified doctor and what a wise child with cancer taught him.

In pre-consultation discussion, play specialists recommended the following:

- Concentrating on free play with medical equipment

It was decided that the most useful play equipment would be real medical equipment (i.e. drips, bandages and syringes etc.), closely supervised by adults to ensure safety of the participants.

- Using a 'blood doll'

These are dolls with Hickman lines, nasogastric tubes and portocaths in place and who have removable hair to represent a child undergoing treatment for cancer.

- Collecting background information from parents

To understand the context of what the children expressed, questionnaires about the child and his/her condition were distributed to parents (see Appendix G*). Where appropriate, information from individual children was shared with parents to ensure that what their child expressed was accurately represented.

Younger children's awareness of their medical needs

These very young children were capable of clearly expressing their experience of living with cancer through their play, body language and spoken explanation. The free play enabled the children to be in control of what they played with and how. Playing with real and Playmobile medical equipment helped them to make links between what was happening to them in hospital and at home. The play and

conversation reported below illustrate young children's awareness of illness and how it is treated.

The children referred to their cancer in simple terms - 'bad blood', 'when your blood is up' and 'bad tummy'. They used colloquial terms to refer to their treatment such as 'wiggly' or 'noodle' for the central venous line. Those children who had a portocath fitted were quite happy to show them to the facilitator. One explained that the port in her arm was for 'the medicine [which] goes in your tummy and makes you better'. One child administered 'treatment' to the facilitator in a calm, efficient and professional manner, putting on gloves, using and then carefully disposing of antiseptic wipes, injecting into a line and taking temperature. Another child's mother reported that her child was capable of asking medical staff specific questions about treatment.

Two children applied 'magic cream' (local anaesthetic cream used for painful procedures) to dolls. The magic cream appeared to be important and great care was taken to rub it in carefully and give it time to work before applying a dressing. The syringes were used to administer medication into the Hickman line or as medicine into the mouths of the dolls. One child described her medicine as 'orange' and she administered it slowly and carefully. One child asked another what else could the syringe be used for. The child replied, 'medicine'. A third child sitting nearby touched his tongue and said, 'Yuk'.

A child asked, 'Where else do we put the tube?' and then placed the tube in the doll's nose. A second child shook his head emphatically and said, 'no!' A third, who had a nasogastric tube inserted, said, 'like me!' and the first child very solemnly said, 'Only on dollies'. The children seemed to be wary of the nasogastric tube.

During another session children were happy to show the facilitator the waiting and consulting rooms and show her where they sit and where the doctor sits.

They lay down on the bed and one used a stethoscope to listen to another's chest. In the treatment room one of the children administered medication to a blood doll. When he forgot to undo the clip, the others reminded him what to do and he rolled his eyes at his own omission.

In their responses to the pre-consultation questionnaire (see Appendix G*), it seems that parents underestimated the level of awareness that their children had about cancer believing they were too young to be able to participate effectively in the consultation. However, it was clear through play that the children did have an awareness and demonstrated quite clearly that they had an understanding both of cancer and of cancer treatment.

Play specialists involved with the consultation did not think that any of the literature for children with cancer that they had seen was suitable for very young children. The books are too wordy and lack the pictures required to help a young child's understanding. There is perhaps a need to develop early years literature on cancer that suitable for very young children.

Relationships with staff

Continuity and familiarity are important to children

The younger children said that continuity is important to them and that separation from main carers worries them. This was reflected in some of the children's discomfort at being separated from their parents and the relief shown when the children recognised individual co-facilitators. One child immediately recognised a member of the hospital's support staff and sought her company while another child who did not know anyone wanted to leave early. They were familiar with particular doctors and appeared pleased when they discovered that another child in the group also knew a doctor by name.

Waiting for treatment

An interesting observation of this consultation process was that children appeared to be used to waiting and accommodating delay. Although anxious to return to their parents, they appeared resigned to the fact that treatment would take a long time.

Key points for services for children aged two to four

- The development of effective listening and observation techniques for supporting very young children with cancer
- Dedicated information about cancer and cancer services in the form of picture books (or other media) for very young children

Children affected by cancer aged 5-7 years

The consultations for children aged five to seven years took place in hospitals in Birmingham and Bristol.

Methods

Sessions for this age group were based on play and creative techniques. A plan for each session was developed with advice from the arts organisation, Wolf+Water, and used as a flexible guide. First, the children were asked to give a name to a cut-out cartoon 'alien' which was used to help establish children's understanding of their illness. This character was used to represent a child with cancer. Participants were asked to imagine this character's favourite activities and to specify its illness. They used the same character to provide information on their experience of being treated for cancer.

Hospital treatment and care

Generally, the children seemed to have few complaints about hospitals and hospital care. Hospital staff received the most praise, and the facilities available for children were also appreciated.

Hospital location and environment

Children were asked to discuss what makes an 'ideal' (or improved) hospital environment. They suggested that an 'ideal' hospital would be located near their home to make visits easier (one child made a drawing of a hospital with his family home inside it) or near a town centre so there would be things to do on family visits. Most children wanted children's wards to be brightly coloured with plenty of toys and art materials available. Children emphasised their wish for privacy ('a door you can lock', 'curtains around you').

Treatment and care

The children discussed the good and bad aspects of their own experiences. Most children were happy with the treatment they had received from hospital staff. Doctors and nurses were described as 'funny', 'friendly' and 'happy'. One child was concerned about hospital staff not getting enough sleep and consequently being grumpy.

'The doctor was funny - that made me happy.' (7 year old)

'The nurses were really kind and friendly. They let me choose a toy from the treasure box after the injection.' (6 year old)

A few children said they liked to know exactly what was going on and knew what all the medical apparatus was used for. The majority, however, preferred discussing their illness in a more opaque way.

'It's best to know all about the medicines and what's happening to you. My mum and dad told me what's going on.' (8 year old)

Reward systems such as letting children play with toys or have sweets after injections or operations were regarded as positive.

Hospital facilities

Play away from the ward

Some children said they wanted to play outside the hospital ward in either separate playrooms or in playgrounds in the hospital grounds. Many said they wanted to have more time outside the hospital building but recognised that this was dependent on their treatment.

Food

Most children enjoyed the food they ate in hospital, although a few said they disliked it and suggested having McDonalds meals instead. Most children of this age said they preferred pizza, burgers and chips to other options.

Communication

Contact with family and friends was considered important by participants. Children thought that patients should be given mobile phones to call home, friends should be able to visit more often, and there should be a school in the hospital.

Key points:

Access to services

- Hospitals should be located nearer children's homes and near other facilities which families could visit.

Hospital facilities:

- Hospital rooms should be colourful and there should be toys and art materials to play with.
- There should be places to play outside the ward including outside play space.

- Young children said they would like to eat pizza, burgers and chips.
- There should be a hospital school.
- Telephones should be made available to patients.
- Friends should be able to visit more often.

Relationships with staff

- Children liked 'funny', 'happy' doctors and nurses.
- They liked to be offered rewards after treatment.
- Some wanted to know about treatment in detail while others did not. Staff should treat children as individuals.

Children affected by cancer aged 8-11 and 12-14 years

The consultations with children affected by cancer aged eight to 11 took place in the Birmingham and Bristol hospitals. It was noted that the group of children in the Bristol group were generally less well than the Birmingham group and consequently there were more breaks in the sessions. One group of young people aged 12 to 14 participated in Leeds. The one young person in this age group who attended the London consultation joined the siblings group. The findings from both age groups are described below.

Methods

A schedule of activity-based exercises was created to be used at each consultation day for these age groups (see Appendix D*). The same schedule was used for both the nine to 11 and 12 to14 age groups. The activities were designed to encourage the children to express their views and to promote discussion between children and facilitators as well as to be enjoyable. Facilitators recorded children's views and comments on flipchart paper.

The activities were planned to involve participants as much as possible and included sessions entitled (see Appendix D*):

- What's Important to You?
- Picture Perfect Diagnosis
- Your Care, Your Views
- The World's Worst/Best Nurse
- Receiving You Loud and Clear

What's Important to You?

The children were asked to identify issues that were of most importance to them in relation to having cancer, write them on post-it notes and stick them to a full-sized body shape. The themes below are those identified as most important by participants.

Family and friends

Children talked about how their family and friends reacted to their illness. Many said that their friends and family members acted differently towards them since their diagnosis, were far too protective of them and did not understand their illness. They also said that they missed friends and family while they were ill and in hospital. Some said their siblings were nicer to them. Many said that they did not like always being asked how they were.

'It is difficult to see my sister 'cause she's scared of hospitals and gets upset.'

School

The main issues children highlighted about school were feeling 'different' and missing school because of clinic appointments, being unwell or undergoing treatment.

'I have to have a stool, cushion and file thing on my desk and no-one else has.'

Older children reported falling behind with schoolwork and getting poorer grades in tests and exams.

Hospital treatment and care

Participants had the most to say about this subject. They talked about being scared, being tired, their hair falling out, having to be on specific diets and being woken up in the night to receive medication.

'*Radiotherapy is scary especially the mask over your face.*'

'People should make chemotherapy so your hair doesn't fall out.'

However, they also made positive comments about hospital staff and effective treatment.

'My portocath has helped because I can swim. It's better than a Hickman.'

Being bored

Overcoming boredom in hospital was an issue raised by a number of participants. They wanted more things to do and to entertain them in the wards including books, Sky TV and games consoles.

'My treatment is boring and annoying.'

'Books for when I can't run about.'

Other people

Many children felt uncomfortable with how they were regarded by adults or other children when they were out or at school. They did not like the way that people stared at them. Girls explained how they were often mistaken for boys because of hair loss and how other children did not want to play with them.

'People in the ladies call me a boy.'

Sports and hobbies

Children said that they were upset that they could not take part in the physical activities that they used to enjoy such as swimming and other sports.

Personal support

Older children talked about the importance of people around them who offered support. In particular, they cited parents and other family members, friends, nurses, doctors and teachers.

Picture Perfect Diagnosis

The aim of this exercise was to provoke discussion about diagnosis and what could be better though the creation of a collage. All comments were recorded. For this session, each group was given a selection of materials, including pens, pictures, magazines and speech bubbles, to create a picture of how and where they thought children should be told they have cancer.

The feelings that a diagnosis of cancer provoked were evident in the pictures created by the children. The facial expressions of their parents (shocked) and themselves (unhappy) demonstrated how emotive diagnosis was for children and families. Different forms of support and solace such as pictures of teddy bears and adults comforting children were also used. One young person said, 'when I got told it was strange because my heart went strange' and drew a picture of a broken heart.

There were broad similarities in the rooms that the children designed as the place where they would prefer to hear about a diagnosis of cancer. The rooms were colourful with sofas or comfortable chairs, pictures and toys. Children did not want to be told in a clinical, impersonal environment. Many stressed that they

wanted to be told in a private room with only their parents and the consultant present.

'Tell, me, my parents and consultant and no one else about my health.'

One child thought there should be a room specifically designated for the purpose of giving diagnosis. Another said that he would prefer to be told in his own bedroom at home with his family.

Children were concerned with who told them and who was present. Some commented that they would like to be told at the same time as their parents although a small number said they would like to be told afterwards by their parents. One wanted her friends to be there as well as her family and another chose her favourite uncle to be with her. Another child wanted a nurse present who would be able to explain medical terms and treatment.

Children wanted to be given an explanation of their illness in simple terms and in a direct manner. One child wrote in a speech bubble, 'OK [child's name], let's get straight to the point…you have leukaemia'. Several children said that they would like to be treated with respect and not like small children who could not understand what was happening. They felt that if they were given a clear and simple explanation they could understand their diagnosis from a very early age.

'Doctors shouldn't explain it [treatment and diagnosis] to someone in a rush and they don't explain enough.'

Some said that a diagnosis of cancer should be explained more sensitively with enough time allowed for questions and answers. It should not be given over the telephone which was one young person's experience. Many had not understood the medical terms or realised that treatment would begin immediately after diagnosis. The older group agreed that having another young person who had

undergone cancer treatment or a liaison nurse to explain and help them understand procedures and terms would have improved their experience. Many children felt that there were too many people involved at the early stages of treatment and all thought that there was too much information to digest.

Some children said that they wanted to be told by a consultant who was friendly, fun and kind. One child thought that a play specialist, rather than a consultant, should have explained her diagnosis to her. She liked the way that play specialists used toys in their work. The pictures created showed how much children need to be supported and comforted by their parents and other hospital staff.

'See a consultant rather than a registrar because they explain things better.'

Many of the children talked about the length of time it took to be diagnosed and how upsetting this was for them and their families. Only one of those in the older age group had been diagnosed quickly. They also talked about when the diagnosis was made. One child, for example, said, 'I spent all Christmas waiting to hear'.

Your Care and Support: Your Views

This exercise was designed to investigate children's experiences of cancer services in hospital, at outpatients clinics and at home. Children were also asked for suggestions for how services could be improved. Children acted as roving reporters and interviewed each other with a questionnaire about their care in hospital, as outpatients and at home and also took part in a group discussion about their involvement in their care

Hospital treatment and care

Location of care

All but one child expressed a preference for being treated at a regional cancer centre rather than a local centre even though it often involved a long journey to hospital. Children thought that the treatment they received at the regional centre was superior, centres were better equipped and staff had more specialist knowledge about cancer treatments as well as knowing children and families better. When being treated in local hospitals children felt that themselves and their family had more knowledge about their condition than the staff. They also liked being in contact with other children who had cancer in the regional centres and felt isolated on general wards.

Some children said that staff in shared care hospitals were ill informed about their current treatment and that they had to explain their needs again and again. They would be happier to attend the local hospital for routine appointments if records were maintained and staff kept up to date with their treatment. They suggested that a specialist liaison nurse attending all appointments could improve communication and that local hospitals kept up to date copies of medical notes.

Hospital environment

The majority of children had stayed on a children's ward and only one on an adolescent ward. All children said that the ward they stayed on was OK or good. The older ones contrasted their experiences of general paediatric wards in local hospitals with specialist wards at regional cancer centres, preferring the latter. The majority of children said that they 'sometimes' had enough space and privacy. They requested more space between beds, more cubicle style rooms and less curtained ones.

Half the children thought that it was 'always easy' for friends and family to visit them and the other half thought it was 'sometimes' easy. They suggested that a separate room should be provided where they could meet with family and friends. They also wanted a mechanism to ensure that friends came to visit when they looked well. Those who lived far from the hospital and whose families had been

able to stay with them or close by appreciated the availability of accommodation attached to the hospital.

Children said that different age groups have different needs and cancer services should reflect this. In particular, older children stated said that they did not like to hear babies crying. Specific activities (such as wig and pyjama parties) and environments designed for teenagers were appreciated as were things like a painted ceiling in one ward which was 'fantastic' for patients who were bed bound.

Treatment

Most children had had chemotherapy and some had had radiotherapy, surgery, bone marrow and peripheral stem cell transplants, biopsies, steroids and lumbar punctures. Children felt that their treatment could have been made better if they had been better informed beforehand, had pre-medication and smaller tablets that were easier to swallow.

'They told me what to do but not what it would do to me.'

Hospital facilities

Food

All the children reported that they were unhappy with hospital food. Nearly all stated that it was 'not good'. They said that it was not hot, it was often burnt and they would like more menu choice. Many children preferred food like burgers and chips while some of the older ones would like the option of lighter meals when they are not very hungry. Children also wanted food to be available all day and not just at mealtimes so they did not have to go to the shop. Older children liked a family Sunday lunch which was provided at some hospitals when families could eat together and requested more opportunities to share meals like this.

Telephone calls

Young people talked about the high cost of making and receiving telephone calls in hospital using Patientline. Although they greatly valued having a personal phone, the cost was prohibitive. It was suggested that costs should be reduced for long-stay patients.

Parking

Children reported that it was often difficult to find a parking space and that car parks were expensive. This sometimes meant that there was a long walk to the hospital making them late for an appointment which added to the family's frustration. Only one child knew about a free parking scheme.

Activities

Children reported that, although there were some activities and things to do available in hospital, such as age appropriate videos, games and Playstation, they get very bored and requested more activities for older children and teenagers including more computers, exercise equipment and art activities.

Education

All the children who had spent time in hospital had done some schoolwork. Children of both age groups said that lessons they had in hospital were not suitable for their age and ability and schooling would have been more effective and interesting if they had been set more demanding work.

Making things better

Each child was asked to think of two things that would make being in hospital better for them. The most common answer was entertainment such as television, Playstation and playing games. Other individual answers were:

- No pain
- Nurses making sure that you went to the toilet to prevent constipation

- Having family around you
- To bring a pet in with you
- Nice nurses, doctors and play specialists
- A garden
- More play ladies

Children were also asked to think of two things that made being in hospital worse for them. Their responses included poor food, the lack of games and things to do and being far from home and/or in isolation.

Outpatient care

Most children received the majority of outpatient care at a regional centre but were sometimes treated locally. All the children estimated that it took them up to an hour to travel to the clinic.

The majority of children said they had weekly or fortnightly appointments. When asked whether they had enough time with staff at appointments, most children said that they did 'sometimes'. The majority said that they were only seen quickly sometimes and usually had to wait. When asked what would improve appointments, they all requested shorter waiting times. Most children wanted more entertainment in the waiting room and requested television and videos or more toys and activities. They also suggested that having more doctors and nurses would speed up the process. One child requested a bed so he could sleep while waiting.

Home care

Children were asked who helps care for them at home, at school and in the community. All the children said their 'mum'. Nearly half also mentioned a community nurse. Other family members, teachers and a summer camp for children were also mentioned.

Involvement in decision-making

When asked how they are be involved in decisions about their treatment for cancer, the children recognised that ultimately they did not have many choices because they were often in a life or death situation.

'Treatment is more important than anything because you could die.'

Many children felt that staff talked to their parents and treated them as though they were invisible. Older children hated being seen as 'cancer victims' and not as individuals living with cancer who were able to make informed decisions.

'Don't treat us like we don't know anything – we know more than they think.'

Children felt that sometimes doctors offered choices to their parents which they felt they could have made themselves. However, some had had more positive experiences of being consulted and listened to.

'I really like that they ask me when they could ask my parents. It makes me feel like I'm the important one.'

Children and young people also talked about times when they did have some choice over their hospital care. One, for example, was asked whether she wanted a portocath or a Hickman line. She said, 'I got a choice but the other children didn't'. Another had been asked whether she wanted her mother to help with her treatment at home. She said she did not and so a community nurse was assigned to her. Others talked about the timing of treatment. Some had good experiences of doctors being flexible about the timing of non-urgent treatment in order to allow children to enjoy holidays or a birthday while others felt that they had been denied opportunities because of a rigid adherence to treatment regimes.

One area in which children felt they would like more say was in the meals provided in hospital and mealtimes. Children in hospital often missed a meal because they were having treatment and so were hungry afterwards. This meant that parents had to bring in food for children. They also requested more choice over when they were able to eat.

The World's Worst Nurse

A cartoon picture of a nurse was drawn on to the flip chart. Children were asked to describe the world's worst nurse. They were also encouraged to describe a nurse's good qualities. The aim of this exercise was to engage participants in a fun discussion about what qualities they would like nurses and other health professionals to have and how they could be supported better. The answers produced were very general but most were related to the experiences of each individual child.

Some children talked about the way nurses explained the treatment they were given and remarked that they were often not told clearly about what was happening to them.

'They do things without explaining and leave you with things you don't understand.'

'Not telling me about the machines.'

Children most commonly said that nurses were either not very helpful or hurt them whilst carrying out treatments. It should be noted that they said that this was more likely to happen at a local hospital rather than at a regional centre.

'Doesn't wash hands – 'cause you might get an infection.'

'Doesn't give you 'magic cream' to stop the needles hurting.'

'Pulls 'wiggly' and it hurts.' [NB: 'Wiggly' refers to the central venous line used to administer medication.]

The second most prevalent topic was how nurses communicate with children. Children said that nurses often did not look at them when they were talking to them or did not listen. They also raised the difficulties they had experienced communicating with nurses whom they did not understand.

'If they are foreign and you can't understand what they are saying'.

Another theme was the amount of time nurses dedicated to children. A number of children said that nurses were either with them too much or too little. They did not want to talk to nurses when they were feeling tired, although they did joke about this aspect.

'Always with you too much or not enough.'

'Always wanting to talk when you're tired.'

Children were unhappy with nurses who talked about them and their illness in front of others on the ward.

Children listed the members of staff who they found most helpful while they were receiving treatment. These were the pain nurse, the community CLIC nurse, ward sister, professor, consultant doctor, surgeon, anaesthetist, physiotherapist, play specialist and home tutor.

Receiving You Loud and Clear

In this session, children were asked to design a website, using a cut out of a computer and collage, to include all the different kinds and ways of giving information they wanted about cancer. Children also discussed the information

they had actually received. All those in the 12 to14 year old group thought that the leaflets they had been given had not been appropriate for their age. They recommended more suitable literature, information aimed at them rather than at parents, guidance on helpful websites, being asked for their opinions on care and treatment and a wider understanding of cancer in children and young people which might be prompted by a storyline in a television soap opera.

Children wanted their websites to contain the following pages:

- My illness
- My messages to you
- Photo board
- Research on leukaemia
- Send me an email
- Message board
- Cancer cartoons
- Other people's stories/diaries
- Link to send messages between hospital and home.
  Access to your results on the screen (accessed by a password, only used by you and your family)
- Chat rooms
- Links to more detailed information on cancer.

Children thought that the best ways of giving children and others around them information were:

- Doctors talking to them/talking to your consultant - '*Consultants give the best information.'*
- Talking to your counsellor
- Receiving good information and then explaining it to your friends.
- Videos on childhood cancer

- Books on childhood cancer
- Leaflets about leukaemia and all types of cancer for children and for adults
- Chatting to other people who have cancer
- Activities for children with cancer
- Camps and special activity days
- Gifts and goodies about cancer – to help children with cancer
- Blood dolls
- In school – advice on how to return to school and what might happen, more education about cancer in schools and information for teachers to tell them about a child with cancer. Asking parents to go to your school and explain to your class what is wrong with you.
- Talking to family – *'Sisters are very good at explaining things'.*
- Using accessible language

Meeting information needs

Just over half the children felt that they had not been given enough information about their illness and treatment. The comments below show that children did not always feel well informed.

'I thought after the transplant it would be over but it wasn't. I still have to have drugs and come to hospital. I didn't know I would have to go into isolation.'

'They didn't inform me that I might go sterile and not be able to have children because of radiotherapy.'

'I was diagnosed aged three but given no information because I was too young. I was given the basics but it was really weird because I was so small so why bother?'

'I wasn't told that I couldn't go to school or it will be a while until I can go out.'

'I did not realise it would take so long for my T cells to come back.'

'You should be told what all the tests are for.'

'I should have been told more about treatment before it started.'

Key points

School

- Schools should be provided with information about a child's condition and how it will affect his or her work.
- Information and education about cancer should be provided for all school students to help overcome children with cancer feeling 'different' from peers.
- Better school/hospital liaison to support children's learning.

Diagnosis

- Hearing about a diagnosis of cancer should take place in a private room, preferably one specially designated for diagnosis which is comfortable and informal and with some toys available.
- Most children requested that only the consultant and their parents should be in the room. Some would have also liked a nurse to be present.
- The diagnosis and its implications should always be explained to the child in simple and realistic terms.
- Further support, such as play therapy, should be available to children and parents.
- Symptoms should be taken seriously and tests undertaken quickly so that a diagnosis can be made as soon as possible.
- Families should be told about a diagnosis of cancer face to face and not on the telephone.

- Some children would like to make contact with another child with cancer of the same age.

Shared versus regional care

- Children did not have confidence in shared care centres. Staff in shared care centres should have better training in specialist cancer care and treatment and have the correct equipment.
- There should be effective communication and information sharing between staff.

Hospital care

- Children want more entertainment in hospital including games and activities specifically for teenagers.
- There should be more resources available on wards such as satellite television, a wide range of books, art materials and games consoles.
- A separate visiting room (like the one at a specified hospital) was suggested.
- Children should be given some say in when they want to see visitors.

Food

- The food available in hospital should reflect what children like to eat (such as burgers and chips as well as more healthy options and lighter meals). Food should also be well cooked and hot.
- Food should be available at alternative times for children who are undergoing treatment or who are 'nil by mouth' and who therefore miss conventional mealtimes.
- Opportunities to eat with family members sometimes.

Education in hospital

- Teaching should be appropriate to children's age and ability.
- Educational activities could help address children's boredom.

Activities

- The provision of a wide range of age appropriate activities and entertainments in hospital including books, videos, computer games, toys and opportunities to play.

Clinic appointments

- Waiting times should be reduced.
- There should be entertainment available in waiting rooms.
- Health professionals should make the necessary time available to discuss issues with children and families.

Children's involvement in decision-making

- Children should be involved in their care whenever possible. Children understand the serious nature of their treatment but should be involved in smaller decisions that could make their lives easier. Where possible, medical staff should accommodate children's wishes which might include acknowledgement of birthdays and other important events.

Staff issues

- All health professionals should explain to children what they are doing to them and what the equipment is that they are using.
- Health professionals should use age appropriate language.
- Nurses should be able to speak and understand English well enough to communicate effectively with children in their care.
- Children should be treated with respect regardless of age.
- When parents are present, health professionals should talk to both the child and the parents and remember that the child is the patient.
- Health professionals should be aware of how a child is feeling and disturb them as little as possible when they are tired or feeling ill.

- Children with cancer are concerned about lack of cleanliness and the risk of infection. Staff should be aware of the importance of scrupulous cleanliness.

Information needs

- All children with cancer should be given adequate information about their illness, treatments (before they begin), how treatment will affect them, how long it will go on for and any side effects.
- Information should be age-appropriate. This is a particular problem for teenagers.
- Children can be given information in a variety of ways:
  - Websites – other people's stories and chat rooms were popular and a useful method of providing information.
  - Information from health professionals provided in clear, fun and sensitive ways.
  - Videos/books/leaflets
  - Cancer related projects and camps
  - Information-based toys such as blood dolls
  - Information support from health professionals

7. The views of siblings

Four sessions for siblings took place at each of the consultation days. Siblings' ages ranged from five to17 years.

Your sibling's illness

The first activity aimed to establish the extent of participants' knowledge about their brother or sister's illness. Despite the inclusion of young children in some of the groups, all participants were able to articulate the name of their sibling's

cancer and what part of the body it affected. They drew around a person's body and marked the areas where they knew their sibling's cancer was.

This exercise showed brothers and sisters' understanding of how their siblings with cancer feel and its effect on these children. The children talked about their sibling's hair loss, big scars on their bodies, pain and its location, the position of the 'wiggly' and marks made by it, the danger of infection, the fact that they may swell or bruise easily, weight loss or gain and other effects of medication. Some explained how illness and medication can prevent them from participating in activities and, in some cases, losing their sense of taste and appetite. They talked about the effect of treatment on their brother or sister and said it could make them scared, grumpy, cry a lot, moody, sleepy, sick, angry, unhappy, different, sad, stressed, mad and 'a pain'.

An 'ideal' hospital

Working in pairs each group was given a selection of pictures and magazines to create a picture of where they thought their brother or sister could have best been treated.

Participants designed colourful rooms with pictures of Scooby Doo and Thomas the Tank Engine on the walls which they thought would make brothers and sisters feel at home. They thought the rooms should be private and comfortable with facilities like DVD and video players to keep their older siblings occupied and more toys and books for their younger siblings. They wanted comfortable chairs for them and other family members as visits lasted a long time.

Siblings proposed that the rooms have free phones so they could make contact with their brothers or sisters whenever they wanted as many said they missed them while they were in hospital. They also suggested having tables and chairs so that their siblings could get out of bed to eat if they were able. They wanted a wider range of food to be available and for it to be of better quality.

Children suggested having baths with bubbles which would comfort their siblings. Some children wanted colourful, soft carpets on the floor and access to swimming pools to allow their siblings to stay fit and not get bored.

They thought doctors and nurses should be available at all times and that siblings should have immediate access to a member of staff especially if they were in pain. They suggested smaller pills be developed which were easier to swallow and tasted better. They wanted someone to look after their brother or sister so their parents could take a break every so often. They wanted staff to explain the medical equipment to them so that they could understand more about their sibling's treatment.

More fantastic suggestions included a stunt show to fly past their sibling's windows, a big wheel to ride on and daily entertainment by clowns. They wanted their siblings to have pets in hospital which would provide comfort and company for them

What makes a difference?

Siblings were asked to write suggestions for things that would make things better for them, their brother or sister and for their parents. Their ideas are listed below:

For siblings

- Somebody to help me with spellings and homework when mummy and daddy are away
- Support from people at school like friends or teachers
- Hospital closer to home so I can see him more
- People to tell me what's wrong
- Have someone to talk to when I'm alone
- Lots of entertainment for children and adults
- Help at home when mum and dad are at hospital

- Someone to go out on my bike with me
- Mum doesn't leave me out if my brother goes to hospital
- More attention for me

For brothers and sisters with cancer

- Better food and advice on diet
- Less painful treatment
- Play specialists so you don't get bored at home or the hospital
- Hospital nearer home
- A study room in hospital
- Toys, games and videos and more things to do in hospital
- For my brother to wait only a year not three years for the 'all clear'
- More doctors and nurses to cut waiting times
- A quicker, easier and less painful way of curing cancer

Parents/carers

- Someone to give my mum or dad a break
- Information sheets for adults to understand
- Having meals delivered free to home
- To be able to get to the clinic quickly instead of having to drive so far
- Helping mum and dad stop being upset

Meeting information needs

Participants were asked to design a web page that would give them all the information they needed for themselves and their family. They discussed what they knew about cancer and cancer treatment and what they wished they had known. Many said they would like to have access to information and support via the internet and suggested how this could be made available with games, quizzes, case studies, information pages, fund raising ideas and chat rooms. They wanted websites that could answer questions and provide links to other useful sites.

Suggestions included:

Games and quizzes

- The battle of the cells - a game where you are a good red blood cell and you have to shoot the bad cells
- Education games such as naming parts of the body
- A wig game
- A quiz about cancer
- Golf with pills. Each pill has a different name to help different cancers
- A bone game
- Create your perfect medicine

They wanted information on:

- Create your perfect medicine
- Cancer – what is it and why do you get it?
- Diagnosing cancer
- Different types of cancer
- Platelets and blood cells
- Medication and how it works
- Cancer research
- Different hospitals
- Curing cancer
- What treatment their siblings have
- What doctors actually do (with pictures)
- How to comfort people with cancer
- How to cope with cancer

Suggestions for support:

- A free support telephone line
- An agony aunt to listen to people with cancer by e-mail or telephone
- Answers to specific questions

- Real life stories
- Meeting other people

Key points:

Access to services

- Hospitals should be located nearer home to enable families to visit easily.

Hospital facilities:

- Hospital rooms should be colourful, comfortable and private and large enough to accommodate enough seating space for visitors and somewhere to eat.
- Rooms should include toys, books, television and DVD players so patients can entertain themselves.
- Hospitals should have luxurious bathrooms and a swimming pool.
- Patients should be able to play with pets.
- Telephones should be freely available to patients.
- Food should be of good quality.

Hospital staff

- Medical staff should be instantly available to patients particularly when they are in pain.
- There should be enough play specialists to provide play opportunities.

Treatment

- Pain should be well managed and pain free treatments developed.
- Smaller, easy to swallow pills should be developed.

Information needs

- Siblings and parents should be provided with information about cancer and cancer treatments via age appropriate websites.
- Siblings should be told about medical equipment and what it is used for.

Family support

- Parents should have access to both emotional and practical support.
- Siblings' need for attention and support should be acknowledged and met.

8. The views of parents and carers

Methods

Parents were invited to share their experiences, views and suggestions in pairs and small groups. Individuals then reported back to the whole group and the data was either written up on a flipchart or in note form by one of the facilitators. Participants were given the opportunity to add to flipchart notes during the breaks. The broad topics covered were diagnosis of cancer, inpatient and outpatient hospital care, information needs, community and home care and family support. Although participants were keen to talk about their own experiences, they were also encouraged to evaluate the treatment their child received, comment on what they thought worked well and what was not helpful and make suggestions for improving services. These topics were covered in all sessions but the level of detail provided by each group varied depending on experiences, time available and the interests of those present. This section covers all four consultations drawing together the issues discussed into broad themes.

Diagnosis

This was an emotive topic for parents, all of whom could remember being told that their child had cancer or leukaemia with horrible clarity. They were, however, able to distinguish between a 'good' diagnosis and a 'poor' one. These were differentiated by the time it took to get a positive diagnosis and how, where and by whom parents and their children were informed. Participating parents had experienced both speedy and slow diagnoses. Some felt these had been handled with professionalism and sensitivity while others described how 'there

were eight or nine doctors in the room'. However, parents also recognised their own important role in the treatment of their child's cancer. One said:

> *'The context of the diagnosis is not that important. It is still going to hurt however you are told. It's who you are that defines what happens next.'*

Time taken

In some cases children were diagnosed on the day that parents took them to their GP while some parents described how they had been made to feel overprotective or neurotic by GPs before they agreed to send them to hospital for blood tests. Many cited delays in the process of undergoing tests and receiving a diagnosis. One mother described how she was told her child was 'lazy' and suffered from glandular fever and had to wait over two months for a correct diagnosis. In some cases it took some time to be referred to specialist services for tests and results of blood tests were sometimes slow in being returned to the GP.

Although immediate diagnosis was a huge shock to some (but not all) parents, being taken seriously by a GP who acted quickly and decisively was much appreciated. In some cases, parents were intuitive and guessed that their child had a serious condition but others found it very difficult to take in because the child seemed so well.

Rare conditions

The experiences of the few participants whose child had a rare form of cancer were particularly painful. One mother waited eight months for a positive diagnosis of her child's cancer after 'fighting' for a second biopsy as the first one undertaken was inconclusive. Another parent who had experienced similar frustrations said,

'I'm still mad at them. I still don't understand why they couldn't admit that they didn't know and consult with more experienced doctors.'

How parents and children were told about diagnosis

All parents found the experience of being told about their child's illness traumatic. It was described as 'a bombshell', 'a bad dream', 'hits you like a ton of bricks' and 'the world falling away'.

Some parents described being told about their child's illness in a private room with only one or two people present. This was preferable to being told in an open paediatric ward which some experienced but being asked to go 'to a quiet room where we can talk' implied bad news to parents. One parent said this was made more difficult by 'the nurse following me in with a box of hankies'. At all sessions parents talked about the emotional shock they felt in trying to get to grips with the diagnosis (even if it had already been suspected). Some were told 'straight' and many struggled to understand medical terms which one father described as 'Latin and Greek'. One described being 'summoned into a room where the consultant was surrounded by staff who weren't introduced'. There they were 'told the science and sent out. It was really awful'. This was some time ago, however, and the parent thought things have now improved. Parents appreciated staff who were approachable and offered to explain things more than once and who seemed to understand that shock made it difficult for parents to take in the details of what they were being told.

Another parent described how distressed hospital staff themselves were to discover that her child had cancer. They knew the child well because he had another condition and were 'upset for us'.

The early stages of treatment – which in many cases started immediately after a child was diagnosed – were also disturbing for parents. Parents described their shock at first going into the cancer ward where seeing 'children with no hair, grey

and ill' helped them take in what they were dealing with. Even seeing the sign saying 'Oncology' was described as a shock.

Parents had a variety of stories to tell about how their children were told they had cancer. Many of the children were very young at the time of diagnosis and parents told them themselves but appreciated staff who explained medical procedures to their children. One parent was advised to tell her child herself. One mother, however, said that her child had been 'stuck on a side ward and avoided' because staff knew that she had leukaemia and were avoiding telling her – perhaps because they were waiting for a more senior member of staff to arrive. Play specialists and other staff helped to explain what having cancer meant to children using drawings, story books, teddies or dolls. In one regional centre, the support offered in how to tell a child was described as 'great'.

Parents agreed that there is no 'correct' way of telling a child but that sensitivity, consideration and showing friendliness towards the child all help to make it easier.

Information and the internet (and see ***Meeting information needs*** on page 60)

Most parents were supplied with literature in the form of booklets from CancerBACUP and other organisations by hospital staff and found this useful although certain rare conditions were omitted from these publications. Many parents whose child had been diagnosed with cancer felt they needed as much information as they could absorb immediately in order to try to make sense of what was happening to them and surfed the internet to find out more. In some cases, medical staff discouraged them from using the internet, warning them that information available there might be inaccurate and anecdotal and that they might find themselves more distressed by an overload of information that they could not make sense of. In others, parents were encouraged to access particular sites which were recognised as reliable. Some staff offered to help

parents interpret information they found. Some parents found information on the internet was useful while others were frightened by it. A mother whose child had a rare condition was encouraged by the consultant to seek information from an American website which provided more detail than the equivalent British one.

The role of other parents

Some parents had found contact with other parents whose children had cancer unhelpful and even distressing at the time of diagnosis. They felt that these 'experienced' parents were too free with their (sometimes negative) advice at a point when they were unable to take in what was happening or too vulnerable to rebuff unwelcome approaches. Others, however, welcomed the support of other parents. One mother described other parents in the cancer ward as 'helpful, supportive and nice' and another parent said, 'the most helpful thing was talking to other parents in similar positions'. One couple described how they found that advice from other parents was useful particularly in relation to making an application for Disability Living Allowance (DLA). Parents at all consultations did, however, say that they had felt that they had needed to be pushy on some occasions on behalf of their child and that their child (and not other people's children) had to be their first concern.

Practical and emotional support

Some parents described practical support that was offered to them and their families at the time of diagnosis, particularly when treatment was urgent and based in another town. One mother appreciated the help of the chaplain in one hospital, for example, who rang her husband at work to tell him and offered a lift in his car. Some parents were offered advice about welfare benefits soon after their child's diagnosis. The timing of this advice, however, was sometimes inappropriate. When parents were trying to come to terms with their child's illness, they felt they could not cope with completing complicated forms.

Parents appreciated 'little personal touches' offered by staff which they felt made a difference to how they felt. They said they took comfort from the smallest examples of encouragement, kindness and 'bits of hope'.

Key points regarding diagnosis

- GPs and Accident & Emergency (A&E) departments should be well informed about symptoms of childhood cancer. Staff at health clinics and A&E departments should take seriously and be responsive to parents' concerns about their child's health in order to aid early diagnosis. Parents do not want to be made to feel neurotic or paranoid by doctors.
- If staff suspect that the diagnosis will be cancer but it has not been confirmed, parents would appreciate some warning which might prepare them for a definite diagnosis.
- When staff tell parents about their child's diagnosis, it should be done in a quiet, private place and there should be plenty of time available. Staff should be sensitive and honest and be prepared to answer questions. There is, however, no ideal or uniform way of 'telling'. All circumstances and individuals are different.
- Staff should be willing to provide information more than once as parents find it impossible to take everything in immediately. They should expect parents to use the internet, suggest reliable websites to visit and be prepared to help them interpret information found there.
- Parents would value access to a telephone number in the period immediately after diagnosis which they could ring for help - medical, emotional or practical.
- Some parents wanted their child to be present and involved throughout the initial diagnosis and for staff to explain it to them in accessible language. Others – particularly those of very young children - felt they wanted to tell their child in their own way. Individual preference should be respected.
- Parents would appreciate some warning about what the cancer ward is like before going there for the first time.

- Many parents suggested that a parent 'buddy' system based in the paediatric cancer ward would be helpful, particularly because nurses are very busy and parents sometimes feel isolated. Parents whose child was already being treated and who were willing could provide advice and support to those whose child was recently diagnosed. Parents of children who had been recently diagnosed would welcome the option to use such support but it should not be thrust upon them.

Hospital treatment and care

Location and access

Although some parents said they would prefer to use services that were closer to home than the nearest regional cancer centre, the majority also felt that care at the regional centre represented the 'gold standard' and preferred their child to be treated there. Regional centres were seen to have more staff in the oncology department and to have greater expertise in treating cancer patients. Some parents reported being prepared to travel much longer distances to ensure their child received the quality and continuity of care available at the regional centre despite the negative impact on other aspects of their lives.

Access to hospital services depended on where families live and, for those living far from regional care centres, could be difficult. One couple, for example, described the long drive from north Norfolk to Cambridge. Public transport is not a feasible option for children who are neutropenic and so access to affordable parking is important to families. One mother said she had no choice but to take her child to London via public transport regardless of the child's blood count or health status. Parents raised concerns about parents who cannot drive or who do not have access to a car as the cost of taxis is high.

Some parents described having to leave home early for appointments in order to find a parking space which 'added stress to an already stressful situation'. Parking half a mile away from the hospital was not feasible for a sick child so one

parent would drop off the child and the other parent (and possibly siblings as well) and then find a space. Parking could also be expensive. Some security staff allowed parents to park in the staff car park at one hospital but others were not prepared to waive the rules.

Parents in more than one centre said that outpatient appointments were not staggered and all patients were asked to arrive at nine a.m. This was difficult for those who lived far away who had to leave home very early and for all families because it meant travelling during rush hour.

Parents who had used hospital transport services had found them unsatisfactory. One reason is that transport is shared and parents are concerned about cross infection with other patients. There are also organisational problems. One mother described, for example, how she had used the ambulance service to go to hospital appointments an hour's drive from home. She and her child had to wait to be picked up and then the journey was extended as other patients were collected from a wide area. The ambulance was then ready to pick them up for the return journey before chemotherapy treatment was completed and there were further delays on the way home. She had since started driving into town for appointments, having got a disabled parking badge which allowed her to park easily. However, she was embarrassed about using the badge when her child was well enough to walk feeling that she was somehow cheating the system. There was a perception from parents at another centre that different wards worked to different protocols on access to transport and this could lead to delayed discharge from hospital.

The hospital environment

There was little discussion of the environment as parents were more interested in discussing staffing and treatment. However, parents in one centre were unhappy with aspects of the environment at the regional centre. This was not a dedicated children's hospital and they did not like the A&E department where at night there

were ‘disreputable types’ in the waiting room. They also objected to having to walk past smokers who hang around outside the hospital and having to wait in clinics with a neutropenic child where ‘all sorts of bugs and germs were being spluttered about’.

Some thought there was not enough space in wards although the cubicle arrangement available in some hospitals was liked because of the space and the privacy provided. Parents at one session described the wards as very clean. The wide age range of children accommodated in the oncology ward was commented upon and it was suggested that there might be a dedicated space for teenage patients. Parents at one session said they would have liked access to showers in the hospital and others said they would like a dedicated resource room for parents where they could learn more about cancer and cancer treatment. Some parents had used a suggestion box located in the ward and thought this was a good idea.

Getting a bed

Most parents preferred their children to be treated at a regional cancer centre (particularly in the early stages of treatment) and, at the centres, wanted them to be accommodated in the cancer ward which they regarded as the right place for them. They valued the specialist care and the contact with other families facing the same issues and were concerned that treatment in other wards or hospitals was not of as good quality as that available in the paediatric cancer ward. Because of caseload sizes, however, it was not always possible for children to have a bed in the cancer ward and parents found this frustrating and, in some cases, were unhappy with the treatment received in other wards. They described having to wait hours in A&E (where their child was exposed to others’ coughs and colds) for a bed when a child was ill and then being disappointed when they were eventually allocated to a different ward. Some parents said it seemed that some families were able to go straight into a ward when a child was sick while others had to wait in A&E and wondered whether this was an issue of protocol or

lack of communication. In one hospital, parents were also frustrated by the effect the lack of beds or a bed booking system had on treatment plans. If a bed was not available, treatment was put off which meant that the plan was not followed.

Continuity of care

As noted above, parents wanted their child to be treated in the cancer ward at the regional cancer centre where they and their child had built up relationships with staff and were confident about the care available. Some felt that staff in other paediatric wards did not understand the needs of their child and some had been offended by comments from staff in other wards who implied that they thought cancer patients were given special treatment which was not 'fair'. Some felt that other parents had not respected their 'space needs' and had generally felt uncomfortable in other wards. In some hospitals there simply is not the capacity to provide beds for all the paediatric cancer patients in specialist wards.

Parents said that the skill with which staff worked was an important aspect of treatment because if chemotherapy was administered well, for example, it caused less distress to a child than being carried out by someone who was inexperienced or clumsy. A shared understanding of policies and procedures for treatment by both staff members and parents was also highly valued by parents. Parents had had mixed experiences of care and treatment in shared care hospitals. Some felt that the nurses were not experienced or skilled enough in medical procedures and therefore did not inspire confidence in them or their children while others thought that shared care was well organised and that staff did have the necessary knowledge and would ring the regional centre for advice if necessary.

Parents reported hospitals having different protocols and techniques for, for example, changing a Hickman line and using different equipment (such as bungs) which caused distress to their children. They reported that the anti-emetic drugs offered to patients also varied between hospitals. Parents had

experienced a variety of advice about bringing newborn siblings into the ward. One mother said that she had been asked not to breastfeed her baby in the ward while another mother who attended another centre said breastfeeding had been welcomed.

Problems raised about the relationship between regional centres and their shared care partners included inflexible protocols that set down, for example, when a child with an infection had to be moved from one setting to another and poor communication and lack of note sharing between partners which resulted in parents – and in one case a child - having to take responsibility for knowing a child's current medication needs. Appointments were sometimes double booked for the same reason.

One parent felt that there was some competition between staff at the regional centre and shared care hospital which was unhelpful. More than one complained about a particular surgical ward where it was felt that there was a lack of understanding of children's needs and a reluctance to seek advice from specialist staff. According to one parent, this resulted in a child being left in pain for much longer than necessary.

Parents did not raise many instances of discontinuity in the specialist cancer ward although they preferred to see the same consultant which was not always possible. One said, 'he is always away. We never see him'. Parents reported that at one hospital the staff rotation system meant that registrars left the ward with no warning which led to lack of continuity.

