Comparison of the expressed experiences of survivors of childhood medulloblastoma with measures of health and quality of life, and with issues identified in consultations

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FINAL REPORT
July 2015

Acknowledgement

The project team wishes to acknowledge the families, practitioners and managers who contributed to the success of the project, and the Christie Hospital Paediatric Oncology Charitable Fund.

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Section 1 Context of the Evaluation

INTRODUCTION

When the study was planned, the Young Oncology Unit at The Christie housed both the adult and paediatric follow-up clinics for patients who have been diagnosed with a tumour of the central nervous system (CNS) in childhood and adolescence or young adult life. Over the past 10 years, the paediatric clinic had developed a true multi-disciplinary team function with input from paediatric oncology, clinical oncology, teenage and young adult oncology, neurosurgery, social work, physiotherapy and clinical psychology. In contrast, the adult clinic (for follow-up of patients diagnosed in childhood and also in late teenage or early adult life) had solely medical input. The clinic for the follow-up of adult survivors of childhood CNS tumours at The Christie had significantly less multidisciplinary team involvement than its paediatric counterpart, and it was postulated that adult patients' needs might not be addressed as fully by the current provision.

This study was designed to develop a process to capture information systematically about patients' problems in the domains of medical, physical, psychological and social wellbeing in order to compare the extent to which these were recognised and supported in the context of the adult and paediatric clinics. Ultimately, this information was expected to inform planning for improvements in survivorship support in both clinics.

BACKGROUND

Children who are diagnosed with a CNS tumour have long been known to face many survivorship challenges that include problems relating to neurological functioning, epilepsy, vision, hearing, short stature, endocrinopathy and psychosocial functioning (Anderson et al, 2001). These findings have been mirrored by our own large audit of the outcomes for 143 patients who have survived a diagnosis of CNS tumour in the contemporary period of 1990-2001 (Panesar et al, 2008).

The survivors of the childhood CNS tumour medulloblastoma, for whom whole-brain radiotherapy is a mandatory part of treatment, serve as an exemplar population for the survivorship challenges that are faced by children diagnosed with a CNS tumour. For children with medulloblastoma, the cognitive sequelae of therapy are well-documented, with problems of attention, memory and intellectual functioning (Mulhern et al, 2004) all leading to poorer educational attainment, adverse employment prospects and peer relationships (Maddrey et al, 2005), an inactive lifestyle (Ness et al, 2009) and poorer health-related quality of life (Bull et al, 2007). Thus, for the health professional in the survivorship clinic, there are a large number of potential variables to address, and the traditional focus on evaluating the medical sequelae encountered may not address all of a patient's needs optimally.

As an example of this principle, we have conducted a large-scale research project of the utility of psychological interventions in relation to the school interface, with benefit for many of the children involved in terms of their educational support and health-related quality of life (Talbot et al, 2008). This psychology input is an integral part of the children CNS tumour follow-up clinic at The Christie, but it is not mirrored in the adult clinic. Therefore, the functionality of the survivorship clinic may be improved if there is a process to capture systematically and prioritise the various domains of the medical, physical, psychological and social functioning that patients experience. This project was intended to be the first step in this process.

In the face of the enormous volume and complexity of information that defines survivorship of childhood CNS tumours such as medulloblastoma, information technology can play a key role in the collation of medical information, identification of patient needs and subsequently, the development of rational care pathways. In the context of this proposal, a huge volume of clinical data from the patient cohort's previous consultations is stored electronically in a semi-structured textual format, as electronic patient records (EPRs) and discharge summaries. This data is amenable to automatic extraction by software through text mining[1]. In addition to identifying relevant information from the EPR and presenting it in a concise form, text mining can aid in making decisions and predictions (Yang et al, 2009). The main advantages of text mining in this context are that extraction and interpretation of data can be done efficiently, objectively and systematically from documents that are scanned from the patient records. Text mining also has the potential to discover knowledge that may not be directly present in any individual document, but which can be deduced or inferred from the whole or part of a collection.

Information technology will be a key to rational care pathway development in the face of the enormous volume and complexity of information that defines survivorship in the context of childhood CNS tumours such as medulloblastoma. EPRs are often complex in terms of linguistic expressions used and are non-uniform, non-homogenous and un-structured (Rao et al, 2006). Moreover, patient data can be highly redundant, incomplete, inconsistent, contradictory and sometimes incorrect. In addition, patient data is not static, it changes over time and thus temporal inferences need to be used as well. Text mining has been used to automate identification for clinical trials (Rao et al, 2006), therapy selection for individual patients, quality assurance and support in emergency units (Zhang et al, 2007). Semantic-based text mining has also been used to retrieve automatically the most similar cases to the current patient for patient management (Zhang et al, 2007), clinical research (Chen et al, 2006) and prediction of a disease status (Yang et al, 2009). When extracted into a structured form, it can be used to perform data analytics and mining. Text mining of clinical data has achieved a degree of success, but the best results have been achieved when research is focussed towards achieving specific sub-tasks within the overall text mining framework (Lewin et al, 2008).