In general, parents reported that they and their children found discontinuity stressful. Reasons given for this were difficulties in finding their way around unfamiliar hospital buildings, having to build relationships anew and learn to trust new members of staff, having to 'tell the story' again and again and getting used to different protocols and ways of carrying out procedures.

Parental involvement in care

Parents reported having limited choices about treatment because of inflexible protocols. Some children had been offered the choice of using a Hickman line or a portocath but others had not.

Parents appreciated it when staff acknowledged their own expertise although, in some cases, they felt that information was not shared with them because it was assumed that they already knew everything. Sometimes they felt that it was difficult to get staff to listen to them because their knowledge was not recognised. One parent, however, said:

'They tell us everything. We can't fault it.'

Parents did not appreciate having to be responsible for informing staff about their child's treatment. One mother described how, when her child was facing liver failure, a doctor asked her, 'how big is his liver normally?' Another was asked by a nurse with regard to medication, 'have I made this up right?'.

Although parents did become well informed about their child's care and were accustomed to monitoring machines, etc., most did not actually take responsibility for administering medication. One mother had been trained to take blood and administer chemotherapy and antibiotics by staff as her very young child was resistant to being treated by anyone else and other parents present admired her for taking such an active part in her child's care. They had not, however, been offered the same opportunities. Another mother had been asked to contribute to induction training for new staff at one of the regional centres and, although she was nervous about addressing a large group, appreciated being asked for her perspective and felt that the views of parents were valued.

Relationships with staff

In general, both mothers and fathers reported having good relationships with members of staff although they agreed that to some extent these depended on personalities and that there was an element of individual preference in developing relationships. In some cases, they felt dependent on a particular person, often a consultant, whom they felt could 'make things happen' when there were problems with their child's care. They trusted staff who were honest about their child's condition and appreciated the kindness, friendliness and thoughtfulness of nurses who would take time to comfort them if they were upset. One parent also said that hospital cleaners were also friendly and supportive to families and another described how staff were 'brilliant' when she was pregnant. Another mother said that as she got to know the personalities, ways and skills of different members of staff she was able to use this knowledge to her child's advantage – for example, she knew that the community nurse was more confident with a portocath than the hospital nurse and so made sure that she was the one to look after it.

Skills in actually treating children were valued and parents reported good treatment in some hospitals. They were not happy with trainee doctors 'poking around' in their attempts to insert a cannula. They felt that the child – who has become familiar with how procedures are carried out - should be listened to more. One parent said that her three year-old child had told a doctor to wash his hands before touching her line! The example of a consultant who had put in a cannula himself was unusual and welcomed by both parent and child.

Some had experienced rudeness and incompetence from doctors and nurses and thought that some nurses had showed favouritism towards some children and families. Others felt that their own knowledge and the fact that they were acting as advocates on behalf of their children had a negative effect on their relationship with doctors. One described being devastated when 'people don't listen and you have to fight to be heard'. Doctors' communication style could worry parents – one wondered, 'what does it mean when they just shake their

heads?' Others talked about the 'mega-egos' of some medical staff in teaching hospitals while others said they had not experienced egotism nor had they felt there were imbalances of power between staff and families. One, for example, was reassured by being given the mobile phone number of her child's consultant.

Relationships with staff may depend on how the child's treatment is progressing. One parent said that the attitude of staff might change if the child is not responding as expected and that this might create a barrier between staff and parents. Others had learned that less senior doctors erred on the side of caution about, for example, sending a child home whereas a more experienced doctor would consider all the child's needs and make a decision based on those. A holistic approach to caring for the child which takes into account his or her wish to lead a 'normal' life and family circumstances was welcomed.

Hospital facilities

Food

The choice, quality and cost of food available for both child patients and other family members were all raised by parents. As families had to travel to the hospital and, in many cases, stayed there, it was not possible for them to eat at home. Food was expensive in hospital and there was no discount available for inpatients' families and, because regional centres were based in town centres, it was not always easy to find a suitable shop to buy reasonably priced food.

Parents were concerned about the quality of the food provided for children saying that it was hard to provide a healthy diet. They also found it difficult to eat with their children without complicated organisation. They welcomed the snack trolley where this was available in the ward. Parents reported some positive experiences of staff being prepared to get hold of a particular food that a child craved outside normal eating times.

Parents complained that in one hospital the parents' kitchen was not conveniently located or secure and food was taken by others. The kitchen was not well-equipped or clean and no one took responsibility for it. Canteen facilities were also criticised for dull, badly prepared food although parents who had stayed at another hospital would have welcomed a canteen as there was only a snack bar which closed in the early afternoon. One mother said that she always brought food with her to hospital or went out to get it locally and found that the limited opening times of restaurants sometimes posed a problem.

The lack of easy access to cold drinking water was mentioned by parents at more than one session.

Telephone

Access to a telephone was a problem for some parents. The patients' phone service – Patientline – was described as good but expensive and not available by the child's bed. Some parents found phones by the beds where they could receive incoming calls only a better and more useful facility. In another hospital, phones were not available in the ward. Parents used the nurses' phone because the pay phone was outside the ward and felt that this arrangement was not ideal.

Accommodation for parents

There was little discussion about the availability and quality of accommodation for parents. One mother described how she shared her child's bed which was the child's wish. Although she enjoyed privacy she was woken in the night by nurses checking the equipment. Others had appreciated family accommodation provided close to the hospital.

Play facilities

Parents appreciated play therapy and play facilities and talked about 'wonderful' play workers. Some, however, said that playrooms were not open at weekends and, in some hospitals, toys were 'tatty' and broken especially those available on

other wards (i.e. not the cancer ward). One mother explained how they brought their own toys into the hospital. Parents whose children were very young at diagnosis said there were few toys available for this age group as did those whose children were aged between eight and 12. Parents thought that there were not enough Playstations or computers.

Hospital education

Many of the children who attended the consultation were very young and so had not been taught in hospital. At one session, mixed views were expressed about the necessity of specialist hospital teachers. It was felt that flexibility was important in assessing individuals' circumstances, capabilities and needs. Some parents felt that teachers had been too forceful in encouraging children to take part in lessons when parents felt they were too ill. One father said that he felt the teaching offered to his child was inappropriate given her age and condition but that it was offered in a friendly way.

Despite thinking that school work was offered initially when her son was too ill, the mother of an older child felt that education was of value as his health improved and that teachers and work engaged him which was a good thing. Liaison between the specialist nurse and secondary school was helpful when he returned as staff and students were informed about his illness (and his hair loss) and could anticipate his needs. A simple arrangement like being allowed to wear a hat in class made the return to school easier.

As there were few older children participating in the sessions, there was little discussion of school/hospital liaison. One family, however, described their child's school as 'useless' because staff there would not make contact with hospital school staff and another father said that his child had got behind at school because of illness but did not blame the hospital for this. It was also thought important that children kept in touch with their peers and this could be done through internet messaging.

Outpatient services

As noted above, outpatient appointments may be preceded by an early start, a long drive and frustrating parking. This might be followed by a long wait in a busy, cramped clinic. Not surprisingly, parents described outpatient appointments as stressful. Some did not understand the appointment system which seemed to book all patients at nine o'clock in the morning. Others said a new system with staggered appointments was being tested at one hospital but that it had not yet made a difference to waiting times. In some cases parents had been asked to arrive at the clinic as early as seven a.m. although their child was not treated until midday. This was seen as a particular problem if the child was not allowed to eat which happened when they were told that 'procedures' would be carried out before treatment. Delays can also occur because medical notes have not been updated.

In some hospitals there was a day unit attached to the ward and this was seen as a good idea. Parents would appreciate somewhere for children to play while waiting.

After the clinic appointment many had experienced waiting for hours for medication to arrive from the pharmacy. Parents thought that doctors did not realise how long this could be. One hospital had a pharmacy in the oncology department and medication was provided more quickly.

Key points regarding hospital care

- Access to free or affordable parking at or near the regional cancer centre.
- Better organised hospital transport.
- An appointment system which takes journey time into account and which does not mean long periods of waiting.
- More capacity at regional centres so that children can be accommodated on the specialist ward.

- A speedy, consistent and clear system for referring ill children to specialist wards.
- Parents would like access to showers in hospital and a dedicated resource room for educational and social purposes.
- Consistent quality of care across all hospital services including general paediatric wards.
- Consistent protocols for treatment across services.
- Opportunities for parents to be involved in children's treatment when appropriate.
- Staff to respect children and treat them accordingly by listening to them, consulting them and taking account of their wishes.
- A range of good quality, affordable hospital foods available to patients and their families. Some flexibility about mealtimes for children undergoing chemotherapy and other treatments. Ward snack trolley welcomed.
- Easy access to an affordable telephone service.
- Play equipment for a wide range of age groups.
- Sensitive, age appropriate teaching and access to educational resources including computers and the internet.
- Effective hospital/school liaison where appropriate.
- Quicker pharmacy service for both outpatients and inpatients.
- Parents appreciated the opportunity to talk about the care their child had received and suggested that more consultation days across the country should be organised which would bring together hospital management staff and parents.
- Suggestion boxes on wards were thought to be a good idea.

Meeting information needs

Parents said they wanted information about 'everything' although they were sometimes frightened by what they learned. Parents learned about cancer and its treatment from the consultant, community nurse, CancerBACUP, the UKCCSG,

the internet and voluntary groups such as the Leukaemia Research Fund. They had received booklets and fact sheets produced by CancerBACUP when their child was first diagnosed. Parents reported that sometimes information (for example, about the time scales for disease development) provided was not consistent. They were, however, positive about the way professionals explained their child's condition and treatment to them. Parents whose child had a rare condition were more likely to feel frustrated with the amount of information available to them.

Parents felt that they were less well informed about the emotional needs of their children and siblings and the need for emotional support for themselves. Some said they had not been told what the side effects of radiotherapy might be.

Accessing information from the internet was seen as both positive and negative. Although some parents had come across misleading information which they felt was potentially dangerous, those whose children had a rare condition found the internet particularly useful. Parents at one session suggested hospitals provide a parents' resource room where parents could access selected websites. This could also be used as a library of information and a place where parents could meet each other informally.

Initially parents felt that professionals were willing to provide as much information as they were able to take in – and repeat it if necessary - which they found helpful. However, a number of parents felt that as their child's treatment progressed and they themselves became involved with the child's health care, information was withheld from them because it was assumed that they were experts themselves and no longer needed it. They also felt that they were told bad news but were less likely to be informed when treatment was progressing well. They found it hard to 'read' blood or bone marrow test results because they did not have baseline information to compare them with so did not know if results were normal or not.

It was felt that information should be available at transitions in treatment and that perhaps there could be checklists available to help normalise adjustments to new aspects of treatment. Parents could be helped to support their child by being well informed about what was going to happen next.

The extent to which parents were provided with information about medication varied and it was felt that this should be consistent across centres. Parents felt they needed to know what effects to expect from medication so that they could effectively monitor their child's response. Some hospitals provided all parents with a loose leafed book which included useful telephone numbers, treatment plans and details of medication. These included sections that could be updated over time (such as blood test results) as well as space to record feelings and note down questions. The book was much appreciated and well thumbed copies were passed around.

Some parents felt that their own knowledge had had an effect on how they were treated by staff. One mother who was training to be a midwife felt that her medical knowledge had a positive effect as did another whose mother worked in an oncology department. Another, whose child had a rare disorder and who had to 'push' for a diagnosis, felt that her specialist knowledge gained from her own work was not respected or welcomed by staff.

Information for children

Parents felt that there was a lack of written information for children and siblings. The parents of children who had leukaemia were provided with (or given information about) a storybook but there was not something similar for other types of cancer. Parents also said that a book on having a 'wiggly' (central venous line) was helpful but that books on other aspects of treatment such as anaesthesia would be useful. Members of staff who drew pictures and took time

to explain to children in simple language what was happening to them were valued.

Sharing information

Children were seen by a number of health professionals both in the regional centres and shared care hospitals. Parents reported lack of communication and information sharing between professionals in different departments, wards and hospitals and described this as both frustrating and potentially dangerous. They described how medical notes and X rays had been lost and it was perceived that professionals 'don't talk to each other'. It is not possible, however, for parents to hold a copy of medical records as these are simply too big.

Parents thought that it was important that GPs were kept informed about a child's treatment which did not always happen. They felt that GPs could advocate on behalf of families when necessary and that, because childhood cancer had an effect on the whole family, GPs should be kept up to date with a child's progress.

It was also suggested that information and support should be available for other family members which would help take the burden of continually having to share information. It would be helpful if grandparents, for example, could access information from professionals.

Parents felt that there should be wider understanding of childhood cancer and its effects which might mean that in general people would be more understanding.

Death

Parents said that it was very difficult for all families when a child died on the ward. They felt that this was exacerbated by staff avoiding talking about it. They thought that this information should be shared sensitively and then parents would be able to talk about it without feeling that they were not allowed to.

Key points regarding the provision and sharing of information

- A wide range of information available on all aspects of childhood cancer including medical, treatment, medication and its effects, emotional, practical and financial.
- Opportunities to ask questions and have information repeated.
- Advice on reliable websites and how to use information from the internet effectively.
- Regular updates on child's progress.
- Interpretation of information about treatment and sharing of positive progress with parents.
- Information in advance especially at points of transition (e.g. before changes in treatment regime).
- Consistent information about medication and its effects.
- More written information for children and siblings including storybooks for very young children.
- Acknowledgement and respect for parents' own knowledge and skills.
- Clear lines of communication between professionals across departments, hospitals and community services including GPs.
- Information available for other family members such as grandparents.
- Wider understanding of childhood cancer.
- A sensitive approach to sharing information about the death of a child.

Community and home care

There was wide variation in participants' experience of receiving home care. Some families had not been assigned a community nurse while others found home services hugely valuable. One mother said she had not used the community service until the end of her child's treatment because she did not know about it. Others had had few visits from a specialist outreach nurse because they lived so far from the regional centre.

CLIC and Macmillan nurses and the 'Diana team' were described as providing excellent services. They treated children at home, liaised with other services such as physiotherapy, offered practical advice to parents, helped access equipment and visited schools to discuss children's health with school staff and fellow students. One mother received support with antenatal care when she was struggling to cope with spending time with a child in hospital and attending appointments herself.

These nursing teams were described as having 'emotional intelligence' which was defined as being understanding, approachable, actively sympathetic and working well together as a team. These qualities were highly valued by parents.

Parents reported a range of experiences with GPs too. As noted above, they felt that GPs should be kept 'in the loop' with information about their child's health, treatment protocol and progress and some valued GPs' role in caring for the whole family and for advocating on their behalf when they experienced problems. Some parents, however, preferred hospital care as they felt 'safer' in secondary care than they did with non-specialist services.

Key points regarding community and home care

- Consistency in the availability of home care services.

Family support

Parents talked about the huge effect cancer and cancer treatment has on the whole family and how normal life is put on hold for the period of treatment. Some suggested that parents need support to make decisions about all aspects of their lives including work, finance and other practical issues as they are so overwhelmed with immediate concerns. Sometimes they felt they needed to be reminded that there is life outside the hospital.

Benefit advice

The availability of advice about welfare benefits was raised at all sessions. In many cases parents felt that they had not been well informed about their eligibility for Disability Living Allowance (DLA), travel allowances and special equipment or, if they had, the advice had been offered at an inappropriate time. Many had received the most useful advice from other parents who had already made an application for benefits. There was inconsistency about the availability of professionals able to provide help with form filling and a feeling that the application process was a lottery. The advice of Sargent social workers was welcomed but these were not available at all hospitals. Nurses in some areas were willing to fill in forms but not all were well informed about how to make a good case.

Parents who had been refused DLA and then reapplied felt humiliated by having to 'prove' how ill their child was. One family had approached the Citizens Advice Bureau because there was no advice available at the hospital.

Siblings

Parents said that there were few services provided for siblings. Some said they would like siblings to have the opportunity to talk to each other. They also wanted advice on behaviour management for their child's siblings. One hospital provided outings for siblings of children who were undergoing treatment and this was much appreciated.

Voluntary groups

One couple said they were involved with a local support group for families and set up by parents. The group raised funds to help with equipment (including pushchairs), parking costs, etc.

Stress

Parents talked about the stress of having a child with cancer. This is caused by constant worry as well as the practical issues of spending most of the time in

hospital while also caring for other children. They said that financial difficulties and pressures from employers exacerbate stress.

There seemed to be regional variations in the availability of social work and psychological support. Some parents were uncomfortable about using social work services because of the stigma it implied for them. It was suggested that hospital social workers are given another title. One mother said she found seeing a psychologist essential while others had not been offered a service. However, parents reported that nurses provided support although they did not always have time. Parents in one hospital had taken part in research on stress and how families cope with childhood cancer and felt that the needs of all family members should be taken into account.

Parents talked about the difficulties they faced. One said she felt like she was permanently living under a cloud. One hospital provided aromatherapy massage sessions for parents one day a week. These were appreciated for giving parents the opportunity to relax but they also liked feeling they were being given 'permission' to take a break from caring for their child. Others had been offered antidepressants by GPs when they felt that what they needed was time to themselves (just to cry or think) and practical support such as child care for their other children, help with housework and someone to talk to.

Key points regarding family support

- Consistent, freely available advice about welfare benefits and access to specialist equipment.
- Support for siblings of children with cancer which could include opportunities to meet each other.
- Easy access to psychological and family support.
- Sensitive, flexible support which could help with practical solutions, emotional support or financial advice.

9. Conclusion

One of the most significant issues raised by the consultation was the difference in quality of services perceived by participants between services available at regional cancer centres and shared care hospitals. This was mentioned by many (but not all) children and parents who both expressed their preference for the care received in regional centres. Parents were prepared for the extra travelling involved in attending a regional centre despite the difficulties this caused but both children with cancer and their siblings were unhappy that the hospital was far from home. The reasons that participants preferred the regional centres were because they felt that staff were better informed and that treatment was more effective and less likely to be painful than in shared care. They also appreciated the resources available and culture of care developed at these centres which included the advice of experienced consultants and senior nurses and support from social workers and psychologists. The specialist nature of the centres means that participants felt they were getting 'the best' services and, having experienced it, were disappointed by the quality of care on offer elsewhere. It may also be that they were initially referred to the regional centre after diagnosis and preferred not to use other services for less tangible, more emotional reasons. One mother, for example, said that her child was not known by name on general paediatric wards either in the regional centre or in her local shared care hospital.

Although children and their parents were positive about the care they had received in general and many were full of praise for hospital staff, another finding of the consultation was that staff sometimes fail to take children's views into account. As can be seen from the consultation's findings, even very young children have an acute awareness of what is going on around them and how they are being treated and young cancer patients should be given the opportunity to discuss their needs and feelings with staff as well as to be informed about their illness, treatment and how it will affect them. Parents' knowledge of their

children's condition and their skills in caring for them should also be acknowledged and valued by staff and they too should be informed about all aspects of their child's treatment and care so that they can actively co-operate with it.

Children at all consultation days said that they get bored in hospital and wanted more and better entertainment. They wanted toys, games and books as well as electronic equipment including Playstations, DVD players and television. Play specialists were much appreciated by children and parents for providing both opportunities for play and accessible information about cancer and cancer treatment to young patients. Participants said that school work had not been appropriate for their age and ability and would prefer to be challenged more by hospital teachers. Parents felt that schools could do more to liaise with hospital staff to support the education of children in hospital.

Food was mentioned by both children and parents. Some complained that hospital food was not to their taste and poorly prepared while parents found eating in hospital could be expensive or difficult because of lack of catering facilities. Children wanted a range of different foods with some flexibility about mealtimes and the opportunity to eat with family members at least some of the time.

Children with cancer, their siblings and parents want consistent, detailed and accessible information about cancer and cancer treatment and for this to be available to siblings and other family members. Children wanted more understanding about cancer amongst peers and the general public in order to normalise their experience as much as possible. Effective hospital/school liaison would support this. Parents' lives are completely changed when their child is diagnosed with cancer and they need access to financial advice as well as practical and emotional support and community health services. Although these are provided in some areas, participants reported that these were not consistent.

Some had had no access to any of these services while others praised them for their helpfulness.

Glossary of terms

A&E: Hospital Accident and Emergency department

DLA: Disability Living Allowance

GDG: Child and Adolescent Guidance Development Group

NCC-C: National Collaborating Centre for Cancer

NICE: National Institute for Clinical Excellence

List of appendices

* All Appendices are available from the NCC for Cancer on request.

APPENDIX E

Teenage Cancer Trust Conference 2004

Analysis of Teenage Cancer Patient Questionnaire Responses

The following is a summary of the responses to an electronic questionnaire carried out at the TCT Conference in 2004.

Demographics

There were up to 271 Teenage and Young Adult (TYA) respondents, aged 14 to 23; 71 % of which were 14 to 18 years old, with 49% male and 51% female (n=271).

22% were 12 or less and 68% were 13 to 18 years old, at start of treatment (n=203).

12% came from Scotland, 33% from the North of England, 26% from the Midlands and 15% from the South East and London. There were few respondents from the South West and Wales (n=261 total).

Diagnosis

Lymphoma	14%
Hodgkins	13%
Leukaemia	30%
Bone cancer	21%
Brain cancer	11%
Testicular cancer	2%
Skin cancer	0.5%
Bowel cancer	1%

Soft tissue sarcoma	4%	
Rabdomyosarcoma	3%	
Breast cancer	0.5%	
Cervical cancer	0.5%	(n=205)

N.B. The incidence of these distributions should be qualified by the likelihood of survival to becoming a TYA from diagnosis, particularly with brain tumours.

Referral and Treatment

Question: Did you visit your GP and, if so, how many times before they referred you to the hospital?

1-2 times	43%	
3-5	28%	
More than 5	29%	(n=193)

17-40% (mean=29.7%) of patients approximately evenly across the age range 14-22 required more than 5 GP visits before referral, with 64% (n=11) of 23+ year olds.

16-35% of haematological cancers require more than 5 GP visits, with considerable variation between leukaemia and Hodgkins lymphoma. E.g. only 15.8% (n=57) of leukaemias are diagnosed with more than 5 visits, as opposed to 34.6% (n=26) of Hodgkins. Furthermore, 52.6% of leakaemias, but only 26.9% of Hodgkins are diagnosed within 2 visits. Lymphoma (n=25) shows intermediary data.

60% (n=15) of brain tumours, 31% (n=39) of bone cancers, 43% (n=7) of sarcomas and 40% (n=5) of rabdomyosarcomas, require 5 or more visits. In addition, only 20% of brain tumours, and 26.9% of Hodgkins, are referred within 2 visits, compared to e.g. 40% of lymphoma and 57.1% of sarcomas.

There is little variation between areas of the country.

Interpretation: Childhood cancers, especially Hodgkins lymphoma, brain tumours and sarcomas, are slow to be identified in primary care.

Question: Were you on your own (12%) or with a parent (88%); n=205.

Question: How long after you first visited hospital did it take for someone to tell you that you had cancer?

Less than 2 weeks	60%	
3-4 weeks	14%	
1-2 months	14%	
3-5 months	5%	
6-8 months	3%	
Longer	5%	(n=212)

80% of 14 year olds know they have cancer within two weeks of visiting the hospital.

25-68% of 15+ year olds know within 2 weeks.

80% of leukaemics know within 2 weeks, but only 25-57% of lymphomas, Hodgkins, bone, brain sarcomas know in 2 weeks.

Generally, 48-62% of patients in the areas know within 2 weeks, with some areas reaching 82-100%.

40% of brain tumours, 33% lymphoma, 34.6% Hodgkins and 25% of bone cancer patients know in more than 4 weeks, with leukaemia at only 10% not knowing within 4 weeks.

Patients knowing in more than 4 weeks varied from 15.4% in London and Home Counties, to 43.3% in the North East.

Interpretation: *There is wide variation in time to confirmed diagnosis of most childhood cancers in secondary care, both in respect to cancer type and area of the country.*

Question: Who told you that you had cancer?

GP	6%	
Hospital Doctor	70%	
Nurse	3%	
Mum	15%	
Dad	6%	(n=212)

Hospital Doctors inform the patient they have cancer 54-83% of the time, with Mum the next frequent, particularly with leukaemia and brain tumours (24 and 36% respectively).

Question: Were you told you had cancer before or at the same time as your parents?

Before	10%	
After	40%	
At the same time	50%	(n=214)

Question: ***Once they told you that you had cancer, how long was it before you started treatment?***

1-2 days	32%
3 days - 2 weeks	44%
3 weeks – 2 months	20%
3 – 4 months	2%

Longer	2%	(n=207)

13-25% (mean= 16.4%; n=137) of patients aged 14-18, started treatment more than 2 weeks after diagnosis; compared to 8-50% (mean= 36.3%; n=58) of 18+ year olds.

Only 6% of leukaemics had not started treatment within 2 weeks, compared to 20-38% of other cancer types.

Scotland, NW England, NE England, E Midlands, London and South England had between 17 and 34% of patients not starting treatment within 2 weeks of diagnosis, whereas in other areas, between 0 and 9% are not treated within 2 weeks.

Interpretation: *There are a significant proportion of patients who do not start treatment within 2 weeks, especially in the 19 to 23+ year olds, and this varies from area to area.*

Question: After you were told that you had cancer, how much help did you get to help you understand what was happening?

Everything I needed	48%	
A bit more than I expected	15%	
What I expected	20%	
A bit less than I expected	12%	
Little or nothing	5%	(n=204)

Question: What choices, if any, were you given for treatment options?

All choices were given	19%	
Some choices were given	25%	
No choices were given	56%	(n=206)

No choice was given in 39-67% of 14-18 year olds, and in 25-86% of 19+ year olds.

Between 33% (rabdomyosarcoma) and 62% (bone cancer) of tumour types offered no choice of treatment.

Across the health regions, between 30 and 67% of patients had no treatment choices.

Interpretation: *There is considerable limitation of choices of treatment in childhood cancers.*

Question: Did you have the choice of entering a clinical trial?

Yes	43%	
No	57%	(n=197)

40-66% (mean= 53.1%) of 14-18 year olds did not have the choice to enter a clinical trial, with 25-86% (mean=64.3%) of 19+ year olds.

48% (bone) to 84% (brain) of patients were not offered a clinical trial.

Variation was essentially independent of the area of the country.

Interpretation: The availability of trials for TYAs is/has been variable.

Question: Did you feel that you were involved enough in making decisions about your treatment and any options that were open to you?

Always	24%
Most of the time	29%
Some of the time	24%
Not much of the time	13%

Never	11%	(n=208)

Between 34 and 75% of all ages were involved all or most of the time in decision making about treatment.

There was little variation across cancer types ranging from 45 to 62% being involved all or most of the time.

There was little variation across areas of the country (43-69%).

Question: What type of treatment did you have?

Chemotherapy	32%	(n=202)
Surgery	28%	(n=173)
Radiotherapy	16%	(n=98)
Other	25%	(n=157)

Where treated

Question: Where did you receive most of your treatment?

A Main Cancer Hospital	49%
A Local Hospital only	13%
A Main Cancer Hospital and a Local Hospital	38%

36-48% (mean= 41.7%; n=132) of 14-18 year olds were treated at both a local hospital and a main cancer hospital, with 40-70% (mean= 24.1%; n=58) of 19+ year olds being treated at a main cancer hospital only.

29-65% of all cancer types, with 29.1% Hodgkins, 30.9% bone, 42% leukaemia and lymphoma and 65% brain, being treated in both local and main cancer hospitals.

Scotland, NW England, E Midlands, E Anglia, South Wales and London and Home Counties had 26-35% patients treated in both local and main cancer hospitals, whereas NE England, W Midlands and North Wales had between 50 and 64% of patients treated in this way.

Interpretation: *There is considerable variation in providing shared care treatment, based on cancer type and area of the country.*

Question: When you were in hospital for your treatment, were you usually treated on a:

Children's ward	53%	
Adult ward	11%	
TCT ward or other adolescent unit	22%	
Children's ward and adult ward	2%	
Children's ward and TCT or other adolescent ward	6%	
TCT ward and adult ward	6%	(n=208)

Question: Was the Cancer Centre ward or day care facility that you were treated on suitable for a person of your age?

Completely	38%	
Mostly	26%	
Partly	20%	
Hardly	12%	
Not at all	4%	(n=194)

Across all ages, the ward was completely or mostly suitable for between 50 and 70% of patients.

Hodgkins and rabdomyosarcoma (42 and 40% respectively) were completely or mostly suitable least, with lymphoma (55%), leukaemia (60%), bone (81%), brain (70%) and soft tissue sarcoma (71%) suitable all or most of the time.

Regional suitability all or most of the time varied between 39% and 46% (E Midlands and E Anglia) to 70% (W. Midlands) and N and S Wales 100% suitable all or most of the time, with other areas in this last group.

Interpretation: *There is considerable variation in suitability of treatment facilities for TYAs and at least 30% of TYAs are treated in facilities that are less than suitable.*

Question: ***If you received any of your treatment at a local hospital, was the ward you were treated on suitable for a person of your age?***

Completely	25%	
Mostly	18%	
Partly	18%	
Hardly	18%	
Not at all	22%	(n=159)

Between 30% and 50% of local hospitals were ages suitable for all or most patients, with 17 to 86% of 19+ patients treated on suitable wards.

Between 20% (Hodgkins) and 57% (bone) of local hospitals were suitable all or most of the time.

In most areas, local hospitals were suitable all or most of the time for 34% to 50% of the time, but North Wales (100%), S Wales (0%) and South England (17%) were outliers for suitability of local hospitals.

Interpretation: *There is considerable variation between local hospitals in suitability for some cancers and in some areas of the country.*

Question: When you received your treatment, did everyone on your ward have cancer?

All	56%	
Mostly	30%	
Partly	9%	
Hardly any	4%	
Only you	1%	(n=193)

Between 79% and 100% of patients of all ages were treated on wards that had only, or mostly cancer patients.

All tumour types were at least 86% cancer patients on the ward, except brain (65%), soft tissue sarcoma (66%) and rabdomyosarcoma (80%).

Most areas of the country had over 92% of patients on cancer or mostly cancer wards, except Scotland (68%) and London and Home Counties (79%).

Interpretation: *There* are outliers in terms of cancer type and region, in the inclusion of non-cancer patients on the same treatment wards.

Question: How long would you be prepared to travel for your treatment?

Up to a couple of hours	37%	
Half a day	8%	
A day	3%	
Travel needing overnight stay	3%	
Any distance any time	50%	(n=189)

Question: If you had a choice about the environment you were treated in, where would it be?

Adult ward	10%
Teenage and Young Adult ward	90%

Children's ward	9%	(n=201)

Question: Were you treated in a single sex area?

Yes	17%	
No	83%	(n=190)

Question: Which would you prefer?

A single sex area	8%	
A mixed sex area	92%	(n=200)

Interpretation: *TYAs prefer mixed sex wards.*

<u>Support and Information Needs</u>

Question: During your treatment, has there been a particular member of staff that you could confide in about your treatment and any concerns you had?

Yes	87%	
No	13%	(n=191)

Question: What was there role?

Activity co-ordinator	7%	
Cleaner	4%	
Doctor	13%	(0-27% across the age range)
Specialist nurse	23%	(9-67% across the age range)
General nurse	31%	(14-53% across the age range)
Physiotherapist	1%	
Play therapist	5%	
Religious representative	1%	
Social worker	7%	
Teacher	1%	

Other	1%
None	8%

Psychiatrist, Psychologist, Radiographer and Volunteer did not score.

Interpretation: Nurses are key communicators and supporters of TYAs with cancer.

Question: How important was it to have a staff member to help you stay occupied with activities, education and interests?

Essential	28%	
Very important	29%	
Quite important	26%	
Not that important	12%	
Unimportant	5%	(n=185)

Question: Was the written or audio information given to you about your cancer easy to understand and suitable for your age?

Aimed at people older than me	27%	
Aimed at people my age	55%	
Aimed at people younger than me	18%	(n=172)

Between 53% and 70% of 14-17 year olds thought the information was directly suitable for their age, with between 20 and 57% of 18+ year olds recording similarly.

Over 57% of most cancer type information was considered suitable for the patient's age, except for testicular cancer and soft tissue sarcoma where the direct age suitability was 33% for both, and more suitable for older patients.

The level of suitability of the information for the patients in the areas of the country was between 46 and 75%, with Scotland at 35% and South England 83% as outliers.

Fertility Counselling

Question: Were you provided with fertility counselling?

Yes	34%	
No	66%	(n=184)

Question: If counselling was given, was it?

Before treatment	29%	
During treatment	38%	
After treatment	33%	(n=100)

Question: Were you satisfied with the counselling?

Yes	52%	
No	48%	(n=110)

Question: Were you told about the risks to your fertility before you started treatment?

Yes	64%	
No	36%	(n=171)

Interpretation: The majority of TYAs did not receive fertility counselling, and when they did, it was not consistent in terms of timing and quality. If this is a reflection of the provision of fertility support and treatment, then it is far from satisfactory in all respects.

Life Issues

Question: What is the most challenging issue you face with cancer today?

Communicating about my illness	20%	
Returning to school	16%	
Dealing with my appearance	26%	
Managing the side effects of cancer	38%	(n=183)

30.7% of leukaemics, with 52.2% of lymphomas, as opposed to 40.9 to 45.7% of Hodgkins, bone and brain, cited managing the side-effects of treatment as the most challenging issue.

Interpretation: Late effects of treatment are a significant burden for many TYAs.

Question:	***Do you have difficulty with?***	
Travel insurance	Yes	41%
	No	20%
	Don't know	39%
Life insurance	Yes	8%
	No	11%
	Don't know	81%
Medical insurance	Yes	16%
	No	11%
	Don't know	73%
Mortgage	Yes	5%
	No	5%
	Don't know	90%
Bank account	Yes	13%

	No	47%	
	Don't know	40%	
Obtaining a place at school	Yes	14%	
	No	56%	
	Don't know	30%	
Obtaining a place at University	Yes	6%	
	No	23%	
	Don't know	71%	
Obtaining a job	Yes	34%	
	No	26%	
	Don't know	40%	(n=168-177)

Interpretation: *There is a notable level of discrimination against TYAs with cancer in terms of insurance, access to education and employment, particularly when adjusted for the numbers who respond yes or no, assuming that the don't knows were age excluded.*

Question: Do you feel that the National Health Service does enough for teenagers with cancer?

Yes	25%	
No	53%	
Don't know	22%	(n=220)

APPENDIX F

Position paper on Compliance

Geraldine Mynors, Head of Projects
Task Force on Medicines Partnership

1. What is known about non-compliance with cancer therapy amongst children and adolescents?

Prescribed medication is the most common from of therapeutic intervention in child and adolescent oncology. New and more effective treatments are constantly being introduced, with an emphasis on developing orally administered agents and moving away from intravenous therapies where possible. Using medicines to best effect is therefore of critical importance in successfully managing child and adolescent cancer. In the past, most attention has been devoted to guiding treatment decisions rather than involving patients in these decisions and monitoring whether the medicines selected are actually taken as prescribed. However, until issues of medicine *taking* are addressed, as well as questions of what to prescribe, a significant proportion of drugs will be wasted and the potential therapeutic gain envisaged by NICE in drawing up its guidelines will not be realised.

The literature on medication compliance in cancer patients is limited, because most treatment is administered in hospital under the direct supervision of health professionals. However, with increasing use of oral therapies, the issue of compliance may become more important in the future. Currently, most research on compliance in cancer patients has been conducted in the context of clinical trials, and uses dropout rates as the measure of 'compliance', which is problematic. Measures and definitions of compliance vary widely between studies. Nevertheless, a review of published studies of compliance in cancer therapy by Partridge et al (2002) review revealed poor compliance in the

paediatric oncology population (see Figure 1 below). The review showed that adolescents were the least compliant cancer patients.

Figure 1: Non-compliance rates to oral antineoplastic agents in paediatric populations

Type of cancer	**Measure of non-compliance**	**Definition of non-compliance**	**Non-compliance rate**	**Number of patients in study**
Leukaemia or non-Hodgkin's lymphoma	Level of drug metabolite in urine	Lower than expected levels	33%	52
Leukaemia, Hodgkin's disease, non-Hodgkin's lymphoma, other malignancies	*Self-report and parent report* Serum bioassay	More than one missed dose per month Not described	35%	46
Hodgkin's disease, acute lymphocytic leukaemia (ALL)	Biological markers	Lower than expected levels	50%	50
ALL	Level of drug metabolite in	Less than expected level	42%	31

	urine			
ALL	Level of metabolites in blood	Less than expected level	10%	327
ALL	Level of metabolites in blood	Less than expected level	2%	496

From Partridge et al (2002)

Research into non-compliance more generally has shown that as much as 50% of medicines for long term conditions are not taken as prescribed (Haynes 2002), and that non-compliance rates can be high even with medicines which are seen as being 'life-saving'. This area, therefore, deserves attention within the NICE guidance being produced.

2. Reasons for non-compliance

The most recent systematic review of compliance by McGavock and colleagues (1996) showed that most non-compliance is almost always the result of conscious choices made by patients rather than simply 'forgetfulness'.

A large variety of factors are predictive of, or associated with, non-compliance with medication regimens (Carter and Taylor 2003). They include:

- Demographic indicators (e.g. age, gender and socio-economic status)
- Medication characteristics (e.g. side effects, complexity of regimen)
- Psychosocial issues (e.g. social support, family functioning, self-esteem).

However, the most consistent predictor of compliance appears to be individuals' attitudes, beliefs and perceptions about their illness and treatment. Horne & Weinman (1999) reported a study which linked patients' beliefs about medication

to compliance. Specific beliefs about particular medications include whether the medication is perceived as necessary for maintaining health, and concerns about adverse consequences such as side effects or becoming dependent.

The authors looked at whether beliefs affected compliance in four different chronic illness groups (people with asthma, cardiac conditions, and renal failure demanding haemodialysis, and oncology patients). They found that specific beliefs about medicines were the strongest predictor of compliance, accounting for 19 per cent of the observed variance. Demographic variables were less significant. Patients who believed that their medication was necessary for good health reported a higher rate of compliance, whereas those who had more concerns about medicine use reported poorer compliance. This study highlights the importance of exploring and addressing patients' beliefs about medication when addressing compliance issues.

These observations are consistent with the limited research which has been done on compliance amongst children and adolescents with cancer. According to Spinetta et al (2002) the reasons for refusal, non-compliance and abandonment of anticancer treatment in children and adolescents include:

- physical discomfort;
- misunderstanding and uncertainty about benefits of medication;
- poor communication regarding diagnosis and regimen;
- frustration with length of treatment;
- fear of side effects; and
- poor understanding of the seriousness of the illness.

The review by Partridge et al (2002) showed that those most at risk tended to have a poorer understanding of their illness than their peers, and to have less perceived vulnerability and higher levels of denial compared to those who were compliant. The relationship between parental involvement and compliance also appeared to be important.

3. The concordance model as the basis of service improvements to improve compliance

Past approaches to improving compliance have focussed on cues and reminders. More recent approaches have instead tried to implement 'concordance' - a two way process of prescribing that recognises that patients are not the passive recipients of prescribing decisions, but have their own views about their condition and treatment. Numerous studies in adults have shown that patients' beliefs and views about medicines are a key influence on whether and how they take them. Patients are much more likely to follow treatment if they have been active partners in prescribing decisions and their views and preferences have been recognised and taken into account (Cassileth 1980). This in turn is only possible if they have sufficient information and understanding about the medicines available to them.

Three elements need to characterise the health system if concordance is to be achieved (see Figure 2 overleaf).

For children and adolescents with cancer, some of the practical aspects of concordance which should be implemented within services are as follows:

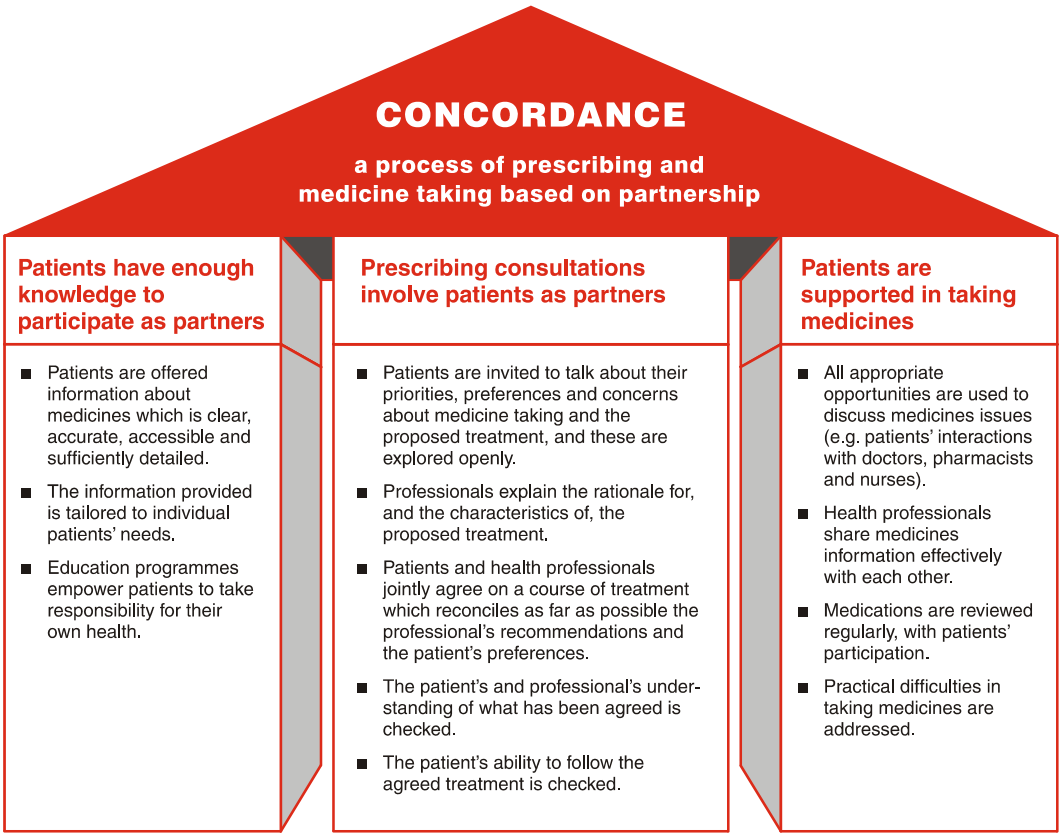
(i) Patients have enough knowledge to participate as partners

Information about different treatment options must be made available both to patients and their families in accessible formats. Patient information leaflets have been shown to be too narrow, too negative and too late to help patients understand treatment options (Raynor et al 2004). The available patient information can be sometimes be particularly unhelpful for children where medicines are prescribed outside their licensed indications, and parents and children need particular help to understand what this means.

Information for patients and families should make clear the potential benefits, but also the risks and side effects associated with treatment, so that these can be fully explored in the prescribing consultation. Such information should also be 'user tested' with real patients to assess whether it meets their needs and what they would like to hear about. (Dickinson 2001)

Patients often appreciate hearing directly from peers who have been through similar experiences, and this can be achieved either by linking up patients on a one-to-one basis or in small groups, or through initiatives such as DIPEx (the Database of Individual Patient Experience), a web-based collection of video clips of patients describing their experiences – which is currently only available for adults but could be extended to children and young people.

Figure 2



(ii) Prescribing consultations involve patients as partners

Consultations between health professionals and patients should explore the extent to which patients want to be involved in treatment decisions and allow them this degree of involvement. In particular, patients should be given the opportunity to discuss their own priorities and concerns within the conversation. Research has shown that many patients do not voice their agendas and concerns during consultations unless these are explicitly brought out. Tools and techniques for doing this should be used where helpful – for example in adults, significant improvements in patient experience have been achieved by using patient self-completed agenda forms prior to consultations (Barry 2000). Similar approaches could be tested with younger patients – perhaps using pictures or simple diagrams with younger children to help them to express their views.

Once patients have voiced their concerns and beliefs, it is essential that this information is shared between different members of the multi-disciplinary team so that all members can work together to build the trust and knowledge of the patient and his or her family.

It is recognised that there are ethical issues surrounding the extent to which a child or young person below the age of 16 can decide to decline treatment considered to be 'optimal', and therefore the extent to which negotiation about prescribing decisions is possible. However, it must be recognised that failing to involve patients fully in prescribing decisions is likely to lead to poorer compliance and poorer outcomes.

Two way communication of this nature is difficult, and it is likely that many practitioners will need additional training and/or opportunities to reflect on consultations in order to review and improve their practice.

(iii) Patients are supported in medicine taking

Once a prescribing decision has been made, it is important that patients are not simply left to take, or not take, the medicine, but have access to ongoing support and additional information, should they need it. For many people, it is only when they start using a new medicine for them that concerns and issues arise – for example if it gives rise to side effects, they don't feel that it is 'working', or the Patient Information Leaflet raises concerns. Ready access to someone at the end of a telephone who can answer questions – even out of hours - and offer support is vital, and in this area hospital pharmacists and nurse specialists can often play a particularly valuable role.

In adolescent medicine, new approaches to ongoing support for patients taking medicines are currently being tried out using new technology such as SMS messaging and by e-mail, focussing on Type 1 diabetes (see http://www.diabetes.org.uk/good_practice/innovative/examples/innovate1.htm). In reviewing such innovations, Medicines Partnership has found that they are most likely to be successful when they offer the opportunity of a two way conversation with someone who the patient knows – rather than being just one way 'prompts' to aid compliance. Nevertheless, these kinds of approaches, which tap into technology with which most young people are now very comfortable, could be a useful adjunct to face-to-face support.

In summary, then, young people with cancer are in particular need of appropriate care and support to enable them to get the most out of medicines. There is evidence that people who are really aware of the risk that cancer presents to them and the possible benefits of treatment are often willing to persist with therapy, despite sometimes unpleasant side effects. Involving patients and their families fully in decisions, proactively offering them opportunities to discuss their fears, concerns and expectations, and supporting them on an ongoing basis through the whole multi-disciplinary team are all important. Approaches which

allow individuals to feel confident and make informed decisions in what may be testing personal circumstances are more likely to promote desirable outcomes

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Background on the Task Force on Medicines Partnership

The Task Force on Medicines Partnership is a Department of Health funded programme, which aims to help patients to get the most out of medications by involving them as partners in prescribing decisions (including those decisions where an informed patient decides to decline the treatment offered) and supporting them in medicine taking where the decision is to accept treatment. The Task Force is a truly multi-disciplinary collaboration of 25-30 members involving doctors, pharmacists, nurses, patients, the NHS, the pharmaceutical industry and academics, supported by the Medicines Partnership Centre, an executive team carrying out the programme of the Task Force. Medicines Partnership was set up at the beginning of 2002 as part of the *Pharmacy in the Future* programme under the NHS plan. More details are available at www.medicines-partnership.org.

APPENDIX G

Position Paper for the Guidance Development Groups Work on Child and Adolescent Cancer in the Specialist Area of Blood and Marrow Transplantation

Dr Paul Veys
Consultant in Charge of BMT

Between 300 and 350 blood and marrow (BMT) procedures are performed on children and adolescents under the age of 18 years in the UK each year. About 2/3 of these procedures are for haematological/oncological malignancies. The number of procedures has remained fairly static over the last ten years. Work is carried out in 19 centres throughout England and Wales. All centres perform autologous transplant procedures whereas 13 also carry out allogeneic BMT procedures. Over a ten-year period 6 of the transplant centres performed over 200 procedures, and to give an idea of the geographical provision of services these major centres are Bristol Children's Hospital, Birmingham Children's Hospital, Great Ormond Street Hospital for Children in London, Manchester Children's Hospital, Newcastle General Infirmary and Royal Marsden Hospital in Surrey. Together these 6 units perform more than 2/3 of the BMT procedures in the UK. The allogeneic work within the UK is closely monitored by the United Kingdom Children's Cancer Study BMT sub-group.

This group is made up of representatives from all the major BMT centres in the UK. It is mandatory that all BMT procedures performed in the UK are reported to this group. The annual report for 2003 from this group illustrates the overseeing activity performed currently by the group (***Enclosure 1****). Most autologous procedures are performed at UKCCSG cancer centres and within National/International collaborative studies, but this work is not as closely

monitored, as for allogeneic procedures, by a reference group,. This is currently being addressed within the general body of the UKCCSG organisation.

The UKCCSG BMT Group provides an “always open” network for communication between professionals for discussion of difficult cases. The Group regularly updates its list of indications for BMT (***Enclosure 2****), and has recently produced consent forms to ensure that full communication has occurred between families and professionals in preparation for these complex procedures (***Enclosure 3****). This process of consent seeks to confirm that adequate information has been given to the family about the procedure itself, storage and/or future use of any stem cells that may have been collected around the procedure, and the use of data collected from any individual BMT procedure.

The UKCCSG BMT group has also produced guidelines governing BMT protocols and the management of post-BMT complications (***listed in Enclosure 1****). Consequently any team in the UK performing paediatric BMT has ready access to nationally agreed guidelines covering the whole area of their practise.

Transplantation procedures will soon come under close scrutiny from the Joint Accreditation Committee of ISHAGE and EBMT (JACIE) accreditation process. The necessary standards required to perform BMT procedures are stringently defined in Section B of the JACIE accreditation manual. (***Enclosure 4****). These standards closely define both the facility and staff requirements to conduct BMT within the immediate multi-disciplinary team as well as necessary supporting teams.

These standards have been modified by the UKCCSG BMT group so that they specifically address the Paediatric population. A remaining grey area concerns adolescents aged 16 and 17 years where, on an individual basis it may be better for such older children to be cared for either in a paediatric BMT unit or indeed in an adult unit. It is not envisaged at this time that there will be sufficient resources

to provide BMT units specifically for adolescent patients. The other remaining grey area is follow-up of children post BMT. It is necessary to continue follow-up of BMT patients lifelong to address a number of late sequelae that may occur many years after transplant. Such sequelae include infertility, growth retardation, neuropsychometric problems, endocrine dysfunction, cataracts, and secondary tumours. It will be necessary to forge links between Paediatric and Adult centres such that long-term survivors can pass seamlessly from one group to the next. Clearly this connection is also very pertinent to Paediatric Haematology and Oncology.

The complex nature of BMT procedures and the considerable expense incurred in performing BMT necessitates that expertise and resources are focussed within designated centres. Currently, in England and Wales, there are 13 centres performing allogeneic BMT and it is likely after the JACIE accreditation process and other on-going reorganisations this number may be reduced to 10. Clearly this means that a number of children may have to travel reasonable distances for their BMT procedure. This may be acceptable for the acute period of hospitalisation, usually 1-3 months in duration, however, travelling distances may complicate the necessity for frequent post-BMT clinic attendances. This is currently being addressed with 2 models of care. Firstly some centres particularly outside London have near hospital accommodation where patients and families may remain outside the BMT ward but close to the Hospital for prolonged periods of time. Secondly some centres have developed comprehensive shared-care models whereby much of the post-BMT care is carried out by local hospitals and community teams in constant liaison with the specialist BMT centre

Further work will be required to ensure that one of these two models is fully operational in all 10-13 Paediatric BMT units.

* If required, Enclosures 1-4 are available from the NCC for Cancer upon request.

APPENDIX H

Nutrition and Childhood Cancer

Mrs Evelyn Ward
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Background

With continued improvement in the treatment of children's cancers the role of nutritional support has become more important. Children differ metabolically from adults and continued growth and development is desired throughout treatment, therefore with more curable children being treated the more children there are subject to the nutritional problems caused by their disease and treatment.

Malnutrition and cancer cachexia are a frequent consequence of paediatric cancer and its treatment. A clear understanding of the metabolic alterations with malignancy leading to nutritional depletion and the value of maintaining nutritional equilibrium are a valuable part in managing these children. [Andrassy RJ, 1998]

The incidence of malnutrition in childhood cancer ranges from 6-50% depending on the type, stage and location of the tumour. [Donaldson SS, 1981, Van Eys, 1979] Malnutrition at diagnosis is often the exception rather than the rule, however malnutrition is more severe in later stages of malignancy, occurring in up to 37.5% of newly diagnosis children with metastatic disease. [Smith DE, 1991] The initial problems resulting from the tumour may soon be compounded by the iatrogenic nutritional abnormalities, the consequence of the treatment and its side effects. [Mauer AM,1990]

As therapies have increased in both complexity and intensity leading to increased survival rates, so to has the severity of the complications including nutritional depletion secondary to prolonged anorexia, nausea, vomiting, mucositis and significant infectious complications. Other common side effects impacting on

nutritional intake include taste abnormalities, dry mouth, constipation, renal impairment and food aversion.

Children who are malnourished at diagnosis have a significant poorer outcome compared with children who are well nourished at diagnosis. [Donaldson SS, 1981]

Malnutrition contributes to a reduced tolerance to therapy. Dose adjustments in chemotherapy have been seen most frequently in patients during a time of malnutrition. [Van Eys, 1979] There appears to be differences in the metabolism of chemotherapy agents between adequately nourished and inadequately nourished patients. [Van Eys, 1984] Malnutrition is associated with a higher risk of infectious complications and higher infection rates have been documented in malnourished children. [Van Eys, 1980]

Nutritional support will therefore improve immune competence, tolerance to therapy, quality of life and promote growth and development. [Van Eys, 1998] Nutritional support must be must be designed to provide adequate protein and calories for all children taking into account their condition and age whether this be oral supplementation, enteral nutrition or parenteral nutrition.

Continued monitoring of nutritional status is an essential component of care. Children at a higher risk of malnutrition include younger children, solid tumour patients, especially abdominal. The risk of malnutrition increases with greater treatment intensity.

It is now well recognised that nutritional support in childhood cancer is an important part of supportive care and the development of ever more intensive protocols highlights the need for aggressive nutritional support. A multidisciplinary team approach is the best way of providing safe, appropriate and effective nutritional support for this group of patients. Along with the

emergence of early and late treatment related morbidity in survivors e.g. osteoporosis, elevated fat mass the role of nutrition remains challenging.

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Service provision

1 – Training

Currently the majority of registered dietitians working in paediatric oncology will have worked in general paediatrics and therefore done basic nutritional support.

The majority will be Senior I grade and will have completed the basic paediatric dietitics module 1 run by the paediatric group of the BDA, but will not necessary have a specific knowledge on childhood cancers and their treatments unless they have completed that part of module 2 run by the paediatric group of the BDA.

It is recommended that they should undertake some of the following training

a) Attend module 2 part, which includes paediatric oncology.
b) Orienteering with different members of the MDT.
c) Support from the paediatric oncology dietitians interest group
d) Spend time with a dietitian who has experience in paediatric oncology.

2 - Recruitment and retention

Generally in dietetics there is a shortfall of trained registered dietitians leading to problems with recruitment and retention. Problems specific to paediatric oncology include:

a) Depending on size of centre the dietitian may not just cover paediatric oncology but other areas of paediatrics making it harder to specialise in oncology.
b) Some centres have rotational posts where dietitians rotate around different specialities and therefore only gain a limited experience in oncology.
c) In most centres dietetic activity has increased due to:
- An increasing number of patients requiring more aggressive nutritional support due to an increase in the use of more intensive protocols.
- Better recognition that nutritional status has a prognostic effect on the outcome of children with cancer.
- Increased monitoring of patients requiring nutritional support due to toxicity affecting tolerance to enteral feeds or parenteral nutrition.
- Increase in the number of Teenage Cancer Trust Units opening requiring dietetic input.

This has often been without any increase in dietetic staffing leading to:

- Less follow up and monitoring of patients deemed to be of a lower nutritional risk.
- Less out-patient follow up
- Less time available to spend on continuing education, research and audit.
- Difficulty finding time to update patient information advice leaflets and other patient orientated documentation.

This can have a adverse effect on staff morale due to an ability to provide a good service to paediatric oncology patients due to increasing work loads and will have an effect on staff recruitment and retention.

3 - Staffing

A recent questionnaire undertaken by a member of the paediatric oncology dietitians interest group showed diversity within UKCCSG centres as to how much dietetic time is allocated to paediatric oncology.

a) Range = 0.4 –1.3 WTE. Mean = 0.65 WTE with 50% of centres having 0.5-0.6 WTE.

b) Bed numbers. Range 8 – 23. Mean = 16. No correlation between dietetic time allocated and inpatient bed numbers. Range 1WTE:9 beds to 1WTE:46 beds. Mean 1WTE:26beds.

c) Input into out patient clinics varies from centre to centre.

Recommended staffing levels

It is difficult to estimate accurately the correct staffing levels and it is possibly easier to estimate on average number of newly diagnosed patients referred to the UKCCSG centre. Ideally a maximum of 80% of available hours should be spent on patient-related work [direct and indirect casework] with 20% on practice-

related work [staff meetings, professional development, training, audit/research, resources and quality assurance].

A reasonable estimate to allow adequate service provision to both in-patients and out-patient clinics would be 1.0WTE per 85 newly diagnosed patients or per 18 beds.

Support staff

As well as members of the MDT other staff involved in the provision of adequate nutrition for the child with cancer include;

a) Diet cooks and catering staff
b) Pharmacy technicians for supply of oral sip feeds, enteral feeds and manufacturing of TPN
c) Milk kitchen staff to make up specialised feeds.

4 - Equipment

In order to provide adequate and effect nutritional support to paediatric oncology the following equipment is needed:

a) A good range of oral sip feeds and calorie supplements suitable for paediatric patients and older children/adolescents

b) Enteral nutrition – Wide range of feeds suitable for infants, young children, older children/adolescents
- elemental
- peptide based
- whole protein standard calorie
- whole protein high energy
- fibre containing feeds

– Nasogastric feeding tubes, nasojejunal tubes

– Gasrostomy tubes

– Enteral feeding pumps

c] Parenteral nutrition – Able to provide tailor made regimens for infants, young children, older children and adolescents.

d] I.T. facilities – to enable patient administration
– audit
– individualised patient information
– research

5 – Multidisciplinary approach to nutritional support in paediatric oncology

A multidisciplinary approach to cancer therapy is well established and this includes a multidisciplinary approach to the nutritional care of the children with cancer. A multidisciplinary approach is the best way of providing safe, appropriate and effective nutritional support.

A team is able to function more effectively than individual members in the following areas:

a) Identification of present or potential nutritional problems
b) Nutritional assessment
c) Recommendations for therapy
d) Supervision and monitoring of recommended therapy
e) Communicating more effectively and quickly with patients, parents and other team members

It is therefore paramount that dietetic staff attend MDT meetings.

6 - Future

The following will have implications for dietetic services in the future:

a) Development of more intensive treatment protocols highlights the need for more children requiring aggressive nutritional support.

b) As the overall cure rate continues to rise early and late treatment related morbidity is coming to the fore. Consequences such as loss in bone mineral mass resulting in osteopenia and osteoporosis, growth problems, altered body composition with a reduction in lean mass and increase in fat mass leading to obesity, type 2 diabetes and ischaemic heart disease can impact on dietetic services.

c) An increasing number of centres are now siting gastrostomies in patients impacting on surgical, dietetic and nursing staff.

d) Increased research need into the role of specific nutrients e.g. glutamine, anti oxidants.

Guidelines for the nutritional management of the childhood cancer

1) *Identification of nutritional risk*

Criteria for identifying children with cancer who are malnourished differ, however determination of the nutritional risk of a child with cancer can be associated with the diagnosis of certain tumours and stages of the disease. Table 1.

The following criteria can be used to identify children with cancer who are likely to require supplementary nutritional support.

a) Total weight loss of >5% relative to pre-illness body weight.
b) Weight for height <90% or BMI < 20 in adolescents aged 18 years and above.
c) Decrease in current percentiles for weight [or height] of 2 percentiles.
d) Food intake <70% of estimated average requirement for more than 5 days.

e) Anticipated gut dysfunction for > 5 days
f) High nutritional risk patients based on tumour type and treatment regimens [Table 1].
g) Mid upper arm circumference and/or triceps skinfold thickness < 5th percentile.

2) Nutritional support

The aim of nutrition support is to
a) Reverse any malnutrition seen at diagnosis.
b) To prevent malnutrition associated with treatment
c) To promote growth and development throughout treatment
d) To enhance quality of life.

Nutrititional support therefore must be designed to provide adequate calories and protein intake for children with cancer by an experienced dietitian with the expertise to tailor make such individualised regimens.

3) Methods of nutritional support

a) Oral feeding

For children of a low nutritional risk, oral feeding is the best method, if they are able to consume sufficient nutrients. However advice with regard to the use of high energy foods and specific advice on eating problems related to the side effects of treatment should routinely be given by the dietitian.

The dietitian can also advise on the use of proprietary sip feeds and calorie supplements with regard to quantity, type and modification depending on the child's age and current oral intake. Due to changes in the child's appetite and taste perception this needs to be frequently reviewed.

b) Enteral nutrition

Whenever nutritional intervention is indicated it is highly preferable to use the enteral route.

Studies report that nasogastric feeding benefits children with cancer and that it is practical, acceptable and tolerated in children with newly diagnosed advanced malignancy who are commencing intensive treatment protocols. It improves their energy intake, wellbeing and their nutritional status as measured by mid upper arm circumference. [Smith DE, 1992]

Even in children undergoing bone marrow transplant where the nutritional insult is complex as is its management, enteral nutrition when tolerated is effective in limiting the nutritional insult leading to a better response and fewer complications. [Papadopoulou A, 1997, Papadopoulou A, 1998]

Generally a whole protein will be tolerated. However following chemotherapy or radiotherapy a hydrolysate or elemental feed may be more appropriate if malabsorption occurs. Careful monitoring of these patients by an experienced dietitian is essential with regard to feed tolerance during and following chemotherpy when it is often necessary to manipulate the feed volume, type and flow rate taking into account the age of the child and clinical condition.

Until recently it would have been rare to place a gastrostomy in a child with cancer as concerns over site infections and placement timing during chemotherapy. However an increasing number of centres are now siting gastrostomies particularly in brain tumour patients, Osteosarcoma patients, Ewing's sarcoma patients, nasopharyngeal tumour patients and adoloscant patients

Enteral feeding can be done easily at home with minimal disruption to the child's normal daytime activities.

c) Parenteral nutrition

Parenteral nutrition is required when enteral feeding alone cannot provide adequate nutrients or for those patients with abnormal gastrointestinal function related to their tumour or following chemotherapy, radiotherapy, bone marrow transplant or high dose therapy and peripheral blood stem cell rescue. This form of nutritional should ideally only be used if the gut is not functioning or accessible. [Pencharz PB, 1998] It is however often the only means of nutritional support in children with severe mucositis.

Parenteral nutrition may be used as an adjunct to enteral nutrition or as a sole source of nutrition. This method of support is expensive, carries a high risk of infection and is not easily carried out at home. [Szeluga Dj et al, 1987] Metabolic complications associated with parenteral nutrition are well documented. [Glynn J, 2001]

Summary

Nutritional support to prevent loss of lean body mass is an integral part of treatment of paediatric oncology patients. It will improve tolerance of therapy, immune competence, quality of life and promote growth and development. [Van Eys, 1998] It is necessary to choose the most appropriate method of nutritional support taking into account the child's age, condition and treatment. It is imperative that the effect on nutritional status is monitored to ensure the optimum support is being given and a multidisciplinary team approach is the best way of ensuring this.

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Table 1

Types of childhood cancers associated with high or low nutritional risk

High Nutritional Risk	**Low Nutritional Risk**
Advanced diseases during initial intense treatment.	Good prognostic Acute Lymphoblastic Leukaemia

Stages III & IV Wilm's tumour Stages III & IV Neuroblastoma Ewing's sarcoma Osteosarcoma Stage IV Rhabdomyosarcoma B-cell Non Hodgkins Lymphoma Acute Myeloid Leukaemia Poor prognostic Acute Lymphoblastic Leukaemia Medulloblastoma Children undergoing BMT or high dose therapy and P.B.S.R.	Non- metastatic solid tumours Advanced diseases in remission during maintenance treatment.

APPENDIX I

THE ROLE OF PSYCHOLOGICAL SERVICES IN SUPPORTING CHILDREN AND ADOLESCENTS WITH CANCER

Dr Deborah Christie
Consultant Clinical Psychologist and Honorary Senior Lecturer
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Biography

Dr Deborah Christie has a Ph.D in neurobiology and a background in experimental psychology before joining Great Ormond Street Hospital as a Leukaemia research funded psychologist for 4 years, documenting the effects of the UKALL10 protocol for leukaemia on cognitive outcome in long term survivors. As part of the research project she provided clinical back up to families who were struggling to access adequate special needs support for their children who had been affected by their treatment. Subsequently Dr Christie worked for 5 years as liaison psychologist to the haematology/oncology and neuroncology services. She has provided short and long term input to young people and families at point of diagnosis, as part of the GOSH late effects services and worked with the palliative care team. Dr Christie is currently Consultant Clinical Psychologist at University College London and Middlesex hospitals where she is a member of the multidisciplinary psychological services team and head of service for paediatric and adolescent psychology. She is a member of the joint hospitals late effects re-design task force.

Acknowledgements

Several colleagues working in paediatric cancer across the country provided information about their service. Several chose not to contribute. Therefore the

opinions, views and suggestions from other psychologists working in paediatric cancer have been incorporated where possible but not the views of those that didn't contribute!!

This document has been written to identify clinical issues and potential gaps providing psychological input to children and families living with cancer. The paper aims to summarise the issues from the perspective of a practising clinician and reflects the personal challenges that are created by current resources and service level planning. The document contains suggestions and ideas, which are entirely those of the author and are based on clinical and personal research experience. It is hoped therefore that the suggestions reflect a general and reasonable perspective on needs. As it has not been reviewed by a committee there may be those that feel that it does not reflect a consensus opinion although it is questionable if one could ever be achieved. This paper does not reflect the view of any institution. The document aims to identify how young people can and would benefit from a psychological service that provides a range of therapeutic support from individuals who are trained in these approaches, methods and techniques and does not specifically identify a particular discipline as being inherently more or less appropriate.

Introduction

What is the difference between a psychiatrist, a psychologist and a psychotherapist? This question may sound like the beginning of a bad joke but it is an often asked question by managers trying to understand the difference - real or imagined - between the many professionals that provide psychological support to young people, families and medical teams dealing with cancer. The main differences are located in training, approaches, method and technique. A child and adolescent psychiatrist (CAP) is a medical doctor who works with children and young people who have serious emotional or mental health problems. CAPs can prescribe medicine for these problems - if this is appropriate - and are often trained in individual and family therapy as well. The therapeutic approach can

range from the biological to a family systems approach. Limited resources mean that the majority of psychiatrists will tend to work with the more severe end of the spectrum of psychological distress. Traditionally the psychotherapist will train in a single therapeutic model that will aim to offer individuals or families a space to explore their thoughts and feelings and integrate their internal and external 'worlds'. As a generalisation the approach taken is non-directive and self-reflective. However once again within this single discipline there are a range of models that inform the kind of questions and approaches that may be used to ameliorate distress in an individual. Finally, clinical psychologists have a scientific training (at a doctorate level) in the understanding of behaviour within the context of normal developmental processes, and the relationships between thoughts, emotions and behaviours. They will be trained in the assessment of emotional distress, behavioural difficulties and levels of cognitive ability. Some have additional specialist training (e.g.; neuropsychology). They can offer a wide range of therapeutic intervention techniques.

To add further confusion family therapists, counselling and health psychologists are also able to provide support and advice to young people and families in distress whilst play/activity specialists and specialist social workers are often key members of the wider psycho-social team. All of these professions offer a combination of unique and overlapping skills using different psychological models and different approaches but have the same goal - that of helping young people and families living with and surviving the challenge of cancer

Cancer Treatment

Around 200,000 children worldwide are diagnosed with cancer each year. There have been significant advances in treatment for cancer over the last two decades and in the developed countries some 70 - 80% of these children will be cured. Although teenagers and young adults are no more likely to survive than they

were 25 years ago, the five year survival rate in younger children has risen[5]. There has also been an increase in the number of paediatric cancer nurses and specialist developmentally appropriate treatment centres, mostly due to the work of the Teenage Cancer Trust. Despite these improvements in medical care and increases in survival rates, a diagnosis of cancer remains a traumatic and terrifying experience for children, young people, parents and other family members. It can also create a significant challenge for the multidisciplinary treatment teams.

So what's the problem?

> *The problem is not that there are problems. The problem is expecting otherwise and thinking that having problems is a problem.* T. Rubin

It is important to acknowledge that many young people and families show remarkable resilience as they begin their journey from diagnosis to treatment and ultimately survival. Some families seem able to cope with minimum intervention and use coping strategies previously developed or learned to 'get on with' their life.

Should we be encouraging families to 'be miserable' and face their worst fears or is denial a useful strategy? Different psychological models would have different responses to this question.

One of the challenges for clinicians is to find ways to acknowledge and respect those that prefer to do without additional support whilst remaining sensitive to coping styles that no longer appear to be helpful. The skill for the clinical team is to be able to offer support that fits for families when they need it. Identifying risk

[5] BMJ 2004 Vol 328 p 540

factors at the beginning of contact with families can help teams identify who may require input. Ensuring that psychological and social support is a resource that is available to all families as and when it is needed can reassure families who struggle at certain times that they should not feel that they have 'failed to cope'.

A comprehensive review of the psychological impact of cancer has been published by Rowland (1990)[6]. The model acknowledges the importance of developmental stages and the impact of cancer on these stages. The disruption caused by illness and treatment is specific to each developmental stage however there are five common sources of problems that Rowland and Holland identify as the 5 D's.

1) *Distance* in interpersonal relationships
2) issues of *Dependence* and independence
3) *Disability* in social or school achievement
4) *Disfigurement* or physical impairment
5) fear or anxiety about *Death.*

Other factors that can affect a family's relationship to cancer will be their developmental stage, communication style and previous experience of illness. Cultural, economic and social factors are additional influences on how a family understand and cope with diagnosis, treatment and the ultimate outcome.

The role of psychological services in supporting people living with cancer

Psychological services have an important role to play at all stages of the patient pathway. This includes at the time of diagnosis, coping with different stages of treatment and providing long term support in rehabilitation and palliative care and

[6] Handbook of Psychooncology: Psychological care of the patient with cancer (1990) Holland, J.C & Rowland, J.H. (Eds)

for those that survive. Input should be available for the young person, their families and the clinical treatment teams, who are sometimes forgotten.

Diagnosis

However well communicated, bad news can be extremely difficult to hear. The distress can have a significant impact on the ability to process and remember information. Parents sometimes say that they were never told the diagnosis or cannot remember information that has been repeated several times.

In some teams a psychological team member will join the consultant in order to hear what the family is told and will then meet with the family again to go through the information and help them assimilate what they have heard. The role here is not to discuss the medical information but to think about the emotional impact of the diagnosis. It also provides an opportunity to assess the thinking style of the family and help them identify ways they have coped in the past with bad news, previous experiences of serious illness and what psychological resources they have available. Cultural and/or religious beliefs about illness and treatment can be important to explore in order that teams have a greater understanding of a families responses to a diagnosis or how they manage whilst on the ward. The communication style of the family can also be considered at this point and then fed back to the clinical team to help enhance future communications with the family.

Coping with treatment

There are a range of difficulties associated with treatment that psychological services can offer specific support for, using a range of evidence based treatment techniques.

These include:

- Procedural distress
  - Treatment refusal e.g.; Chemotherapy, surgery, medication compliance
  - Needle Phobia

- Medication tolerance and associated distress e.g.
  - Anticipatory anxiety
  - Anticipatory vomiting
  - Food Aversion
- Body Image
  - Hair loss
  - Amputation
- Emotional distress
  - Depression
  - Anxiety
  - School avoidance

There is evidence to suggest that Cognitive Behavioural Therapies are the most effective in the support and treatment of these conditions. Cognitive behaviour therapy (CBT) is an approach that aims to help an individual identify underlying negative thoughts that have developed in response to their environment. These thoughts create distressing emotions which then drive behaviours that are usually maladaptive or unhelpful. CBT includes a range of techniques that can be helpful in many different anxiety related conditions. A child who is refusing to allow chemotherapy to be started on time or who becomes acutely distressed when they need to have catheters inserted or bloods taken can be helped to use relaxation techniques (including guided imagery and hypnotherapy). Graded exposure is another way to reduce the distress associated with procedures. CBT has also been shown to be effective in the management of panic attacks, eating disorders, body image, anxiety disorders and depression and is a relatively brief, symptom focussed, practical and effective therapy.

In addition to CBT, family based solution-focused approaches are being increasingly seen as helpful in the clinical setting. These view the patient as the expert and focus on “what works” e.g. identifying “what helped” during periods when their illness was in control. The approach is non-pathologising and

normalising. The author has found it to have significant promise with young people with a wide range of chronic illness who are reluctant to engage with more traditional psychological approaches. Solution focussed therapy begins from where the young person and their family wish to be in the future and identifies strengths, abilities and resources that can be used to achieve the preferred future. It introduces the possibility that no matter how difficult the situation, change is possible. Families are invited to see themselves as experts who already have solutions but just need support in recognising and identifying exceptions to the problem being the rule.

There is a small but growing number of research papers describing these approaches in children[7] and increasing clinical evidence that these approaches are effective and liked by families.

> *'a 12 year old girl had not expressed any concern about hair loss to her family or the medical team however was withdrawn and resistant to treatment at the beginning of each treatment cycle. In a conversation she was able to identify bravery as the ability that allowed her to challenge the effects of her disease and its treatment on her life. She told me she had needed bravery be able to go out in public without her hair'.*

Bravery was therefore identified as a unique strength and ability and discussions followed that helped her identify how she could use this ability to help her in other aspects of her treatment. Members of her nursing team were interviewed and invited to join her bravery team and were asked to look out for any times they spotted bravery hanging out on the ward with her.

[7] Viner, R., Christie. D., Taylor, V. and Hey, S., (2003) Motivational and solution-focused intervention improves HbA_{1c} in adolescents with type 1 diabetes Diabetic Medicine 20, 739-742

The meaning of the long term consequences of treatment can also impact on how a young person copes with the acute treatment phase. An example of this would be discussions about limb amputation. The effect on a young person's life will be unique and have a meaning that is specific to their hopes and ambitions. For example a young teenager with an ambition to be footballer that loses a leg or a musician who loses an arm may believe that they no longer have any future and that it is not worth surviving. Thoughts about the amputation become increasingly catastrophic and are accompanied by increasing distress and depression. CBT can focus on addressing these catastrophising thoughts and help young people think about creating alternative futures. It is important to point out that these are just two approaches and some families will find other models of psychotherapy, general support and non-specific counseling to be as helpful. The role of psychological services is to work with the clinical team to find out what works best for whom and fit the intervention to the family, not the other way round.

Communication problems

Difficulties in communication during treatment can arise between the child and their family, between the clinical team and the family, and very often within the team where different views about how best to treat the child are held.

In any complex system it is important to take an approach which can consider the views of the different members of the wider system. It may be important to think about the role of school, the church or local community. Religion, race, and cultural views may influence a family's attitude to treatment and how they communicate with or are perceived by staff (e.g.; Jehovah's Witness).

Some of the above issues may be embedded in communication difficulties between different parts of the system. The role of the psychological team is to enable and understand where the difficulties are located and to think about how to enable more productive conversations between these different parts.

Systemic approaches offer a perspective which allows an understanding of the family's relationship to help and, when problems occur, offer opportunities to think in a positive way about who wants help from whom[8].

Relationships

Perhaps one of the most immediate consequences of treatment is its impact on dependence-independence. The specific nature of the impact is influenced by the child's developmental stage. In healthy children there is a gradual and complex renegotiation of their relationship with their parents as they grow from new-born to young adult. For young children the parent is seen as having complete authority and control whilst a key developmental task of adolescence is achievement of independence and autonomy.

The process of separation in the healthy family begins slowly as children begin to feel confident with being left with other carers, start school and learn to take responsibility for personal care. There is an increasing need for physical and emotional privacy. Successful negotiation requires parents to allow adolescents to make safe mistakes, reviewing and revising consistent and reasonable negotiated boundaries, which adolescents are required by their adolescent job description to ignore and step over!

Personal privacy is often difficult to maintain on busy wards. Weakness or incapacity requires levels of personal and intimate care that may not have been required since early childhood. Teenagers used to personal space and privacy find cancer hijacks their journey towards independent living as they once again become dependent on parents and nurses for physical and intimate care.

Psychological services can offer a family meeting to explore these anxieties and offer space to think about the way that cancer has knocked them off track. Family

[8] Christie, D., and Fredman, G. (2001) Working Systemically in an Adolescent Medical Unit: Collaborating with the network Clinical Psychology 3: 8-11

therapy creates opportunities to explore previous strengths in families that can be built on to help them recreate previously negotiated independence.

Unpleasant but necessary treatment may be 'enforced' by parents and medical teams, who 'know what is best'. Ultimately decisions about treatment may challenge the ability of parents (and doctors) to allow children choice. Parents can feel their role as protecting children challenged when they are required to encourage children to accept treatments that are unpleasant or distressing.

In family meetings parents can explore their feelings of powerlessness in the face of cancer and complicated medical decisions. Young people may find it difficult to ask for or accept independence post treatment. Therapy can focus on uncovering how young people were previously successful and introduce possibilities of rediscovering these previous successes and ways to get their life back on track.

Social relationships

Cancer is usually a condition that is unpleasant and debilitating, that requires treatment in hospitals, perhaps a long way from home and can cause dislocation from family, friends and peer groups. There are particular time points, (transition stages) in a child's life that increases the difficulties, for example the child just about to begin reception or transfer from junior to secondary school. Children may miss the critical first class and the opportunity to establish new developmentally appropriate peer relationships. Absence from school will mean difficulty establishing or maintaining peer networks.

For many young people returning to school during treatment can feel like an impossible hurdle. Treatment induced sickness, pain or discomfort may result in not wanting to meet friends or play with siblings in between treatment cycles. Hard won friendships may be dislocated and hard to repair. Young people talk about feeling like an outsider.

R was 16 when she was diagnosed with a brain tumour. After a year off school for treatment she felt her friends had all moved on without her. When she went to school she didn't have the confidence to approach groups of girls and believed that nobody was interested in her. R was encouraged to find ways to answer back these negative thoughts about people not wanting to talk to her. We also decided that confidence had gone into 'hibernation' and strategies for waking confidence up and getting it to help her reawaken old friendships were tried out.

Research has shown lower social competence in children treated for brain tumours[9]. These problems may be due to deficits in physical appearance (e.g. hair loss) increased physical limitations (surgery or treatment effects) and missed school days and social activities. Cognitive deficits may also underlie social skills deficits due to white matter damage secondary to cranial radiation therapy.

A group intervention Social skills training (see Barakat et al) has been found to be potentially effective in mediating these effects and could be delivered in all services both as part of on-treatment and follow up clinics.

Late effects.

The impact on long term cognitive functioning has been extensively documented in the research literature[10]. Monitoring the impact of treatment and liaising with educational services is a key function of cancer psychological services. A team in

[9] Barakat et al (2003) Evaluation of a Social-skills training group intervention with children treated for brain tumours: A pilot study Journal of paediatric psychology 28 (5) 299 -307

[10] See for example Christie, D., Leiper, A.D., Chessells, J.M. and Vargha-Khadem, F. (1995) Intellectual Performance after presymptomatic treatment for lymphoblastic leukaemia: effects of age, time since treatment and sex. Archives of Disease in Childhood**.** (73) 136 - 140
Christie, D., Battin, B., Leiper, A.D., Chessells, J.M., Vargha-Khadem, F. and Neville, B.G.R. (1994) Neuropsychological and neurological outcome after relapse of Lymphoblastic Leukaemia. Archives of Disease in Childhood. (70): 275 - 280

Birmingham is currently running a regular workshop for teachers of recently diagnosed children to inform and educate them about the acute and long term effects of treatment.

However, very few services have sufficient resources to offer a regular screening programme. In the majority of clinics neuropsyschological assessment can only be offered some years after treatment when problems become severe and remediation or rehabilitation is much more difficult. Butler and Copeland (2002) have recently developed a cognitive remediation programme that has reported improvement in attention and concentration skills[11]. At the present time this is an expensive resource however the increasing number of children surviving cancer demands that the cost of cure must also now be addressed as intensively as the search for new and more effective treatments has been. A challenge for cancer services must be to ensure that current levels of cognitive functioning at time of diagnosis are determined and monitored over time. In this way treatment induced deficits can be addressed quickly and appropriate special needs support can be put in place. The establishment of specialised rehabilitation and remediation programmes is also essential.

Palliative treatment

Despite heroic efforts by clinical teams, for a number of children and young people treatment is not successful. Death remains a possibility for a percentage of children who are diagnosed.

[11] Butler and Copeland (2002) Attentional processes and their remediation in children treated for cancer: A literature review and the development of a therapeutic approach. Journal of the International neuropsychological society (8) 115 - 124

Failure to respond to treatment may occur early on in the treatment cycle whilst for others there is an ongoing risk of relapse and decreasing treatment options. This 'sword of Damocles' feeling is often reinforced by the need to attend long term survivor or late effect clinics where continual tests 'just to check' do little to reassure children that they no longer have cancer.

There are many views as to how and when families should be told about treatment 'failure' and who should be involved in this process. These views are influenced by personal beliefs and professional experience as much as evidence based practice. Decisions as to when to introduce palliative care teams differ widely across services and may be different within a single service[12].

The developmental stage of a child and their family may determine the kind of anxieties that surface in response to bad news. For younger children the concerns may be specific and concrete.

> *J was 5 and suffering from terminal skin cancer. In conversations with him about what might happen if he didn't get better he talked about needing to be sure that someone was going to look after his goldfish and his play station and how he knew his mummy and daddy were going to be sad that he was going to heaven without them.*

His parents felt able to begin having these conversations with him once they were reassured that **he** could tolerate the idea of not getting better. They also asked for the psychologist to talk to his sister about what was happening to get sense of what she understood and was able to talk about with them.

For teenagers a need to protect themselves from distress may mean they choose not to talk or acknowledge what is happening and throw themselves into activities

[12] Goldman A. and Christie D. (1993) Children with cancer talking about their own death with their families. Paediatric Haematology and Oncology. (10) 223-231

whilst they are still physically able to do so. In contrast other young people may find it helpful to discuss their anxieties about unfulfilled futures and loss of future relationships and unlived lives. The psychological team can often have a role here reassuring the clinical team that the young person has made a personal choice about how they have chosen to live with their potential death. Their way of coping may fit for them and our ideas about what should or shouldn't happen may not always be helpful[13] (see Griffin and Christie, 2004 for a description of working with a palliative care nursing team).

The psychological team can also offer input to bereavement services and work with bereaved parents and siblings.

> *L's husband had died of cancer 12 months before her oldest daughter was diagnosed. E died on the ward. Meeting with L helped her think about what strengths and abilities she needed to cope with this second terrible loss and how to support E's younger sister.*
> *J's parents asked for his sister to meet with the psychologist after he died. She talked wanting to feel it was OK to be happy without being disloyal to J..*

The state of play

Psychological services across the country offer a range of in patient and out patient services which include;

- Individual, family and parental therapy
- Individual counseling and support for staff
- Clinical training to the Multi-disciplinary team
- Staff Consultation to the Multi-disciplinary team
- Young person and parent groups

[13] Griffin,A., Christie,D. (2004) Trails and Triumphs, stories and solutions; Using systemic therapy techniques to facilitate paediatric and community nurses groups. Journal of child health care Vol 8 (1) 67-76

- Staff Groups (for nurses, play specialists, junior doctors)

There appears to be very little consistency around provision of psychological services to child and adolescent cancer services in the UK. The majority of services primarily use clinical (or health) psychology and/or social workers as the main discipline. Very few have dedicated child psychotherapy or psychiatry although most can access or refer to if needed. The majority of services respond to specific referral requests and few are able to see all the young people admitted to the service. This role is often left to specialist social workers. The number of psychology sessions specifically allocated to haematology/oncology ranged from 0 to 10 (full time) at larger specialist teaching hospitals. However even at some of the specialist centres there is a relatively small number of sessions of dedicated psychological input although support may be provided through the general paediatric psychology service.

It was very difficult to obtain an accurate breakdown of referral patterns across services however about half of the referrals seem to be for emotional and behavioural problems, including procedural anxiety. About 25% are for parental support (including bereavement). The remainder were for cognitive assessments, sibling problems or school problem. A number had been seen for long term emotional sequelae. The majority were referred during the initial hospital admission although services that offered specialist late effects service often had larger number of referrals for rehabilitation and cognitive support.

One of the challenges expressed by those who work in different services is the paucity of thought given to managing communication between shared care teams. There is also a feeling among psychologists who work in cancer that there is an enormous amount of potential work that could be being provided. This includes being able to work proactively rather than just responding to distress, offering a range of group work and supervision of other members of the clinical team (e.g. communication skills with nurses, teaching procedural anxiety

methods to play specialists/activity co-ordinator). There are also opportunities to oversee the broader aspects of psychological care to ensure coherence and clinical governance that psychology expressed a wish to be involved in. For others the frustration is that limited sessions can make it difficult to carry out research unless they have dedicated funding.

A gold standard service

The ideal psychological service should be able to offer flexible and creative ways to think about how families are living with a diagnosis of cancer. Patients should have access to individual and family interventions that fit their way of thinking and is coherent with their cultural and religious beliefs.

The psychological team should be able to offer

- A range of individual approaches including brief solution and cognitive behavioural therapies
- Systemic consultation and family therapy
- Anxiety management
- Guided imagery and visualisation
- Hypnotherapy
- Relaxation training
- A range of groups (age appropriate and family based) including social skills training.
- Neuropsychological assessment, cognitive rehabilitation and remediation support

Managers must recruit individuals that have the skills and training to offer these services rather than focussing on specific disciplines.

Different clinicians have different views as to what works best for their service. Should the psychologist meet and greet every child (usually the remit of cancer

charity funded social workers) or should they only provide targeted interventions specifically requested by the clinical team?

In an ideal world this is not an either/or situation. Initial screening and assessment of a family's beliefs, strengths abilities and potential risks should inform a positive watch and wait approach. Teams should be transparent and open with families informing them of the range of support that is available and is as much a part of the treatment as the chemotherapy.

Job plans for psychological services should follow a planned activity format where regular supervision, professional development and training are incorporated into the job plan.

Planned activity should include (in order of priority)

- Attending ward rounds and multidisciplinary team meetings,
- Offering assessment and consultation to families and the team
- Brief and long term therapy
- Working with a family consultation/family therapy team
- Monitoring treatment effects
- Educational liaison
- Neurocognitive rehabilitation treatment programme (acute and long term treatment effects)
- Audit, Research and Service Development

What is the formula?

Sadly there is no magical formula that allows us to say for 'x' number of patients 'x' sessions of psychological support will be able to offer all of the above. The staff on wards have a fairly clear view of what they want from their psychological

support teams[14]. However resources will dictate the model that can be provided. When patient numbers and allocated time are well balanced a process consultation model allows a rapid, flexible, and frequent service. In contrast limited resources may mean that an indirect consultation model can only offer indirect consultation at ward rounds with minimal direct patient contact.

The end – or the beginning?

Guidelines for patient care often tread a middle ground once the extremes of evidence have been sifted through. A basic minimum of psychological delivery should be agreed and incorporated into the care pathways just as certain medical procedures are agreed and provided for. However, recommendations often say 'all patients should have access to psychological support' but do not say what for or what that support should look like. We should ask the young people and families that we work with what they found helpful and what they would have liked more or less of. It is the patients that are the experts not us. Different interventions will be relevant at different times in a young person's treatment and what seems to fit for us as a clinical team may not make sense to them.
In some teams introducing different ideas or working in a different way rather than increasing the number of sessions can be helpful. We should be identifying effective models of good practice and build on what works.

As clinicians the guidelines you are developing will provide an opportunity to develop creative and collaborative answers to the challenges created by cancer for children and adolescents.

[14] Christie, D. and Wigley, K. (1999) "3 into 100 won't go". Awareness and satisfaction of Nursing Staff towards a Ward Liaison Paediatric Psychology Service. Clinical Psychology Forum (125) 6-9
Christie, D. and Daycock, L.J. (2003) Evaluating a psychological liaison service: Easier said than done. Clinical Psychology 31, 13 - 18

APPENDIX J

NICE Child and Adolescent Cancer. Special Report on Late effects and long-term follow up of young people treated for cancer

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Summary Points

- Long-term morbidity risks in childhood and adolescent cancer survivors largely relate to treatment modality and the challenge remains to further improve survival rates whilst reducing the incidence and severity of such treatment-induced late effects.

- Treatment-related morbidity is diverse, with potential effects on the endocrine system (growth, puberty, fertility, pituitary, thyroid and other disorders), cardiovascular, second tumours, pulmonary and renal complications, and cognitive, educational, psychological, social and quality of life manifestations.

- Morbidity can be anticipated and monitored to optimise prevention and treatment – ideally through multidisciplinary follow-up.

- Evidence-based and graded recommendations provide a basis for the effective, informed and pragmatic follow-up of a cohort of patients who, it is estimated, will make up 1 in 715 of the adult population by the year 2010. The further development of evidence-based, therapy-based guidelines for follow-

up are an important prerequisite for an effective and cost-effective follow-up strategy.

Introduction

The incidence of Childhood cancer is 100 – 130 per 10^6 per annum and 1 in 600 children under the age of 15 years will develop cancer which is now curable in 65 – 70% (Campbell et al, 2003). It has been estimated that by the year 2010 1 in 715 of the adult population will be a long-term survivor of childhood cancer. Leukaemia makes up approximately one third of childhood cancers and brain and spinal tumours about one quarter. Childhood cancers are diverse in their site of origin and histological type but long term morbidity in survivors relates more to the treatment – surgery, chemotherapy, radiotherapy, bone marrow transplantation – than the cancer type or site.

With increasing understanding of the effects of these treatment modalities on tissues and organ systems, many of these treatment-related sequelae are predictable – and many are preventable or treatable with informed and careful follow-up. For the majority of those treated for cancer in childhood and adolescence, the goal is not merely long-term survival but high quality of life.

Nevertheless there is still an 11-fold increased overall risk of death in five year survivors of childhood cancer (Mertens et al 2001, Moller et al 2001) with still higher risks in females (18.2- fold), those diagnosed under the age of 5 years (14-fold) and those with an initial diagnosis of leukaemia (15.5-fold) or CNS tumour (15.7-fold). The commonest cause of death amongst 5 year survivors is a second malignancy (19.4-fold increased risk). Other common causes include cardiac problems (8.2-fold) and pulmonary problems (9.2-fold). Whilst cancer recurrence is the cause of death in about two thirds between 5 and 9 years after diagnosis, treatment related causes of death account for about 1 in 5 deaths (second cancer, cardiac toxicity, pulmonary complications).

Treatment-related morbidity is diverse with potential effects on the endocrine system (growth, puberty, fertility, pituitary, thyroid and other disorders), cardiovascular, pulmonary and renal complications, and cognitive, educational, psychological, social and quality of life manifestations (Wallace 1996, Bath et al 1998).