RESEARCH AIMS

1. To structure a process to capture systematically and prioritise the various domains of the medical, psychological and social functioning which patients with medulloblastoma experience.
2. To elicit the narrative experience of patients who have been diagnosed with medulloblastoma in childhood, and that of their carers, with particular emphasis on key transition points and family health, and also social, psychological and educational functioning.
3. To extract, via text mining, the explicit and intuitive information in the patient hospital records and family narratives that relate to the patients' medical and psychosocial functioning as defined above.
4. To identify discrepancies in the themes of health and outcome as identified from family narratives and the hospital records as a pilot step towards the development of an informatics-based system that can inform more effective follow-up strategies.[2]

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[1] Text mining involves the pre-processing of document collections (text categorization, information extraction, term extraction), the storage of the intermediate representations, the techniques to analyze these intermediate representations (such as distribution analysis, clustering, trend analysis, and association rules), and visualization of the results (Feldman R. and Sanger J., 978-0-521-83657-9 - The Text Mining Handbook: Advanced Approaches in Analyzing Unstructured Data, Cambridge University Press, 2007).

[2] It was envisaged that the patients' narratives would identify additional factors that were not identified from either the paediatric or the adult clinic records, so additional analysis was planned. It was hoped that this supplementary knowledge would inform service developments for both clinic settings.

Section 2 Study Design

OVERVIEW

Three sources of data were accessed: measurement of health status, quality of life and psychological well-being; interviews with patients; and documentary data from patients' medical records. Eligible patients attending the clinics were approached by the oncologists and then directed to speak to the researcher (Professor Long) to discuss participation and to arrange a home visit for data collection. Interviews were recorded and transcribed. Copies of the relevant parts of corresponding medical records were anonymised and processed for text mining together with the interview transcript by Dr Dehgan, Dr Nenadic and Professor Keane.

SAMPLE

Twenty-seven children and adults aged between 7 and 40 years of age, who were undergoing follow-up at the Christie Hospital adult or paediatric clinic, and who were at least two years following the diagnosis of a medulloblastoma diagnosed before the age of 16 years were identified from the North West Children's Cancer Registry. Seven children less than 18 years were recruited from the paediatric clinic, and 14 patients aged 18-40 years were recruited from the adult follow-up clinic. Of these two populations, purposive sampling was employed such that all seven patients from the paediatric clinic were in the 11-16 age range and seven of the older patients were in the 18-24 age range. This was to ensure that patients were included at the time of key transition points (leaving school and starting employment). The remaining patients were between 26 and 38 years of age. One patient agreed initially but withdrew later as a result of life challenges, and five more patients were lost to the study either as no response to the request to participate (n=3) or living too far away (eg 180 miles) (n=2) and not returning to clinic within the timespan of the study. This usually resulted from a cancelled clinic appointment. The total sample was 21 patients. Patients and their carers were invited to participate in the study by means of a letter jointly from Dr Estlin, Dr McCabe & Dr Gattamaneni or invited directly during clinic visits.

INSTRUMENTS AND MEASUREMENT

For consideration of the survivorship needs of medulloblastoma patients, a raft of instruments was adopted to ascertain health status, health-related quality of life, and psychological wellbeing as per the recommendations of the CCLG CNS Tumour Quality of Survival Group. Health status and health-related quality of life are often measured by the same instrument.

Health Status and Health-Related Quality of Life (HRQL)

HUI2 and HUI3

The Health Utilities Index (HUI®) is a family of generic preference-based systems for measuring comprehensive health status and health-related quality of life (HRQL). HUI provides descriptive evidence on multiple dimensions of health status, a score for each dimension of health, and a HRQL score for overall health. Health dimensions include vision, hearing, speech, ambulation/mobility, pain, dexterity, self-care, emotion and cognition. Each dimension has 3-6 levels. The Health Utilities Index (HUI) evolved in response to the need for a standardized system to measure health status and HRQL to describe the experience of patients undergoing therapy; long-term outcomes associated with disease or therapy; the efficacy, effectiveness and efficiency of healthcare interventions; and the health status of general populations.

HUI currently consists of two systems, HUI2 and HUI3, which together describe almost 1,000,000 unique health states. Each includes a generic, comprehensive health status classification (a profile) system and a generic HRQL utility scoring system (Furlong et al, 2001). For most applications, HUI3 should be specified as the measure for primary analyses. It has the more detailed descriptive system of the two systems, full structural independence, and population norms available. HUI2 also offers distinct, independent attributes including self-care, emotion that focuses on worry and anxiety, and fertility. The two systems are independent but complementary, adding valuable information and with the HUI2 providing an efficient source of data for secondary sensitivity analyses.

The HRQL scoring systems provide utility (preference) scores on a generic scale where dead=0.00 and perfect health=1.00. The health status classification and HRQL scoring systems are generic in terms of applying to all people aged 5 years and older in both clinical and general populations but have been found to be acceptable and useful in UK studies of children and young people with CNS tumours (Glaser et al, 1999) in conjunction with the Pediatric Quality of Life Inventory (PedsQL).

PedsQL

The 23-item PedsQL™ Generic Core Scales were designed to measure the core dimensions of health as delineated by the World Health Organization, as well as role (school) functioning. The 4 Multidimensional Scales are: Physical Functioning (8 items), Emotional Functioning (5 items) Social Functioning (5 items), and School Functioning (5 items). These then contribute to 3 summary scores: Total Scale Score (23 items), Physical Health Summary Score (8 items) Psychosocial Health Summary Score (15 items)

A Brain Tumour Specific Version had also been developed (Palmer et al, 2007) along with a complementary measure of family stress and function (Family Impact Module). The PedsQL™ Family Impact Module (Varni et al, 2004) was designed to measure the impact of paediatric chronic health conditions on parents and the family. The PedsQL™ Family Impact Module measures parent self-reported physical, emotional, social, and cognitive functioning, communication, and worry. The module also measures parent-reported family daily activities and family relationships.

EORTC

For adult patients the EORTC have developed the QLQ-C30 survey instrument of HRQL for cancer patients that covers five functional scales (physical, role, cognitive, emotional and social) and three symptom scales (Fatigue, pain, nausea/vomiting) and serves as a global HRQL scale also (Aaronson et al, 1993). This scale has also been validated in long-term cancer patients (Apolone et al, 1998).

Emotional Wellbeing and Distress

HADS

The Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith, 1983) measures depression and anxiety in adults within and outside hospital and community settings. It is based on an easy-to-use questionnaire which allows the researcher to establish the presence and severity of both anxiety and depression simultaneously, whilst giving a separate source for each. It offers cut-off points to indicate whether someone is 'within the normal range', or in a 'mildly', 'moderately' or 'severely' disordered state. HADS contains two scales that provide an estimate of irritable mood disorder and help to identify particular areas of anxious concern.

PI-ED

The Paediatric Inventory of Emotional Distress (PI-ED) (O'Connor et al, 2010) is based on the HADS and can be used with a child population in a range of clinical and educational settings. The scale asks children and young people about their symptoms of anxiety and depression. A concrete cut-off score then identifies children most in need of further clinical assessment and intervention. Like HADS, PI-ED is a valid, reliable and clinically useful screening measure. It remains the only UK-standardised measure of emotional distress that is psychometrically robust in a large non-clinical and clinical sample across the 8-16 age range.

INTERVIEWS

The narrative experiences of the patient and their carers (one or both of the parents or guardians as decided by the family) were captured in focused interviews to ensure that the discussion addressed the common problems of neurological sequelae, endocrine problems, visual issues, hearing problems, psychosocial areas of memory and attention, school performance, educational experience and attainment, independent living, employment and impact on carer health and well-being. Families were asked to describe the issues that had been important to them at different times, and whether any needs that they had identified were met through the follow-up clinic over time.

Some families preferred to be interviewed as a family unit. This required particular skills in order to elicit the individual perspectives of all participants, but the research team had used this approach in a number of studies, including two evaluations of services for families in Blackpool (Livesley et al 2008, Ravey et al 2008). All interviews were conducted at the patient's home or in a private space close to the clinic, and the digital recordings were transcribed professionally. The transcripts were then anonymised and submitted for analysis by text mining techniques. Separate qualitative content analysis (Elo & Kyngas, 2008) was undertaken of the interview material for lessons to be learned by the research and clinical teams.

DATA EXTRACTION AND TEXT MINING FROM MEDICAL RECORDS

A huge volume of relevant clinical and medical data is stored in a semi-structured textual format, such as patient electronic records and discharge summaries. The main aim of text mining is to identify relevant information in such texts and to present it in a concise form so that it can be used to support understanding of documents as well as making decisions and predictions (Yang et al, 2009; Spasic et al. 2010). This was undertaken with the clinical information that can be gained from the transcribed family narratives and the Royal Manchester Children's Hospital and Christie hospital records in relation to the known medical, psychological and social-educational outcomes for the patient population. The main advantage was that extraction and interpretation of data could be completed efficiently, objectively and systematically from documents that were scanned from the patient records.

Text mining also aims to discover knowledge that may not be directly present in any individual document, but which can be deduced or inferred from the whole or part of a collection. It is therefore essential to identify medical terminology in documents. Terminology is typically prone to many variations and ambiguity, so term classification (such as patient, carer, symptom, drug), identification and mapping to medical controlled vocabularies (for example, UMLS, IDC-9-CM, IDC-10) was undertaken. The research team had used a similar approach in previous clinical text mining projects to extract and predict information about patients from their hospital discharge summaries (Yang et al, 2009; Spasic et al. 2010).