Growth and endocrine function following treatment of childhood malignant disease and the effects of chemotherapy are also reviewed by Wallace (1996) and Wallace and Kelnar (1996a, b) respectively

Whilst there is still a dearth of prospective longitudinal interventional large-scale studies of therapies designed to prevent, modify or treat morbidity in long-term survivors, there is an increasing body of evidence (descriptive, case control or cohort studies) on which scientifically sound recommendations for monitoring and follow-up can be based. The development of the SIGN guideline “Long term follow-up of survivors of cancer in children and young people” (SIGN No 76, 2004) in which I was involved as chair, has provided a systematic review of the evidence in many (although not all) of these areas. Its evidence-based and graded recommendations provide a basis for the effective informed and pragmatic follow-up of a cohort of patients who, it is estimated, will make up 1 in 715 of the adult population by the year 2010.

Areas covered by the SIGN guideline are 1) the assessment and achievement of normal growth, 2) the achievement of normal progression through puberty and factors affecting fertility, 3) the assessment of thyroid function, 4) the early identification, assessment and treatment of cardiac abnormalities and 5) the assessment and achievement of optimal neurodevelopment and psychological health. Important areas not covered by the SIGN guideline include second malignancy, renal, respiratory and liver dysfunction.

Thus long-term morbidity risks relate to treatment modality and the challenge remains to further improve survival rates whilst reducing the incidence and severity of such treatment-induced late effects. These can be anticipated and monitored to optimise prevention and treatment – ideally through multidisciplinary follow-up involving paediatric oncologist, paediatric endocrinologist, paediatric neurologist, radiation oncologist, paediatric neurosurgeon, clinical psychologist, specialist nurse and social worker.

Growth impairment

Long-term effects of radiotherapy (RT) and chemotherapy (CT) on growth and endocrine function have become more obvious and important as survival following childhood cancers has improved (Sklar et al 1993). Adverse effects on growth may result from radiation-induced hormone deficiencies, impaired spinal growth from spinal RT (and from CT), primary hypothyroidism from spinal RT, precocious or delayed puberty from abnormal gonadotrophin secretion, gonadal failure from RT or CT, and problems with nutrition or obesity (Didi et al 1995, Shaw et al 2000, Reilly et al 2000).

At diagnosis of acute lymphoblastic leukaemia (ALL), there is already low bone turnover with reduced levels of collagen formation and resorption markers (PICP, PIIINP and ICTP) (Crofton et al 1998). In remission, there is further bone synthesis suppression (low levels of PICP and PIIINP) and growth suppression (Ahmed et al 1997, 1999, Crofton et al 1999, 2000) which probably relates to glucocorticoid (prednisolone) and high dose methotrexate therapies. This suggests that there may be an increased risk of long term osteoporosis and fractures. Comparison between countries suggests that the degree of growth impairment is proportional to the intensity of the CT regimen. CT has a disproportionate effect on spinal growth impairment perhaps because of the large numbers of spinal epiphyses. High dose cranial irradiation is associated with a significant potential height deficit because of the combined effects of precocious puberty and an impaired pubertal growth spurt.

The hormone deficiency effects of RT will depend on the site of irradiation, total dose of irradiation, fractionation schedule and the child's age at treatment. Growth impairment will result from RT to the hypothalamo-pituitary axis (the hypothalamus is more radiosensitive than the pituitary and the GH axis the most radiosensitive followed by the gonadal axis). RT to the spine (in the treatment of medulloblastomas, ependymomas, germinomas) will result in late pubertal growth failure (the spinal growth spurt occurs towards the end of secondary sexual development) and primary hypothyroidism due to a direct effect on the thyroid gland. CT (glucocorticoids, methotrexate) will also impair growth.

RT doses of >24 Gy will be associated with precocious (especially in young girls) or delayed puberty and GH deficiency within 5 years (Ahmed et al 1986). Higher RT doses (eg ~54 Gy used in craniopharyngioma) will cause GH deficiency within 2 years. Lower doses (<24 Gy) may be associated with precocious puberty, an impaired pubertal growth spurt due to relative GH insufficiency in that context (Crowne et al 1992) and reduced pubertal spinal growth. Total body irradiation (TBI) used as preparation for bone marrow transplantation (~7.5 – 15.75 Gy) may also be associated with pubertal GH insufficiency, thyroid dysfunction and a radiation-induced skeletal dysplasia.

The same total dose of RT given in several fractions minimises GHD and growth impairment and fractionated TBI produces less damage to normal tissues. Younger children (especially girls) are more likely to develop precocious puberty and a pubertal growth spurt can be mistaken for 'catch-up' growth. Obesity can normalise growth at the expense of disproportionate bone age advance and reduced height prognosis.

Clinical growth assessment should consist of the regular measurement of sitting and standing height, skinfolds, weight and calculation of BMI, and puberty staging. It is recommended that all children who have survived childhood cancer

should have their height and weight measured regularly, on and off treatment, until they reach final adult height. Sitting height should be measured in children who have received craniospinal irradiation (SIGN Grade B recommendation). Chemotherapy is also likely to have a deleterious affect on spinal growth which may be particularly manifested by growth failure in late puberty.

Children with impaired growth velocity should have growth hormone levels measured after appropriate stimulation tests (SIGN Grade C recommendation). Other causes of poor growth, including potential deficiencies of other pituitary hormones or problems related to early or delayed puberty, should be considered and treated as necessary (SIGN Grade B recommendation). Children with craniopharyngioma should be tested at presentation for growth and other pituitary hormone deficiencies and at regular intervals thereafter (SIGN Grade B recommendation). Young girls receiving cranial radiotherapy should be closely monitored for signs of precocious puberty (SIGN Grade B recommendation).

Children who have been treated with low dose cranial radiotherapy are at risk of precocious puberty and growth hormone insufficiency (GHI), while those treated with higher doses are at risk of an evolving endocrinopathy with GHI developing early and in some children gonadotrophin, thyroid or cortisol deficiency developing later on (Shalet et al 1988). Thus laboratory assessment (baseline free thyroxine, cortisol, testosterone / oestradiol, IGF-I etc), physiological profiles (GH, GTs, cortisol etc) and dynamic tests (insulin hypoglycaemia, GNRH, hCG, TRH, synacthen etc) will be relevant. Nevertheless, integrating clinical and anthropometric information (plotting on appropriate growth charts, calculation of height velocity, calculation of body mass index and plotting on age-related BMI standards) as a prelude to appropriate investigation and treatment is an important role for the paediatric endocrinologist in a multidisciplinary team. Much information can be gleaned from careful anthropometry and pubertal assessment in the context of knowledge about the anti-cancer treatment received so as to minimise investigations in children who have already been through many

unpleasant treatments and investigations. Interpretation of biochemical (hormonal) information must be on a background of thorough understanding of growth and puberty so that treatments can be used timeously and appropriately. Available treatment modalities include the use of GH for growth failure, pubertal suppression and thyroxine, glucocorticoid and sex steroids as indicated.

If a child has a good prognosis from the underlying condition two years from treatment, GH therapy should be given when indicated on biochemical and anthropometric grounds. (SIGN Grade B recommendation). There is a high relapse rate in the first 2 years after diagnosis and it seems inappropriate to treat children with daily injections if the prognosis is poor or whilst the chance of relapse is still high. There is no evidence that GH is associated with reactivation of the primary lesion (Swerdlow et al 2000) but GH may well be 'blamed' for any relapse. Where the cause of growth impairment is unclear, a trial of GH may be appropriate (SIGN Grade C recommendation). In cranipharyngioma there is every reason to start GH therapy without delay once deficiency is identified – the response is excellent and on a par with that seen in other causes of GH deficiency. Management of the growth disorders secondary to treatment of childhood cancers is reviewed by Bath et al (1998).

There is accumulating evidence that childhood cancer survivors (particularly of leukaemia, but also of brain tumours and craniopharingioma), however they were treated, are at risk of obesity in adolescence and adult life (Davies et al 1995). The aetiology is likely to me multifactorial (nutritional, psychological, lifestyle including lack of exercise, endocrine and neuroendocrine) and is difficult to prevent or treat. There are potentially severe consequences: childhood obesity may affect educational attainment and interpersonal relationships adversely, especially in boys (Wake et al 2000, Gortmaker et al 1993), may persist into adulthood and is associated with an increased risk of hypertension, stroke, myocardial infarction or type 2 diabetes mellitus, osteoarthritis, breast and bowel cancers, skin disorders and asthma and other respiratory problems.

Hypertension, dyslipidaemia and hyperinsulinaemia are increasingly found in obese children with two or more risk factors found in 58% of obese children (Freedman et al 1999) with significantly increased Odds ratios for raised diastolic BP (2.4), raised LDL cholesterol (3.0), raised HDL cholesterol (3.4), raised systolic BP (4.5), raised triglycerides (7.1) and high fasting insulin (12.6).

Treatment for childhood cancer may result in reduced bone mineral density (Nysom et al 1998). An increased fracture rate remains to be demonstrated, but the observed decrease in bone mineral density would be expected to predict for increased fracture risk.

Children who are about to undergo head and neck cancer treatment, should be advised about the possible effects (particularly from radiotherapy) on oro-facial growth and teeth – eg facial growth, tempero-mandibular joint function, enamel defects, mineralisation and development of crowns and root stunting. Specialist dentists have a role in the care of these children (SIGN Grade D recommendation). Whilst levels of decay seem no worse than control children, treatment such as radiotherapy which reduces saliva may increase caries risk (see SIGN Dental Caries Guideline 2000).

Thyroid disorders

Abnormalities of thyroid gland structure and function may occur following treatment for childhood cancer either due to primary damage to the thyroid gland itself, particularly from neck irradiation, or secondary to damage to the hypothalamo-pituitary-thyroid axis. Chemotherapy is an independent risk factor for thyroid dysfunction.

Groups particularly at risk of thyroid dysfunction include those treated for thyroid cancer (which is very rare in childhood) and survivors of neuroblastoma who have received 131-I-MIBG. All will require thyroxine replacement therapy. Children with Hodgkin's disease treated with radiotherapy to the neck have a

significantly increased risk of hypothyroidism, thyroid nodules and thyroid cancer compared to those treated with chemotherapy alone. Transiently abnormal thyroid function tests are common in the first few years after treatment but hypothyroidism may develop many years later.

Children treated with craniospinal radiotherapy are also at increased risk of primary hypothyroidism. Cranial radiotherapy is not associated with an increased risk of primary hypothyroidism but may cause 2ry / 3ry hypothyroidism by damage to the pituitary / hypothalamus.

In the past children were treated with low-dose radiotherapy for a variety of non-malignant disorders (eg skin conditions or lymphoid hyperplasia). The risk of thyroid cancer in such groups is significant (<10% over 35 years (Pottern et al 1990, Favus et al 1976)). That radiation is indeed an important cause of thyroid cancer in children (Brill & Becker 1986) has been demonstrated by the effects of the short-lived radioactive fallout from the1986 Chernobyl nuclear power plant accident (Shibata et al 2001).

The prevalence of thyroid dysfunction in survivors treated with total body irradiation seems variable, may be transient and can be secondary to thyroid or hypothalamo-pituitary dysfunction (Borgstrom and Bolme 1994, Katsanais et al 1990, Thomas et al 1993).

Survivors who have received radiotherapy to the neck, brain or spine should have their thyroid function checked after completion of treatment and regularly thereafter – surveillance should be life-long (SIGN Grade B recommendation). There are no good quality studies which address the question of screening for thyroid nodules or second primary thyroid cancers. Although ultrasound may detect more abnormalities than simple clinical examination, their clinical significance is unclear. Survivors at risk should be advised accordingly and asked to seek urgent medical advice if they notice a palpable neck mass.

Thyroid hormone replacement is safe and effective in a dose of approximately 100mcg /m^2/day. Although there is no high quality evidence to support or refute the use of thyroxine in compensated primary hypothyroidism (clinical euthyroidism with normal free T4 but raised TSH levels) it is arguably sensible to treat such patients with thyroxine as persisting high TSH levels may theoretically predispose to malignant change due to thyroid hyperstimulation in these patients.

Puberty and fertility problems

The impact of combination cytotoxic chemotherapy on gonadal function is dependent on gender and age of the child undergoing treatment and the nature and dosage of the drugs received. Drugs known to cause gonadal damage include procarbazine, cytosine arabinoside, and the alkylating agents, particularly cyclophosphamide, chlorambucil, mustine, melphalan, busulphan, and the nitrosoureas. Both the testis and ovary are vulnerable to radiation damage (Waring and Wallace (2000)).

High dose (>24 Gy) radiotherapy to the hypothalamus / pituitary (eg for brain tumours) may result in delayed puberty whereas lower doses (<24 GY) are more commonly associated with early / precocious puberty especially in children treated when they are very young (Quigley et al 1989). Thus early puberty (in boys) and precocious puberty (in girls) are common sequelae in young children who have received cranial irradiation as CNS directed treatment for ALL. The pubertal growth spurt can be mistaken for 'catch-up' growth.

The majority of childhood cancer survivors are fertile. There are low risks of infertility following chemotherapy for Wilms' tumour and ALL and following cranial RT <24 Gy. Abdominal, pelvic and total body irradiation may all result in ovarian damage (Saunders et al 1996). The human oocyte is sensitive to radiation (LD_{50}<2Gy) and the risk of ovarian failure increases with increasing doses of radiotherapy (Wallace et al 1989, Wallace et al, 2003). Infertility or subfertility is

common after CT for Hodgkin's disease RT (Thomson et al 2002). Ovarian failure after TBI is common with the risk relating to age at treatment (younger children are at lower risk). Sex steroid replacement therapy is necessary if there is evidence of ovarian failure, from puberty through to at least the fifth decade, for bone mineralisation and cardiovascular protection.

In young adult women, physiological sex steroid replacement therapy (Critchley at al 1990,1992) improves uterine function (blood flow, endometrial thickness) so that these women could potentially benefit from assisted reproductive technologies (Bath et al 1999, 2001). However they have reduced uterine distensibility with increased risk of small-for-gestational-age infants and miscarriage or preterm delivery (Saunders et al 1996). They should be counselled appropriately and managed as high risk pregnancies by an obstetrician aware of the potential problems.

In boys, the germinal epithelium is much more sensitive to radiation than Leydig cells – 1.2 Gy to the testis will result in azoospermia, whereas >20 Gy (in prepuberty) or >30 Gy (post puberty) is necessary before Leydig cell function is damaged significantly (Shalet et al 1985). Thus spontaneous progression through puberty does not necessarily indicate subsequent fertility. Permanent azoospermia is likely in most patients receiving more than 4Gy.

The current management of ALL in children in the UK includes cyclophosphamide. Although the long term fertility for this group of patients is not known, the available evidence suggests that the total dose of cyclophosphamide (2-3g/m^2) is unlikely to be sterilising (Wallace et al 1993). Treatment for Hodgkin's disease in the UK with "ChlVPP" (Chlorambucil, Vinblastine, Procarbazine, Prednisolone) is known to cause gonadal damage particularly in the male and the agents implicated are chlorambucil and procarbazine. In a recent long-term follow-up study 89% of the males treated before puberty had evidence of severe damage to the germinal epithelium and recovery of

spermatogenesis is unlikely. Around 50% of girls treated for Hodgkin's disease prepubertally with 6 or more courses of ChlVPP had raised plasma gonadotrophin levels, but longer follow-up is needed to determine whether these women have recovery of function or go on to develop a premature menopause (Mackie et al 1996).

As part of their monitoring, childhood cancer survivors should have routine assessment of gonadal function. Counselling is necessary for young people at high risk of infertility and sperm cryopreservation must be made available for post-pubertal boys at risk of infertility before treatment starts. Ovarian cortical strip cryopreservation may allow preservation of ovarian function in the future but remains entirely experimental (Wallace et al. in press). The Royal College of Obstetricians and Gynaecologists and the British Fertility Society have provided reports from working parties on the storage of ovarian and prepubertal testicular tissue (refs) providing standards for best practice in the cryopreservation of gonadal tissue. Strategies to protect the prepubertal testis from damaging effects of CT or RT are under investigation (Meistrich et al 2000, Kelnar et al 2002).

Cardiovascular morbidity

Cardiovascular disease can occur as a consequence of cancer treatment and contribute significantly to the late morbidity and mortality of disease-free survivors (Truesdell et al 1994). The majority of cardiovascular damage is the result of a direct effect by radiation and chemotherapeutic agents (particularly anthracyclines), but an indirect contribution can occur from injury to other organs.

There are no randomised controlled trials examining the cardiotoxic effects of chemo- and / or radiotherapy in the treatment of children and young people with cancer. However there is strong evidence that anthracyclines such as daunorubicin and doxorubicin cause cardiac damage in a cumulative dose-related fashion (Pihkala et al. 1996). The mechanism appears to be focal myocyte death with replacement fibrosis (Truesdell et al 1994). There is probably

no ‘safe’ dose – cardiac dysfunction can occur with relatively low anthracycline doses – and adverse cardiac effects increase over time (Goorin et al 1990, Nysom et al 1998, Sorensen et al. 1995, 1997). Younger age at treatment and female gender appear to be independent risk factors (Lipshultz et al.1995). Higher antracycline doses seem particularly to be associated with prolongation of the QT interval (Mladosievicova et al. 2000).

Mediastinal irradiation increases the risk and incidence of coronary artery disease and myocardial infarction. Specific risk factors are high dose (>30Gy), minimal protective cardiac blocking, young age at irradiation and length of follow-up (Hancock et al. 1993). Patients receiving Total Body Irradiation for BMT conditioning must also be considered at risk. Whilst mediastinal radiotherapy appears to induce atheromatous lesions of the proximal coronary arteries (and similar lesions can be seen in the carotid bulb after cranial irradiation) there is no strong evidence that radiotherapy alters HDL blood lipid levels. Radiation damage has an additive effect to anthracycline cardiotoxicity.

The balance between useful and pragmatic assessment for cardiac dysfunction in those at risk is not easy to determine. The literature supports echcocardiographic assessment at diagnosis and at regular intervals during treatment.

Children who have satisfactory left ventricular function on simple echocardiographic measures, and who have received modest cumulative anthracycline doses (<250 mg/m^2) may benefit from three-yearly echocardiogram surveillance. There is no evidence on which to base recommendations for the monitoring of patients who receive larger doses.

Survivors of childhood cancer who are pregnant, considering becoming pregnant and those wishing to take part in competitive

sports should have a detailed cardiological assessment.

Protective drugs (such as ICRF) are under investigation and may improve the prognosis in subclinical cardiotoxicity (Wexler et al.1996). The data currently available do not support the routine treatment of the damaged heart with angiotensin converting enzyme (ACE) inhibitors such as captopril or enalapril. Although short term improvements have been demonstrated, studies are uncontrolled and not blinded and long term outcomes are unknown (Wexler et al. 1996).

Lifestyle changes (smoking cessation, improved diet, appropriate exercise) should be encouraged. There is no evidence to suggest restricting employment or limiting activities is beneficial. However the risks from competitive sporting activity and pregnancy are likely to be considerable and pre-pregnancy counselling is important so that women patients understand the risks involved.

Renal morbidity

Renal toxicity after successful treatment of childhood cancer is common and leads to a wide range of manifestations of variable severity, and may be irreversible. There are many causes of nephrotoxicity in children treated for malignancy, including the disease itself, chemotherapy, radiotherapy, surgery, immunotherapy, and supportive treatment. Assessment of renal toxicity should include both glomerular and tubular function. The two most commonly implicated agents are ifosfamide and cis-platinum. Ifosfamide nephrotoxicity usually affects predominantly the proximal tubule (causing a Fanconi syndrome), but may also impair glomerular function. Platinum nephrotoxicity (commoner after cis-platinum than carboplatin) causes glomerular impairment and hypomagnesaemia due to tubular damage. Unfortunately an incomplete understanding of the pathogenesis of ifosfamide or platinum nephrotoxicity has hindered attempts at developing protective strategies.

Cognitive, education, social, quality of life and psychological outcomes

Although during the course of cancer treatment children can miss substantial amounts of schooling, a decline in cognitive function is neither a frequent nor inevitable consequence of treatment for childhood cancer (Eiser 1998, 2002). There is a strong observed association between cranial irradiation and structural brain abnormalities (disruption of frontal lobe / basal ganglia connections, temporal lobe calcification and cortical atrophy). Their functional significance is more difficult to determine but impairment may be associated with vasculopathy, calcification and EEG abnormalities (Mulhern et al. 1999). Both structural abnormalities and cognitive impairment correlate positively with dose of brain irradiation and negatively with age at irradiation.

Thus, in the treatment of childhood cancer, cranial irradiation is an important risk factor for cognitive decline particularly in high dosage and young children. Regular review for such deficits should be part of follow-up for patients at risk (SIGN Grade D recommendation). This is likely to have significant resource implications. Screening annually using the Wechsler Intelligence Scale for Children (WISC) may be practical – if a problem is suspected, the patient's cognitive function should be assessed more comprehensively.

The treatment of childhood cancer is likely to impact on educational, psychological and social functioning and thus the impact on overall quality of life may be considerable. Studies addressing these issues are largely observational and outcome measures assessed range from formal psychiatric and psychological assessments through self-completed questionnaires to socio-demographic variables (eg marriage or employment). Adverse outcomes with regard to employment and marriage are, indeed, common findings but the risk of bias in the studies is high. Frank psychiatric disorders seem uncommon but survivors do seem to be at risk of anxiety, low mood and low self-esteem. Again,

brain tumours and treatment with cranial irradiation are frequently reported risk factors for adverse psychological and social outcomes.

There are currently no prospective studies using standardised assessment measures which address particular interventions for preventing or managing adverse quality of life outcomes in these groups of patients.

2nd primary tumours and tumour recurrence

Current knowledge of the longer term risks of second cancers are based on treatments used many years ago, and there will be an inevitable delay before we can assess the longer term consequences of current therapies with confidence. Nevertheless, in the UK, there is a 1 in 25 risk of childhood cancer survivors developing a second primary cancer within 25 years of the primary diagnosis – an approximately 6-fold increased risk (Hawkins et al 1987). It is likely that this relates both to carcinogenic effects of anti-cancer therapies and genetic predisposition to cancer development. Thus the excess risk after all childhood cancers (except retinoblastoma) is related to the carcinogenic effects of radiotherapy and alkylating agents (Hawkins et al 1996, Tucker et al 1987a) and there is likely to be some element of genetic predisposition which would include, for example, constitutional mutations of the p53 gene (Neugut et al 1999).

The large second cancer excess after heritable retinoblastoma is attributable to the carcinogenic influence of both constitutional mutations in the RB gene and exposure of bone to radiotherapy and alkylating agents (Hawkins et al 1996, Tucker et al 1987a).

Second primary bone cancer affects about 1 in 100 survivors by 20 years from the original diagnosis (Hawkins et al 1996). Bone cancers, mostly osteosarcomas, are the most common solid second cancers observed after both heritable retinoblastoma and all types of childhood cancer except retinoblastoma (Hawkins et al 1996). About 7% and 0.5% (respectively) of these two groups of

survivors are affected by 20 years from diagnosis of the original childhood cancer. This corresponds to about 380 and 25 times the expected number of bone cancers, respectively and is attributable to the carcinogenic influence of both constitutional mutations in the RB gene and exposure of bone to radiotherapy and alkylating agents (Hawkins et al 1996, Tucker et al 1987a).

Second primary leukaemia is diagnosed in about 1 in 500 of UK survivors of childhood cancer by 6 years from diagnosis of the original childhood cancer, about 8 times the number expected (Hawkins et al 1992). Increased cumulative exposure to alkylating agents (Tucker et al 1987b) or epipodophyllotoxins (Hawkins et al 1992) increases the risk of subsequent leukaemia. In addition other topoisomerase II inhibitors, including the anthracyclines, appear leukaemogenic .

Second cancer is the leading cause of death in long term survivors of Hodgkin's disease, with exceptionally high risks of breast cancer among women treated at a young age. Breast cancer risk increases with increasing radiation dose up to at least 40 Gy. A radiation dose of 4 Gy or more delivered to the breast was associated in one study with a 3.2 fold (95% CI 1.4-8.2) excess risk. The risk increased to 8 fold (95% CI 2.6-26.4) with a dose of more than 40 Gy (Travis et al.2003). Young age at treatment has a major effect on risk of second malignancy after Hodgkin's disease (Swerdlow et al.2000). Although absolute excess risks are greater for older patients, relative risks of several important malignancies are much greater for patients who are treated when young.

There is still considerable uncertainty concerning the long-term risks of the adult carcinomas observed most commonly in the general population, including carcinomas of the lung, large intestine and breast.

Follow-up of childhood cancer survivors

With improving survival rates there is an urgent need for effective and cost-effective long-term follow-up strategies to be developed (Wallace et al. 2001). There is good evidence of wide variation in the extent to which survivors of childhood cancer are discharged from hospital follow up (Taylor et al. 2004).

Much of the evidence base in these areas is, necessarily derived from descriptive longitudinal studies. Such studies are handicapped by the lack of appropriate control groups and small numbers of patients in individual studies. Whilst this introduces much greater risks of bias than from conclusions drawn from and recommendations based on well conducted randomised controlled studies, this should not devalue the importance of the recommendations derived from such studies. Indeed, many of the studies are distinguished by meticulous attention to detail and report patients enrolled into national and international clinical trials – high quality information describing potential late effects of childhood cancer therapies is available. A corollary of the current dearth of high quality interventional studies to prevent, modify or eradicate such late effects is that collaborative research will, in the future, need to be on a national or international scale.

Who should these patients be seen by? How often should they be seen? How should they be assessed and investigated? Adult cancer specialists are overwhelmed by the large numbers of patients with breast, lung and bowel cancers. In addition, the expertise for dealing with such problems is very different from that required for the appropriate follow-up of childhood cancer survivors.

It will be clear from the above discussion that the degree and nature of adverse long-term morbidity risk will depend on the site of the underlying malignancy, the type and intensity of the treatment given and the age of the child at treatment. Whilst most childhood cancer survivors will require long-term follow-up, this has major practical (eg geographical) and resource (eg expertise and financial) implications. The British Cancer Survivor Study has been developed to obtain

estimates of the risks of particular adverse health outcomes amongst survivors and their offspring and to investigate the variation in risk in relation to the types of treatment received. Such national population-based studies will provide a basis for the further development of long-term clinical follow-up strategies. Clinically-based research will require the maintenance of regular patient contact.

In the context of such developments it is likely that appropriate follow-up strategies will vary between patient / treatment groups. At one extreme, there are survivors for whom the benefit of clinical follow-up (beyond 5 years from treatment completion which equates with “cure”) is not established and for whom annual or even 2 yearly postal or telephone contact may be all that is necessary. Such patients would include those treated with surgery alone (eg stage I or II Wilms’ tumour survivors, some germ cell tumours) or low risk chemotherapy (eg single system disease such as Langerhans Cell Histiocytosis) – level 1 follow-up (table 1).

At the other extreme would be patients who have received radiotherapy (other than low dose (<24GY) cranial irradiation), bone marrow transplantation, or megatherapy (eg brain tumours, stage IV patients of any tumour type). They should be seen in a medically supervised late effects clinic at least annually, and, until final height is achieved 3 to 4 times per annum – level 3 follow-up (table 1). The majority of patients on current protocols (eg chemotherapy-treated or those who received low dose (<24GY) cranial irradiation) would fall somewhere in between. In theory, nurse- or primary care-led follow up on an annual basis might be appropriate – level 2 follow-up (table 1).

What is clear is that if late adverse effects are to be anticipated and monitored to optimise prevention and treatment outcomes this requires a wide spread of expertise. Multidisciplinary follow-up involving paediatric oncologist, paediatric endocrinologist, paediatric neurologist, radiation oncologist, paediatric neurosurgeon, clinical psychologist, general practitioner, specialist nurse and

social worker is necessary but, with so many health care professionals potentially involved, it would seem logical that there should be particularly important role for a key worker for each patient. The primary area of professional expertise will vary with the nature of the patient and their treatment, the intensity (level) of follow-up required and local resources and practicalities. It could be a hospital specialist (eg paediatric oncologist), primary care doctor or specialist nurse. The latter could be a particularly appropriate co-ordinator for many of these patients but there is currently no formal training programme or career structure fore such an individual.

The further development of evidence-based, therapy-based guidelines for follow-up (Kissen and Wallace 1995) are an important prerequisite for an effective and cost-effective follow-up strategy. Further information to guide and inform the future follow-up and management of childhood cancer survivors will come from national population-based cohort studies and large multi-centre clinical studies. Future randomised childhood cancer treatment trials should address systematically not only survival outcomes but also long-term treatment morbidities.

Follow-up outcomes should be audited carefully. As knowledge accumulates, it will be increasingly possible to determine and deliver appropriate levels of surveillance in relation to clinical need so as to deliver high quality care in a targeted, and thus effective and cost-effective, manner.

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Table one

Possible levels of follow up more than 5 years from completion of treatment.

Level	Treatment		Method of Follow-up	Frequency	Examples of Tumours
1	♦ Surgery alone ♦ Low risk chemotherapy		Postal or telephone	1-2 years	♦ Wilms' Stage I or II ♦ Langerhans Cell Histiocytosis (Single system disease) ♦ Germ cell tumours (surgery only)
2	♦ Chemotherapy ♦ Low dose cranial irradiation (<24Gy)		Nurse or Primary Care led	1-2 years	♦ Majority of patients (eg ALL in first remission)
3	♦ Radiotherapy, except low dose cranial irradiation ♦ Megatherapy		Medically supervised late effects clinic	Annual	♦ Brain tumours ♦ Post BMT ♦ Stage 4 patients (any tumour type)

APPENDIX K

Position paper to the Guidance Development Group for Child & Adolescent Cancer in the Specialist Area of Paediatric Endocrinology

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Title

Key issues and concerns raised by the Scope within the specialist area of paediatric and adolescent endocrinology and transition to adult endocrine needs.

Areas of particular input requested on:

- **Composition of MDT's and their skill mix**
- Age transitions and transitions between different patient pathways.
- Continuity of care
- Communication between professionals as well as families.
- Involvement in decision making
- Access to information (for professionals & patients).

Biography:

Dr Spoudeas has 15 years experience in Paediatric Endocrinology in a tertiary centre and has supported the endocrine late effects of cancer survivors at 3 centres (Great Ormond Street Hospital, University College Hospitals and The Royal Marsden Hospital) over that time. She completed her thesis on this topic in 1995 and continues to work in this area leading the neuro-endocrine support service to oncology at the North London Cancer Network, and solely servicing some 1000 patients with a large cohort of brain (>700), bone (100) tumours and bone marrow transplant (>50) patients.

SCOPE

Background

The Institutes' service guidance states it will cross reference other documents as well as those mentioned in 2B of the Scope. It is important that these include:

- The NICE technology appraisal guidance No 42 (May 2002) on the use of human growth hormone in children with growth failure.
- NICE technology appraisal 64 (August 2003) on the use of human growth hormone in adults with growth hormone deficiency.
- Guidelines on adult clinical osteoporosis currently being developed by NICE
- All the Type II Diabetes clinical guidelines (NICE) (Feb-Oct'2002) and the accompanying technology, appraisal for insulin infusion, glitazoma and insulin glorgine (Dec 2002-Aug 2003).
- NSF Framework for children and adolescents – www.dohh.gov.uk/nsf/children/htm
- Guidelines on rare endocrine tumours currently being developed by multidisciplinary groups under the auspices of the UKCCSG and BSPED (available from Dr Spoudeas) to be published end 2004.
- Thyroid cancer society guidelines on thyroid cancer [1,2]

- Royal College Physicians guidelines – pituitary tumours – which include references to childhood disease [3].
- Fertility preservation strategies 2003 – **report of BFS multidisciplinary group**[4].

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Comments

- The current SIGN guidelines on long term follow up after childhood cancer (released 2004), although helpful in many areas, do not, in my opinion include adequate representation from the multiple stake holders, professional organisations and patient groups consulted by NICE currently.

- As a result, although largely robust, they carry some important areas of potential disagreement in the recommendations made, particularly with what I would see as the ideal patient care pathway for the endocrine & neuro-endocrine follow up of childhood cancer survivors.

- These discrepancies pertain to the key areas of age appropriate transitional adolescent and adult services, which groups of patients should be routinely assessed in an endocrine or reproductive setting (both paediatric and/or adult), and at which point in the patient pathway.

- In particular, the Endocrine input to SIGN appears limited to the involvement of one individual who has also functioned as the methodologist for that group, without wider endocrine paediatric and adult specialist consultation. Given that 85% of the late effects witnessed in cancer survivors are potentially (neuro) endocrine or reproductive in origin[1,2] and can be pre-symptomatically detected and treated, this is a potentially important omission.

- Since this is an adult survivors issue [3,4], the apparent absence of representatives from adult specialist experts or stake holders and allied professionals (particularly psychology, psychotherapy, educationalists, occupational therapists and physiotherapists), is also a weakness of the SIGN guidelines, although there is good representation from nurses and primary care.

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Summary of this Position Paper

Needs-Led Services:

- To service the needs of an increasing and accruing number of young adult survivors of childhood cancer, an appropriate age transition needs to be effected with collaboration from dedicated adult ambulatory services.

- Ambulatory and multi-disciplinary one-stop cross-sectional and/or prospective assessment needs to be made available equitably to all cancer survivors as determined by their needs and requirements.

- Effective community, educational and job employment advice needs to be co-ordinated and facilitated through carers, counsellors, youth workers and social workers in these services for young adults.

- Endocrinology, fertility and secondary consequences of obesity are the most important and largest health-related consequences of cancer survival and need to be prioritised in age-appropriate endocrine and reproductive settings.

- Neuro-disability and/or cognitive impairment are particular challenges to long term mental health, independence and employment and appropriate rehabilitative services need to be developed to detect and support those with brain injury, at an early stage.

- To facilitate prospective and complete detailed outcome data for future audit, investment needs to be prioritised towards developing electronic health care records available to multiple users at multiple sites, (including the patients

themselves and doctors in primary care), facilitating electronic health follow up by questionnaire and developing appropriate educational tools for professionals caring for these patients.

- Both professional and patient information needs enhancing and should be targeted to age-appropriate groups as videos, CD ROM's, advertisements and web-enabled (as well as written) information for multi-disciplinary collaboration.

- Governance issues and special standards frameworks preclude the follow up of these children indefinitely in paediatric oncology centres. The development of new ambulatory services bringing in multi-professionals and interfacing with both those adult survivors treated as adults, as well as those adult survivors treated as children, needs to be effected in tertiary/quaternary cancer centres.

- All children with cancer should be assessed by endocrinologists and/or reproductive health specialists at least once in their childhood (peri-pubertally 11-12yrs) and once in their young adult (at adult height 16-18yrs) lives. Preferably this should again occur at the end of their treatment and at adult transition (19yrs), so that patients can fully understand what benefits may accrue, and have contacts which they can re-activate as necessary.

- In my opinion **every** high risk patient who has a brain or CNS tumour, has received craniofacial , spinal, pelvic or thyroid irradiation, bone marrow transplantation, high dose or multiple therapy for relapse should be seen prospectively and regularly from diagnosis or the end of treatment on a 6 monthly basis to adult height in an endocrine setting. This should be mandatory for all patients with tumours involving an endocrine gland or situated close to it (including all central, optic, sella/suprasellar, pineal, hypothalamic tumours).

- All minors (<16yrs) multiply-treated or receiving known significantly gonadotoxic agents, as defined by UKCCSG recent guidelines, should be seen and counselled by an appropriately trained professional for fertility preservation techniques before treatment and followed subsequently in an endocrine and reproductive setting at the end of treatment, and at intervals thereafter, for reproductive counselling and HRT.

- The ideal multi-disciplinary team should include age-appropriate professionals (paediatric, adolescent and/or adult), facilitating transitions at ages 12-13yr and 20-25yrs, and access to appropriate services. The core team should ideally include neurological, (neuro) endocrinological, (neuro) psychological, oncological, reproductive, cardiac and renal medical expertise, appropriate diabetic, psychotherapeutic, occupational and physiotherapy support, a dedicated CNS practitioner trained and experienced in endocrine, reproductive as well as, oncological counselling issues., and allied professionals (including alternative and homeopathic therapists as appropriate), providing play therapy, career counselling, schooling advice and social work. Age appropriate psychiatry would be a helpful addition and should at least consist of access to an identified individual with appropriate family therapy, systemic or cognitive behavioural skills, responsible for supporting those at risk of significant mental health issues and/or family break-down.

CONCERNS RAISED BY SCOPE

Redesign of services to meet need

- Infants, young children, adolescents, and young adults in their early 20's are considered in the scope. Thus, by definition, patients may present to adult specialists, making a better collaboration between paediatric and adult specialists in a multi-disciplinary setting, vital. This collaboration might exist in

both acute oncological care and rehabilitation. It is my impression that these two aspects of healthcare are better separated into 2 settings. On occasion they will need to occur hand in hand, as is the case in young people with brain tumours, (particularly those centrally positioned), those receiving high dose gonadotoxic chemotherapy (where fertility preservation and counselling will be required), and those undergoing heavy treatments (such as bone marrow transplants or high dose therapy). In others, it is possible that an end of treatment or 5 yr MOT type assessment will suffice. Importantly endocrine referral should be virtually routine at, 1) end treatment, 2) pubertal age, 3) end of growth and 4) adult transition.

- Professionals involved in acute oncological care may wish to be part of the late MDT rehabilitation team. This makes for good continuity of care but can sometimes inadvertently prevent non-oncological specialist and expertise information being equitably accessed by all patients. To avoid this, it is likely that those participating in the late follow up MDT require adequate funding and recognition of their roles, under an identified key late effects co-ordinator; this could be a paediatric late effects nurse or late effects consultant, responsible for the necessary timely referrals for specialist expertise. Once the referral has been made, certain basic recommendations for follow up should be made (eg: 5 yearly assessments for those who are at least risk, cross sectional assessments at age appropriate transitions, or annual (6-12 monthly) intervals for those at sufficient risk of growth and endocrine abnormalities (see appendix 1*). However it is likely that endocrinologists would prefer to follow patients on a 3-6 monthly basis to obtain a more detailed picture of the risk-benefits balance of earlier intervention, particularly in high risk patients.

- Benign as well as malignant tumours are considered as part of the scope, where treatment is complex, including chemotherapy and radiotherapy. This will include many tumours which currently present to neuro-surgeons or

endocrinologists rather than paediatric oncologists, (eg craniopharyngiomas, thyroid tumours, adrenal tumours and rarer pituitary and parathyroid tumours which form part of the MEN syndromes). These tumours are the current topic of our multidisciplinary collaboration to develop consensus best practice guidelines between the BSPED and UKCCSG aimed at improving the outcome and survival of these rare cases. In these cases it may be more appropriate that the key co-ordinator leading the treatment care pathways and late effects pathways is an Endocrinologist, (rather than an oncologist) collaborating with oncology. In other cases, radiotherapists and/or neuro-surgeons may wish to take the lead, but a multi-disciplinary team, also able to deliver ophthalmological, auditory and neuro-psychological and neuro-developmental assessments is important.

- This multi-professional contribution is at least as vital, if not more so, than the co-ordination of care by a key worker and currently remains unrecognised and unfunded in the complex cancer care service provision. More resources should be allocated up-front towards rehabilitation as well as acute therapy.

- Another model is the empowerment of the patient himself to drive his own rehabilitation pathway through appropriate information and/or electronic health record as necessary, or through a primary care physician.

- It would seem appropriate that the information technology develops to a capacity to support the centralisation of this very important aspect of cancer care and survival. This model needs considering under the ideal health care setting and services provision & whether or not this is delivered in secondary, tertiary or community care. I would favour a “spoke & wheel” service (see later).

Key areas of Clinical Supportive Management

It is an omission in the current SCOPE that paediatric endocrinologists, endocrine surgeons and neurosurgeons are not mentioned as practitioners involved in the diagnostic services. Their particular relevance to those children presenting with brain tumours involving the pituitary area or other endocrine tumours, can-not be underestimated.

Similarly under oncology treatment services, specialised endocrine/reproductive and late effects nursing is not mentioned. This omission underlines the reasons for the under funding of such support services to acute and chronic oncological care and requires rectification.

CLINICAL NEED FOR THE GUIDANCE:

Age – Appropriate and Needs – Led Service

a) General Comments

This Scope clearly defines the importance of community support staff and social care and the need for specific age appropriate services.

- Age appropriate services are also a stated aim of the Children's NSF framework with an increasing awareness that children (<12yrs), adolescents (12-20yrs) and, possibly also young adults (20-30yrs), should be seen in their own specific age-appropriate and needs-led services, in separate areas from older or younger age groups. The current practice of seeing survivors of childhood cancer in paediatric services, usually paediatric acute oncology services, I would see as inappropriate both for their long term specialist and non oncological rehabilitative needs and for meeting those needs in age appropriate services.

- There are clearly governance issues if paediatric specialists continue to see patients well into adult life without appropriate training or support services in those areas, or without adult colleagues and their specialist expertise. The current SIGN guidelines suggest that patients' needs are not adequately met solely by adult specialists, specifically adult oncologists, and highlight the need for appropriate training in this area, but they do not adequately consider the establishment of one-stop, age appropriate multi-disciplinary specialist, rehabilitative surveillance services, which I would wish to see endorsed. The skill mix of such a service should be targeted to meet patient needs - based on disease and treatment related criteria (***see later***). For example psychological and/or psychotherapeutic services (with access to psychiatry and fertility) are an important part of the rehabilitation service for patients and their families whilst dedicated neuro-rehabilitative support to those with significant brain injury from disease, treatment or treatment-related complications, carry evidence based-benefits in the longer term. Such early rehabilitative support has long been advocated for children & young people with brain and spinal tumours [(1)] and more recently in adults with brain injury [(2)], but access to such services is likely to be confined initially to specialist tertiary centres with dedicated funding streams. Equitability of access for all eligible patients becomes of real ethical concern, but patients themselves could be better empowered and demand these services through information about service provisions & follow up/surveillance choices.

- The NHS needs to prioritise funding for better multi-disciplinary preventative and rehabilitative care of such patients and develop collaborative services between adult and paediatric sub-specialists and between primary, secondary and tertiary care. In this way, better training and research in this area can be effected, patients needs can be identified and patient choice can be enhanced.

- Prospective national registries and audits of detailed, comprehensive, functional, endocrine and quality of life outcomes should be encouraged, to better determine levels of need, treatment-and-disease related toxicity and the as yet undefined contribution of social adversity. Such prospective outcomes have been lacking to date in UKCCSG clinical trials which have prioritised survival alone, without addressing the quality of that survival. In the absence of such data changes to cancer therapeutic protocols have been driven by assumption rather than evidence as to treatment-related toxicity [3&4]

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A) AGE – APPROPRIATE TRANSITIONAL SERVICES;

I would support the development of:

a) Integrated, needs led and assessment based services for specific groups of childhood cancer survivors according to degrees of late injury.

b) In age-appropriate one stop, multi-disciplinary settings in order to maximise knowledge and efficiency.

c) Such services should be delivered from tertiary centres in a "spoke & wheel" design with centralised data collection for the purposes of audit, education and treatment recommendations to primary and secondary care centres.

d) These services could conceivably well interface with late effects/neuro-rehabilitative services for young adult survivors of cancer treated in adulthood as well as childhood; such adults may have lesser levels of need, but likely similar issues and experiences.

e) Since long term cancer survival is very much poorer in adults than in children, the large majority of survivors of adult onset disease will have been treated for breast cancers, lymphomas and leukaemias and will have similar late effects issues to young adults treated in childhood for these conditions. An example of such a pilot development service is attached (appendix 2*).

f) An interface between the adult and paediatric services, with professionals from both represented in the multi-disciplinary ambulatory setting separate from acute oncology, would be ideal for future research, service provision and training of future health professionals. Such a service might be better termed a "SUCCESS" service (SUrviving Childhood Cancer, Empowerment, Surveillance & Support) than a "Late Effects" or "After-Cure" service which lacks specificity.

B) CHILDHOOD & ADOLESENT POPULATION

Schooling, Neuro-disability Liaison & Endocrine Need (85% of the "late" problem):

- A single screening tool devised to identify endocrine and (neuro-) psychological needs in a dedicated multi-disciplinary "after-cure" rehabilitative

and surveillance service should be a necessary part of the **end of treatment** assessments for **all** children with cancer. Leaving these assessments for a period of 5 years until "cure" has been defined, may be too late for some, compromising quality of life issues such as (HRT) hormone replacement therapy, obesity, peak bone mass, or age-appropriate puberty(1) sexual(2), reproductive(3) potential and schooling & employment(4).

- Given that survival rates are high(5) each child cured of cancer has a further 68 yrs of potentially reduced quality of life ahead (compared with 10yrs for each adult). The vital importance of early (neuro)-rehabilitation back into their community, education & future employment can-not be understated. This process is currently neither streamlined according to need nor equitable for all groups of patients across the UK. The "after-cure" care services for long term survivors have traditionally been seen as of secondary import to those of acute cure and received consequentially less attention, NHS prioritisation and funding (6). This needs to change.

- Inparticular the liaison with schools and with further educational colleges for the identification and support of specific learning needs, (up to the age of 19), should be a vital part of services for every child(4).

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- Those deemed at particular risk, high dose therapies, identified from screening assessments(1), or brain-injured, (ie: CNS tumours, cranially irradiated or multiply treated) should be prioritised for additional support and assessments from psychologists, (eg: clinical, educational or neuro-psychologists) physiotherapy and occupational therapy, visual and hearing assessments to aid in targeting support and concessions in school and prepare them for adult transitions. These assessments should be prioritised at important age and maturational transitions, such as entry into primary and secondary schooling, prior to GCSE's and between the ages of 16-19yrs for young adult transitions. (*see example of pathway for neuro-oncology appendix1*).*

- Career advice and support, from youth workers and/or career counsellors or social workers is vitally important at these adolescent transitional stages above. The input to adult transition is currently woefully absent, many patients and families struggling to access neuro-disability services, sheltered accommodation and structured employment, retraining and rehabilitation schemes(2,3).