All selected data was anonymised to remove any reference to personal information as informed by the *Data Protection Act 1998*. The specific types of personal information removed from the data included *Names* such as personal names of doctors, patients and their relatives; *ID* numbers such as patient identification or reference numbers; *Dates* specifically, date of birth of patients; *Location* information such as patients' home and school addresses; *Contact* information such as patients' telephone numbers; and *Institution* information such as school and hospital names.

Subsequent to the data extraction and anonymisation process, text mining methods were developed and validated to extract mentions of clinical events automatically, including medical problems (eg: mentions of signs or symptoms, disease or syndrome), treatments (eg: mention of therapeutic or preventive procedures, medication), and tests (eg: diagnostic procedure). Specifically, state-of-the-art data-driven (Conditional Random Fields) Named Entity Recognition (NER) methods were developed to this aim (Kovacevic et al, 2013;

Dehghan, 2015). Additionally, a knowledge-driven (dictionary and rules) method was developed and validated to extract mentions of health-related quality of life (HrQoL) concepts (Dehghan, 2015). For comparison between hospital records and interviews, descriptive statistics and visualisation techniques (bubble and pie charts) were used to analyse any similarities and differences.

ETHICAL REVIEW AND MONITORING

The study proposal was reviewed by a National Health Service Research Ethics Committee (reference number 11/NW/0662) and by the University of Salford Research, Innovation and Academic Engagement Ethical Approval Panel (reference HSCR 12/05). Research governance review and monitoring was conducted by The Christie NHS Foundation Trust under study ID 11-DOG10-04.

Section 3 Findings: Demographics

SEX OF PARTICIPANTS

No attempt was made to recruit equal numbers of male and female patients, though purposive sampling would have addressed a noticeable imbalance in the sample had it become evident. However, there were more males than females in the younger transition group, and more females than males in the adult transition group (Table 1).

Table 1: Sex of participants by age group at recruitment

	Male	Female
11-16 years group	5	2
18-24 years group	2	5
25-40 years group	3	4
TOTAL	10	11

AGE OF PARTICIPANTS

The age at diagnosis and the age at recruitment was recorded in order to capture the variance in length of time as a patient. The youngest age at diagnosis was six months, and the oldest age at diagnosis was 11 years. At recruitment, the participants ranged from 11 to 39 years (Figure 1).

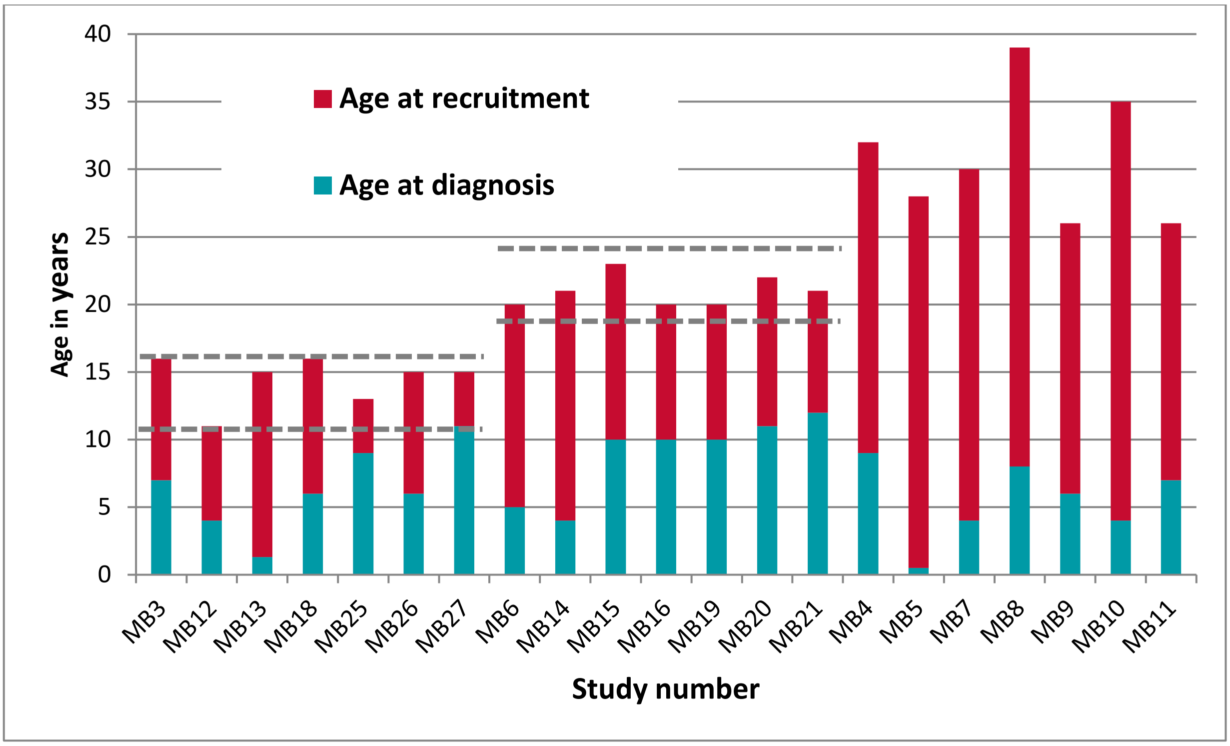


Figure 1: Age at diagnosis and at recruitment (with transition age bands)