- To achieve the independence and employment which survivors require, certain high risk individuals and their families may also need accessible child & family psychiatric/psychological/ psychotherapeutic support and/or a period of inpatient assessment and neuro-rehabilitation in a dedicated special facility, particularly in young adult life.

- Those most in need with motor, sensory and neurological deficits, multiple pituitary deficits, visual and hearing impairments, cognitive impairments, which may progress with time, will require longitudinal assessments as they mature and particular help at adult transition.

- In some late evolving or severe cases, appropriate assessments and support will be necessary in young adulthood and access to the appropriate brain injury and neuro-disability services, currently extremely difficult, could be better enhanced for those with significant brain injury and/or other physical and psychiatric disabilities. This small but important and growing group of brain injured survivors (some 30-40%of all survivors) has repeatedly been neglected in the many reports of late effects, which have largely concentrated on cross sectional hormonal and growth assessments in other survivors, (potentially treatable and preventable) rather than more in-depth analysis of causation of specific organ dysfunction [1-3]

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C) NEED Adult Population of Survivors:

- Prospective national data registries of, endocrine, quality of life, functional & neuro-psychological assessments from diagnosis to adult life which include all relevant treatment protocols and account for missing data are becoming increasingly important to identify needs for “rare” tumours, such as children’s

cancer generally, not withstanding specific very rare tumour groups within this category. This is especially the case as cancer treatments become more intensive and prolonged with heavier chemotherapy and its multiplicity of late side effects, as well as the potential for additive toxicity where two treatment modalities co-exist. There is a widespread belief that systemic chemotherapy has no central neuro (endocrine) toxicity, but this is clearly not the case as exemplified by growth hormone abnormalities[1] and platinum ototoxicity in children with brain tumours. All children will require careful endocrine surveillance beyond adult life as deficiencies evolve[5]. There is assumption that chemotherapy alone has little neuro (endocrine) toxicity, but this is clearly not the case[1&4].

- Whilst we are in a position to only accrue retrospective data on those who survive and are seen comprehensively in the above tertiary endocrine and rehabilitative settings, we are unable to document the size of any problem or determine the best interventional therapy. New treatment strategies change the picture; fertility protection strategies are increasingly available on an "*ad hoc*" basis but they are currently not funded and their risk benefit profile or cost efficiency is unknown[6&7]. Randomised studies in this area have never been performed and without adequate documentation of the outcome of both those who do and those who do not undergo such treatments, we will be unable to determine this in the future. The current endeavour (Nov'03) by the late effects group of the UKCCSG to collect such data prospectively over 1 year is therefore important, particularly as only 30% accrual to a national retrospective study of fertility after Ifosfamide regimen, has been achieved, but at best this can only be a single cross sectional snapshot of what exists at a given time. This is why prospective very long term longitudinal data should be encouraged and collected anonymously.

- In the absence of prospective detailed endocrine and neuro-psychometric evaluations from diagnosis, before and after each therapeutic modality,

much late toxicity in this area is blamed on the treatment itself, particularly cranial irradiation[8&9] . Arguably, however, it could be just as much due to the tumour itself, the surgery employed, the peri-operative complications and circumstances of psycho-social adversity, and inadequate rehabilitation as to the chemotherapy or radiotherapy employed to effect a cure. Recent studies have not confirmed previously held beliefs[10] as to the causation of endocrinopathies observed after cranial irradiation.

- The "moving baseline", of constantly changing cancer therapies and the long lead time necessary for documenting late post-maturational organ toxicity[5], means that much of today's evidence of late effects come from already outdated treatment regimens used 10-20 yrs ago in existing adult survivors. This demonstrates the importance of comprehensive prospective longitudinal data collection on all treated patients whether or not they be survivors or on randomised trials within the UKCCSG, ensuring detailed treatment datas are available to all professionals caring for these survivors. The BSPED is working currently to try to achieve more comprehensive endocrine & QoL data collection through national endocrine registries for endocrine and brain tumour with the UKCCSG.

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D) MAKE UP OF MDT

- Depending on the mix and needs of patients, different MDT's could be established. The role of a new specifically trained CNS Practitioner to this service with oncological, endocrine and reproductive diagnostic counselling skills should be developed[(1)]. Additional medical and allied professional expertise in neuro-oncology, neurology, including epilepsy, neuro-rehabilitation, (neuro)-surgery, (neuro)-endocrinology, (neuro)-psychology, occupational and physiotherapy as well as psychotherapy and career advice

from a youth worker, teacher or educational psychologist may all be required for brain injured survivors of CNS tumours or CNS directed therapy (eg: intrathecal or systemic chemotherapy, cranial irradiation and/or cerebrovascular accidents, high dose or multiple intensive therapy) with subsequent cognitive impairment, and potential neuro-psychiatric disorders. The latter has been particularly under investigated but is potentially treatable, and is the subject of a recent grant proposal submitted to the CRUK from UCLH (Paediatric Endocrine Dept) Cambridge (Department of Neuro-psychiatry) and Birmingham (British Childhood Cancer Survivor group).

- Specific rehabilitative inpatient assessment units (eg: the multi-disciplinary teenage assessment service at UCLH) need to be made more available to specific groups of needy individuals, with a view to better in-depth assessments of need, and enhancing independence.

- The ultimate aim of this service guidance is to improve quality as well as quantity of survival. As children mature it is clearly important to obtain their own perspective, (increasingly shown to be different from predicted by parents' and professionals) and ascertain their needs.

- I would argue that a paediatric endocrinologist (and ultimately a reproductive or adult endocrine specialist) should be involved from an early stage in the assessment of growth, development, puberty, reproductive health after cancer and that any multi-disciplinary team supporting long term rehabilitation and surveillance for childhood cancer survivors should at least include these two members of the team

E) PROFESSIONAL/SPECIALTY EXPERTISE – THE IMPORTANCE OF ENDOCRINOLOGISTS:

- Whilst it is clear that many paediatric oncologists are developing expertise in the area of late toxicity, given that 85% of late effects are hormonal in nature and affect up to 70-90% of survivors, most patients would benefit from an independent expert assessment of growth, puberty, fertility and future bone and reproductive health, from an endocrine and reproductive specialist at least once.

- To be equitable, these services should be accessible to all survivors at preferably at 4 cross sectional periods (eg; a) at the end of treatment, b) onset of pubertal age, c) at the end of growth, d) as a young adult), this being combined with a psychological /psychotherapeutic, Quality of Life or functional assessment questionnaire at the same time. In other words, certain MOTs could be performed in the tertiary setting ("spoke") with recommendations made to secondary and primary care ("wheel"). (*Recommendations for assessment and earlier referral could be made as per appendix 3*).*

- Other more intensively treated high risk groups, brain tumours, especially where centrally positioned, those receiving high dose therapy, bone marrow transplantation or cranial irradiation) should be prioritised for expert assessment from an endocrinologist soon after treatment has finished and at least annually thereafter.

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- Those with tumours in endocrine glands or very closely situated to them or tumours positioned centrally in the brain, which can have life threatening effects from pituitary dysfunction [(2)] should be assessed **at diagnosis** by endocrinologists as well as oncologists; a collaboration to achieve better

registration and treatment for these rare diseases is in currently in progress (UKCCSG rare tumours group and the BSPED endocrine tumours group) & will report by the autumn of this year.

- Those whose treatment protocols put them at significant risk of sub-fertility should be pre-pubertally assessed and counselled regarding fertility preservation techniques by someone suitably trained in all the pertinent areas of counselling, pubertal assessment , legal aspects of consent and gamete cryopreservation & storage(3).

- As children mature, their information and development needs change. By nature of their speciality, endocrinologists are very used to issues of adolescent transition, Quality of Life (as opposed to life saving decision making), and counselling (eg: with respect to short stature and infertility), the dangers of hypothalamic hypopituitarism, thirst & sleep disorders, obesity and secondary glucose intolerance. The latter is a likely consequence for the large majority of survivors(4&5).

- Adult endocrine transition services for monitoring consequent endocrine problems which include adult growth hormone deficiency(6) , osteopenia, hormone replacement therapy, assisted reproductive technology, secondary insulin resistance & diabetes, panhypopituitary, cardiac, renal & other health-related risks are necessary to alleviate and potentially prevent these important consequences (see attached articles for review).

- Hypopituitarism and hormone replacement therapy also impact on quality of life. HRT even in the pre-symptomatic patient, can prevent decline in health & well being, decrease mortality from hypopituitarism and its related complications(7&8). The effects of hormone deficiency are subtle and difficult to recognise and the interpretation of endocrine tests in different centres, using different methods of assessing hormonal reserve, require interpretation

by those appraised of its difficulties and pit falls[9&10]. Equally, to the untrained eye, advanced rates of growth from precocious sexual maturation, or obesity, can be mistaken for catch up growth and their underlying endocrine implications missed depriving the patient of potentially beneficial treatment intervention to enhance growth and sexual reproductive capacity.

- Many of the late effects of treatment relate to oncology therapies; however they can equally reflect primary as well as secondary neuro-endocrine issues which might pre-date treatment as well as result from it. Thus specialised endocrine nurses are likely to be of enormous help particularly in the management of neuro-endocrine conditions in the MDT setting (eg: in the assessment of the adipsic and hypopituitary patients, in the assessment of growth & puberty and in the education of patients and families in the management of hypopituitarism and diabetes insipidus and emergency Hydrocortisone rescue. Age appropriate and adult specialist endocrinology and endocrine nursing support should be a part of the MDT late effects service. This is especially required for brain tumours, tumours of the endocrine glands or treatment related endocrine toxicity, or where high dose steroids are used as part of treatment and may cause adrenal suppression [1&11].

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F) INFORMATION TECHNOLOGY DEVELOPMENT: - The need for audit:

Audit – Suggested Requirements to Identify need:

- The audit of long term quality of survival and toxicity issues after childhood and adolescent cancer needs to be prospective and long term. Most existing

studies lack the denominator of patients and are retrospective in nature. There are few, if any, detailed prospective and longitudinal outcomes and these registries are currently being discussed for rare endocrine tumours with pump-priming pharmaceutical company support. Since there are only about 300 brain tumours and endocrine rare tumours annually in the UK, it is not an impossible task to collect endocrine data on all such patients over a 5-10yr period at 6 monthly intervals. This could potentially be combined with functional and quality of life measurements at appropriate longer intervals. Such endocrine outcomes would include detailed weight, height, sitting height, skin fold thicknesses, puberty staging, thyroid, gonadal and pituitary function tests and their relation to functional status, health-related quality of life, HRT treatment, adult bone mineral density, body mass index, reproductive status, thyroid and pituitary function, systolic cardiac function, lipid status, & potential for atherogenesis. Quality of life & tubular renal function would be added because of their relationship to adult growth hormone deficiency and osteoporosis respectively. Semen analysis, ovarian and uterine size and other pelvic assessments are appropriate in adult life in patients contemplating pregnancy and pre-pregnancy risk assessment. Specific other endocrine data will be required on patient with adrenal disease and/or panhypopituitarism.

- The whole service could be better streamlined with a specifically tailored, web-enabled and password protected, electronic health care record for the centralisation of all data, the standardisation of any treatment recommendations, streamlined access to the personal cancer treatment history, for multi-disciplinary professionals (potentially at many sites) and providing information to patients, schools, employment and community networks. This needs to be given priority as a currently achievable goal, given the NHS IT strategy. This is a recommendation of the NSF Diabetes framework, another long term chronic illness with similar issues of transition and self empowerment.

- This endeavour would support standardised prospective and longitudinal data collection of outcomes of interest to ascertain the best interventional strategies for preventing and/or curing late organ dysfunction. Specific endocrine, cardiac, renal, respiratory, quality of life, psychometric and psychiatric screening evaluations, undertaken nationally in a few dedicated centres, could improve the future quality of survival, just as national collaboration through the UKCCSG has improved the quantity of survival (An example of a data driven form for such services viewed in a paediatric setting (appendix 4*) and adult setting (appendix 5*) are included.

- The recognised difficulties in collecting prospective data over many years mean that innovative ways of collection through collaboration with interested other societies and their professionals (eg the endocrine, fertility and neurological societies) could benefit patients, particularly those at greatest risk. The latter could most helpfully be considered as those who are most intensively or multiply treated or where disease involves a vital organ such as the brain or reproductive tract.

Palliative care

- It is my understanding that palliation includes the alleviation of suffering in those living with, as well as dying from, an incurable disease. Several patients surviving brain tumours, high dose therapy, total body or spinal, craniofacial or pelvic irradiation, will have incurable and possibly painful secondary consequences which require long term support. This potentially palliative service should not be underestimated in its importance. An increasing number of survivors are being recognised to suffer from chronic fatigue symptoms (anecdotal case reports), (currently unexplained) severe hypothalamic disturbances [(1)], obesity, thirst and water imbalance and the secondary consequences which follow, (diabetes and heart disease being of

particular concern) [2&3]. Services established to support these disabilities might also be considered palliative, particularly where suffering becomes extreme, limits quality of life, self esteem and adult independence and perhaps causes the suicides noted[4]. This is a particularly under-researched area of cancer survival and one which has potentially very important funding implications. Inpatient or ambulatory assessment services may need to be developed to support those with chronic disease and the role of complimentary or alternative therapies is still to be explored.

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Support services

Information & Access:

- Better community, primary and secondary care liaison and information is necessary. If we are to enhance ultimate independence in those most at risk, we might require access to family therapy, behaviour and intervention strategies, improve school awareness of the needs of children, enhance access to neuro-disability services & social housing, career advice & job opportunities in adulthood. An example of how this might look for a child with

a brain tumour is attached(Appendix 1*). This aspect of transition is particularly poor are present, many families being unaware and unable to easily access specialised services for the visually or hearing impaired or distressed young adult. Part of this arises from the fragmentation of care of survivors in different specialities and in age inappropriate settings (an example of endocrine and sperm banking leaflets are in *Appendix 6&7*)*

- Although we know survivors are less likely to be employed, form sexual and peer relationships or become parents, and lack self esteem and confidence (1), the necessary career counselling and psychometric evaluation to inform and support future employment & rehabilitation in the adult work place is severely lacking. Long term adult depression is a potential concern in those cognitively impaired (2) but it is treatable and could improve function and quality of life. More psychotherapeutic, diagnostic and therapeutic strategies are necessary to effect adult transition and independence. A specific information-giving MDT assessment is potentially beneficial at this stage for all young adult survivors.

- These MDT assessments are not easily available or appropriate in the paediatric oncology departments where these patients are currently, predominately seen. It is my view that a few tertiary/quaternary specific young adult services need to be developed which should be reasonably accessible through self or GP referral (eg via, newsletters to survivors or Internet) . These services could provide information (see earlier IT section also) and perform needs assessments to deliver appropriate information and rehabilitative strategies to survivors, including hormone replacement, +/- career advice, , independence skills and access to information, and social health care. Individuals could thus be empowered to make their own choices at times appropriate to them.

- The voluntary sector may be able to assist in some of these areas and better information and links need to be established to specific existing units.

- In addition, 5 yearly 'MOT' assessments could be offered to all patients, if their record of care could be centralised and accessed remotely by primary and secondary care services. The necessary IT software could be developed to support this and the data collection, questionnaires and standardised hormone assessments, and treatment could be forwarded from the centre to the primary and secondary services as necessary, according to individual patient preference and funding. The most important development which would need to occur to facilitate this for **all** survivors, (rather than just those with high priority) is the development of a web-enabled, password protected, electronic health care record which meets the data protection criteria of the European union, is accessible by patient, primary, secondary and tertiary health care professionals, and carries levels of alert to the physicians involved. This could also provide information for the patient and feed data back through a centralised collection system to primary care so that appropriate appointments can be sent out as necessary for those patients deemed needy. A similar model has been proposed for diabetes. A local grant proposal has been submitted from our Trust but at present no funding has been identified and this model does not exist elsewhere.

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* Appendices are available from the NCC for Cancer upon request

APPENDIX L

Paediatric palliative care
A position statement

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Sources

This document draws largely on two sources of evidence:

1. That provided by families and professionals to the Association of Children with Terminal or Life-threatening illnesses and their families (ACT) in collaboration with the Royal College of Paediatrics and Child Health regarding palliative care for children, and for adolescents and young people (1-4).

2. Selected data from a recent regional research project (5) regarding children needing paediatric palliative care in Wales. The data (table 6, figures 1 and 3) included children referred for specialist paediatric palliative care, those who were reported by paediatricians to need palliative care but were not referred, and those who were referred to a Children's Hospice. The full report is being prepared for publication as a scientific paper and can be made available to the Committee if required.

1. The nature of paediatric palliative care

"Palliative care for children and young people with life-limiting conditions [LLC] *is an active and total approach to care, embracing physical, emotional, social and spiritual elements. It focuses on enhancement of quality of life for the child and support for the family and includes the management of distressing symptoms, provision of respite and care through death and bereavement." (3)*

Palliative care for children differs significantly from the adult specialty. Where adult services predominantly focus on cancers, children's palliative care services must cater for a wide spectrum of very different conditions. Furthermore, in comparison with adults, the palliative phase in childhood is usually characterised by greater uncertainty, and often by intermittent or fluctuating need for specialist

involvement over many years or decades. The families of children with life-limiting conditions may in effect experience many 'terminal' phases.

2. The nature of children who need it

2.1 Who are they?

The range of conditions needing specialist paediatric palliative care is very wide. The Royal College of Paediatrics and Child Health, in association with the Action for Children with Life-Threatening diseases (ACT) has identified four groups of conditions (table 1). Cancer is an important example, and the single commonest condition, but a greater number (table 6) of children have life-limiting conditions that are not malignant (5).

By reviewing the available evidence, the ACT/RCPCH Guide concludes that around 10:10000 children aged 0-19 in the UK have life-limiting conditions (3), of whom 10% per annum will die from their condition. This approximates to 12000 children in England and Wales of whom 120 are likely to die from the condition in any one year. According to the Guide, 40 will die of cancer, around 20 from heart disease and 120 from other conditions. These figures are generally accepted, but may even be an underestimate (6-12).

A recent study (5) suggests that many are not recognised. The study also confirmed the ACT/RCPCH finding that for every one child with cancer, there are three with non-malignant life-limiting conditions with palliative care needs. Even among those who were recognised, for over one third of patients (36%) their paediatricians felt palliative care was inadequate. The usual reason was too little respite provision.

2.2 Where are they?

Flexible palliative care means that care should be available to children in whatever environment they find themselves. In practice, there are four main areas: home, school, hospital and children's hospice (table 2). A specialist paediatric palliative care team should ideally be able to support all carers in all environments (fig 2). **The exact nature of support will vary according to the needs of the individual child, family and team of professional carers (2).**

2.3 What do they need?

A 1998 pilot palliative care project for the Department of Health (13) identified 17 specific needs for children and families with LLC (table 3). Paediatricians identified similar issues in their patients (5):

1. Coordination of services in community (97%)
2. Physical symptom control (95%)
3. Emotional etc. support for family (95%)
4. Respite (82%)
5. Discussing prognosis with family (79%)

These findings emphasise the multidimensional nature of PPC, and consequently the need for an approach that combines the skills of many different disciplines having in common expertise in working with children for whom cure is not, or is no longer, possible.

The RCPCH/ACT Guide authoritatively set out the scope of conditions that may limit the life of a child (table 1) but left some definitions unclear. It did not, for example, distinguish respite from other aspects of palliation, or define 'active' or 'specialist' paediatric palliative care. A working glossary for this document is given in table 4.

Doctors and nurses have a long history of working closely together in paediatrics

and child health. Children's palliative care in particular is a field in which the skill sets of doctors and nurses often overlap. It is often assumed that symptom control is the purview of doctors, and psychosocial issues are best dealt with by nurses. In managing children with LLC, while doctors are often skilled at physical symptom control and nurses at other aspects of holistic symptom management, both sets of skills can be found in members of both professions.

The skills in Table 4 therefore apply equally to nurses and to doctors. They do not abolish professional boundaries; doctors remain accountable to their profession for their own prescribing practice, irrespective of the degree of expertise of the nurse advising them.

2.4 How should we ideally meet their needs ?

After wide consultation throughout England, Wales, Scotland and Northern Ireland, the RCPCH and ACT made recommendations regarding the ideal provision for paediatric palliative care in British regions (1, 3), which are summarised in table 5.

In summary, they emphasise the need for a network with three key elements:

- A sound community children's nursing infrastructure.
- Skilled medical support from general paediatricians with an interest and some training in paediatric palliative care (one per NHS trust) and from tertiary specialists in paediatric palliative care (one per region).
- Coordination and continuity of care through:
  - a system of named key workers and dedicated coordinators liaising with primary, secondary and tertiary care and also between statutory and voluntary providers of paediatric palliative care.
  - multi-disciplinary teams
  - an appropriate documentation system based on parent or patient-held

records.

3. What do we have in the UK at the moment?

Services were recently audited against these ACT/RCPCH recommendations (table 5) in Wales. It seems likely the results are representative of the UK as a whole, suggesting that while there are some areas which are progressing, there are others in which improvement is still needed even five years on.

3.1 Strengths	3.2 Weaknesses
• A small number of tertiary specialists in paediatric palliative medicine. • A small number of general paediatricians developing a special interest in PPM. • A small number of general practitioners working within Children's Hospices developing a special interest in symptom control. • Well-developed subspecialty outreach models of care, involving highly trained and experienced Clinical Nurse Specialists in a variety of sub-specialities including oncology, respiratory, neurology and neonates. • A network of 20 – 30 Children's Hospices providing high-quality respite care, generic and semi-specialist palliative care (2). • Diana Teams in many parts of the UK, providing nursing care to dying children at home (14). • Beacon developments in community provision of generic	• Still very few consultant specialists in paediatric palliative medicine. • Weak research and evidence base: practice often anecdotal or drawn from adult practice. • Very small academic base. There is only one medical senior lecturer in paediatric palliative medicine in the UK, and few academics from nursing or other disciplines. • Very few training opportunities for doctors, nurses or other disciplines wanting to make care of dying children their main interest. • Tendency of each provider of palliative care to children to see themselves as the only, or the main, providers rather than as part of a group of services. • Inconsistent provision of community paediatric nursing teams across the country. • Slow (though often well coordinated) access to continuing care funds. • Little specific provision for adolescents. • Clinical record-keeping and audit

palliative care such as the Avon Lifetime Service (10, 15). • Numerous voluntary and charitable services providing resources that overlap, sometimes substantially, with palliative care. • Unified community and hospital trusts in many health districts, facilitating transfer of inpatients into home. • Well-developed adult palliative care services serving as a model from which the paediatric specialty can learn. • Increasing numbers of paediatric palliative care training programmes for nurses and one course for doctors. There is a larger selection of resources for adult palliative medicine which are of less relevance but may still be useful to those working with children.	system do not cross boundaries. • Inconsistent professional supervision of carers. • Lack of regional or national databases of children needing palliative care. • Lack of consistent child psychiatric support in most regions. • Lack of child bereavement facilities • Lack of therapeutic supervision for carers

4. What could we have in the future?

4.1 Opportunities for development

4.1.1 Paediatric palliative medicine

The multi-dimensional nature of palliative care means that it cannot be provided exclusively by any one profession. Traditionally it is an area of care that nurses have taken more seriously than their medical colleagues. Increasingly, however, doctors are recognising the role that they can play in supporting nursing and other colleagues in caring for dying children. There are currently six consultant specialists in paediatric palliative medicine, of whom one is a nurse and five are doctors. They are dispersed as follows: Great Ormond Street, London (2), Cardiff (1), Alder Hey, Liverpool (1 nurse, 1 doctor) and St.James' University Hospital, Leeds (1). There is a paediatric oncologist in Southampton with two sessions dedicated to palliative medicine in children with cancer. There is an increasing number of consultant paediatricians taking on palliative medicine as a special interest, and some Children's Hospice GPs who are undergoing further training in order to become 'GPs with a special interest'. Numbers are not exactly known, but based on current membership of the British Society for Paediatric Palliative Medicine are probably around 40 or 50.

4.1.2 Education

There is no recognised postgraduate clinical training for nurses in paediatric palliative care. There are, however, academic MSc courses at Oxford Brookes, King's College and Newcastle among others. Most of these are open to doctors but in practice do not meet the needs of those wishing to specialise in paediatric palliative medicine.

Postgraduate training for paediatricians is available in two centres: Great Ormond Street London, and Cardiff. Both are 12-month SpR posts for paediatric trainees who have completed their core paediatric training. There is post in Children's Hospice medicine at Helen house Children's Hospice designed for GP trainees, which could also be accessed by paediatric trainees at the discretion of their Deanery.

There is a paediatric option for the distance-learning Diploma in Palliative Medicine based in Cardiff.

4.1.3 Research

Both paediatrics and palliative medicine rely heavily on the use of therapeutic approaches that may have little evidence base. The use of medications for unlicensed indications illustrates this and is common in both (16-18). It is doubly difficult to find a good evidence base on which to build good palliative medicine in children, yet a rational and therefore compassionate approach requires that one should be found. There are very few academic centres of research excellence in palliative care in children, and only one consultant senior lecturer.

The need for education and research emphasises the importance of developing academic support for PPC and indicates **a need for academic departments that combine both nursing and medical academics (1-3)**.

4.1.4 Children's Hospice

Children's hospices represent an important resource for children with LLC (2). Between 25% and 50% of children referred for specialist palliative medicine (fig 1) have also used a children's hospice (2, 5). Children's hospices can provide high-quality specialist respite in a comfortable 'home from home' environment, and are staffed in such a way as to allow them to address many of the psychosocial and spiritual needs of the child and family that are difficult to approach in more acute hospital settings. They therefore provide much-needed generic and semi-specialist palliative care. With appropriate specialist medical and nursing input, Children's Hospices can also potentially provide specialist palliative medicine for cases where families choose not to remain at home during the palliative phase.

Properly supported, hospices could potentially provide a wide range of other

services such as child bereavement services and complementary therapies. Links with hospital and community services could be developed further, particularly with hospices that offer a community nursing service. Allowing nurses on a community paediatric nursing team to rotate through children's hospices would increase flexibility and continuity as nurses could be deployed there according to the child and family's need. The experience of staff at children's hospices is a resource for developing palliative care education.

The hospices themselves are a resource which should be valued and developed through political and professional dialogue. Robust and fair contractual arrangements should take account of regional needs. Simplified access to continuing care packages that include hospice care would improve utilisation.

4.2 Practical challenges to developing good palliative care in children

4.2.1 Commissioning

The impact of suboptimal care for a dying child is very significant, most obviously on the family but also on professionals and society as a whole. Most individual PCTs will, however, encounter relatively few children with life-limiting conditions and there is a risk that they will accord paediatric palliative care an inappropriately low priority for commissioning. Like many tertiary specialities in children, paediatric palliative care services may need to be commissioned on a different basis from their adult equivalents (1-3).

4.2.2 Continuing care

Current mechanisms for accessing continuing care funds are unwieldy and can result in a delay in providing services (1-3). The main cause for this is the lack of a tripartite funding agreement between Social, Education and Health services.

All children with LLC need all of medical, educational and social support (19).

Much time and effort are wasted drawing arbitrary distinctions between them. While the exact proportions are of course different for each child, they are to a large extent predictable on the basis of the diagnosis. Incomprehensibly, the presence of a medical condition is sometimes taken to imply less of a need for educational or social service funding. Furthermore, there are particular difficulties getting funding across administrative boundaries, especially where the three are not coterminous.

One solution is a nationally centralised Tripartite Fund for children with LLC, to which all three budgets would contribute on the basis of average need in the region. Access should be automatic at diagnosis of a LLC, and be based largely on the anticipated needs of a child with that condition.

4.2.3 Record-keeping

The nature of paediatric palliative care is that it takes place in a number of different clinical environments. The same child may need the same sort of care to be delivered at home, school, hospital and hospice within the same week. CNS and consultants in paediatric palliative care, other CNS and consultants, GPs, and therapists will all need to add to the notes. It is essential that a robust system of record-keeping be developed that allows the records to be available to all professionals who are involved in the care of the child. The best way to achieve this is through patient-held records, paper or electronic. In order to minimise the risks as part of Clinical Governance, these must be subjected both to clinical review as necessary by the responsible consultant and to regular audit.

4.2.4 Access

In practice, access of children to palliative care services is also potentially limited by a number of other factors including:

Culture

Palliative care services for children, particularly children's hospices, are often poorly accessed by those from ethnic minorities (unpublished data), who because of inherited conditions are over-represented especially in RCPCH group III. Culturally-appropriate support for families with children with LLC should be developed.

Geography

Children in rural areas in particular may be difficult to access by specialist nurses and/or doctors. A flexible system for supporting local primary care teams is therefore particularly important (fig 2). The importance of geography in influencing the likelihood of an appropriate referral (fig 3) has been shown in Wales (5).

4.2.5 Clinical governance - the need for specialist support and training

Nurses caring for dying children can acquire considerable experience and many become *de facto* prescribers through their advice to medical colleagues. This raises important issues regarding clinical governance. It is likely that NHS trusts would be criticised for allowing nurses to remain in this vulnerable position without providing them with appropriate specialist paediatric palliative care training and support. Furthermore, there is little formal provision for clinical supervision. To address these clinical governance issues, it will be necessary to develop adequate structures for training and supervision, as well as patient care pathways and guidelines. These will need to originate from specialist nurses and paediatricians in paediatric palliative care.

4.2.6 Lack of facilities for adolescents

It is becoming clear that there is a dearth not only of respite facilities for those over eighteen, but also of services for adolescents and young adults with more specialised palliative care needs. This has been the subject of a recent detailed survey by ACT (4). Its recommendations are summarised in table 7.

4.3 Summary

Children with life-limiting conditions, malignant and otherwise, have needs that are in common, and mark them out as a group distinct from other children. It is our experience that this is not yet widely recognised, and that boundaries too often cut across their care. The result can be provision that is poor and patchy or simply delayed by division and duplication.

Paediatric palliative care is expanding in the United Kingdom. There is considerable semi-specialist skill among doctors and nurses, and enthusiasm among a wide range of other professionals who have a part to play in developing a seamless service that is flexible enough for the needs of individual children and families. At the same time, interested doctors and nurses are increasingly taking forward more specialist skills. Such development could be threatened by a weak infrastructure in generic medical and nursing skills, particularly by poor community paediatric nursing provision.

Respite provision by children's hospices is of extremely high quality but can cater for only relatively small numbers. Children's hospices are rarely the sole providers of palliative care to a family. There is a need to consolidate the position of children's hospices in the wider framework of care to children with LLC, through professional contact and fair funding arrangements, and to expand statutory home and inpatient respite provision.

5. Recommendations

5.1 Urgent needs

- Establishment of commissioning arrangements for a palliative care network for children.

- Establishment of multidisciplinary teams in paediatric palliative care as per ACT/RCPCH model (table 8).
- Expansion of consultant-led tertiary services as per ACT/RCPCH model
- Establishment of multi-professional academic departments of paediatric palliative care in order to:
    - Expand teaching opportunities for doctors and nurses in particular
    - Expand research and evidence base for palliative care in children
- Expansion of SpR training opportunities:
    - More numbers
    - Broader training to include Children's Hospice work as well as specialist palliative medicine in children.
- Expansion of nursing training opportunities:
    - To combine paediatrics and palliative medicine
    - Beyond oncology ('non-cancer oncology outreach nurses')
- Establishment of community paediatric nursing teams (including further Diana Teams) where they do not exist currently
- Robust and fair funding arrangements with children's hospices, including appropriate sessional commitments from professionals employed in statutory sector such as specialist physios, speech and language therapists and consultants.
- Formal provision of clinical supervision and psychology services for those working with dying children.

5.2 Long term goals

- Development of adolescent and young people's palliative care services.
- Record-keeping and audit system that crosses home/hospital/school/hospice boundaries.
- National register of children needing palliative care
- More inpatient respite provision
- Development of primary care paediatrics
- Review of continuing care funding arrangements

- Education resources for professionals allied to medicine, educationalists and other carers
- Development of child-specific chaplaincy
- Expansion of child psychology and psychiatry service
- Unification community and hospital paediatric service.

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7. Tables

***Group 1*:** Life-threatening conditions for which curative treatment may be feasible but can fail. Palliative care may be necessary during periods of prognostic uncertainty and when treatment fails (e.g. cancer, cardiac anomalies).	***Group 2:*** Conditions in which there may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal childhood activities, but premature death is still possible (e.g. cystic fibrosis, muscular dystrophy).
Group 3: Progressive conditions without curative treatment options, in which treatment is exclusively palliative and may commonly extend over many years (Batten's disease, mucopolysaccharidosis).	***Group 4:*** Conditions with severe neurological disability which may cause weakness and susceptibility to health complications, and may deteriorate unpredictably, but are not considered progressive (e.g. severe cerebral palsy).

Table 1: Conditions that may need paediatric palliative care (1, 3).

Environment	**Royal College Groups**	**Main Primary Carers** (not exhaustive)	**Palliative care offered**
Home	All	Parents District nurses CNS (groups 1 & 2) GPs SW	Respite Generic palliative
School (Special and mainstream)	1, 2, 3, some 4	Teachers LSAs Nurses (SNs, CCNs) NNEB Community therapists Community paediatricians (groups 2, 3 & 4) SW	Respite
General inpatient paediatric unit **Specialist inpatient paediatric unit**	Mainly 1 & 2 3 & 4 occasional visits	General/subspecialty paediatricians Ward paediatric nurses Hospital therapists CNS Clinical psychologist SW	Generic palliative Semi-specialist palliative (especially symptom control)
Children's Hospice	2 & 4 Occasional 1 & 2	Carers Children's nurses GP Often SW	Respite Generic palliative

Table 2: Clinical environments in which children needing palliative care may be found (CNS – clinical nurse specialist, SN – school nurse, LSA – learning support assistant, NNEB – nursery nurse, SW – social worker).

- Normalisation of life as far as possible, e.g. continued access to play and education, and contact with friends and peers
- Information and advice about the condition and its treatment
- A 24-hour helpline
- Benefits advice
- Practical help in the home
- Help with transport
- Psychological support
- Support for siblings
- Pre and post-bereavement advice
- Continuity of staff, at least one of them, i.e. the key worker
- Education and/or special education
- Social work input
- Play, art, music etc. therapies
- Record keeping mechanisms and facilities
- Administrative, clerical and information and communication technology support
- Provision of aids, equipment, housing modifications etc.
- Complementary therapies

Table 3: Main needs of children with LLC summarised from pilot projects in England (13).

Paediatric palliative care – the sum total of respite and all forms of palliative expertise in children.

Symptom management – the preferred use of this term is in a holistic sense to include attention to physical, psychosocial and spiritual aspects of symptoms. In many documents, the term is often used in a more restricted fashion to mean only control of the physical aspects of symptoms. To distinguish between the two this document uses the term 'holistic symptom management' for the former and 'symptom control' for the latter. Thus symptom control is a part of holistic symptom management, as are psychological and bereavement support.

Respite care – care whose main function is to relieve the family of the burden of care by providing support in the home or an alternative 'home-like' environment such as a children's hospice. Respite care will often incidentally address some aspects of holistic symptom management.

Generic palliative care – palliative care skills that might be expected to result from medical or nursing training in paediatrics or child health.

Semi-specialist paediatric palliative care – skills in holistic symptom management that may be expected from any specialty paediatrician or paediatric clinical nurse specialist with expertise in one particular condition or a narrow range of conditions. Examples include paediatric oncologists, CNS in cystic fibrosis. Adult physicians with palliative medicine training would be a special category within this.

Specialist paediatric palliative care – skills in holistic symptom management that may be expected from paediatricians with specialist training in palliative medicine, or CNS in paediatrics and palliative care.

Table 4: Glossary of terms used to describe some of the skills needed in paediatric palliative care.

Recommendations by RCPCH/ACT	**Current Provision (0 – no provision**

	12 – full provision)
1. The needs of children with life-limiting conditions have been thoroughly assessed in a number of recent studies. Commissioners should use this research which is unlikely to need further validation for local applicability (section 5 introduction).	No unnecessary research being undertaken. (6/12)
2. Children with life-limiting conditions should be recognised as a discrete group and commissioners should work with NHS trusts to set up a robust local database of these children (sections 4.3.1,8.1.1).	No specific database, but overlap with Special Needs Registers being developed in different regions (5/12)
3. A flexible children's palliative care service, recognising each family's individual needs, should be provided in each district. It should be available over a long term as well as when death approaches (section 8.3.4).	Flexibility very limited: Inconsistent community paediatric nursing team cover 'Palliative care' not well defined Palliative care team often poorly defined. (6/12)
4. Every district should have a senior paediatric professional as coordinator of children's palliative care. The need for coordination of the network of services should be included in purchasing specifications (section 8.3.1 - 8.3.6).	Coordinator for continuing care but not palliative care. Multidisciplinary specialist paediatric palliative care 'teams' not yet in a position to take on this role. Specialist nurse(s) in paediatric palliative care needed. (6/12)
5. Community children's nursing teams are essential for the management of children with palliative care needs. Commissioners should facilitate their establishment and/or development to address the needs of this caseload (section 9.3.1 iii).	Established CCNS in Gwent and Bro Taf., under development in Iechyd Morgannwg. (8/12)
6. Occupational therapy and physiotherapy are a crucial part of children's palliative care and children should have access to them in the community (section 9.3.1 iv).	Inconsistent provision in the community. Better for children in III and IV as often coordinated through special school. Play specialist available to oncology children at Llandough. (5/12)
7. In addition to respite provided by social services and social work departments, respite with medical and nursing input should be available locally or within a short distance. Commissioners should ensure a choice of such health-based respite by purchasing or commissioning a variety of services both within and outside the district's boundaries (sections 9.8, 9.8.3).	Very little residential respite care. Monday to Friday, Ty Hafan and Ty Gobaith/Hope House provide inpatient 'specialist respite' (6/12)
8. Each child and family should have a care plan, drawing together the provision of all	Most patients have a care plan, but often fragmented in absence of coordinated palliative

components of care; where appropriate, voluntary agencies should be recognised as integral to the care plan. (section 9.2.1).	care team. (5/12)
9. Each family should have its own named keyworker who is responsible for the coordination of the care plan, ensuring that total care, not just healthcare, is available (sections 8.4.1,9).	Inconsistent. Many specialist services (especially neonates, oncology, respiratory and renal paediatrics) have Clinical Nurses Specialists who are experienced, but not usually formally trained, in palliative care (4/12)
10. Continuity of care is vital. Speedy communication and clear documentation of management plans and treatment sheets are essential; parents should hold the record in case emergency care is required (section 8.4.2).	Inconsistent. Usually poor. Patient-held records not widely used, but are being developed. (5/12)
11. NHS trusts, purchasers and general practitioners need to develop a clear mechanism for the accessibility and funding of medications, disposable medical and nursing supplies and medical equipment (section 9.3.3iii).	Continuing care guidelines clear but access often too slow for urgent palliative care needs. (4/12)
12. Families should not be caught up in a financial and bureaucratic trap caused by disagreement between health, education and social services/social work departments over the provision and funding of aids, respite care and housing adaptations. The agencies must not delay in reaching urgent agreement over funding and timely provision (section 9.3.4).	Still an obvious problem in most areas. Local implementation of flexibilities under the Health Act 1999 (e.g. pooling of resources) likely to improve current situation eventually. (2/12)
13. Services should be developed to meet the needs of older teenagers and young adults requiring palliative care. Partnerships between children's and adult services should be developed to help with the transition (section 9.8.5).	Few teenagers access adult palliative care services. Access to specialist paediatric services inconsistent (NB many teenagers still seen by adult physicians e.g. haematology) (1/12)
14. A tertiary network of children's palliative care specialists should be developed as a resource for local professionals and a basis for advice, training and research (section 10.2.2v).	Single tertiary specialist in post. His academic and clinical roles cover all of Wales, in direct clinical or advisory capacity. (6/12)
15. Children's palliative care courses for nurses need to be developed. Training should be provided for all other professionals in the field (sections 10.2.3, 10.2.4).	Medical diploma available. Nursing diploma under development (4/12)
16. All staff working closely with families should have formal psychological support and supervision written into their job descriptions	Psychologists generally supportive, but constraints on staff numbers mean arrangements are informal and ad hoc (4/12)

(section 10.2).	

Table 5: Recommendations of ACT/RCPCH guidelines (3) for Paediatric Palliative Care Services, and how, in the view of a multidisciplinary group, services matched up to them in November 2002. Scores are out of 12, where 12=fully met and 0 =not met at all. Although this study was done in Wales, it is likely that the results are illustrative of the situation in the United Kingdom as a whole.

It can be seen that the most pressing concern was services for teenagers, adolescents and young adults.

	Reported by paediatrician to have palliative care needs, but not referred for specialist PPM.	Referred to specialist paediatric palliative medicine service	Total
Group I	4	35	29
Group II	3	15	18
Group III	10	5	15
Group IV	16	13	29

Table 6: Diagnosis of patients reported by paediatricians in the WPSU study or referred to specialist PPM (5). Patients in group 1 (mainly cancer) are an important group, but more children did not have cancer. Children with non-malignant conditions were less likely to be referred for specialist palliative medicine.

- Recognition as a distinct group
- Involvement in decision-making
- Multidisciplinary, multi-agency services
- Link and key workers
- Joint Health/Social Service planning
- Psychological, spiritual services
- Transition planning from children's services
- Specific training in
    - palliative care
    - management of young people

Table 7: recommendations made by Joint Working Group of ACT and National Council for Hospice and Specialist Palliative Care Services for palliative care among adolescents (4).

- Senior lecturer/consultant in paediatric palliative care and SpR hold the ring by advising, supporting and training locally, regionally and centrally.
- Community or other sub-specialised paediatrician with special interest in paediatric palliative care, and admission rights
- Clinical nurse specialist in paediatric palliative care (when available)
- Community children's nurse(s) with wide networks with other nurses working with children in community, hospitals and hospice
- Clinical nurse specialists, eg. oncology, respiratory, neonatal care
- Clinical psychologist
- Chaplaincy
- Therapists
- Social worker
- Others, such as NNEBs, healthcare and care assistants, etc. according to need
- Administrative, clerical and ICT support staff

Table 8: Potential members of a multidisciplinary team in paediatric palliative care.

8. Figures

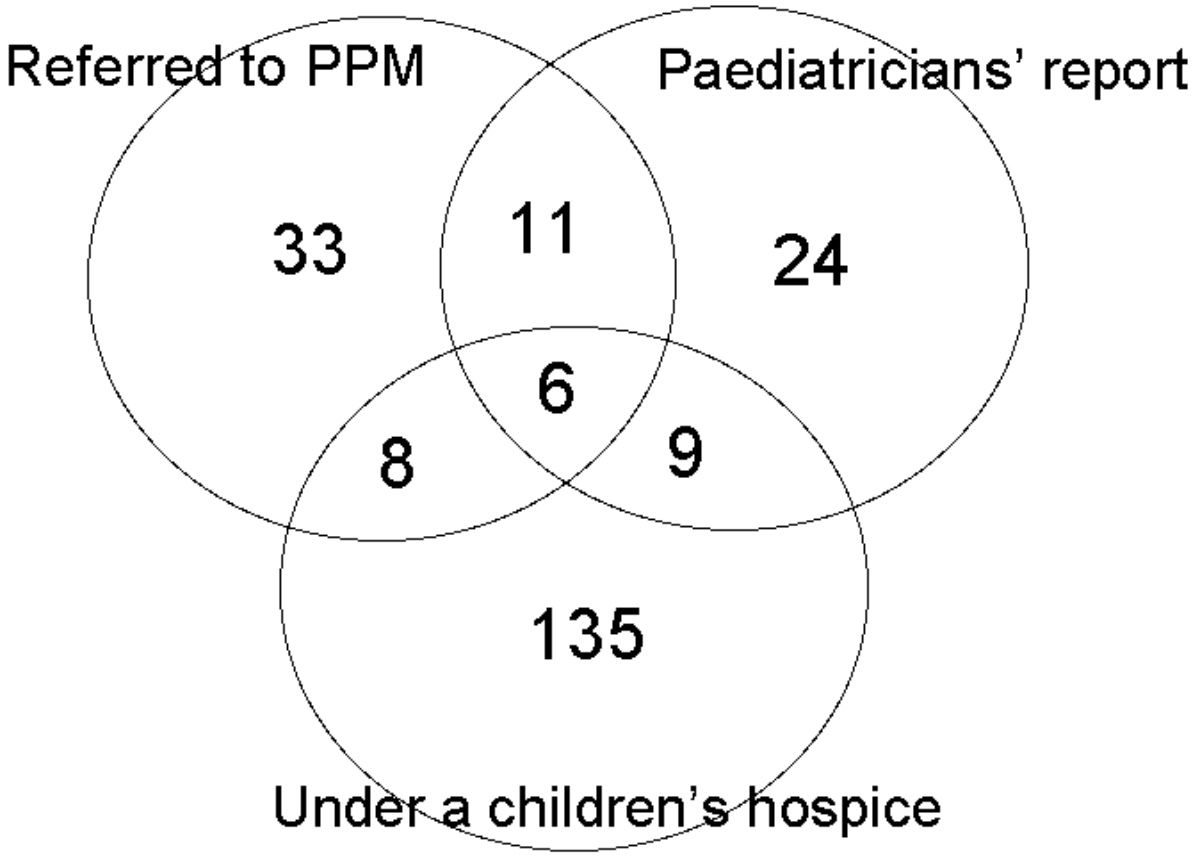

Fig 1: Children needing palliative care. It can be seen that although the skills of general paediatrician, children's hospice and specialist palliative medicine paediatrician (PPM) overlap, they are far from the same.

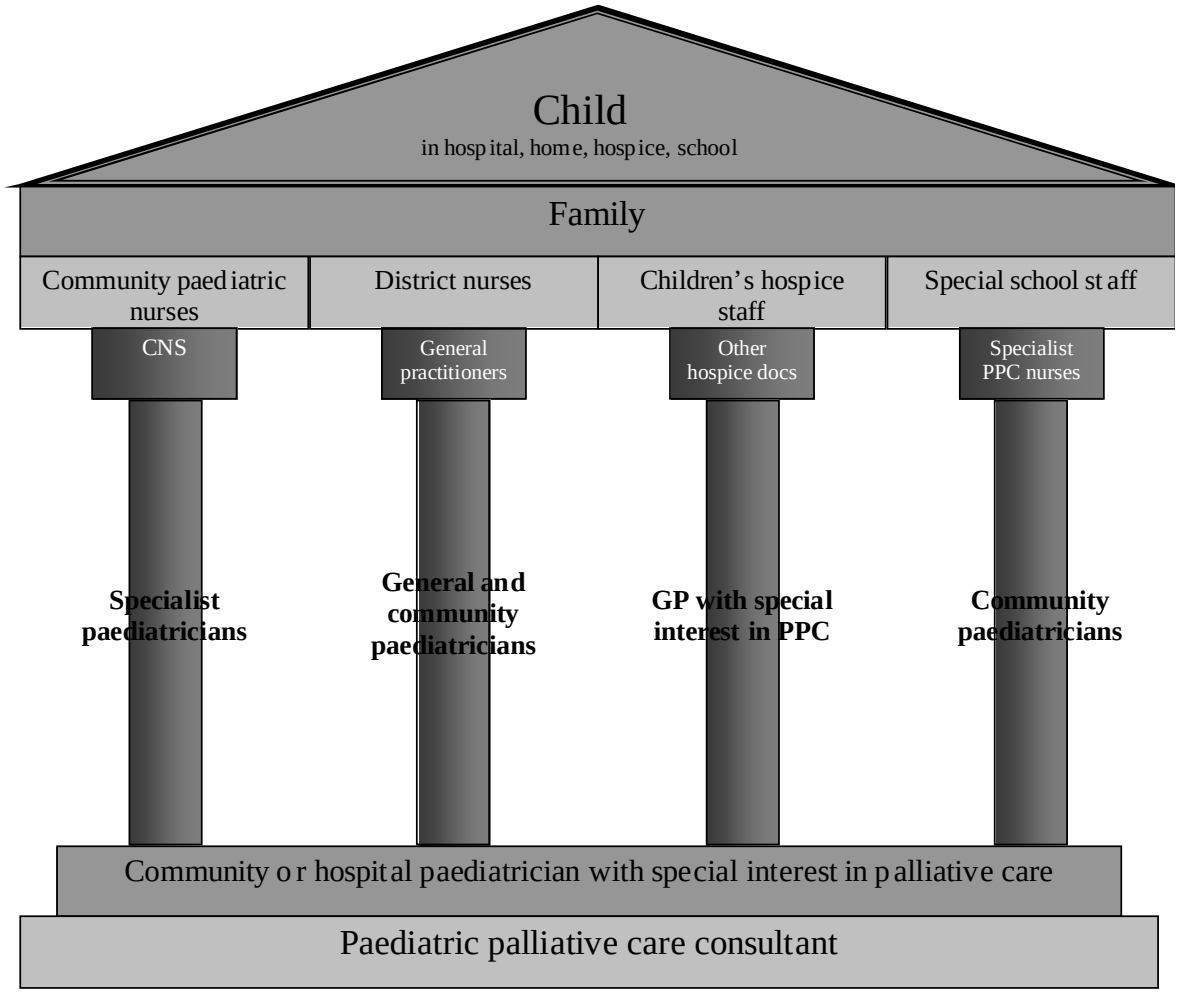

Fig 2: Supporting the supporters. The challenge for specialist paediatric palliative care is to provide support to many different professionals in several different clinical environments. Current interprofessional, interdisciplinary and internecine boundaries mean that families are typically cared for by a number of teams working independently. While this model works well for some children, for others the specialist can optimise the delivery of palliative care in part by providing common ground on which to base these vertical supports.

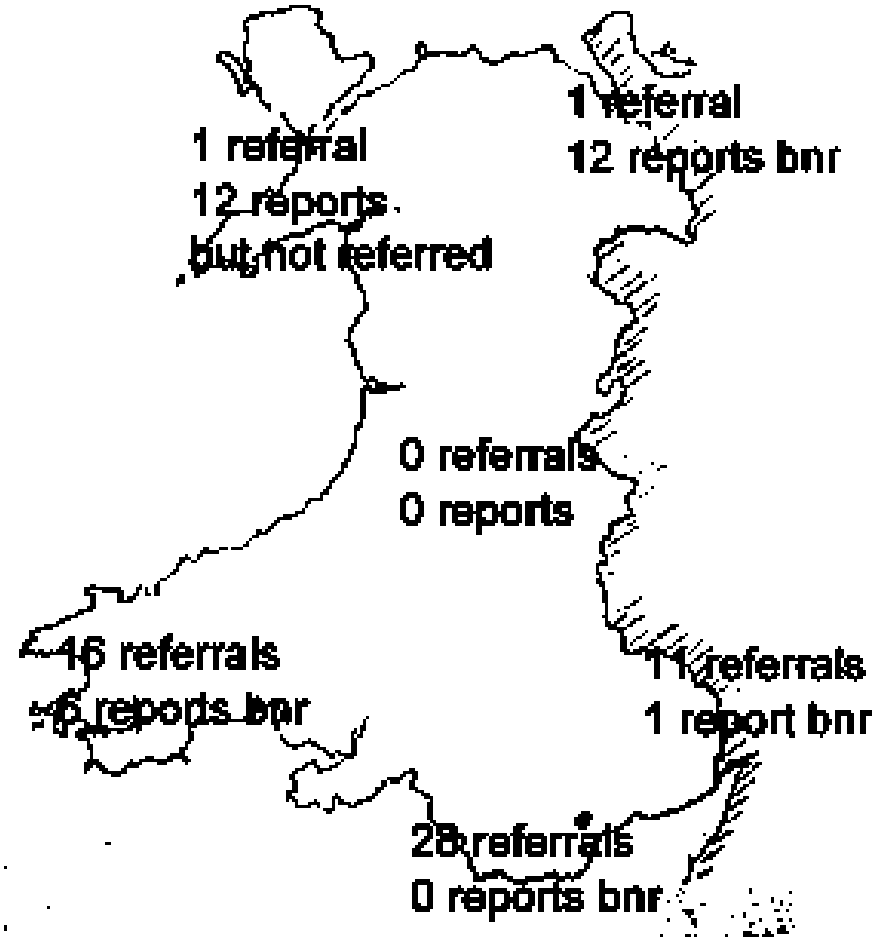

Fig 3: Relationship of referrals to geographical location in Wales. The ratio of children reported by paediatricians to have PPC needs but not referred (bnr) to those actually referred for specialist PPM is related to distance from Cardiff, (•) where specialist services are based. Children with palliative care needs who are at some distance from Cardiff are less likely to be referred even when their needs are recognised. This supports the ACT/RCPCH recommendation (1, 3) for a national network of tertiary specialists.

Appendix 2

Draft 3

Analysis of the Potential Economic Impact of the Guidance: 'Improving Outcomes in Children and Young People with Cancer'

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Acknowledgements

The authors wish to thank all those colleagues who have given us their valuable time support and information including

- All members of the guidance development group
- NCC-C staff.
- Staff from the NHS, particularly finance managers, MDT co-ordinators, clinicians and nurses,
- Staff from Royal Colleges and
- All those people we contacted in connection with assessing financial support for the NHS from charitable sources

We also had very helpful discussions with Sue Ward at The School of Health and Related Research (ScHARR) at the University of Sheffield, an author of the economic implications of previous service provision guidance.

Executive summary

A detailed costing exercise was conducted in order to estimate the cost implications of implementing the key recommendations of the "Guidance on Cancer Services: Improving Outcomes in Children and Young People with Cancer" in England and Wales. The potential costs for inclusion was vast, the economic analysis focuses on those aspects of the key recommendations which are likely to have significant costs implications. It is acknowledged that there is an element of uncertainty around the estimates presented and that there will be variation between cancer networks. Further assessments will be needed at cancer network level and/or NHS Trust level to determine exact cost implications.

The major impacts on costs fall into the following broad areas, Table 1.

Table 1: Cost Summary (All costs in £million per year)

	Mid Point	**Range** (± 25%)	
Additional staffing at principal treatment centres			
Low scenario*	**6.4**	4.8	8.0
Moderate scenario**	**14.5**	10.9	18.2
Multi-disciplinary teams			
Low scenario	**2.0**	1.6	2.6
Moderate scenario	**3.0**	2.3	3.9
Training and Education			
CPD for additional staff	**0.2**	0.1	0.3
Key worker			
	3.3	2.5	4.1
Place of Care			
10 additional units for young people	**7.5**	5.6	9.3
Total Low Scenario*	**19.4**	**14.6**	**24.2**
Total Moderate Scenario**	**28.5**	**21.4**	**35.7**

* Low scenario – considers the cost of employing at least one whole time equivalent (WTE) key professional at each principal treatment centre: and weekly MDTs meeting of 1 hour duration

** Moderate scenario – considers the cost of employing at least 2 WTE key professionals at each principal treatment centre: and weekly MDTs meeting of 1 hour duration

Staffing at principal treatment centres

Two scenarios have been used to estimate the costs required for additional staffing at the 17 principal treatment centres and 8 Teenage Cancer Trust (TCT) units in England and Wales. The mid point estimate for the low scenario is £6.4 million with a range of between £4.8million and £8 million. This would ensure a minimum staffing level of at least 1 whole time equivalent (WTE) of each key professional at each centre. The moderate scenario mid point estimate is £14.5 million with a range of between £10.9 million and £18.2 million. This would enable each centre to have at least 2 WTEs. The key professionals include paediatric clinical oncologists, clinical psychologists, paediatric oncology outreach nurses (POONS) and play specialists among others.

Multidisciplinary Teams (MDT)

It has been estimated that the annual costs in England and Wales for MDTs as outlined in the guidance would be between £1.6 million and £2.6 million for 1 hour meetings once a week or between £2.3 million and £3.9 million for 2 hourly meetings, again weekly. The costs will vary across cancer networks in line with existing MDT configurations and staffing levels.

Training and educational needs

The estimated costs associated with the training and education of the additional staff detailed above would be between £146,000 and £257,500 in England and Wales. This calculation assumes a basic professional skill level as a baseline.

Key worker

The cost of employing new staff as designated key workers in England and Wales, based on a nursing salary plus on-costs, would be in the order of £3.3 million, with a range of £2.5 million and £4.1 million. This calculation is based on one key worker per 30 patients at principal treatment centres.

Place of care

The estimated cost of planning, building and equipping 10 additional units dedicated for young people with accommodation for both in-patients and day cases would require expenditure of between £5.6 million and £9.3 million.

Staffing each additional unit would entail recurring annual costs of between £0.6 million and £0.75 million per TCT unit, based on information from 2 NHS Trust managers.

Financial support for the NHS from charitable sources

Members of the GDG considered it important to assess the degree of support that charities gave to NHS services for children and young people with cancer. Therefore, although this is not an economic implication of the guidance it has been included here.

The financial contribution to services for children and young people with cancer from charitable sources is estimated to be between £35 million and £62million per annum. This comprises estimations that between £19.3 million and £32million is raised from charitable sources annually to specifically support children and adolescents with cancer, a further £13.5million to £26.5 million is contributed to children and young people as a result of fund raising by cancer charities focusing on people with cancer; a further £2.2 million to £3.6million supports hospice services. It is often the case that staff working in specialist centres and in the community are funded by charities, such as Children's Leukaemia in Childhood (CLIC) or paediatric oncology outreach nurses (POONs) often funded by Macmillan Cancer Relief. This charitable source of funding forms an essential aspect of the service provision for children and young people with cancer for both staffing and equipment.

1. Introduction

Guidance has been developed to improve the service provision for children and young people with cancer. This section serves to inform commissioners and Trusts of the resource and cost implications of implementing the recommendations of this guidance. The Centre for the Economics of Health (CEI) at the University of Wales, Bangor has been commissioned to support this process by analysing the potential cost implications.

1.1 Scope

In line with previous NICE service provision guidance the objective of this economic analysis is to:

- Identify different possible models of implementation, which will vary depending both on the baseline position and on the chosen means of achieving the targets set out in the guidance;
- Identify the key economic issues and cost drivers of guidance implementation;
- Estimate the costs of implementing the guidance according to the different models identified, and in so doing provide a structure and methodology that Trusts may use to do their own analysis;
- Estimate the national cost implications of adopting the cancer guidance;
- Estimate the national current contribution made by charitable organisations for the support and provision of services for children and young people who have had a diagnosis of cancer

The analysis does not aim to:

- Provide a definitive answer to the cost implications of the guidance for specific UK Children's Cancer Study Group (UKCCSG) cancer centres or tcts (but to produce an indication of the scale of costs involved for different paradigms);
- Analyse the health outcome measures of meeting the guidance;
- Estimate the cost-effectiveness, or otherwise, of implementing the guidance recommendations.

2. Process and Methods

2.1 Integration of economic review with the cancer guidance

The research into the cost implications of the guidance was developed in parallel to the production of the Improving Outcomes Guidance for Children and Young People with Cancer. Members of CEI attended all the Guidance Development Group (GDG) meetings, facilitating a full understanding of the guidance as it developed.

2.2 Literature and data searching

Literature searches were carried out by both the NCC-C researchers, information specialists and by the Health Economics team at Bangor. Searches were conducted in order to identify any existing costing exercises, audits of cancer activity, cost of illness studies or models of treatment pathways. All literature was screened for economic content. Any emerging economic literature was referred to the economics group for appraisal.

In addition to the specific research questions raised by the GDG, searches were conducted of the economic literature relating to both

- General issues of children and young people with cancer, and
- Specific issues relating to the key recommendations of the guidance.

The databases searched were MEDLINE, Cinahl, NHS EED, HTA and DARE. No filters were used to restrict the searches, however limitations to the searches included

- Studies in English
- Publicly funded health services, i.e. similar systems to NHS
- Publications after 1990

Unpublished data was obtained as a result of direct contact with members of the GDG, other expert clinicians and finance directors from cancer centres and Trusts. The finance directors of each UKCCSG and TCT were invited to participate in a survey requesting current levels of investment in services for children and young people with cancer, reported in Section 3.

2.3 Costs

Procedural cost data was obtained using Healthcare Resource Group (HRG) costs from Payment by Results[1]. HRG costs are produced by every Trust in the country, using a very detailed bottom up method which costs all elements of patients' care including: theatre time, laboratory tests, pathology tests, minutes of nursing time, minutes of consultant time, physiotherapy, x-rays, ultrasound, pharmacy and overheads (administration, heating etc.)[1]. Data are available for inpatient elective and non-elective cases, as well as day cases. Where HRG costs were not available, financial managers at NHS Trusts were consulted.

Staff salary pay-scales, obtained from the Department of Health (DH) website[2] were used, wherever possible to calculate current salaries. Further advice on calculating staffing costs was provided by the payroll managers of 3 NHS Trusts.

For each professional grade, a mid-point scale was chosen, upon which 20% employment on-costs plus a London weighting were added as appropriate. In some instances, salaries were neither available from standard sources[3] nor from the DH. Often these posts were funded by charities. In the case of play specialists, for instance, many are currently paid on nursing grades B or C. Calculations for additional staff costs for play specialists are based on the National Association of Hospital Play Specialist recommendations[4]. Salaries for the social workers and teachers were based on Local Authority rates, again using salary scale mid points.

Where calculations are based on hourly rates, salary and on-costs, leave and sickness were taken into account by assuming a 42 week year[3].

The impact of the Working Time Directive is not clear at this time, and has not been taken into account. However it will need to be considered by commissioners, as will the Agenda for Change and similar government initiatives[5].

Very little recent costing data was found in the literature for the UK. There are some non-UK studies of costs, but treatment patterns and cost structures are likely to be quite different so have not always been included.

2.4 Discussions with clinicians and other key professionals

Advice from clinicians on the GDG was sought to ensure that appropriate assumptions were made for future activity, to identify data sources and to assist in the interpretation of data. A number of clinicians and finance managers from individual TCTs and UKCCSGs were contacted to discuss activity and resource implications of future staffing levels and various aspects of the guidance. Several specialist nurses and co-ordinators were also contacted to discuss their roles in multi-disciplinary teams (MDTs) and in patient-centred care. Information and advice was sought from the DH and Royal Colleges concerning current workforces. Data collected as part of the NPHS needs assessment was used to estimate future staffing requirements[7].

2.5 Identification of key cost issues

CEI used the guidance, GDG discussions and a formal survey of GDG members, preliminary data analysis and consultations with both clinicians and finance managers to identify and prioritise the key cost issues. A proforma was produced to collate information on the extent to which key economic issues had been dealt with in previous guidance, and the extent to which literature was available for key questions relating to this guidance. Current annual budget estimates for both UKCCSGs and TCTs were sought through a questionnaire administered to finance managers as part of the NPHS needs assessment, copy in appendix A.

2.6 Cost analysis

For each of the key issues identified, an estimate of the national and local cost consequences has been made wherever possible. The approach adopted for each issue is detailed in the relevant section.

The costs for each cancer network will vary dependent upon population base, health service facilities, staffing levels and local patient flows. Estimates are based on broad working assumptions concerning future staffing configurations.

Commissioners and Trusts will need to make further considerations of staffing levels based on their local situation..

2.7 Sensitivity analysis

For each cost we chose a range of ± 25 % to reflect uncertainty in the estimate. For consistency, it seemed important to use the same method to consider uncertainty throughout the document rather than a variety of different solutions relevant to each section. There will be uncertainty in our estimates, for example, in existing configurations or frequency of MDT meetings, current and future staffing levels and costs relating to any additional facilities. In addition there will be cost savings as a result of the guidance that are not possible to quantify at this time.

3. Costs associated with current service provision for Children and Young People with Cancer in England and Wales

In order to set the context of this report we start by giving estimates of the current levels of annual investment in children and young people with cancer services in England and Wales. The data was collected in co-operation with the needs assessment conducted for the GDG as reported earlier in the guidance[7]. A short financial questionnaire was sent to finance directors of all United Kingdom Childhood Cancer Study Group (UKCCSG) centres and Teenage Cancer Trust (TCT) units as part of the initial needs assessment exercise. Finance directors questionnaire included in Appendix A.

Finance directors were asked to provide cost information in respect of the level of investment in child and/or adolescent cancer for the financial year 2002/3 focusing on staffing, drugs, radiotherapy and other NHS costs; a question relating to the contribution from non-NHS sources was also included. The reported costs are therefore from a NHS and charity perspective. Costs falling on wider society and in particular the families of children and young people with cancer are beyond the scope of this report. More information regarding the data is detailed in Appendix A.

Unfortunately the response rate was disappointingly low, with only 5 responses from 17 UKCCSGs and 7 TCTs. From the data collected we estimated that the current level of NHS investment in cancer services for children and young people under 25 treated at either UKCCSG centres or TCTs in England and Wales was between £45 and £74million per annum. Broadly the range of costs per bed per annum was between £160,000-270,000; however there are a range of services available at the different centres.

Using cost data supplied by finance directors, we estimated that a further £1 to £3 million per centre per annum would be required for medication and other NHS services, and £0.5million for radiotherapy Table 7. This would increase the estimated expenditure range to between £53 and £80 million per annum.

An additional budget of between £1 and £2million from charitable contributions was managed by NHS financial directors. This charitable funding was spent in a range of ways including staff, (both nursing and social workers) and equipment. The current level of annual investment by TCTs for young people with cancer was estimated to be £0.7 million per annum which was likely to increase to £1.8 million per annum over the next ten years[6]. This is expanded upon in section 7.

3.1 Employment Cost at UKCCSG centres and TCT units in England and Wales based on Needs Assessment for children and young people with Cancer[7]

In view of the paucity of information from the questionnaire to finance officers, further calculations based on the information provided by a needs assessment exercise on actual staffing levels were made. The costs of employing staff members directly involved in the care and treatment of children and young people with cancer has been calculated based on the needs assessment exercise analysis[7]. Griffiths et al[7] acknowledge that there was a variation in the way respondents completed the questionnaire. In order to reduce this variation, the calculations are primarily based on the information provided by 16 of the 18 responding UKCCSG and TCT centres in England and Wales.

3.2 Methods

Current staffing levels, activities and grades, obtained from the needs assessment exercise, together with staff salary pay-scales, obtained from the Department of Health (DH) website[2] were used to calculate the current staffing cost of UKCCSG and TCT centres. See Section 2 for further information.

In cases where the employee's grade was not stated by the responding UKCCSG/TCTs, the mid point of the most commonly reported grades were used to calculate the cost.

Two centres that provided data appeared as outliers and were excluded from the costing analysis for the following reasons:-

- Centre No. 4 because it appeared that many additional shared staff were included , and
- Centre No.18 had missing data and was the only TCT centre to be included

separately from a neighbouring UKCCSG

3.3 Sensitivity analysis

The cost scenarios presented reflect the staff costs for providing 2 scenarios for each principal treatment centre :

- Low cost scenario - the costs of increasing current staffing levels to a minimum of 1 WTE for each key professional group as described in the needs assessment
- Moderate cost scenario - the costs of increasing current staffing levels to a minimum of 2 WTE of each key professional group

3.3 Cost of staffing at UKCCSG/TCT Centres

The costs related to the staff employed at the UKCCSG/TCT Centres in England and Wales are shown in full for all the 18 centres that provided data, Table 3.1. However the 2 outliers have been excluded from further analysis. Tables 3.2-3.4 breakdown the total staff costs shown in Table 3.1.

The number of new patients treated at the 16 centres was 1658, (range 60-172) using the 228 available beds. The employment costs of the medical, nursing and other staff caring for those patients was £43.6 million, (Table 3.5), with a mean staffing expenditure at each centre of £2.7million. This does not include ancillary, catering, administration and other staff. The mean cost per bed was £200,000 (range £120,000-260,000). This does not include treatment given at non-UKCCSG centres.

Extrapolating from this data, we estimate that the cost of employing the staff involved in the care of children and young people with cancer to be approximately £50 million per annum in the 17 Centres and 8 TCT units in England and Wales. Applying the ± 25% margin, the range of costs is estimated to between £42.5 and £62.5 million.. This would allow for uncertainties such as, staff who may not be dedicated to cancer patients alone.

It is of note that economies of scale may operate, as evidenced by the following:-

- Centres that appear not to offer specialist services have the fewest beds

(centres 1 and 16).

- One centre which has low annual patient activity has a cost per bed that is above the median.
- The centre with the highest cost per bed also has a low number of beds.
- One centre with low staff cost per bed staff has the highest number of beds.
- The centre with the highest patent activity that offers specialist services is one of the most costly. However such specialist centres treat the people with severe disease.

3.4 Suggested additional staff at UKCCSGs and TCTs based on the needs assessment exercise[7]

There is a marked variation in the numbers of AHPs employed in the centres, (Table 31 of the needs assessment[7] costs table 3.2). Some centres have limited or no access to physiotherapists, speech and language therapists, occupational therapists, play specialists or clinical psychologists.

Respondents commented that staff shortages prevented services such as a 24 hour on call system for palliative care from operating. An increased level of MDT working and the named key worker will create a further burden on staff time. These services can only be sustained if there are adequate numbers of appropriately trained staff.

There are national shortages of generalist AHPs; currently there are 63 vacancies for dieticians, and over 600 vacancies for both occupational therapists and physiotherapists in England[8]. Considering scientific and technical staff, there are just under 200 vacant positions for speech therapists and for clinical psychologists plus 172 for operating theatre staff. At a consultant level there are 11 clinical oncology vacancies and 128 anaesthetists; within nursing there are 397 vacancies. These vacancy figures are taken from Department of Health vacancy figures 2004 and are vacancies of 3 month duration or more as at 31 March 2004 which Trusts were actively trying to fill[8]. **Recruitment of specialised staff to enable the guidance to be fulfilled will therefore not be immediate, it will take some time to achieve.**

There are national shortages of diagnostic and therapeutic radiographers, not just those with a paediatric speciality, there are 488 vacancies for generalist diagnostic and 144 therapeutic radiographers nationally[8]. From the needs assessment we know that only 5 of the 17 paediatric principal treatment centres employ dedicated paediatric diagnostic or therapeutic radiographers.

The Cancer Group Workforce Team is working to improve the current recruitment and retention issues involved in tackling the AHP, consultant and nursing staff shortfall. Initiatives are in place to improve the career progression for radiographers. Provision of 3 additional histopathology training schools will expedite the training of SHOs/SpRs, as well as piloting extended roles for biomedical scientists. It will be some time before these initiatives start to have an impact on staff numbers. In the meantime it will be difficult for commissioners to recruit staff in line with the guidance, although this will vary according to the local situation. The economic impact calculated here is based on the assumption that the staff are available.

The numbers of additional staff detailed here should be regarded as an absolute minimum requirement for MDTs, key workers, and 24 hour on-call systems as suggested in the guidance to be either instigated or maintained. The method of calculating the costs has been detailed above.

3.5 Costs of additional staff

Additional funds required to enable each principal treatment centre to have minimum staffing levels of each key professional discussed above is outlined in Table 3.6, Appendix B.

An additional expenditure of £3.3 million would give each Centre at least 1 WTE AHP, speech therapist, clinical psychologists (including provision in palliative care), adolescent support worker, data managers and social workers. A further £1 million would employ a minimum of 1 WTE Paediatric Oncology Outreach Nurses (POON), specialist nurses and research nurses to be employed at each centre. It is of note that Macmillan are one of the current funders of POONS. A

similar calculation has not been possible for all nursing staff as the situation is far more varied.

We note that there is a marked variation in the numbers of nurses employed in the different centres. Guidance from the Royal College of Nursing recommends that in paediatric oncology units, a third of the beds should be staffed to high dependency levels, that is at a ration of 1:2 nurses to patients. The remainder of the beds should be staffed at a ratio of 1:3. Further investigation of staffing would need to be conducted at, at least, cancer network level, to estimate additional nurses by grade, that need to by in post.

When considering consultant posts, taking into account the vacant posts, we have estimated that an additional £2 million would be needed to give each centre at least 1 WTE of each specialist. £2.8 million would be required to ensure that each centre has a minimum of 2 WTE of the listed consultants at each centre.

Radiographers have been excluded, based on the assumption that radiographers, both diagnostic and therapeutic, would mainly be required at those centres that administer radiotherapy treatment.

We estimate that for each of the principal treatment centres for children and young people in England and Wales to have at least 1 WTE complement of the staff detailed above would cost around £6.39 million, (mean of £0.37 million per principal treatment centre) and for each to have 2 WTEs there would be a cost requirement of £14.5 million, (mean £0.85 million per principal treatment centre), excluding any further additional nursing staff.

3.7 Employment costs for all centres

Based on the calculations outlined in this section, we estimate that the current level of investment in staffing cancer services for children and adolescents under 25 treated at either UKCCSG centres or TCTs in England and Wales is between £42.5 million and £62.5 million per annum. **Using the ± 25% margin the range of costs required for the additional staff at these 17 principal treatment centres, including the 8 TCT units, is between £4.8 and £8 million for the**

minimum staffing level and between £10.9 and £18.2 million for the moderate level of 2 WTE.

We acknowledge that not all children and young people are treated in specialist centres; approximately 90% of all children are treated at UKCCSG centres[9]. Caring for children and young people with cancer has an impact on shared care centres at district general hospitals, particularly in terms of staffing costs. The costs of staffing at shared care centres has been too complex to include in this analysis. It is further anticipated that additional costs as a result of the guidance will primarily be at principal treatment centres.

One of the uncertainties that the 25% margin attempts to control for is that even at principal treatment centre level it is not always possible to separate costs of the care for young patients in general from those patients with cancer, or from young adults and other adults who are hospitalised. In addition, it has not been possible to assess the additional requirement, if any, of staff not mentioned above, for example, hospital pharmacists, scientific staff or clerical staff, or indeed any cost savings as a result of the guidance.

We further acknowledge that the estimated cost implications includes staff such as social workers, play specialists and some palliative care nurses who are predominantly funded by charities, the total estimated cost of employing these members of staff is just under £3million.

4. Multidisciplinary Teams

In this section the key costs involved in implementing the recommendations for the suggested levels of MDT working at principal treatment centres and at other treatment centres for children and young people are estimated.

A key recommendation of the guidance states that

> *"Care should be delivered throughout the patient pathway by multidisciplinary teams (MDTs), including all relevant specialist staff. Membership and governance of these teams should be explicit and include clearly defined responsibility for clinical and managerial leadership".*

The costs outlined below are based on the suggested staff membership as detailed in Tables 4-6 of the guidance. As stated in the guidance the membership of the MDTs for children and young people with cancer will vary according to need and whether the meeting is diagnostic, treatment, psychosocial or palliative. We have attempted to estimate the employment costs of the three MDTs suggested in the guidance, an average core MDT, a late effects MDT both based at the principal treatment centres and also the core MDT based at other sites. We acknowledge that the estimates will be imprecise. In addition to these MDTs, there are specific tumour types which need consideration by either a specialist MDT or through liaison with other sub-specialists, as listed in the guidance. The costs of such teams are beyond the remit of this report.

4.1 Methods

The methods of calculating the salaries are outlined in section 2. Annual meeting costs are derived by estimating the time spent attending meetings by suggested membership multiplied by their hourly rate (salary and on-costs, leave and sickness taken into account).

At present no calculation has been made to estimate any savings the use of teleconferencing would present. The cost of petrol and parking has not been taken into account.

The guidance suggests that the teams meet weekly and the estimated costs reflect this. We have assumed that for each meeting one hour of preparation might be required for all members plus 3 hours for the supporting clerical officer/secretary. We anticipate that the duration of weekly MDT meetings would be either 1 or 2 hours, both scenarios are included in the Tables 4.1 and 4.2.

The costs are estimates and will vary depending on the actual make-up of the MDT. The costs are based on the existing 25 principal treatment centres in England and Wales, comprising 17 UKCCSG Centres plus a further 8 teams to facilitate young people treated at TCT units. Estimates for the costs of MDTs at non-principal treatment centres with shared care are based on the assumption of 1 or 2 per cancer network.

The costs presented are compared with:-

i. a baseline of no MDTs at present, and
ii. additional costs from a baseline of 40% (assumption that currently 40% of centres have fully functioning MDTs). This will not be the case for all MDTs.

The costs are based on meetings being held in normal working hours. This has not been the case in the past with 91% of respondents reporting that meeting were held either at lunch time or outside normal working hours[10].

Sensitivity analysis

The cost scenarios that are presented are for employment costs for those attending meetings. Two scenarios are presented

1) Low cost scenario – where each principal treatment centre has a weekly MDT of all types described in the guidance that take 1 hour
2) Moderate cost – where each MDT has a duration of 2 hours

4.2 Costs of MDTs based at principal treatment centres

The estimated costs relating to the suggested core members of MDTs are given in Table 4.1, Appendix C. The costs will vary across cancer networks in line with

existing provision and local arrangements, and indeed on the exact configuration of the MDT.

The estimated annual cost per principal treatment centre is £66,200 to support the salaries of each member of the 2 suggested MDTs for an hourly weekly meeting. This would rise to an estimated £98,000 to support 2 hourly meetings. This estimate is expected to be the same for teams considering either young people or children.

An estimate of between £1.1million and £1.7million would be needed to provide England and Wales with the suggested requirement of MDTs to consider paediatric cancers at 17 principal treatment centres. The costs of the same MDT provision for each principal treatment centre, that has a facility for young people, currently 8 in England and Wales is estimated as between £0.5 million and £0.8 million. This does not include an estimate for young people who are treated in an adult setting. We acknowledge that :-

- the principal treatment site for young people might have a different location from those that treat children; and
- it is possible that not all principal centres will require all the teams described above.

It is assumed that resources will need to be made available to allow staff to attend MDT meetings and to cover any absences. It is further assumed that additional staff would be needed to facilitate this. This is considered separately.

4.3 Core MDT at other treatment sites.

In order to estimate the costs relating to the core MDT at other treatment sites, we have again given estimates for both hourly and 2 hourly meetings, Table 4.2. It is estimated that the range for hourly to 2 hourly meetings annually would incur an estimated salary cost of between £24,800 and £36,500 for each treatment centre.

The guidance recommends that local hospitals that treat children and young people with cancer enter into shared care arrangements with principal treatment centres. For the purposes of this estimate we have assumed that there would be 1

or 2 such arrangements per cancer network or between 37 and 74 MDTs based at other treatment centres in England and Wales, (assumption based on the needs assessment excercise[7]).

The estimated cost of employing the staff involved in the core membership of MDTs at 37 other treatment sites in England and Wales is estimated to be approximately £0.9 million per annum for hourly meetings, including an element for travel and £1.3 million for 2 hourly meetings, again inclusive of some travel Table 4.2 Appendix C. The cost for 2 MDTs per network, one for children and one for young people, would be between £1.8 million for 1 hour meetings and £2.7 million for 2 hourly meetings.

4.4 Total estimated MDT costs

The costs involved in establishing the MDTs for children and young people with cancer, at the existing 25 principal treatment sites, is estimated to be £1.6 million for 1 hour meetings and £2.4 million for 2 hour meetings. The estimated cost of all teams, including 74 shared care MDTs (2 per cancer network) the estimate would be £3.5 million for hourly meetings rising to £5 million for meetings lasting 2 hours. This would vary in line with existing local arrangements/requirements.

4.5 Net costs of MDTs

The cost analysis has been presented above on the assumption that no MDTs currently exist within the cancer network. This is not the case, however there is uncertainty about the number that are currently meeting weekly with a full complement of staff. The most recent Audit Commission/CHI[10] report indicated that 22% of Trusts had MDTs in place for children and young people with cancer, although not all of them were fully functioning. We would anticipate that this figure has improved in the intervening years, if we assume that 40% currently are fully functional then we estimate that the annual net costs in England and Wales will be between £0.9 million for 1 hour meetings and £1.4 for 2 hour meetings at the existing 25 sites. The assumed net costs, using the same assumption that 40% currently meet weekly with a full compliment of staff, then for 74 shared care MDTs (2 per network) the annual net costs would be £1.1million for hourly meetings rising to £1.6million for meetings lasting 2 hours, inclusive of some

travel, as detailed above. **The annual salary cost estimate for the 3 teams, as detailed in the guidance, meeting weekly in England and Wales will be between £1.6 and £2.6 million for hourly meetings rising to between £2.3million and £3.9 million for meetings lasting 2 hours**. (Estimate includes the ± 25% to allow for uncertainty).

The costs will vary across cancer networks according to the current frequency and attendance at MDT meeting. Factors such as the number of teams serving the cancer network, which team members travel and the distances travelled (or the cost package for video conferencing charges) will impact on the annual cost of running MDTs and should be investigated independently by each cancer network.

4.6 Additional Staff Requirements

It may be that in some centres where there will need to be an increase in the current frequency of MDT meetings, then staffing issues maybe significant. It may be that increased staff will need to be employed, particularly with additional time spent in meetings and additional travelling requirements, in order to cover MDTs members leave, both holiday and sickness. It should be noted that for some members of the MDT, in particular the key worker, funding may not be required as this post may already be carried out by another member of the MDT, e.g. the clinical nurse specialist.

Shortages of specialist neurosurgeons, radiologists, histo/pathologists, oncologists psychologists and AHPs will hamper development of full MDTs in the short and the long term. Methods may need to be considered to share neighbouring expertise when there is a shortage of personnel.

Many MDTs currently lack administrative support which will also need to be addressed by local managers. In order to ensure that all units and centres have fully operational MDTs in accordance with the guidance it is might be that a dedicated MDT co-ordinator/ administrator post is required in each centre/hospital. This will be an issue for local decision.

4.7 Discussion

The establishment of MDTs for cancer services for children and young people where none currently exist will have significant resource implications. The cost of service re-configuration for an individual cancer network will vary according to the existing MDT configuration and staffing levels, as well as the future MDT configurations. The base case assumes that meetings are held weekly, as suggested by the guidance. Factors such as the number of teams serving the centre, which team members travel and the distances travelled (or the price package for video/tele-conferencing line charges) and how, or if, MDTs combine will impact on the annual cost of running MDTs and should be investigated independently by each cancer network.

Teleconferencing offers the advantages that any travel time is eliminated, allowing for more efficient use of scarce specialist staff. This would impact on the shared care teams principally.

The cost analysis has explored a limited number of the potential variations; costs would obviously vary in line with local requirements. The cost analysis is intended as a guide rather than being definitive. It takes into account the age-specific MDTs and not the disease specific MDTs. Nor does it take into account costs of providing facilities, parking, public or private transport or specialist equipment.

Staffing issues will be significant and the development of MDTs will need to evolve gradually over a number of years. Additional staff will need to be recruited to allow existing staff the time to attend meetings, as well as to provide holiday and sickness cover. Currents shortages of staff will hinder both the development and operation of the MDTs. Furthermore, feedback from the needs assessment would indicate that some centres are stretched to provide current services. For the guidance to have any impact on outcomes additional resources will need to be in place.

5. Continuing Professional Development (CPD)

The workforce development section of the guidance states that:-

- *All staff should be trained and competent to undertake specific tasks and address the specific care needs of patients and families. They should also undertake relevant CPD to maintain their competence and stay abreast of scientific and technological advances.*

- *The need for trained specialist staff across all disciplines, able to work with children and young people with cancer, should be included in workforce development plans by cancer networks, to ensure the provision of a sustainable service.*

- *Specific attention is required to address the shortage of allied health professional expertise in this area and the need to develop robust evaluation of the contribution of such services.*

- *There should be access for nurses and other healthcare professionals to appropriate post-qualifying specialist education in the care of children and young people with cancer*

5.1 Background

'Continuing Professional Development. Quality in the new NHS'[11] was published by the Department of Health in July 1999. CPD had been defined in the earlier document 'A First Class Service : Quality In The New NHS'[12] as "a process of lifelong learning for all individuals and teams which meet the needs of patients and deliver the health outcomes and healthcare priorities of the NHS and which enables professionals to expand and fulfil their potential". CPD was envisaged to extend to the majority of health professionals, and would include learning from clinical audit, work based learning through the processes of coaching, mentoring, job rotation and shadowing, learning sets and work based projects in addition to attending courses. Education providers would be encouraged to deliver flexible, modular education and training with students studying in the workplace, home and classroom. The report noted that CPD is financed from a range of sources such as local training and development budgets, charitable and educational trusts and industry sponsorship with health professional sharing financial responsibility for their own professional development. In 'Working Together –Learning Together. A

Framework for Lifelong Learning for the NHS'[13] it is noted that most professions are "considering arrangements for some form of mandatory re-registration or re-validation, or strengthening their current requirements".

5.1 CPD in Medicine

The discussion of CPD in the field of medicine has been primarily focused in the post graduate training of doctors to equip them to work as general practitioners and specialists in the NHS. After attaining a first medical qualification, to qualify as a paediatric oncologist requires a further 2 years of general professional training in paediatrics as a Senior House Officer followed by a further five years as a Specialist Registrar during which an oncology speciality would be developed. On successful completion of this training the candidate becomes a consultant. CPD for consultants involves activities such as attending lectures, leading grand rounds, writing and presenting papers and attending meetings and symposia. For these purposes they are allocated an annual budget by their Trust of approximately £3,000 and time to carry out these activities. (see Table 5.1).

5.2 CPD in Nursing

Nurses who train in the UK can either undertake a Diploma in Higher Education (Nursing Studies) over three years of full time study or a Bachelor of Nursing (Hons) degree course over three to four years. Both courses are structured so that the first year is a Common Foundation Programme during which the basic principles of nursing are introduced. Students then branch into a speciality choosing between adult, children's, mental health or learning disability nursing.

Nurses in the UK are required to maintain their registration by meeting the post-registration education and practice (PREP) standards set by their regulatory body, the Nursing and Midwifery Council (NMC). This contains two components, a PREP practice standard which specifies the minimum hours of practice required to renew their registration and PREP CPD which requires nurses to undertake and record learning activity since they last registered. Both unstructured/informal learning activity and structured/formal learning from seminars, courses and clinical supervision are acceptable.

Until 31 March 2001 the English National Board for Nursing Midwifery and Health Visiting (ENB) was responsible for approving programmes that led to recordable qualifications. That role is now shared between the NMC the Department of Health and the Higher Education Institutions. A large range of post-registration courses are offered, encompassing diploma, degree and masters level courses. However there is no longer a central registry of courses kept and nurses are advised by the NMC to contact individual education providers or to search for courses using the internet.(Table 5.2). In addition Higher Education Institutions and organisations such as The Royal College of Nursing offer short courses on specialist topics which do not lead to a recordable qualification.

The loss within the UK of the ENB 240 course on the 'Care of the Child with Cancer' removed a stable benchmark of specialist education from within the speciality. Those wishing to undertake specialist education at degree level now have a limited number of options, with a degree in either child health or in cancer, at one of those few institutions which provide specific modules or programmes which address the care of children and young people with cancer. There is a new framework for a degree in Children's Cancer Nursing, which is available as a distance learning course.

Masters level education and preparation is increasingly recognised as essential for nurses taking on Clinical Nurse Specialist or Nurse Practitioner roles, as, for example would be required in developing Nurse Practitioners in Long Term Follow-up. This is inevitably undertaken on a very individual basis. There are an increasing number of Masters courses and 'student designed' awards which focus on independent clinical practice that can provide appropriate preparation for such roles.

5.3 CPD for Allied Health Professionals

Some courses offered to nurses by Higher Education Institutions are available for interprofessional learning. Other professions that would have contact with paediatric oncology patients, such as radiologists, have their own requirements for CPD. In addition multi-disciplinary study modules are available at NHS trusts. The Royal Marsden NHS Foundation Trust, for example, has 40 such courses, including a 10 day module "Caring for a Child/Teenager with Cancer in a Non-

Specialist Setting" at £480 per module[14] .

5.4 Training for Play Specialists

A Professional Diploma in Specialised Play for Sick Children and Young People is available in 10 colleges in the UK [4]. The fees are approximately £600-1000. The training is one year part time and it offers a minimum placement of 200 hours in a hospital setting for people who are not already in employment. The qualification is designed to give a competency in the provision of play for children and teenagers.

5.5 CPD costs

An estimated cost for CPD has been calculated on the basis of a mean cost per module of £800 for key nursing, AHP and scientific staff. This does not include staff time as time for training has been included in the costs of employing the additional staff. The CPD for clinicians was estimated to be £3000, as above.

The estimated costs associated with training and education for CPD of the additional staff detailed in section 3 would be between £146,000 and £257,500 in England and Wales (Table 5.3). This assumes a basic professional skill level as a baseline. Further costs relating to the basic training costs of AHPs, clinicians, nurses can be accessed in Netton and Curtis[3.]

6 Other Potential Cost Implications

6.1 Specialist Paediatric Diagnostic Services

The guidance states that

"Histopathological diagnosis of paediatric tumours can be difficult due to their relative rarity, the overlapping morphological phenotypes and the increasing use of small core biopsies for primary diagnosis and the different interpretation of pathological features in the context of paediatric as opposed to adult cases. Many tumours are unique to children and specialist knowledge is essential.

The requirements for the histopathological diagnosis of tumours in young people are very similar. There is clearly an overlap with tumours of the paediatric age group, but also the other tumours that are increasingly common in the teenagers and young adults (such as lymphomas, bone tumours and germ cell tumours) all require very specific expertise for their correct diagnosis and assessment". (p39)

6.1.1 Background

The Department of Health is currently modernising pathology services, and the workforce issues in particular are being considered by The Cancer Workforce Initiative[8]. It has been recognised that there are currently too few trained paediatric pathologists[8]. As a result, various initiatives are in place to increase training opportunities. It will take some time for these improvements to have an impact on current shortages.

Although pathologists involved in the delivery of the clinical service (eg. paediatric surgical or autopsy pathology) usually contribute to the MDTs (multidisciplinary team meetings) the current severe shortage of paediatric pathologists limits the time available for MDT activities, particularly when travel is required[15]. Pathologists with a University appointment who contribute to service delivery, also contribute to the MDTs, in the usual way. Still more staff would be required to meet the expectations set in the guidance

6.1.2 Current workforce

In England there are 28 paediatric pathologists, including 2 academic posts, of which 7 are single handed, and in addition there are 7 part timers. There are currently 12 vacancies. In Wales there are 2 academic paediatric pathologists and no vacancies. Currently in England there are 7 pathologists training to have a specialty in paediatrics, and 4 vacancies. There are none in Wales. The situation will be further exacerbated in the next 5 years as 5 will be retiring[16].

The MDT requirement for paediatric pathologists to attend weekly diagnostic MDTs meeting at each principal treatment centre hours, would be between 1 and 2 hours a week at each centre (for costs refer to Table 4.1). This equates to an additional requirement of between 0.5 WTE and 1 WTE paediatric pathologists across England and Wales. Discussions with representatives from the Royal College of Pathologists acknowledge that in view of the current shortage not all diagnostic MDTs have a participating paediatric pathologist[15]. Local commissioners need to take this into account, particularly in areas where there are single handed or part-time pathologists.

The guidance recommends that all paediatric tumours are reviewed by specialist paediatric pathologists. Again local commissioners need to consider if this is this current practice.

In addition, the diagnostic MDTs for young people will require input from histo/cyto-pathologists, some of whom will be diagnosed via the disease specific MDT. This will have local impact in areas of extreme shortage of pathologists with a high workload.

The guidance on Improving Outcomes in Haematological Cancers [17] has relevant costing information concerning pathology laboratories.