Of those who were lost to the study, one declined to participate after initial agreement (young transition group, aged 15); two lived too far away for a home visit and had no clinic appointment for a further year (one in the older group, aged 38; and one in the adult transition group, aged 22); and the remaining three could not be contacted or did not respond (one in young transition group, aged 16; and two in the older group, aged 28 and 37).

Section 4: Health Status, Quality of Life and Psychological Well-Being

HEALTH STATUS

Overall health and quality of life

HUI3 focuses particularly on full structural independence, while HUI2 offers insight into attributes of self-care and emotion (including worry and anxiety). The score normally ranges from 0.00 (indicating "dead") to 1.00 (indicating perfect health). Negative scores up to -0.3 (HUI2) or -0.36 (HUI3) indicate a state of health "worse than dead". The responses from one participant in the 11-16 group indicated "perfect health", although other problems were identified which were not assessed by HUI2/3. For all other participants in this group, the results showed severe impairment of health. For respondents in the 18-40 years group the responses were only moderately better (Figures 2 and 3). There was one negative HUI2 score of -0.03 (Figure 2).

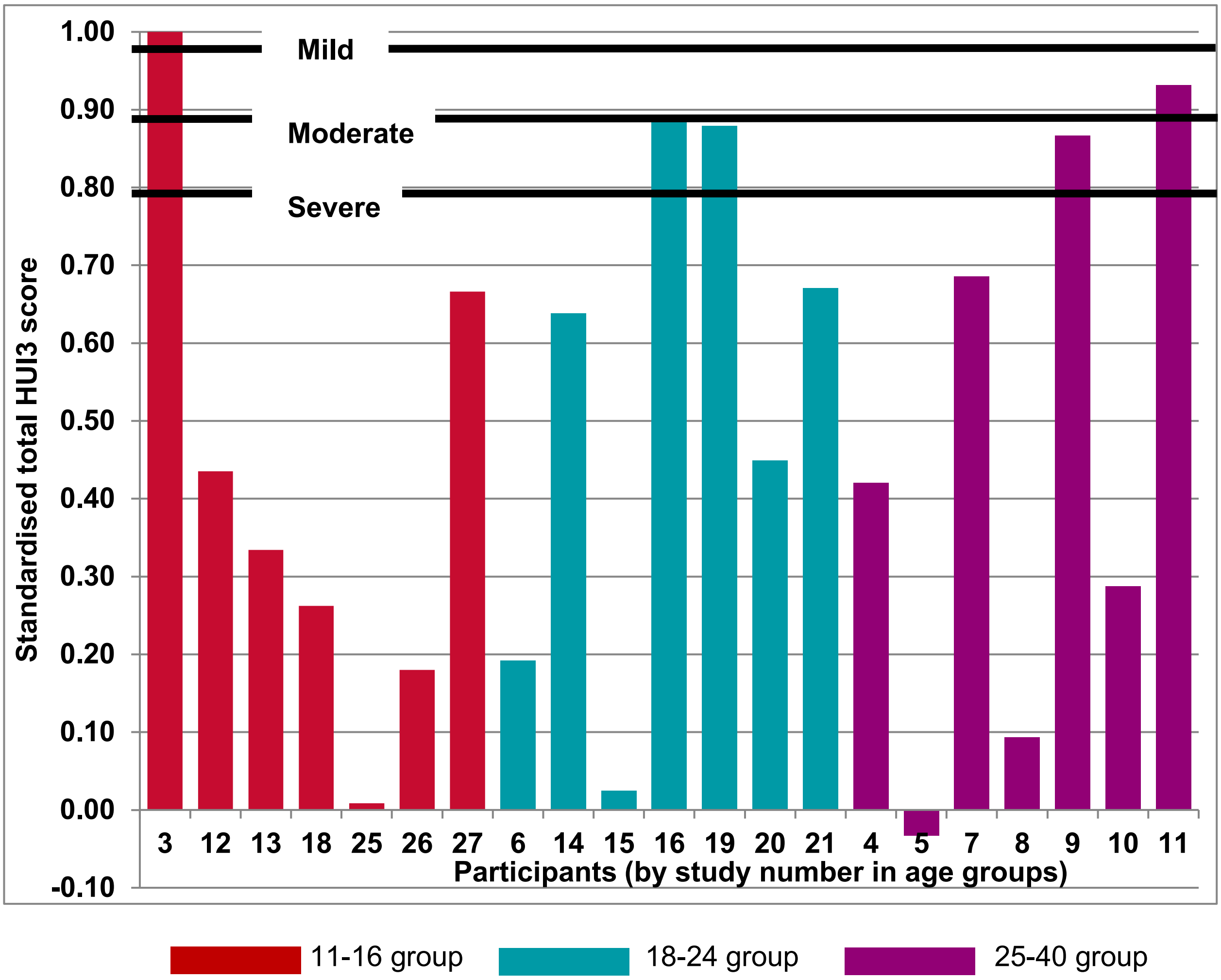


Figure 2: HUI3 total scores

Despite these scores showing seriously impaired health for most participants, most patients reported overall good or very good health. With one exception (MB25), all in the 11-16 group indicated good or very good health. All patients in the adult transition group expressed their health to be good or very good, with one declaring health to be excellent (MB 16). In the 25-40 group, two participants claimed only fair health (MB4, MB8), with the other reporting good or very good health (Figure 4). With the exception of the participant who reported no problem at all, all of the 11-16 group reported severe problems with cognitive ability.