6.2 Imaging

The guidance states that

"The provision of MRI scanning should be sufficient to ensure that suspected cases of CNS and other malignancies can be investigated rapidly".

There are currently 223 MRI scanners and between 200 and 230 CT scanners in the UK; the capital costs per scanner are £0.75million and £0.45 respectively. Costs per MRI scan recorded in the National Tariff are £352, and CT scans for radiotherapy planning are listed as being £96[1]. Additional costs would be incurred for younger patients as a result of the guidance recommendation that a play therapist or key worker be in attendance.

It is not anticipated that the guidance will have any impact on provision as such scans are currently routinely conducted. As with other areas of service provision for child and young persons with cancer, staff shortages is the area of concern for service delivery. In 2002 the Royal College of Radiologists anticipated that a further 228 additional radiologists per year would be required in England and Wales by 2005; this did not include any provision for 24 hour service availability. Annual training provision currently is nearer 80 per year.

6.3 Protected theatre time and access to paediatric anaesthetists

The key recommendations of the guidance state that

> *"Theatre and anaesthetic sessional time should be adequately resourced for all surgical procedures, including diagnostic and supportive procedures, in addition to other definitive tumour surgery. Anaesthetic sessional time should also be assured for radiotherapy and painful procedures. The paediatric surgeon with a commitment to oncology should have access to emergency theatre sessions during routine working hours".*

6.3.1 Costs related to theatre service

Literature searches for costs of theatre time for children and young people with cancer using Medline, NHS EED and HTA were unproductive. The National Tariff does not include costs for central venous line access as a separate procedure for costing within HRGs, although it has been included in some of the larger procedures[18].

The following theatre costs are based on information from 5 finance directors (August and October 2004),

i. the costs of inserting a central venous line in theatre, is estimated to be £2,633 and
ii. the estimated cost per hour of medical oncology theatre time is £423

The costs relating to BMT for children range between £20,000 to 100,000 depending on the nature of the transplant[1].

6.3.2 Access to paediatric anaesthetics

The needs assessment found that at least 6 of the centres had inadequate numbers of paediatric anaesthetists. Two reported having 0.3 or 0.4 WTE seeing 300-400 new patients annually, and a further 3 (where there were surgeons) reported having no specialist paediatric anaesthetist. There was a minimum of 5 paediatric anaesthetists available at the remaining centres. It was reported that theatres are operational between 8a.m. and 8p.m. at a 98% capacity.

Therefore it is estimated that between 3 and 7 additional paediatric anaesthetists would be needed to provide additional cover at existing centres. The lower figure would give all sites at least one WTE and the upper would give a minimum of 2. The cost of employing the additional anaesthetists would be a range between £276,000 and £700,000 Table 3.6 (included in chapter 3).

6.4 Computerised prescribing

The guidance states that

"Agreed protocols across cancer networks and within shared care arrangements, ensuring the safe administration of appropriate and timely treatment, should:

- *result in better clinical outcomes in terms of response, survival and symptom control*
- *minimise complications and errors in the prescription and administration of chemotherapy*
- *improve patient and family confidence.*
- *improve quality of life*

Reduction in risk may be achieved by:

- *computerised prescribing*
- *patient held records*
- *information for parents and families*
- *training and education for staff in all care settings in which chemotherapy is given*
- *designated pharmacist as part of MDT"*

(p 49)

6.4.1 Background

Computerised prescribing enables a secure electronic transfer of prescriptions between the GP or secondary care clinical, pharmacy and the Prescription Pricing Authority and has the potential to reduce errors while ensuring that all prescribing is recorded.

A national program of improvements to information technology has been commissioned[19]; the government has committed £6.2 billion to the strategy which will include the introduction of computerised prescribing and electronic transmission of prescriptions (ETP). Some UKCCSG centres have a dedicated computerised chemotherapy prescribing system. The National Program for Information Technology aims for all prescribing to be electronic by 2010 and for all

hospitals providing chemotherapy should have an electronic prescribing system in place by 2006[19].

The costs involved are detailed below for reference, it is a subject for debate as to whether this expenditure is part of the cost implications of the guidance or should be considered as a cost relating to the national strategy for the whole of the NHS.

6.4.2 Methods

An electronic search of NHS EED, HTA and DARE was performed. It did not reveal any oncology related papers on electronic prescribing systems. A review article from the US was identified but not considered sufficiently robust or of sufficient relevance to this analysis[20].

Enquires were made with the designers/producers of an electronic prescribing system (ChemoCare) to estimate costs of ETP. **Costs of ChemoCare installation has been estimated to be between £240,000 and £500,000 per principal treatment centre[21]**. Other systems are also commercially available. The lower estimate is based upon 1 principal treatment with 50 concurrent users and the upper estimate would involve 1 hub (principal treatment centre) and 5 spokes, 100 concurrent users.

The cost of an electronic prescribing system would be dependent on a number of variables:

- Single hospital or cancer network
- Number of concurrent users in each cancer centre
- Number and complexity of interfaces to other hospital system:
    - Pathology
    - Patient Administration System
    - Radiotherapy scheduling system
- Installation which will include costs for:
    - Project management
    - Training
    - Implementation

The costs can therefore be seen to be highly variable. However, the costs above were based on a system recently supplied in a large teaching hospital and a large Cancer Network[21].

It is not possible to say at the moment how many principal treatment centres do have electronic prescribing in place, therefore no attempt has been made to estimate costs for England and Wales. As with other areas of the guidance we suggest further considerations need to be taken at a local level.

6.5 Key workers

The guidance states in the key recommendations that

"Appropriately skilled, professional key workers should be identified to support individual children and young people, and their families by:

- *coordinating their care across the whole system and at all stages of the patient pathway*
- *providing information*
- *assessing and meeting their needs for support".*

Literature searches did not identify any evidence concerning key workers in this area. However, key workers are employed to ensure continuity of care of disabled children. Two different models of key worker interventions are found in the UK, either a specifically employed individual is employed to be a key worker for up to 30 children (range 12-40) or an existing worker is nominated to be a key worker for upwards of 3 children (range 1-35)[22]. The economic impact would depend on which model is adopted.

A key worker for each patient would provide a structured route for co-ordination and continuity of care as the patients moves through the patient journey, including between centres and shared care. It has been shown that there is an inadequate provision of staff who are likely to fulfil this function. The guidance suggests that the key worker would change during the course of the patients care.

Within UKCCSG centres the role of key worker will most usually be undertaken by a Paediatric Oncology Outreach Nurses (POONS). At present the case load

carried by POONS varies from centre to centre, but as the needs assessment exercise demonstrated there is a shortage in many centres. For other patient the key worker may be the paediatric or young persons CNS.

Some centres have established other models of care and support have been adopted. Here the role of the key worker could be taken by other nursing staff, such as Nurse Specialists for Leukaemia, for Neuro-oncology, or for Bone Marrow Transplant patients[23]. They could operate either across the length of the patient pathway through treatment, or for defined episodes of care.

There is a general lack of identified key workers for young people, although there are Nurse Specialists for Teenagers and Young Adults in a number of both UKCCSG and TCT centres[23]. This is a role which needs to become more established, and clearly this will have an economic impact.

The UKCCSGs and TCTs have around 1900 new patients per year[7], with a range of 60-172 per centre. If the model adopted was to employ a new member of staff to be a key worker responsible for 30 patients then 95 new members of staff would be required across England and Wales, with a range of 3-8 per centre. This calculation is based on the assumption that for every 20 new patients there would be a further 10 on the existing caseload[23]. Therefore, the cost of employing new staff as designated key workers in the principal treatment centres in England and Wales, based on a nursing salary plus on-costs as before, would be in the order of £3.3 million. The 25% margin would give a range of between £2.5 million and £4.1 million. When considering the costs at individual centre level the range would be between £103,500 and £276,000 between centres based on the range of 60-172 new patients per centre.

The actual cost of employing key workers will vary according to the model adopted. It should be noted that for some principal treatment centres extra funding may not be required as this post may already be carried out by another member of staff e.g. the clinical nurse specialist. Further work concerning the economic impact will need to be conducted at the local level. This analysis does not

consider the economic implications of key workers at the shared care centres because the situation is far more complex.

6.6 Protocol based care

It is possible that an increase in protocol based care may be likely to result in cost savings as a result of improvements in outcomes. It is not possible to estimate at present the level of this potential saving.

6.7 Place of care

The key recommendations of the guidance states that

> "*All care for children and young people under 19 years must be provided in age appropriate facilities. Young people of 19 and older should also have unhindered access to age appropriate facilities and support when needed".*

The guidance does not anticipate any increase in provision of age appropriate facilities for children at present, not does it estimate a projected number of additional units for young people. However, there is a need for increased provision of age appropriate facilities for young people adjacent to areas of existing expertise in adjoining paediatric or adults oncology centres. Exact location would need to be considered by commissioners at a local level.

The Teenage Cancer Trust (TCT) estimates that it costs £750,000 to plan, build, and equip each new unit with accommodation for both in-patients and day cases.[6]. The TCT aims to increase the current provision of 8 TCT units in England and Wales to 18 units to enable all young people with cancer to be treated in age appropriate facilities[24]. A further 10 units would require further expenditure of approximately £7.5 million in capital costs alone; the ± 25% to allow for uncertainty about the exact number required would give a range of between £5.6 and £9.3 million.

Staffing an additional unit would entail recurring annual costs of between £0.6 and £0.75 million per TCT unit, based on information from 2 NHS Trust managers. This estimate includes nursing, activity co-ordinators and some medical support. However, it is acknowledged that the medical staff would be shared with other age groups.

As with other aspects of the guidance the costs relating to any additional facilities for young people would not be an immediate cost and the revenue required would vary across cancer networks in line with existing provision, activity or need and expertise.

It should be noted that some of the costs outlined above could be offset against income generated for the TCT units from other service providers who were not able to offer age-appropriate facilities in their localities. The relevant HRG tariff for in-patient stays is around £2000 and around £900 per day case. There would be cost savings in other areas of cancer services where young people have been inappropriately treated, such as adult or paediatric wards. However it is not possible to quantify those savings.

The costs of services provided by TCT for young people with cancer are explored further in the next section.

7. Financial support from Charitable Sources in Cancer Service Provision for Children and Young People with Cancer.

7.1 Objective

The objective of this section is to identify charitable funding contributions to children and young people with cancer services in England and Wales in 2003 in order to make a broad estimate of the current extent of charitable support in the provision of these services.

7.2 Contextual overview

The knowledge that charities provide considerable financial contributions towards the provision of health care services in the UK is not new. In 2000, it was estimated that in London alone, charities contributed £500 million a year to health care with £200 million of this going directly to hospitals[25]. Funding from charities provided approximately 10% of the London NHS budget. The greater proportion of charitable contributions supports research, refurbishment and expansion of hospital buildings and clinical equipment.

Identifying the vital role charities play in supporting NHS services is no easy task. The current exercise was an attempt to quantify the degree of charitable funding to children and young people with cancer services in England and Wales.

The Charity Commission lists around 200,000 registered charities in England and Wales, of which around 650 are raising funds for cancer[26]. Cancer charities range from large national organizations with paid workers to small local charities relying on volunteers. Their aims and activities also vary widely and may involve funding hospices, contributing to capital projects, providing psychosocial support to patients and their families, supporting research or financing posts for health and allied professionals working in hospitals and community services, to name but a few. A similarly large number of charities work to promote the interests of children and adolescents and some of their funds will undoubtedly be directed to young cancer patients.

Cancer research in 2000 attracted £180 million per annum in funding across the UK, of which £20 million came from the government[25]. Two hundred and fifty charities are estimated to support cancer research in the UK[27].

It was hoped to establish the degree to which service provision for children and young people with cancer in England and Wales relied upon charitable support in one year. However, many difficulties were encountered and are described in detail in the discussion section. What is reported here is an attempt to estimate the financial contribution made by charities to children and young people with cancer services in England and Wales in 2003.

7.3 Methods

To assess the funding contribution made by charitable donations to cancer services for children and young people in the England and Wales, charities involved in fundraising for children and adolescents with cancer and for hospices providing institutional and or community-based care for these children and adolescents were identified.

The lists of hospices and charities were compiled from several sources. Firstly, members of the Child and Young People with Cancer Guidance Development Group were surveyed for contact details of charities operating in their area. Next, Macmillan Cancer Relief, National Alliance of Childhood Cancer Parent Organisations (NACCPO), CancerBACUP and the Charities Commission websites that list support groups for children and young people with cancer and their families and fundraising charities were consulted. Keyword searches were undertaken on the Charities Commission Register using common cancers of childhood such as leukaemia, brain tumours, neuroblastoma and non-Hodgkin's lymphoma, Hodgkin's disease and retinoblastoma.

A further search was also made on the internet for hospices in England and Wales which care for children and adolescents. Finally, each charity that was contacted was asked to name any other charities they were aware of working in the field.

For ease of analysis the charities identified were subdivided into three groups:

- charities that were primarily established to support children and adolescents with cancer
- charities established for other reasons but who direct some of their expenditure to supporting children and adolescents
- charities established to support the running of a hospice caring for children and adolescents.

For the first two groups, the Charities Commission website[28] was used to identify the total expenditure for each charity in 2003 or for the closest preceding year. In order to establish the proportion of those funds that directly supported young people with cancer, an annual report or statement of accounts was sought from which to extract data, either via the internet or by email with follow–up by phone. By comparing the reported total expenditure with the funds directed to supporting children and young persons with cancer, an estimate of the proportion of funding supporting these services was made.

7.4 Results

7.4.1 Charities established primarily to support children and young people with cancer.

The first category of charity surveyed was those established primarily to provide support for children and young people with cancer, and their families; this group constituted the majority of charities identified. Forty two charities were identified in this group and a brief description of the objectives and the total expenditure for 2003, or the closest preceding year, of each of these charities is shown in Appendix E, Table 7.1.

The total expenditure reported by each charity on the Charities Commission register includes monies spent on raising funds and overheads. Total expenditures for each charity were summed to yield a total expenditure of £30,340,220; with each charity contributing £740,000, on average.

Charities were contacted directly and annual financial reports were sought on the internet in order to establish how much of the total expenditure was directly supporting cancer services for children and young people. This data suffered from a low response rate and detailed information was only obtained for 15 charities (36%) in this category. In addition, extrapolation from the internet at times involved averaging over several years. Charities were also asked where possible to break expenditure down to show financial contributions to staff costs, capital projects, accommodation, care grants, research and education. In order to adequately represent the variation among the charities a brief description of each follows Table 7.2 in Appendix E.

By comparing the results from the survey of the Charities Commission register with information drawn from annual reports web sites and responses from individual charities we estimated that an average of 85% of expenditure goes to funding cancer services with the remaining 15% of expenditure covering costs incurred in raising these funds.

A total expenditure of £30,340,220 was reported in the Charity Commissions register by the 42 charities we identified whose primary objective was to assist children and young people living with cancer and their families. Applying the estimate that 85% of the total expenditure goes directly to supporting this patient group, while acknowledging the uncertainty of the estimate, we calculated that an estimated £25,790,000 was directed by this group of charities in 2003 to support the provision of services for children and adolescents living with cancer. Using the previously applied range of ± 25% to allow for uncertainty in factors such as the percentage of expenditure supporting service provision and the contribution from those charities not identified, this resulted in a range of £19,340,000 to £32,240,000. A summary of results for charities established primarily to support children and adolescent with cancer is shown in is shown in Appendix E, Table 7.3.

7.4.2 Other (non-hospice) charities which support children and adolescent with cancer.

The second group to be considered was those charities that allocated a portion of their funding to children and adolescents with cancer. This encompassed a large range of organisations and included those charities established to support cancer patients in general, charities involved with a specific form of cancer, or with diseases relating to a specific body organ and charities supporting illness in childhood. Again the scale of these charities varied between very large and established national organisations to small localised charities. We estimated that as many as 1000 charities undertook activities which could potentially benefit children and adolescents with cancer. It was not feasible to locate details for all of these so our analysis was based on a limited sample of 104 charities from this group. Appendix E, Table 7.4 shows the total expenditure in 2003 for charities who direct part of their expenditure to supporting cancer services for children and young people.

The second area of uncertainty was the amount these charities could be expected to direct to support children and adolescents with cancer. Both the needs assessment "An Assessment of Need for Child and Adolescent Cancer Services in England and Wales"[7] and official statistics indicate that the incidence of cancer in the under 25 age group is in the vicinity of 1%.

Charities were asked to estimate the amount which they estimated had been directed to supporting services for children and young people with cancer. Most charities found it extremely difficult to isolate this component from their overall activities and were unwilling to attempt to do so. Consequently, an exceptionally low response rate was achieved with only three estimates. Estimates of the percentage of the total expenditure which these three charities directed to support children and young people ranged between 0.3 to 3% and is shown in Appendix E, Table 7.5 and is followed by a brief description of each charity.

Combining this information to provide a meaningful summary required numerous assumptions to be made, involving a degree of uncertainty. We used a 1% incidence rate of child and young people with cancers as the basis to apportion expenditure by charities while acknowledging the uncertainty of this estimation method.

Total expenditure for the 2003 year in this group of charities ranged from £50 to £301,318,327 (mean: £5,646,824; SD: 31,394,003). When charities with a total expenditure greater than £20,000,000 were excluded, mean expenditure was reduced to £604,182 (SD: 1,723,277). This figure was used to avoid overestimation of the contributions of the remaining 900 charities for which no data were available. Assuming these charities direct 1% of their annual total expenditure to support cancer services for children and young people, we estimated the total charitable contribution of these groups to be £6,041,820 (± 25% £4,531,365 to £7,552,275). To this was added the known contribution of £9,000,000 from Cancer Research UK resulting in a final figure ranging between £13.5m and £26.6m. A summary of results for other (non-hospice) charities which support children and adolescent with cancer is shown in is shown in Appendix E, Table 7.6.

Another source of funding is the New Opportunities Fund which was established as a lottery distributor in 1998 when the UK National Lottery was established. This has recently been renamed as the Big Lottery Fund. Cancer related funding opportunities vary between England and Wales. In England there are 2 funding streams for palliative care one for adults and one for children with total programme awards for £20.6 million and £40.7 million respectively. In Wales there is one funding stream for palliative care worth £3.5m. Since the New Opportunities Fund is not a registered charity its contributions were not included in this section.

7.4.3 Hospices

The third group is charities which support the work of hospices for children and adolescents some of whose patients will have a diagnosis of cancer. We located thirty one hospices (administered by 28 organisations) in England and Wales providing care for children and adolescents. Several other hospices which are still in a pre-operational establishment phase were excluded since they could not supply the required data. We contacted all 28 administering organisations and received ten responses (36%) to our request for information. Table 7.7 in Appendix E shows the results of the survey.

In trying to establish the total charitable contributions supporting the activities of all hospices working with children and adolescents it was necessary to make several assumptions on the basis of the sample. Firstly, it was assumed that one in 10 hospices working with children and adolescents would not have any oncology clients, leaving 27 sites from which hospice care was provided. Secondly, whereas a large proportion of adult hospice patients will have a cancer diagnosis, comparatively low numbers of children and adolescent hospice patients have a diagnosis of cancer, which we estimated to be 6% of the client base. Thirdly, on the basis of the cancer client base a proportional estimate was made of the average charitable funding directed to oncology patients however, this does not allow for variation in resource use and costs by diagnosis related group. This figure was extrapolated to the 27 hospice sites providing care to children and adolescents with cancer, summed and adjusted by ± 25% to form an estimated total annual charitable funding of hospice services ranging between £1,948,759 to £3,247,931 in 2002-03 and £2,203,969 to £3,673,283 in 2003-04 to support children and young people with cancer.

7.5 Discussion

In total, we identified, 42 charities directly involved in supporting children and young people with cancer, 104 charities that direct some of their resources to supporting this group of service users and 28 organisations involved in running hospices in England and Wales. Combining the collated information to provide a meaningful summary value proved to be extremely difficult. A number of complications and methodological problems were encountered in estimating the charitable contributions made to the provision of cancer services for children and young people in the England and Wales. These included:

- double counting errors caused by some charities distributing part of their funds on to other charitable organisations; for instance the Katie Trust contributed £6,000 to Sargent Cancer Care for Children; Children with Leukaemia passed £10,000 to Children's Leukaemia in Childhood (CLIC), and the Lisa Thaxter Trust directed funds to the UKCCSG

- difficulty isolating the component directed to children from cancer charities or identifying the amount directed towards children with cancer from charities supporting ill children
- no adjustment was made for charities that extended their activities beyond England and Wales
- different financial years were used by different organisations
- charities sampled were not selected randomly and may not be representative of all charities involved in supporting children and adolescents with cancer
- extrapolating from the sample required numerous assumptions to be made

7.6 Conclusions:

Our investigation of charitable funding contributions to cancer services for children and young people with cancer in England and Wales in 2003 identified a number of methodological difficulties and is a broad estimate rather than a definitive measure of the current amount of charitable support in the provision of these services.

We estimate that between £19.3 million and £32 million is raised from charitable sources to specifically support children and adolescents with cancer, a further £13.5 to £26.5 million is contributed to children and young people as a result of fund raising by cancer charities focusing on people with cancer; a further £2.2 to £3.6 million supports hospice services for children and young people with cancer. Therefore the estimated contribution to services for children and young people with cancer is estimated to be between £35 and £62 million per annum.

It is likely that the actual amount will be in the mid to upper range as there are other donations that have not been taken into account in this analysis. Examples could include equipment presented or cash donations direct to hospitals and cancer centres all of which might not been included in this estimate.

8. Conclusions

Implementation of the guidance is likely to have a significant cost impact. It is estimated that the total additional cost per year of caring for children and young people with cancer once the guidance is implemented will be around £19 million for the low scenario estimate and £28.1 million for the moderate scenario. The low scenario was based on at least 1 WTE key professional at each principal treatment centre with weekly 1 hour MDT meetings. The moderate scenario was based on 2 WTE key professional at each centres with the MDT meetings having a duration of 2 hours. The range for the lower estimate is between £14.3 and £23.7 and between £21.1 million and £35.2 million for the moderate scenario. The level of uncertainty surrounding these estimates is high and there will be significant variability between cancer networks. We have attempted to allow for the uncertainty of the estimates by applying a ± 25% margin. However it is likely that the low cost scenario will be exceeded.

The most significant resource implication is likely to be the workforce requirements, as a result of overcoming existing shortages of staff. The costs of employing additional workforce personnel will vary across cancer networks in line with existing provision and local arrangements. The costs are likely to between £6.4 million and £14.5, plus a further £2.9 million in order to provide additional key workers

Re-structuring of services into multi-disciplinary teams is suggested in the guidance and in many cases this recommendation may constitute a significant change to current practice. The estimated annual cost of MDT activity is likely to be between £2million and £3million. This is based on 2 teams at the principal treatment centres (25 comprising 17 paediatric and 8 for young people) and 1 team based at non-principal treatment centres (74 or 2 per network). This assumes core team members attending weekly meetings during the working day.

In line with the National Service Frameworks the service provision guidance for children and young people with cancer does recommend age-appropriate facilities. The 8 existing facilities for young people have been primarily provided by the Teenage Cancer Trust. If this provision were increased to 18 units in England

and Wales as recommended by the TCT then a further capital cost of between £5.6 and £9.3 million would be required plus recurring annual costs of between £0.6-0.75 million per TCT unit for staffing.

Respondents to the recent needs assessment exercise[7] commented that staff shortages prevented services such as a 24 hour on-call systems for palliative care from operating. An increased level of MDT working, need for improved 24 hour on call system for palliative care, and the named key worker will create a further burden on staff time. Such services can only be sustained if there are adequate numbers of appropriately trained staff. Recruitment of specialised staff to enable the guidance to be fulfilled will not be immediate, in view of the current shortages of many specialised staff from play specialists, clinical psychologists, specialist nurses to consultants in pathology, oncology and anaesthetics the cost implications of the guidance will be gradual over some years.

The costs implications of the guidance have focused on the costs at principal treatment centres, it is likely that other cost implications will fall in shared care facilities that have not been possible to estimate here. However it is also the case that there are potential cost savings as a result of changes the guidance will introduce.

The value of charitable contributions to cancer services for children and young people cannot be understated. The estimated contribution to services for children and young people with cancer is estimated to be between £35 and £62 million per annum, this is similar to our estimates for the costs of employing nurses, AHPs, psychologists and clinicians at principal treatment centres. It is likely that the actual amount of charitable contributions is nearer the upper margin as there are donations from members of the public direct to hospitals, and smaller charities that have not been included in this analysis.

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Appendix A

Details of Survey of UKCCSG/TCT Financial Directors

Financial Survey of finance directors of all UKCCSG and TCT centres as part of the initial needs assessment exercise.

Five responses from the 17 UKCCSG centres surveyed in England and Wales were received (response rate 29%) and none from current TCTs. Data from the survey is presented in the following Tables, a copy of the letter and questionnaire sent to the directors follows.

Table 1.i Best estimate of investment in children and young people with cancer for financial year 2002/3

Centre	Staff (%)	Drugs (%)	Radiotherapy (%)	Other NHS (%)	Charity (%)	Total
A	£3,076,020 (73)	£613,474 (15)	*	£395,775 (9)	£122,816 (3)	£4,208,085
B	£1,179,000 (45)	£552,000 (21)	*	£858,000 (33)	£14,000 (1)	£2,603,000
C	£1,364,700 (44)	£611,800 (19)	£23,700 (1)	£1,141,900 (36)	£5,000 (<0.2)	£3,147,100
D	£3,084,504 (64)	£1,707,479 (35)	*	0	£47,594 (1)	£4,838,577
E	£1,084,000 (74)	£250,000 (17)	*	£131,000 (9)	Not completed	£1,465,000
F**	£1,800,000 (34)	£2,300,000 (42)	*	£1,062,000 (20)	£200,000 (4)	£5,362,000

* Centres unable to estimate radiotherapy costs; treatment takes place elsewhere and not invoiced.

** Centre not in England or Wales

Table 1.ii Best estimate of annual costs (as above) per patient and per beds based on the NPHS survey

Centre *	New patients per annum	Beds	Care on site	Deaths per year (%)	Total estimated cost/year (£)	Estimated Cost per bed per year (£)	Estimated Cost per patient per year (£)
A	102, 10 over 15, all under 18 years	19, of which 4 teenage beds + 2 day case	BMT	26 (25)	4,208,085	221,478	41,255
B NPHS 5	60 3 over 15, max age 16	14 + 4 day case beds	BMT and neurosurgery	15 (25)	2,603,000	185,928	43,383
C NPHS 3	90-100 All under 18 years	10 dedicated + 2 shared care	BMT and Retinoblastoma	15 (15)	3,147,100	262,258	33,127
D NPHS 2	172, 8 over 15 all under 16	14 + 4 teenage beds	BMT, neurosurgery and Retinoblastoma	39 (22)	4,838,577	268,865	28,137
E	106 All under 16 years	9 **	Some radiotherapy and specialist bone and sarcoma surgery	8 (7)	1,465,000	162,777	13,820
F	80-90 All under 16 years	22	BMT, specialist bone and sarcoma surgery,	20 (23)	5,362,000	243,727	63,082

* NPHS survey code given where possible from Griffiths, Fone and Sandifer, Interim Report into Child and Adolescent Cancer Services Needs Assessment for England and Wales, October 2003. Centres A and E responses were received after the Interim Report was completed.
** Additional beds were available for those over adolescents over 15 but no dedicated beds

Table 1.3 Costs from a NHS Trust of radiotherapy services for children and adolescents with cancer during 2003.

Patients	Number	Total estimated cost/year (£)	Estimated Cost per child (£)	Range per child (£)
Children up to age of 15 years:	15	£52.6k	£3.5k	£336.00-£5,580.00
Young people 16 to 23 year olds	12	£39k	£3.24k	£900 - £5,000

Example information letter and questionnare sent to finance directors

11th September 2003

Dear Director of Finance

FINANCIAL QUESTIONS FOR THE GUIDANCE ON CANCER SERVICES: IMPROVING OUTCOMES IN CHILD AND ADOLESCENT CANCER

You may be aware that the National Collaborating Centre for Cancer (NCCC) have been commissioned by the National Institute for Clinical Excellence to produce **Guidance on Cancer Services: Improving Outcomes in Child and Adolescent Cancer.** My colleague Rhiannon Tudor Edwards and I, from the Centre for the Economics of Health, are working together with the NCCC and the Guidance Development Group to assess the economic consequences of the guidance.

We propose to start the process by finding out what the current investment budget for child and adolescent cancer is. This information together with the information gathered from the enclosed questionnaire pack will enable us to estimate the current service provision costs for child and adolescent cancer services.

We are interested to know the **level of investment in child and/or adolescent cancer for the financial year 2002/03** at your centre or unit. We realise that this s not a straightforward exercise, particularly for adolescents, many of whom might be referred to adult services, but a best estimate would be helpful. For those centres that see a mixed age range, it would be helpful to have separate information for children under 15 years of age and those between 15 and 23 years of age. If this is not possible, then please could you give an approximate level of investment for children and young people under 23. **Please could you complete the enclosed form and return it preferably by email to** p.linck@bangor.ac.uk**, or a hard copy to the above address by 17th October 2003 .**

All information that is collected during the course of the research will be kept strictly confidential and will not be used for any other purpose. All data will be held securely.

We would be very grateful for your assistance in this important research. If you have any queries or comments then please do not hesitate to contact me.

Yours faithfully
Pat Linck
Research Officer
Email p.linck@bangor.ac.uk

Centre for the Economics of Health
Institute of Medical and Social Care Research
Wheldon Building
University of Wales Bangor
Gwynedd LL57 2UW

Tel: Pat Linck 01248 382397

FINANCIAL QUESTIONS FOR THE GUIDANCE ON CANCER SERVICES: IMPROVING OUTCOMES IN CHILD AND ADOLESCENT CANCER

Name of the UKCCSG Centre: .....

Or Teenage Cancer Trust Unit: .....

Please would you give a contact name and email address for further financial information.

Name: .....

Email: .....

Level of investment in child and/or adolescent cancer for the financial year 2002/3

	Children aged 0-14 years	**Young people, aged 15-23 years**	**If separation is not possible, 0- 23 years**
Staffing			
Drugs			
Radiotherapy			
Other NHS			
Estimate of non-NHS revenue funding from charity/voluntary sector			
Total If a breakdown of NHS costs is not possible please give a best overall estimate			

Thank you very much for you assistance. If you have any queries or comments then please do not hesitate to contact me.
Pat Linck, Research Officer Phone 01248 382397
Email p.linck@bangor.ac.uk

Appendix B

Tables from Section 3 – current estimates of employment costs at principal treatment centres based on needs assessment

Table 3.1 Annual cost of employing all staff according to professional groups employed at UKCCSG/TCTs, showing all 18 responses of Needs Assessment[7]

Centres	AHPs (£)	Clinicians (£)	InPatient Nurses (£)	Day/OP nursing posts (£)	Other nurse posts (£)	Palliative Care (£)	Total staff (£)	Staff cost by finance directors (£)
1	92,466	1,339,577	392,107		119,781	190,972	2,015,122	
2		2,014,941	1,076,250	242,886	527,658	278,909	3,612,987	3,084,504
3	518,582	1,707,306	488,610	61,099	178,463	188,731	2,964,327	1,364,700
4	1,478,475	11,374,543	1,201,075	325,369	199,407	308,619	14,688,081	
5	954,801	1,698,322	735,189		170,477		3,388,312	1,179,000
6	112,107	1,426,786	647,154	185,390	142,345	214,587	2,586,025	
7	750,608	2,119,516	709,274	165,857	353,583	271,518	4,016,772	
8	137,092	1,318,865	733,961	220,077	285,156	363,324	2,773,319	
9	95,766	766,619	439,639	197,972	161,471	128,929	1,628,926	
10	93,331	1,446,438	420,466	157,267	97,591	102,289	2,219,792	
11	388,883	1,011,308	1,094,255	227,796	385,775	267,211	2,989,452	
12	61,241	1,891,874	524,496	141,392	98,538	108,677	2,727,679	
13	136,465	2,295,303	769,847	146,249	277,610	353,397	3,701,260	3,076,020
14	259,477	1,387,100	606,666	205,148	279,587	371,001	2,829,392	
15	191,464	921,303	557,855	144,283	189,754	175,158	1,990,063	
16	249,656	1,117,835	547,704	32,498	64,996	81,646	2,029,339	1,084,000
17	209,569	507,038	951,571	28,601	453,500	448,582	2,145,362	
18		154,923	410,657		68,989	66,600	632,180	
Total	**5,729,984**	**34,499,596**	**12,306,774**	**2,481,888**	**4,054,682**	**3,920,151**	**58,938,392**	
Mean	**791,124**	**1,916,644**	**683,710**	**165,459**	**225,260**	**217,786**	**3,274,355**	
S.D.	**387,106**	**2,425,740**	**250,738**	**80,280**	**134,318**	**123,427**	**2,964,432**	

* NB Centres Nos. 4 and 18 included for information here, excluded from further analysis

Table 3.2 Annual cost of employing all AHPs employed at UKCCSG/TCTs, showing all 18 responses of Needs Assessment[7]

Centre	Physios* (£)	Occupational Therapist (£)	Clinical Psychologist (£)	Speech Therapists (£)	Dietician (£)	Play Therapist (£)	Adolescent Support Worker (£)	Radiographer (£)	Data Manager (£)	Research Nurses (£)	Total (£)
1	31,493	144	21	1,102	15,747	18,764			25,196		92,466
2											
3	13,287		190,131	66,147	21,591	43,156		164,724	19,545		518,582
4	27,105	39,228	88,970	141,114	37,792	100,260		944,796	66,712	32,498	1,478,475
5	16,609					58,167		827,115	52,911		954,801
6	7,873		5,219		15,747	38,674		3,149	25,196	16,249	112,107
7	18,896		298,534		34,643	37,527		149,464	142,386	69,158	750,608
8					15,747	67,549			25,196	28,601	137,092
9					31,493	18,764			30,235	15,275	95,766
10			15,657		12,597	37,527			13,249	14,301	93,331
11	35,747		36,534		86,366	72,093	19,337		36,106	102,700	388,883
12					25,195	24,444	11,602				61,241
13			10,438		12,597	37,527			45,352	30,550	136,465
14	12,597		78,287			57,102			50,391	61,099	259,477
15	47,240		26,096		15,747	18,764			37,793	45,825	191,464
16	33,217	29,530	130,478		29,530	21,862			5,039		249,656
17			52,191		62,986	75,054	19,337				209,569
18											
TOTAL	**244,064**	**68,902**	**932,556**	**208,364**	**417,777**	**727,235**	**50,277**	**2,089,248**	**575,307**	**416,255**	**5,729,984**
MEAN	**24,406**	**34,379**	**77,713**	**103,631**	**29,841**	**45,452**	**16,759**	**417,850**	**41,093**	**41,626**	**832,750**
S.D.	**12,529**	**20,352**	**89,945**	**70,065**	**21,234**	**24,152**	**4,466**	**433,950**	**33,526**	**28,569**	**390,526**

* Where grade not given mid point of most commonly stated grade/s used
NB Centres Nos. 4 and 18 included for information here, excluded from further analysis

Table 3.3 Annual cost of employing all medical staff employed at UKCCSG/TCTs, showing all 18 responses of Needs Assessment[7]

Centre	Paediatric oncologists (£)	Paediatric Haematologists (£)	Paediatric Surgeons (£)	Paediatric Anaesthetists (£)	Associate Specialists (£)	Staff Grades (£)	Specialist Registrars (£)	SHOs (£)	Total Clinicians (£)
1	161,880	95,224	380,894	571,342		52,387	41,971	35,879	1,339,577
2	418,984	380,894	761,789		64,024		209,853	179,397	2,014,941
3	342,805	76,179	352,327	666,565	-	-	125,912	143,518	1,707,306
4	295,193	228,537	6,456,160	3,746,096	268,261	104,774	167,882	107,638	11,374,543
5	228,537	171,402	190,447	952,236	-	-	83,941	71,759	1,698,322
6	276,148	95,224	285,671	571,342	-	78,581	83,941	35,879	1,426,786
7	380,894	190,447	476,118	952,236	-	-	83,941	35,879	2,119,516
8	209,492	-	380,894	476,118	-	52,387	92,335	107,638	1,318,865
9	285,671	142,835	95,224	38,089	64,024	20,955	83,941	35,879	766,619
10	161,880	95,224	476,118	571,342	64,024	-	41,971	35,879	1,446,438
11	238,059	190,447	190,447	28,567	-	52,387	167,882	143,518	1,011,308
12	95,224	47,612	1,047,460	571,342	-	52,387	41,971	35,879	1,891,874
13	190,447	95,224	57,134	1,714,025	64,024	-	120,630	53,819	2,295,303
14	333,283	95,224	190,447	476,118	-	52,387	167,882	71,759	1,387,100
15	285,671	285,671	-	-	64,024	52,387	125,912	107,638	921,303
16	114,268	-	285,671	476,118	128,048	-	41,971	71,759	1,117,835
17	190,447	-	95,224	-	-	-	41,971	179,397	507,038
18	66,657	-	-	-	-	52,387	-	35,879	154,923
Total	**4,275,540**	**2,190,143**	**11,722,025**	**11,811,535**	**716,431**	**571,020**	**1,723,906**	**1,488,995**	**34,499,596**
Mean	**450,057**	**292,019**	**1,379,062**	**1,574,871**	**179,108**	**103,822**	**191,545**	**156,736**	**3,631,536**
S.D.	**98,390**	**92,829**	**1,547,745**	**930,336**	**76,954**	**21,593**	**53,358**	**51,164**	**2,425,740**

NB Centres Nos. 4 and 18 included for information here, excluded from further analysis

Table 3.4 Annual cost of employing all palliative care staff according to professional groups employed at UKCCSG/TCTs, showing all 18 responses of Needs Assessment[7]

Centre	Macmillan Nurses (£)	CLIC * (£)	Other Pall Nurses (£)	Social Workers (£)	Clinical Psychologist (£)	Consultants (£)	Total Pall Care Staff (£)
1	-	72,982	32,498	33,300	52,191	-	190,972
2	182,456	-		66,600	29,853	-	278,909
3	72,982	-	32,498	83,250	-	-	188,731
4		-	130,089	16,650		161,880	308,619
5	-					-	-
6	-	48,747	48,747	16,650	5,219	95,224	214,587
7	129,993			141,525	-	-	271,518
8	-	36,491	145,965	66,600	-	114,268	363,324
9	-	68,989		59,940	-	-	128,929
10	68,989	-		33,300	-	-	102,289
11	130,681	-		136,530		-	267,211
12	32,498	-			-	76,179	108,677
13	174,470	52,740	32,498	83,250	10,438	-	353,397
14	-	-	97,591	99,900	78,287	95,224	371,001
15	138,667	36,491			-	-	175,158
16	64,996	-		16,650	-		81,646
17	32,498	32,498		66,600	31,315	285,671	448,582
18		-		66,600	-	-	66,600
TOTAL	**1,028,231**	**348,940**	**519,886**	**987,345**	**207,304**	**828,445**	**3,920,151**
MEAN	**165,459**	**21,809**	**74,269**	**65,823**	**13,820**	**48,732**	**217,786**
S.D.	**66,972**	**27,511**	**49,474**	**39,556**	**23,898**	**80,792**	**123,427**

* Childrens Leukaemia in Childhood

NB Centres Nos. 4 and 18 included for information here, excluded from further analysis

Table 3.5 Annual staff costs calculations at UKCCSG/TCT Centres in England and Wales (excluding outliers), ordered by costs per bed

Centres	Facilities on site	Total staff cost per year	No. of new patients per year	Staff costs per new patient per year	No. of beds	Staff costs per bed per year
9	Radiotherapy, BMT	1,628,925.81	115	14,164.57	14	£116,351.84
11*	BMT	2,989,452.49	122	24,503.71	23	£129,976.20
15	BMT, neurosurgery	1,990,063.19	150	13,267.09	15	£132,670.88
6*	Radiotherapy, neurosurgery	2,586,025.27	110	23,509.32	17	£152,119.13
17	BMT	2,145,361.80	82.50	26,004.39	14	£153,240.13
8		2,773,319.42	75	36,977.59	15	£184,887.96
14	Radiotherapy, BMT	2,829,392.01	145	19,513.05	15	£188,626.13
13	BMT	3,701,260.26	102	36,286.87	19	£194,803.17
10	Neurosurgery	2,219,791.51	77.50	28,642.47	10	£221,979.15
16		2,029,339.22	106	19,144.71	9	£225,482.14
12	Neurosurgery, some BMT	2,727,679.42	72.50	37,623.16	12	£227,306.62
7	Radiotherapy, BMT, neurosurgery	4,016,772.44	108	37,192.34	17	£236,280.73
5	BMT, neurosurgery	3,388,312.25	60	56,471.87	14	£242,022.30
3	BMT, Retinoblastoma	2,964,327.22	95	31,203.44	12	£247,027.27
1		2,015,122.34	65	31,001.88	8	£251,890.29
2	BMT, neurosurgery, Retinoblastoma, specialist bone surgery	3,612,986.88	172	21,005.74	14	£258,070.49
	TOTAL	**43,618,132**	**1,658**	**456,512**	**228**	**3,162,734**
	MEAN	**2,726,133**	**104**	**28,532**	**14**	**197,671**
	S.D.	**699,076**	**32**	**10,943**	**4**	**47,922**

* These centres do not have separate oncology beds, all beds are shared with general paediatrics

Table 3.6 Annual costs of employing additional staff at 17 UKCCSG centres and associated TCTs

Staff member	Additional staff for a minimum of 1 WTE at each centre	Approximate salary + on-costs of previous column (£)	Additional staff for a minimum of 2 WTE at each centre	Approximate salary + on-costs of previous column (£)
AHPs and scientific staff				
Physiotherapists	9.05	285,013.42	13.50	425,158.15
Occupational therapist	15.00	472,397.94	24.00	755,836.70
Clinical psychologist	9.20	480,159.30	18.20	573,176.17
Speech therapists	14.00	617,375.19	18.50	582,624.13
Dietician	6.75	212,579.07	19.50	614,117.32
Play therapist	1.00	18,763.60	6.80	161,897.09
Adolescent support worker	14.40	278,455.68	31.40	607,187.95
Data manager	4.20	105,821.52	13.60	293,275.79
Clinical psychologist, palliative care	14.4	751,553.68	31.00	1,617,928.07
Social worker, palliative care	4.5	149,768.10	12.70	422,678.86
Nursing staff				
Research nurse	8.50	259,672.45	28.00	965,851.49
Paediatric outreach nurse*	-		2.00	68,989.39
Paediatric outreach nurse, palliative*	9.00	310,452.26	20.00	689,893.92
Nurse specialist/practitioner	6.20	213,867.12	16.20	558,814.08
Macmillan Nurse	6	218,947.20	16.00	583,859.20
Medical staff				
Paediatric oncologist	-		2.40	228,536.64
Paediatric haematologists	3.70	352,327.32	14.40	1,371,219.84
Paediatric surgeons	1.40	133,313.04	5.40	514,207.44
Paediatric anaesthetists	2.90	276,148.44	7.30	695,132.28
Paediatric palliative care consultant	13.20	1,256,951.52	29.30	2,790,051.48
Total (to nearest whole number)	**143**	**£6,393,567**	**330**	**£14,520,436**

* estimates given where staff shared between palliative and non-palliative care, salary estimate for nurses mid point G-H

Appendix C

Tables from Section 4 – estimates of the costs of age appropriate MDTs

Table 4.1 Annual staff employment costs of suggested core membership of MDTs at principal treatment centres

Core MDT for children or young people	Hourly Rate (£)	MDT duration 1 hour plus + 1 preparation (£)	MDT duration 2 hours plus + 1 preparation (£)
2.5 consultants	60	300	450
3 specialist nurses (Mid Pt. Gde G-H)	22	132	198
2 allied health professionals, (Mid Pt Senior 1-2)	40	160	240
3 of the 6 listed below (Mean hourly rate)	23	135	203
Specialist pharmacist (Mid pt Band 7-8) Dietician (Mid Pt Senior 1-2) Play specialist; activity coordinator/youth worker Psychologist mid Pt Gde A-B Teacher, Local Authority Scale Mid Pt M3-6 Social worker			
Clerical support + 3 hours preparation	14	56	70
Total salary cost centre per meeting		783	1,161
Annual salary costs of weekly meetings per centre		40,716	60,346
Late effects MDT			
Key worker (assume G or H Gde nurse)	22	44	66
Lead clinician (expertise in late effects)	60	120	180
Nurse Specialist	22	44	66
Endocrinologist	60	120	180
Psychological services professional	33	66	99
Appropriate AHP	20	40	60
Secretarial support + 3 hours preparation	14	56	70
Total salary cost per meeting per centre		490	721
Annual costs of weekly meetings per centre		25,480	37,492
Annual salary costs of both teams based at 1 principal treatment centre		66,196	97,838
Annual salary costs of both teams based at 17 paediatric principal treatment centre in ENGLAND AND WALES		**1,125,332**	**1,663,246**
Annual salary costs of both teams based at 8 TCT units in ENGLAND AND WALES		**529,568**	**782,704**

Overall Total Cost for 25 teams		**1,654,900**	**2,445,950**

Table 4.2 Annual staff employment costs of suggested core membership of MDTs at other treatment sites

Core MDT at other treatment sites	**Hourly Rate (£)**	**MDT, 1 hour plus + 1 preparation (£)**	**MDT, 2 hours plus + 1 preparation or 1 hr.mtg + travel (£)**	**2 hour mtg with 1 hour travel**
Lead clinician/oncologist/haematologist	60	120	180	240
Key worker (G/H Gde nurse)	22	44	66	88
Nurse Specialist	22	44	66	88
Specialist pharmacist	23	47	70	93
Ward nurse/community (Mid ptG-H)	22	44	66	88
Appropriate allied health professionals (x2), (Mid Pt Senior 1-2)	40	80	120	160
Social worker	21	42	63	85
Clerical support + 3 hours preparation	14	56	70	84
Total salary cost per meeting		**477**	**702**	**926**
Annual salary costs of weekly meetings		**24,804**	**36,478**	**48,152**
Annual cost in ENGLAND AND WALES (37 teams or 1 per network)		**917,748**	**1,349,686**	**1,781,624**
Annual cost in ENGLAND AND WALES (74 teams or 2 per network)		**1,835,496**	**2,699,372**	**3,563,248**

Appendix D

Training and education

Table 5.1: Post-registration course and CPD for clinicians.