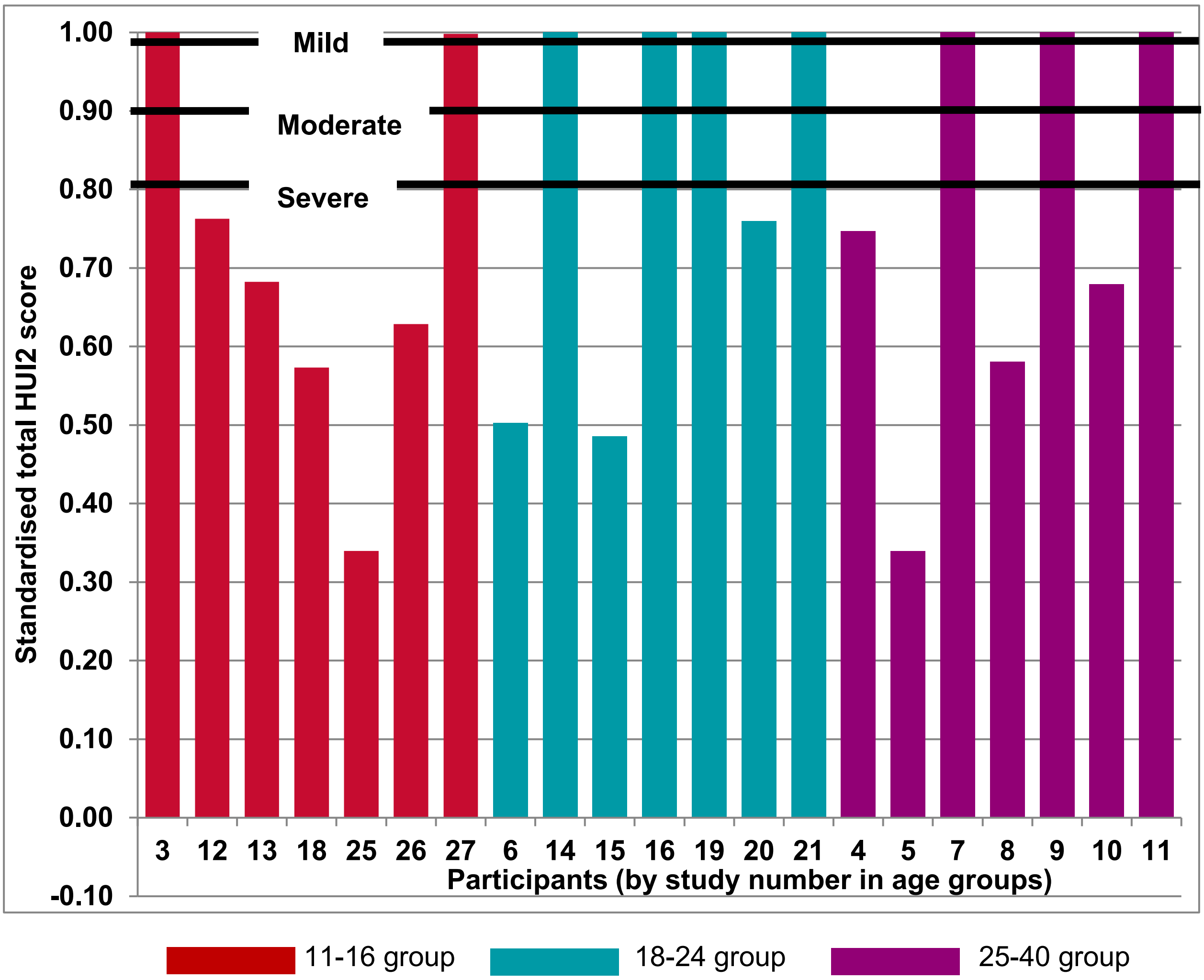


Figure 3: HUI2 total scores

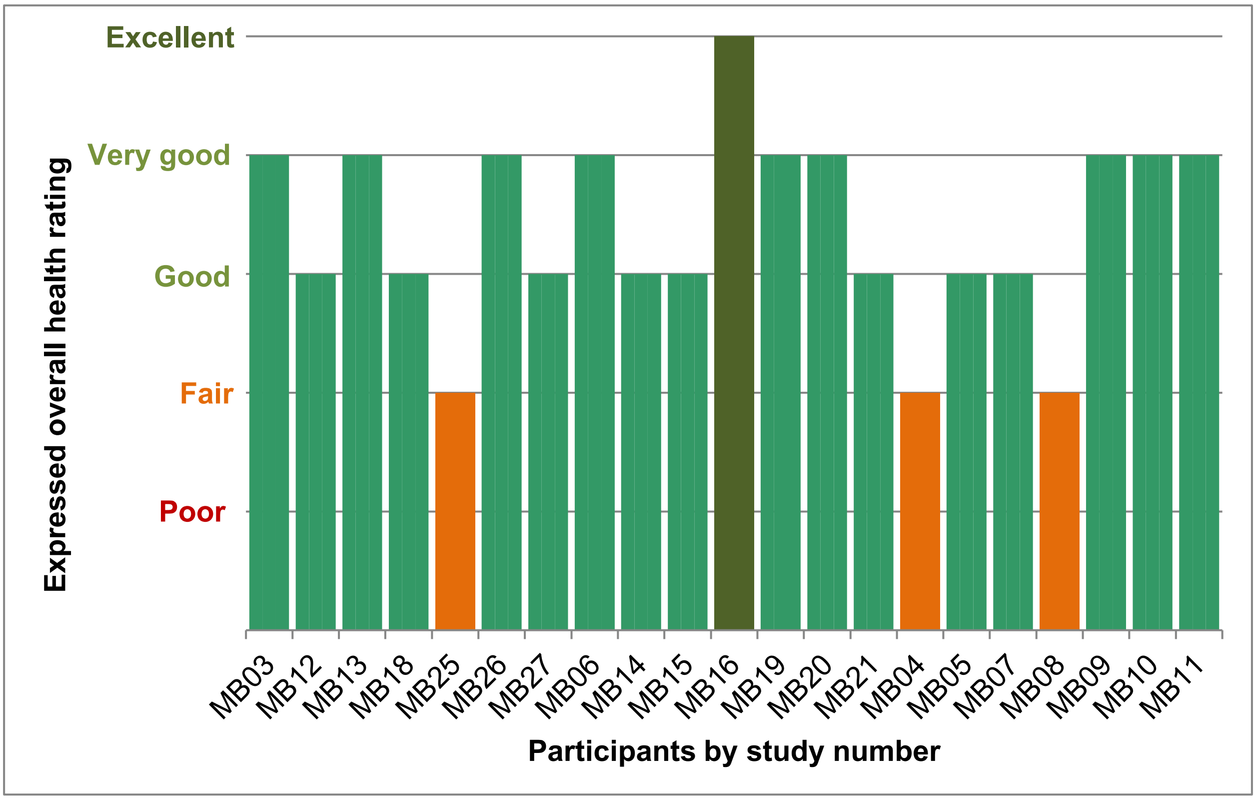


Figure 4: Expressed overall health rating by participants

QUALITY OF LIFE

PedsQL Measures For Quality Of Life In The Under-18 Group

Three instruments from the PedsQL range were used to indicate quality of life in this group. Scores are transformed to a 0-100 scale, with higher scores indicating better quality of life. There is no cut-off point, but in this study scores of less than 66 were taken to indicate reduced quality of life, and scores of 50 or less were taken to indicate seriously impaired quality of life. Diminished quality of life was indicated for most patients in most areas. Major negative impacts on families were also revealed. Table 2 details the summary scores for the three instruments.