Offered by	Course Title	Duration	Cost
Royal College of Paediatrics & Child Health	Paediatrics and Child Health.	2 years general professional training in paediatrics then 5 years in higher specialist training.	Approx £50,000[1]
Local NHS Trust	CPD for consultants	Ongoing	Approx £3,000 pa + leave.

1 From phone conversation Royal College of Paediatrics & Child Health

Table 5.2 Post-registration courses offered by Universities for nurses working with children and young people with cancer

1. CATS: Credit Accumulation and Transfer Scheme. This is a UK-wide scheme used across Further and Higher Education to signal how 'hard' and long courses are. 1 'CAT' is equivalent to a notional 10 hours of study so *60 CATS* indicate around 600 hours study.
2. Varies depending on choice of topics.

Education Provider	Course Title	Cost of BSc Degree	Cost per module
School of Nursing and Midwifery Studies, University of Wales College of Medicine	BSc (Hons) in Nursing Children and Young People with Cancer	£2,110	Module 1 20 CATS[1] £370 Module 2/3 30 CATS £530 Module 4 40 CATS £680
School of Nursing and Midwifery. University of Southampton	BSc (Hons) Clinical Practice or Healthcare Studies BSc (Hons) Child and Adolescent Cancer Care	From £5,346[2]	Type A 20 credits £ 593 Type B 20 credits £ 791 Type C 20 credits £1,190
Faculty of Health and Social Care. London South Bank University	BSc (Hons) Professional Practice: Childhood Cancer Nursing	£4350	Level 3 15 credits £450- £600
School of Nursing. The University of Nottingham	BSc (Hons) Health Care Studies, Child Health Care (Oncology)	£2,556	10 credits from £213

Table 5.3 Costs of training additional staff at 17 UKCCSG centres and associated TCTs

Staff member	Additional staff for a minimum of 1 WTE at each centre	Approximate annual training cost at 1 module per year (£)*	Additional staff for a minimum of 2 WTE at each centre	Approximate annual training cost at 1 module per year (£)*
AHPs and scientific staff				
Physiotherapists	9	7,240	14	10,800
Occupational therapist	15	12,000	24	19,200
Clinical psychologist	9	7,360	18	14,560
Speech therapists	14	11,200	19	14,800
Dietician	7	5,400	20	15,600
Play therapist*	1	800	7	5,440
Clinical psychologist, palliative care	14	11,520	31	24,800
Social worker, palliative care	5	3,600	13	10,160
Nursing staff				
Research nurse	9	6,800	28	22,400
Paediatric outreach nurse*			2	1,600
Paediatric outreach nurse, palliative*	9	7,200	20	16,000
Nurse specialist/practitioner	6	4,960	16	12,960
Macmillan Nurse	6	4,800	16	12,800
Total	**122**	**82,880**	**271**	**181,120**

* The cost given above for training play specialist is the also the average cost for training unqualified play specialists or additional modules under CPD for existing staff

Table 5.4 Costs of training additional consultants at 17 UKCCSG centres and associated TCTs

Consultants	Additional staff for a minimum of 1 WTE at each centre	Approximate annual CPD cost at 1 £3000 per year (£)*	Additional staff for a minimum of 2 WTE at each centre	Approximate annual CPD cost at 1 £3000 per year (£)*
Paediatric oncologist	0	0	2	7,200
Paediatric haematologists	4	11,100	14	43,200
Paediatric surgeons	1	4,200	5	16,200
Paediatric anaesthetists	3	8,700	7	21,900
Paediatric palliative care consultant	13	39,600	29	87,900
Total	**21**	**63,600**	**59**	**176,400**

* The calculation is based on the cost of CPD for consultants detailed in Table 5.1

Appendix E

Charities Supporting Cancer Research and Cancer Services for Children and Young People

Table 7.1 Total Expenditure for 2003 or closest preceding year by active charities established with a primary goal of supporting children and young people with cancer, as reported on the Charities Commission website.

	CHARITY	Area of Operation	FINANCIAL YEAR	Total Expenditure	DESCRIPTION
1	Anthony Clifford Trust Fund	England & Wales	06/02/01 to 05/02/02	30,663	Relieve poverty of families of children under 16 with leukaemia and advance education about cancer and leukaemia in children.
2	Camp Quality UK	England & Wales	1/01/03 to 31/12/03	54,203	Promotion of welfare of children and young people with illnesses, especially cancer, through appropriate holidays and activities.
3	Camp Simcha - Children With Cancer	England & Wales & outside	01/01/02 to 31/12/02	87,959	Relief of suffering of Jewish children with cancer or other life threatening illness
4	Cancer and Leukaemia in Childhood (CLIC)	England & Wales & beyond	01/01/03 to 31/12/03	7,040,000	Relief for children and young people with leukaemia or cancer and further research into problems arising from these diseases.
5	The Candlelighters Trust	West Yorkshire	01/03/03 to 29/02/04	822,295	Relief from suffering from a malignant disease or life threatening heamatological disorder.
6	Charlie's Challenge	Kent	01/07/02 to /30/06/03	57,836	Relief of children with brain tumours and support related research at the Maudsley Hospital and Guy's Hospital.
7	Childhood Cancer Organisation for Parents and Relatives (COPARS)	Cambridgeshire	01/04/02 to 31/03/03	6,454	Alleviate the hardship and distress of childhood cancer to patients, family and friends and provide education and support for the same.
8	Childhood Cancer Unit Parents' Association (Ccupa)	Southeast England	01/05/02 to 30/04/03	7,591	Provides advice, information and support to parents and educates the public and those working with children with cancer.
9	Childhood Eye Cancer Trust (CECT)	England & Wales	01/07/02 to 30/06/03	133,768	Relief for children with retinoblastoma through grants, services, research and information.
10	Children's Cancer Support Group (CHICs)	Merseyside	01/12/02 to 30/11/03	119,501	Relief of children with leukaemia and associated disease and support of their parents.
11	Children's Leukaemia Society	Wales	01/02/02 to 31/01/03	34,276	Relief of children under 16 suffering from leukaemia.
12	The Children's Leukaemia Trust	England & Wales	01/07/02 to 30/06/03	54,236	Provide facilities and staff for children with leukaemia undergoing bone marrow transplantation and support research in this area.
13	CHIN-UP	Northeast England and Cumbria	01/04/02 to 31/03/03	1,967	Fundraising for children's hospice for NE England
14	Chris Lucas Trust	England & Wales & beyond	01/04/02 to 31/03/03	2,128	Funding of research post into rhabdomyosarcoma at times and places deemed appropriate by the trustees and dissemination of results.
15	Christian Lewis Trust Cancer Care For Children	England & Wales	01/09/02 to 31/08/03	619,016	Relief of neuroblastoma and related childhood cancers through research and the comfort of sufferers.
16	The Claire Lemmon Fund	Dorset	01/04/02 to 31/03/03	15,038	Outings and holidays for children with cancer and leukaemia &

					families.
17	Claire Sadler Trust Fund	Dorset	01/04/03 to 31/03/04	9,510	Relieve teenagers and young adults suffering with cancer particularly by provision of holidays, trips and outings.
18	DJMF Child Cancer Concern	Essex, greater London	01/04/02 to 31/03/03	3,260	Relief for children and young persons primarily with non-Hodgkins lymphoma, research funding, holidays and relief of financial hardship.
19	Doctor John Owen Holiday Trust for Sick Children	Wales	01/08/02 to 31/07/03	2,028	Relief of children with cancer or leukaemia in particularly holiday accommodation for the benefit of these children and their families.
20	Edwards Trust	West Midlands	6/04/02 to 5/04/03	471,494	Support research, provide relief and advance education by increasing awareness of complementary approaches to childhood cancers.
21	The Foundation for Children with Leukaemia	England & Wales & beyond	01/01/03 to 31/12/03	9,674,837	Relief of children suffering from leukaemia and promotion of research into causes, treatment and cure for the disease.
22	Help Adolescents with Cancer (HAWC)	England & Wales	01/11/02 to 31/10/03	26,871	Funding counseling and social activities for adolescents with cancer and their families,and friends.
23	The Joshua Gilbert Rhabdomyosarcoma Appeal	Greater London	Financial statements not posted on Charities Commission's online Register.		Relief of children with rhabdomyosarcoma tumours and research into causes and treatment of these tumours.
24	The Katie Trust	England & Wales	01/04/01 to 31/03/02	22,278	Relief from financial hardship for child cancer victims and their families and to advance research into child cancer.
25	Laura Crane Trust	England & Wales & beyond	01/04/02 to 31/03/03	108,999	Relief of persons suffering from cancer primarily aged between 15-25 and particularly by the provision of funding for research.
26	Leucan	West Sussex	01/04/02 to 31 03/03	3,735	Relieve suffering of children and young people with leukaemia or cancer and their families including bereavement.
27	LIFT (Basildon and Thurrock)	Essex – Basildon	31/05/02 to 30/05/03	3,089	Relief of children with cancer or leukaemia undergoing treatment at Basildon Hospital.
28	Lisa Thaxter Trust	England & Wales	01/04/02 to 31/03/03	88,468	Relieve children suffering from cancer by provision of treatment facilities and financial assistance for research.
29	Llandough LATCH	Llandough	01/01/02 to 31/12/02	390,766	Relief of children suffering with cancer and leukaemia and their families.
30	The National Alliance of Childhood Cancer Parent Organisations (NACCPO)	England & Wales	01/04/02 to 31/03/03	1,261	Information and support service for children with cancer and their families, and raising public awareness about childhood cancer.
31	Neuroblastoma Research Fund	Merseyside	01/01/03 to 31/12/03	28,337	Relief of sickness and promotion and dissemination of research.
32	The Neuroblastoma Society	UK	01/01/03 to 31/12/03	147,104	The relief of children suffering from neuroblastoma
33	North of England Children's Cancer Research Fund	North of England	01/04/02 to 31/03/03	633,545	Promote and support research into causes and treatment of childhood cancer by funding research posts and projects at Newcastle University
34	Parents Association For Seriously Ill Children (PASIC)	Derby/Lincoln/ Nottingham hires	01/09/02 to 31/08/03	27,483	Relief of suffering of children with cancer and relief of financial needs among their parents and families.

35	The Rainbow Centre for Children	Gloucestershire, Kent, Somerset	01/09/01 to 31/08/02	169,050	Relieve suffering of cancer or other life-threatening illness patients, especially children, and their families by advice and counseling.
36	Royal Orthopaedic Bone Tumor Service (Rohbts)	Shropshire	01/06/02 to 31/05/03	7,354	Relief of sickness for children with bone tumours, provision of holiday facilities for them and families,,relief of poverty and promote research.
37	Samantha Dickson Research Trust	Hampshire	01/04/03 to 31/04/04	425,070	Research into causes of childhood brain tumours particularly gliomas and relief of patients suffereing from malignant gliomas.
38	Sargent Cancer Care For Children	England & Wales	01/04/02 to 31/03/03	6,992,145	Alleviate suffering of children and young people with cancer and provide care and assistance to them and their family.
39	Society Of Parents Of Children With Cancer (Spocc)	Shropshire, West Midlands	01/04/02 to 31/03/03	29,569	To promote the relief of children with cancer particularly by psychological and social support, travel, outings holidays.
40	Teenage Cancer Trust	England & Wales	01/07/02 to 30/06/03	1,338,618	Relief of cancer and related diseases in young people by provision of capital costs, research costs, funding nurses, seminars and conferences
41	Trent Regional Health Area Parents Association of Children with Tumours and Leukaemia (PACT)	Derbyshire, Lincolnshire, Nottinghamshire, South Yorkshire	01/04/02 to 31/03/03	182,605	Relief care and welfare of children with leukaemia and tumours particularly those who reside in the Trent Regional Health Authority area.
42	United Kingdom Children's Cancer Study Group (UKCCSG)	England & Wales & beyond	01/12/02 to 30/11/03	465,813	Promotion of research at 22 centres in UK funded by Ca Research UK
	TOTAL			**30,340,220**	

Table 7.2 Funding contributions in the UK, 2003 from national & local charities established specifically to support cancer services[1] for children and young people.

Charity	Staff costs	Capital Projects	Accommodation	Care Grants	Research	Education	Other	Total
National Charities								
Children with Leukamemia[2]	£60,000	£1,250,000	£783,350	£250,503	£3,563,930	2,091,545	£182,058	£8,181,386
Christian Lewis Trust cancer care for children[3]	£96,813							£229,212
CLIC[4]	£3,160,000	£1,580,000	£1,106,000	£632,000	£869,000		£553,000	£7,900,000
The Joshua Gilbert Rhabdomyosarcoma Appeal[5]					£52,000			£52,000
The Laura Crane Trust[6]								£57,143
The Lisa Thaxter Trust[7]		£1,000		£10,370	£28,804		£24,100	£64,274
The Neuroblastoma Society[8]								£131,450
Sargent Cancer Care for Children[9]	£2,000,000		£1,000,000					£3,000,000
TCT[10]	£95,000	£500,000					£66,666	£661,666
National Charities sub-total	*£5,411,813*	*£3,331,000*	*£2,889,350*	*£892,873*	*£4,513,734*	*£2,091,545*	*£825,824*	**£20,277,131**
Local Charities								
Candlelighters[11]	£121,000	£95,458	£10,000	£60,434	£442,046			£800,000
COPARS[12]								£6,454
The Katie Trust[13]								£45,203
LATCH[14]	£47,648	£113,187	£10,961	£109,119	£8,174		£69,223	£358,312
NECCR[15]					£300,000			£300,000
PASIC[16]								£30,000
Local Charities sub-total	*£168,648*	*£208,645*	*£20,961*	*£169,553*	*£750,220*	*£0*	*£69,223*	**£1,539,969**
TOTAL	*£5,580,461*	*£3,539,645*	*£2,910,311*	*£1,062,426*	*£5,263,954*	*£2,091,545*	*£895,047*	**£21,817,100**

1 Note that some of these charities do not confine their activities within England and Wales but rather operate throughout the UK.
2 From annual report to Dec 2003 at www.leukaemia.org/annualreport2003.pdf.
3 From Trustees Report and Financial Statement
4 From annual report to Dec 2003 at www.clic.org.uk/resources/documents/clic_annual_review.pdf
5 From Charity Update, 2003 at http://www.jg-rabdo.com/ Includes all of UK
6 From information at www.lauracranetrust.org/main.htm Based on average annual fundraising over 7 years used to fund research and support for patients.
7 From personal correspondence with Geoffery Thaxter 20/08/04. nb 'other' revenue expenditure was mostly information provision for families. Grants made to UKCCSG, Studentship,
8 From annual report to Dec 2003 at http://web.ukonline.co.uk/nsoc/finances2003.htm
9 From personal correspondence
10 From personal correspondence
11 From personal correspondence with Sally Amos 27/07/04.
12 From information at http://members.lycos.co.uk/copars
13 From information at www.katietrust.org
14 From personal correspondence with Denise Henderson 03/08/04
15 From telephone conversation to NECCR
16 From telephone conversation to PASIC

NATIONAL CHARITIES

Children with Leukaemia is the national charity dedicated to conquering childhood leukaemia. The four goals are to support research into the causes of childhood leukaemia and into more effective treatments with fewer side effects, assist in the development of six centres of excellence and support of patients and families. Support activities includes funding nursing posts, supporting other charities, participating in a programme of treatment for children from eastern Europe and providing hospital and holiday accommodation.

Cancer and Leukaemia in Childhood (CLIC) funds research, and supports patients and families. Its activities include coordinating teenage social support groups, providing hospital and holiday accommodation, giving care grants to families to cover bills and funding staff who work within the NHS setting. Currently,
across the UK, CLIC funds 37 nursing posts, 9 play specialists at Regional Oncology Centres, 9 doctors and have endowed funding for the Chair of Paediatric Oncology at Bristol Childrens Hospital,

Christian Lewis Trust Cancer Care for Children is a national charity supporting children with cancer and their families every day in every way. The charity's aims are to make a difference in the life of a child with cancer and their family by improving their quality of life and by providing emotional and practical support to their affected families during this enormously stressful period.

The Joshua Gilbert Rhabdomyosarcoma Appeal. Rhabdomyosarcoma is a rare type of cancer which predominantly occurs in children. It is highly aggressive and results in the death of about 30% of those children who develop this type of tumour. The appeal raises money to provide research fellowships for worthwhile research projects which seek a cure for this form of cancer.

The Laura Crane Trust funds research into cancers which occur among 13-25 year-olds. The trust also supports measures to improve the quality of life for teenage cancer patients, both during and in the aftermath of their illness such as funding a hospital-based social worker, a hospital-based activities coordinator and recreational equipment and contributing to the cost of a purpose built Teenage Cancer Trust Unit,

The Lisa Thaxter Trust funds research projects including one in Africa. It supports International Childhood Cancer Day and funds the publication which provides information, advice and support to the families of children and young people with cancer. It also supports an organisation for the siblings of children with cancer and funds holidays for them.

The Neuroblastoma Society The sole purpose of the Society is the relief of children suffering from neuroblastoma, and to achieve this it raises funds for medical research into improving both diagnosis and treatment of the disease. The Society also offers an opportunity for parents to give each other mutual help, support and comfort.

Sargent Cancer Care for Children funds 90 Sargent care professionals, who provide a range of services. These include social workers, family support workers, grants, residential accommodation for families, mentoring and youth programmes.
Sargent care professionals and care staff work in partnership with medical and nursing staff and become part of the multi disciplinary team.

Teenage Cancer Trust (TCT) builds, adapts and equips specialist units within the NHS setting to care for adolescents with cancer. The TCT will be expanding the current funding stream over the next 10 years. They forecast that £4m will be spent in their Centre's in Glasgow, Leeds, London and Cardiff, a further £2.6 million has been allocated to NHS posts, giving a total amount of £6.6 million. A further £11.1m is currently under negotiation, comprising, £40,000 per annum into a patient and family support pilot in London and Home Counties via UCLH, and the appointment of a Professor of Adolescent Cancer Medicine and team attached to one of the Schools of Medicine (yet to be determined) at £250,000 pa plus inflation for 10 years.

LOCAL CHARITIES

Candlelighters is a Yorkshire based charity at the Specialist Unit at St. James's University Hospital in Leeds. It is dedicated to promoting research into childhood cancer and supporting the 150 children and teenagers referred each year with cancer and their families. Candlelighters raises funds to improve the facilities for children being treated at the Unit in Leeds; to purchase the latest medical equipment for the ward, outpatients clinic and bone marrow transplant unit; toys, books, games and videos which make the children's time in hospital more pleasant; to encourage support groups for parents, siblings and teenagers and to provide holiday breaks.

Childhood Cancer Organisation for Parents and Relatives Support (COPARS) was established at Addenbrookes Hospital in Cambridge as a support group for the families of children with cancer on Ward C2, both during and after treatment. Volunteers visit families at the ward and there is an organised programme of events throughout the year providing an opportunity for COPARS families to meet and to have fun together.

The Katie Trust is focused in the North-East of England and Cumbria and aims to support research and patients and their families. It funds two PhD studentships at the Northern Institute of Cancer Research in Newcastle upon Tyne into cancers which affect children. It has provided financial assistance with transport and funeral costs for families in hardship, and has provided mobility aids for patients. The Trust has financially contributed towards a care worker's post, based in Newcastle upon Tyne and is planning to make a contribution towards funding for a specialist teenage years care worker to operate in the North-East later this year.

Llandough Aims to Treat Children with Cancer and Leukemia with Hope (LATCH) is a local charity based in Cardiff, providing support, maintenance and development of the oncology units at Llandough Hospital. It has assisted with the purchase of research equipment and funding for a research doctor as well as funding social workers and community nurses in the development of a community support team. The charity assists children and their families emotionally and practically with financial aid and with travel, clothing, holidays, accommodation for parents at Llandough Hospital.

North of England Children's Cancer Research Fund (NECCR) aims to raise money to continue research into causes and treatment of childhood cancers. The NECCR supports a team of doctors, scientists, statisticians and research nurses at Children's Cancer Unit in the University of Newcastle upon Tyne

The Parents Association for Seriously Ill Children (PASIC) is a registered charity, formed in 1977 for children who had been diagnosed with cancer. It is there to provide support to any families in that situation, especially in the East Midlands, but also nationally. It was felt that the best kind of support would come from parents who had been in a similar position with the help of consultants from the Queen's Medical Centre, Nottingham. It now supports more than 250 families with practical help, financial help or information and advice. It helps to pay additional bills (eg. heating, telephone, travel expenses) for families and any additional equipment costs; it organises social events for families, including siblings; it offers specialised bereavement support; special support for teenagers; it provides a network for parents to stay in touch and provide mutual support; also a regular newsletter keeps families updated.

Table 7.3 Summary of estimated expenditure for charities established primarily to support children and adolescent with cancer.

Number of Charities identified	41
Total Expenditure during 2003	£30,340,220
Range of Expenditure	£1,261-£9,674,837
Mean Expenditure	£740,005
Estimated % of total expenditure for charitable purpose	85%
Estimate of total expenditure for charitable purpose	£25,789,187
Estimate of total expenditure for charitable purpose ± 25%	£19,341,890-£32,236,484

Table 7.4 Active charities directing some support to cancer services for children and young people as reported on Charities Commission Website for 2003 or closest preceding year.

	CHARITY	Area of Operation	FINANCIAL YEAR	Total Expenditure	DESCRIPTION
1	Action for sick children	England & Wales	01/01/03 to 31/12/03	75,606	Promote the welfare of sick children at home and in hospitals and provision of information and liaison services.
2	The Adrian Pope Charitable Trust	Staffordshire	05/10/02 to 04/10/03	5,000	Relief of patients with brain tumours by grants research support.
3	Afiya Trust	England & Wales	01/04/02 to 31/03/03	348,975	Advance of health of persons from minority groups.
4	African Caribbean Leukaemia Trust	England & Wales	01/08/02 to 31/07/03	165,189	Relief and support of leukaemia sufferers, public education and promotion of research.
5	Aintree Hospitals Leukaemia Fund	Merseyside	01/01/03 to 31/12/03	20,936	Financing the work at the Haematology Department of the Aintree Hospital. Research and treatment of patients suffering from leukaemia.
6	Andrea's Gift	West-Yorkshire and Leeds	01/01/03 to 31/12/03	602	Promote research into causes and cures of paediatric and adult brain tumours and provide relief for patients and their families and carers.
7	Annette Fox Leukaemia Research Fund	West-Yorkshire and Bradford	01/04/02 to 31/03/03	90,013	Relief of sickness and promotion of research for leukaemia, lymphoma and other related haematological diseases.
8	Association for Children with life-threatening or terminal conditions and their families (ACT)	England & Wales	01/04/02 to 31/03/03	153,581	Information & education service, publish leaflets for families & campaign for palliative care.
9	The A -T Society	England & Wales & beyond	01/01/03 to 31/12/03	176,711	The relief of sickness among people suffering from Ataxia Telangiectasia.
10	Atkinson Morley's Hospital Neurosciences Research Foundation	Greater London	01/04/02 to 31/03/03	137,155	Undertake and promote research in neurosciences and publish results.
11	Birmingham Heartlands and Solihull NHS Trust (Teaching) - Leukaemia	West Midlands	01/04/02 to 31/03/03	2,787,000	A subsidiary fund ot the Birmingham Heartlands and Solihull NHS Trust (Teaching), for any charitable purposes relating to the NHS.
12	Birmingham Heartlands Leukaemia Support Group	Birmingham	01/04/03 to 31/03/04	41,135	Relief of suffering by provision of advice
13	Bob Champion Cancer Trust	England & Wales	01/07/02 to 30/06/03	334,773	Research into testicular cancer and other malignant disease including Hodgkins disease and non-Hodgkins lymphoma particularly children and young adults and patient care at the Royal Marsden Hospital.
14	Boston Cancer and Leukaemia Fund	Lincolnshire	01/04/02 to 31/03/03	568,429	A subsidiary fund of the United Lincolnshire Hospitals NHS Trust Charitable Fund for any charitable purposes relating to the NHS.
15	The Bournemouth Leukaemia Fund	Dorset, Hamphshire	01/01/03 to 31/12/03	35,033	Education and research into leukaemia and support for work at the Royal Victoria Hospital Boscombe, Bournemouth and other hospitals.
16	Brain & Spine Foundation	England & Wales	01/04/02 to 31/03/03	439,950	Research into neurological disease

17	Brain Tumour Foundation	Surrey, West Sussex	01/04/02 to 31/03/03	27,858	Public education about brain tumours and relief of sickness and distress of people suffering with brain tumours and their carers and families.
18	Brain Tumour Research Campaign (Way Ahead)	England & Wales & Europe	01/04/02 to 31/03/03	6,810	Promote research and its publication.
19	Brain Tumour UK	England & Wales	01/01/03 to 31/12/03	148,797	Promote research and provide advice, support and financial assistance to people with the disease and their family and carers.
20	The British Brain Tumour Association	England & Wales	06/04/02 to 05/04/03	288	Relieve sickness and distress of people with the disease and promote research and its dissemination and publication.
21	British Liver Trust	England & Wales	01/04/02 to 31/03/03	426,077	Research into disorders of the liver
22	British Thyroid Association	England & Wales & beyond	01/04/02 to 31/03/03	93,763	Provide advice, support, relief and treatment of people with thyroid disorders, and to support research.
23	Bromley Area Leukaemia Support Group	Greater London – Bromley	01/11/02 to 31/10/03	22,035	Relief of sickness and poverty of persons with leukaemia.
24	Cancer Black Care	Greater London	01/01/02 to 31/12/02	429,983	Relief of need among cancer patients from specified ethnic groups and education in causes and prevention of cancer.
25	The Cancer/Leukaemia Fund	Merseyside	01/04/02 to 31/03/03	165,400	Subsidiary of The St Helens and Knowsley Hospitals Charitable Fund for any charitable purpose relating to the NHS.
26	Cancer Research UK	England & Wales	01/04/03 to 31/03/04	301,318,327	Promote health by research into nature, cause, diagnosis, prevention, treatment and cure of all forms of cancer.
27	Carol's Smile	England & Wales	01/03/03 to 29/02/04	44,025	Relief of suffering from lymphoma, particularly Hodgkin's disease and promotion and dissemination of research into the same.
28	Changing Faces	England & Wales	01/04/03 to 31/03/04	698,397	Assist those with facial and other physical disfigurement by counselling and training and promote knowledge about disfigurement.
29	The Co-operative Clinical Cancer Therapy Trust Fund	England & Wales	01/04/02 to 31/03/03	680,751	Maintain co-operation between clinics by recording and disseminating results of clinical therapy and research into malignant disease.
30	Cornwall Leukaemia Trust	Cornwall-Carrick	01/01/03 to 31/12/03	8,836	Relief from leukaemia and related disorders by provision of buildings, equipment and facilities at the Royal Cornwall Hospital Treliske.
31	Cure Leukaemia	West-Midlands - Birmingham	01/11/01 to 31/10/03	881	Relief of persons suffering from leukaemia and undertaking research into causes, prevention and treatment.
32	Derbyshire Leukaemia Research Fund	Derbyshire	01/04/03 to 31/03/04	58,562	Provision of facilities for research into leukaemia and related diseases.
33	Donna's Dream House Charity	Cheshire, Cumbria, Lancashire, Nth Yorkshire Merseyside,	22/05/02 to 21/05/03	72,652	Relief for children with life-threatening illnesses by the provision of holiday accommodation for such children and their families
34	East Kent Blood Trust	Kent-Thanet-Margate	01/04/02 to 31/03/03	11,992	Promotion and dissemination of research into blood diseases including haemophilia, leukaemia and AIDS.

35	Elimination Of Leukaemia Fund	Kent, Surrey	01/07/02 to 30/06/03	452,067	Promote research into leukaemia, relieve suffering, hardship and distress of patients and their dependants, and support other charitable organisations with similar objectives.
36	The Exeter Leukaemia Fund	Devon	01/09/02 to 31/08/03	372,521	Relief of persons with leukaemia and related diseases in Devon.
37	The Friends of the Leukaemia and Lymphoma Unit (The General Infirmary at Leeds)	North-Yorkshire, West Yorkshire, Leeds	01/12/01 to 30/11/02	25,954	Relief of persons with leukaemia, lymphomas and related disorders.
38	Gordon McLeod Leukaemia Fund	Hertfordshire – Worcestershire	01/09/03 to 31/08/04	7,275	Research into and education of the public about leukaemia and related malignancies and relief of patients with these disorders.
39	Gwynedd Haematology and Cancer Relief Fund	Conwy and Gwynedd	01/04/02 to 31/03/03	10,304	Relief of patients with haematology disorders, promotion of research, training of staff, purchase of publications and equipment.
40	Gwynedd Haematology and Cancer Unit Fund	Conwy, Gwynedd, Anglesey	01/04/02 to 31/03/03	1,173,017	Subsidiary of North West Wales NHS Trust Charitable Fund for any charitable purposes relating to the NHS.
41	Haematology and Leukaemia Trust Fund General Charity	South Yorkshire	01/04/03 to 31/03/04	329,236	Subsidiary of Rotherham General Hospitals NHS Trust General Charity, for any charitable purposes relating to the hospital or NHS.
42	Haematology Care Group	West Bromwich	01/04/02 to 31/03/03	10,954	Welfare of people suffering from leukaemia, lymphoma and other blood disorders and their families.
43	Haematology Fund Charity	East Sussex – Kent	01/04/02 to 31.03/03	712,000	Subsidiary of Maidstone and Tunbridge Wells NHS Charitable Fund, for any charitable purposes relating to the hospital or NHS.
44	Horace Hayhurst Memorial Fund	England & Wales	17/02/03 to 16/02/04	17,623	Research into leukaemia and related diseases of the blood.
45	The James Orton Fund for Leukaemia Relief	West-Midlands – Birmingham	01/06/02 to 31/05/03	285	Relief of sickness and need for those with leukaemia or a related disease.
46	Jeremiahs Journey	Southwest England	01/07/02 to 30/06/03	36,497	Bereavement support for children.
47	The Jessie May Trust	Bath, NE Somerset & Bristol	01/04/02 to 31/03/03	248,589	Palliative care in the community for children and young persons with terminal diseases and education of health professionals.
48	Karen Morris Memorial Trust	UK and beyond	01/06/02 to 31/05/03	9,664	Relief of people with leukaemia and promotion of research
49	The Kay Kendall Leukaemia Fund	England & Wales	06/04/02 to 05/04/03	1,096,818	For research into and treatment of leukaemia.
50	The Kent Leukaemia and Cancer Equipment Fund	Kent - Maidstone	01/04/02 to 31/03/03	151,075	Relief of cancer and leukaemia patients at the Maidstone General and other Kent hospitals including provision of screening facilities.
51	The Laura Centre (COPE)	Leicester-Allexton	01/09/02 to 31/08/03	237,213	Intergenerational bereavement service.
52	The Lee Smith Research Foundation	Greater London	01/07/02 to 30/06/03	5,118	Support for medical research into treatment and cure of leukaemia.
53	The Leicester Haematology Research Fund	Leicestershire	01/10/02 to 30/09/03	5,299	Promotion of research and meeting the cost of additional care facilities not available under NHS for patients with leukaemia/related diseases.
54	Leuka 2000	England & Wales	01/01/03 to 31/12/03	205,434	Promote research and provide and maintain specialist units for treatment of leukaemia patients.
55	The Leukaemia and Lymphoma Research	Tyne and Wear	01/04/02 to 31/03/03	927,249	Subsidiary of City Hospitals Sunderland NHS Trust Charitable Funds

	Fund				for any charitable purposes relating to the NHS.
56	Leukaemia Busters	Hampshire	01/04/03 to 31/03/04	90,758	Equipping, staffing and maintenance of antibody unit at Southampton General Hospital and support research into leukaemia and lymphoma.
57	Leukaemia Care Society	England & Wales & beyond	01/04/03 to 31/03/04	487,671	Promote the welfare of people suffering from leukaemia and allied blood disorders and relieve the needs of patients and families.
58	Leukaemia Fund	Lancashire	01/04/03 to 31/03/04	801,000	Subsidiary of Pennine Acute Hospitals NHS General Charitable Fund, for any charitable purposes relating to the NHS.
59	Leukaemia Fund Charity	Greater London	01/04/02 to 31/03/03	25,358,000	Subsidiary of Greater Ormond Street Hospital Children's Charity.
60	Leukaemia Research Appeal for Wales	Wales	01/04/01 to 31/03/02	61584	Relief for persons with leukaemia resident in Wales and promotion of research at University Hospital of Wales.
61	Leukaemia Research Fund	England & Wales	01/04/02 to 31/03/03	21,717,474	To encourage, promote and assist research into leukaemia and its related conditions.
62	The Leukaemia Society (UK)	England & Wales	01/04/02 to 31/03/03	65,195	Relieve those with leukaemia and bone marrow diseases, educate the public, promote research and maintain a bone marrow donor data bank.
63	Leukaemia Survivors Research Charity	Greater London	01/04/02 to 31/03/03	25,358,000	Subsidiary of Greater Ormond Street Hospital Children's Charity.
64	The Leukaemia Unit Appeal for the Metropolitan Borough of Dudley	West-Midlands, Dudley	01/04/02 to 31/03/03	45,234	Relief of sickness by construction and maintenance of units in Dudley for treatment of residents with leukaemia and related diseases.
65	LINC The Leukaemia and Intensive Chemotherapy Fund	Gloucestershire, Herefordshire, Worcestershire	01/04/02 to 31/03/03	29,767	Relief of patients undergoing chemotherapy at Gloucestershire Oncology Centre, advancement of staff education and support research into leukaemia and related conditions.
66	Lymphoma and Leukaemia Fund (Wales)	England & Wales	01/03/02 to 28/02/03	52,776	Relief for persons with lymphoma or leukaemia resident in Wales particularly at Singleton Hospital, Swansea and promotion of research.
67	Macmillan Cancer Relief	England & Wales & beyond	01/01/03 to 31/12/03	82,231,000	Assist cancer sufferers by grants, further cancer education, grants to hospitals, hospices, nursing, convalescent or holiday homes, any other activities to lessen suffering and preserve health of patients & others
68	Marie Curie Cancer Care	England & Wales & beyond	01/04/01 to 31/03/03	72,077,000	To promote the welfare and relief of people with cancer and investigate the causes, distribution and treatment and to promote its cure.
69	The Mark Ridgwell Leukaemia Trust	England & Wales	06/04/02 to 05/04/03	89,043	Purchase of equipment and promotion of research into leukaemia.
70	Mary Obolensky Underwood Foundation for Leukaemia Research	England & Wales	01/10/02 to 30/09/03	8,455	Relief of patients, promotion of research and public education for leukaemia and other malignant diseases.
71	The Medical Academic Festival Orchestra and Choir	Greater London	01/11/02 to 31/10/03	4,643	Promote research into a range of diseases including leukaemia.
72	Musgrove Leukaemic Group Somerset	Somerset -Taunton	01/03/02 to 28/02/03	86,676	Relief for persons with leukaemia resident in Wales particularly at Musgrove Park Hospital, Taunton and promotion of research.
73	National Cancer Alliance	England & Wales & beyond	01/05/02 to 30/04/03	175,700	Public education on cancer and its treatment and the relief of sickness by developing services and resources.

74	North Tees and Hartlepool NHS Trust Leukaemia Fund	North Yorkshire Stockton-on-Tees	01/04/02 to 31/03/03	516,000	Subsidiary of North Tees and Hartlepool NHS Trust General Charitable Fund, for any charitable purposes relating to the NHS.
75	The Pat Broadbent Leukaemia Fund	Greater Manchester	01/04/03 to 31/03/04	38,693	Relief for people with leukaemia at Manchester Royal Infirmary by provision and maintenance of facilities, grants and research promotion.
76	The Paul Vander Molen Foundation	England & Wales	01/02/03 to 31/01/04	3,500	Relief for people with leukaemia, promotion of research, advance of public education and provision of recreational facilities.
77	Penelope Tanner Samaritan Charitable Fund	Buckinghamshire, Gloucestershire, Oxfordshire	01/04/02 to 31/03/03	10,549,225	Subsidiary of Oxford Radcliffe Hospitals Charitable Fund and other related charities. Specifically to support patients with leukaemia whilst at the hospital and at home.
78	Philip Simons Memorial Charitable Fund	Buckinghamshire, Gloucestershire, Oxfordshire	01/04/02 to 31/03/03	10,549,225	Subsidiary of Oxford Radcliffe Hospitals Charitable Fund and other related charities. One specific aim is to support research into leukaemia at the hospital.
79	The Pinderfields Leukaemia and Haematology Fund	West Yorkshire	01/04/02 to 31/03/03	2,208	Relief of people with cancer, leukaemia and other blood disorders particularly by provision of equipment at Pinderfields Hospital.
80	The Plymouth and District Leukaemia Fund	Cornwall, Plymouth	01/01/03 to 31/12/03	63,176	Relief for people with leukaemia in Plymouth and district by promotion of local research and provision of equipment and facilities.
81	The Rainbow Trust Children's Charity	England & Wales	01/07/02 to 30/06/03	2,349,860	Respite care for families with children with life threatening diseases or bereaved. Outreach teams to visit families and education of the public.
82	R B Gray Charitable Trust	England & Wales	06/04/02 to 05/04/03	11,052	Supporting charities established for relief of a range of illnesses including leukaemia and charities conducting research into these.
83	The Richard Thomas Leukaemia Fund	Hertfordshire	01/08/02 to 31/07/03	19,130	Support work at Byrd Ward of Northwick Park Hospital including staff training and purchase of equipment facilities and services.
84	Royal Hallamshire Hospital Leukaemia and Myeloma Research Trust	South Yorkshire – Sheffield	01/04/02 to 31/03/03	22,039	To finance research into leukaemia and myeloma at the Royal Hallamshire Hospital Sheffield and disseminate findings.
85	Royal Liverpool Children's Charitable Fund	Merseyside	01/04/03 to 31/03/04	694,508	Comprised of 40 subsidiary funds including the Alder Hey Leukaemia Research Fund.
86	Royal Liverpool University Hospital Andria Butler Fund	Lancashire - Merseyside	01/04/02 to 31/03/03	1,328,260	Subsidiary of Royal Liverpool & Broadgreen University Hospitals NHS Trust Charitable Funds for any charitable purposes relating to the NHS.
87	Royal Liverpool University Hospital Leukaemia Fund	Lancashire - Merseyside	01/04/02 to 31/03/03	1,328,260	Subsidiary of Royal Liverpool & Broadgreen University Hospitals NHS Trust Charitable Funds for any charitable purposes relating to the NHS.
88	Simon Cohen Memorial Trust	England & Wales	01/08/03 to 31/07/04	350	Relief of sickness and advancement of education about leukaemia.
89	The Spencer Bourn Foundation for Leukaemia Research	Herefordshire	01/10/02 to 30/09/03	1,424	Promotion of research into leukaemia at the County Hospital, Hereford.
90	Stoke Mandeville Hospital Carl Todd Oncology Charity	Bedfordshire, Buckinghamshire, Hertfordshire.	01/04/02 to 31/03/03	844,000	Subsidiary of Stoke Mandeville Hospital Charitable Fund for any charitable purposes relating to the hospital or NHS

91	Stuart Martin Memorial Fund	Greater London, Kent, Dartford	26/07/02 to 25/07/03	50	Relief of sickness particularly amongst children with leukaemia at GOSH and other hospitals and other local charitable purposes.
92	The Sue Harris Bone Marrow Trust	England & Wales	01/08/01 to 31/07/02	33,909	Promote research into leukaemia and disseminate results, relief especially for those of the Jewish faith, relief of poverty.
93	The Tracy Sollis Leukaemia Trust	England & Wales	01/01/03 to 31/12/03	23,988	Prevention and relief of leukaemia primarily through research.
94	T.R.E.B.L.L.E Trust Fund Charity	Berkshire, Buckinghamshire	01/04/03 to 31/03/04	380,806	Subsidiary of Heatherwood and Wexham Park Hospitals Trust General Fund for any charitable purposes relating to the NHS.
95	The Tyneside Leukaemia Research Association	Cumbria, Tyne and Wear, Newcastle upon Tyne	01/01/01 to 31/12/03	1,113,018	Encourage and support research into causes and treatment of leukaemia. To raise funds and co-operate with organisations with similar objectives.
96	Umuada Ngwa Charitable Trust	England & Wales & beyond	01/09/02 to 31/08/03	1,392	Relief of need, sickness and distress in children in Africa, relief of sickness for children with leukaemia in the UK and promotion of African languages and culture in the UK.
97	United Kingdom Brain Tumour Society	England & Wales	01/01/03 to 31/12/03	148,797	Research into cause and cure, advice, support and financial assistance.
98	The University of Newcastle upon Tyne Development Trust – Jeffcock Medical Research Fund	Tyne & Wear - Newcastle upon Tyne	01/04/02 to 31/03/03	1,803,554	Subsidiary of The University of Newcastle upon Tyne Development Trust Charities. Specifically to fund research into cancer and leukaemia through research fellowships and studentships.
99	Wessex cancer trust	Wessex	6/04/02 to 05/04/03	904,764	Relief from cancer, promote research and educate public.
100	West Yorkshire Forget Me Not Trust	West Yorkshire	1/01/03 to 31/12/03	17,684	Support of children & their families in West Yorkshire with extraordinary medical needs.
101	The Williams Haematology Charitable Fund	Greater London	01/04/02 to 31/03/03	296,800	Subsidiary of The Lewisham Hospital NHS Trust Charitable Fund specifically for research into and staff training about leukaemia.
102	World Cancer Research Fund UK	England & Wales & beyond	01/10/02 to 30/09/03	8,238,934	To promote, fund, support and encourage research as to the causes and treatment of cancer for the general benefit of the public.
103	Yeovil Hospital NHS Leukaemia Research Trust	Dorset – Somerset	01/04/02 to 31/03/03	341,000	Subsidiary of East Somerset NHS Trust General Charitable Fund specifically for research into leukaemia.
104	Young minds	England & Wales	01/04/02 to 31/03/03	1,274,412	Education about and relief of children and families suffering psychological or emotional disturbance.
	TOTAL			**£587,269,708**	

Table 7.5 Charities established to support cancer patients in general, those with a specific form of cancer, diseases relating to a specific body organ or charities supporting illness in childhood that distribute a portion of their funding to children and adolescents with cancer.

Cancer Charities	**Year**	**Expenditure to Children**	**Total Expenditure**	**% to canc**
Cancer research UK	01/04/03 to 1/03/04	9,000,000	301,318,327	
Macmillan	01/01/03 to 1/12/03	245,000	82,231,000	
Non-cancer Charities	**Year**	**Expenditure to Children**	**Total Expenditure**	**% to canc**
Changing Faces	01/04/03 to 1/03/04	156,846	576,205	0.3 %

Cancer Research UK supports and undertakes a comprehensive programme of research in institutes, hospitals, universities and medical schools throughout Britain and Northern Ireland. The research portfolio targets all aspects of cancer and covers work in four broad subject areas: the biology and causes of cancer, developing cancer treatments, cancer prevention and improving quality of life.

Macmillan Cancer Relief is a UK charity that works to improve the quality of life for people living with cancer by funding specialist Macmillan nurses, doctors and other health professionals to deliver care, building cancer care centres, giving financial support to those in need and providing a range of information and support services, including a telephone helpline, publications and local cancer information centres and assisting local self help and support groups.

Changing Faces has a primary focus on disfigurements affecting the face, but their work also has relevance to disfigurements of other parts of the body. The charity addresses disfigurements of any origin, whether present at birth or acquired later in life. The charity supports and represents people with disfigurements by working to build their self-confidence and self-belief, ensuring they receive effective health care and rehabilitation and working to increase public awareness and knowledge about disfigurement.

Table 7.6 Summary of estimated expenditure by other (non-hospice) charities which support children and adolescent with cancer.

Number of charities sampled
Total expenditure for sampled charities during 2003
Range of Expenditure for sampled charities
Average Expenditure for sampled charities

Average Expenditure for charities with total expenditure < £20,000,000	
Estimated % of total expenditure to children and adolescent with cancer	
Estimated total number of charities	
Estimate of total expenditure for charitable purpose	
Estimate of total expenditure for charitable purpose ± 25% + £9,000,000 from Cancer Research UK	

Table 7.7 Results of survey of annual charitable contributions to costs and estimate of percentage of oncology patients at hospices for children and adolescents.

Hospice	Feature[1]	2002-03	2003-04	
Hospice a	Multiple sites	£2,199,000	£2,672,666	
Hospice b		£ 580,000	£ 580,000	
Hospice c		£2,778,000	£2,798,000	
Hospice d	Multiple sites	£1,894,346	Unavailable	
Hospice e	Community care	£ 342,000	Unavailable	
Hospice f		£1,056,834	£ 872,509	
Hospice g		£2,820,000	£2,820,000	
Hospice h		£1,515,200	£1,704,600	
Hospice i		£1,250,000	£1,250,000	
Hospice j		£ 637,000	£ 952,000	

1 Assumed to be a single organisation administering a single hospice unless otherwise stated.