Table 2: PedsQL transformed scores (0-100) for Core, Brain Tumour Module and Family Impact Module (scores in red indicate seriously impaired quality of life)

	PedsQL 4.0 Generic Core Scale			PedsQL Brain Tumour module		PedsQL Family Impact Module		
	Physical Health	Psychosocial Health	Total	Cognitive Functioning	Pain & Hurt	Parental HRQL	Family Functioning	Total
MB3	94	93	93	93	100	84	94	80
MB12	25	58	47	14	50	70	53	60
MB13	72	37	49	21	58	60	34	47
MB18	38	52	47	43	42	70	38	57
MB25	9	35	26	21	25	41	47	44
MB26	75	55	62	18	75	50	53	54
MB27	53	50	50	61	50	36	59	40

PedsQL 4.0 Generic Core Scale

A ***Total Quality Of Life*** score is calculated from all of the items, and two aspects of the core scale are also derived: physical function and psychosocial functioning. Five of the seven patients showed a score of 50 or less, with a sixth patient scoring only 62. The two contributing sub-scales also showed problems with quality of life. The ***Physical Health*** summary score showed seriously impaired quality of life for four of the seven patients (the remainder showing reasonably good scores). In contrast, all but one of the patients reported seriously impaired ***Psychosocial Health***. This is arrived at from the Emotional, Social, and School Functioning Scales.

PedsQL Brain Tumour Module

No summary score is calculated for this module, so six dimensions of quality of life are reported: *Cognitive functioning, Pain and hurt, Movement and balance, Procedural anxiety, Nausea,* and *Worry*. Deficits were common in all sub-scales other than nausea, but two subscales showed dramatic results. For *Cognitive functioning,* one patient had excellent quality of life, another had reduced quality of life, and the remaining five showed particularly seriously impaired quality of life. This was consistent with the interview narratives. Although two patients reported *Pain and hurt* as never being a problem, all of the others reported seriously impaired quality of life because of pain.

PedsQL Family Impact module

A *Total score* and two subsidiary scores are calculated: ***Parent HRQL Summary Score***, and ***Family Functioning Summary Score***. Other than the same patient who reported having no problems, the six remaining families reported reduced (n=3) or seriously impaired (n=3) quality of life because of the impact on the family. The *Parent HRQL Summary Score* contributed to this, with one family reported reduced quality of life and three reporting seriously impaired quality of life due to parental health-related quality of life. The *Family Functioning Summary Score* exerted a more substantial impact, with three families each reporting reduced or seriously impaired quality of life as a result of interrupted family functioning.

EORTC QLQ-C30

This adult patient scale, incorporating functional scales, symptom scales, and single items, offers a global health status/QoL score – standardised to a range of 0-100, a higher score indicating higher quality of life. Interpretation of scores remains problematic. There is no pre-defined cut-off for poor QoL, but King (1996) provides helpful guidance from a review of studies in which this instrument was used. She concluded that a mean (standardised) score of 66 was a high mean, while 50 was considered to be a low mean. In this study, the mean standardised GHS was 73.21 – a relatively high mean score. However, two patients in the 25-40 group had scores considerably lower than a "low mean" score: MB4 at 33.33, and MB8 at 41.67. These related directly to declarations of only "fair" overall health status in the HUI2/3 responses and moderately severe or severe health impairment in HUIs and HUI3. Two other patients were borderline at 58.33, also reporting severe or moderately severe impairment in health status in HUI2 and HUI3, but offering an expressed overall quality of life as "good" (Table 3).

Table 3: Comparison of HUI2/3 scores with QLQ-C30 Global Health Score for four patients

	HUI3 standardised score	**HUI2 standardised score**	**HUI2/3 Overall rating of health**	**QLQ-C30 Global Health Status**
MB14	Severe	Moderate	Good	58.33
MB4	Severe	Moderate	Fair	33.33
MB5	Severe	Severe	Good	58.33
MB8	Severe	Severe	Fair	41.67

EORTC QLQ-BN20

This short instrument which complements the QLQ-C30 is designed to establish quality of life deficits for adult patients specifically with a brain tumour. While individual patients indicated a variety of problem areas, two symptom scales showed more widespread deficit. The *Motor dysfunction scale* demonstrated that five of the fourteen patients were troubled by associated physical disability either "quite a bit" or "very much", while the same number of patients reported diminished quality of life because of a *Communication deficit*.

PSYCHOLOGICAL WELL-BEING

The Paediatric Index of Emotional Distress (PI-ED), developed from the Hospital Anxiety and Depression Scale (HADS), is a standardised measure of emotional distress. Individual scores above gender-specific cut-off values indicate the likelihood of clinically significant distress. These cut-off values are 10 for boys and 11 for girls. HADS produces separate outcomes for anxiety and for depression (without differentiation between sexes), interpreted as normal (0-7), mild (8-10), moderate (11-14), and severe (15-21). Moderate and severe are taken to indicate clinically significant concern. Tables 4 and 5 illustrate the outcomes for each instrument.

Although the two scales are not directly comparable, and certainly not statistically, comparison of the number of cases above and below the cut-off points for each indicates a much greater problem in the young transition group. The narrative data explained to some extent that patients came to accept their (increasing) health problems and reducing quality of life, but the lack of connection to consequent depression was remarkable. However, during data collection, a number of patients expressed that the instrument did not really capture the nature of the emotional distress that they were experiencing as time went by.

Table 4: Outcomes of measurement of emotional distress by PI-ED. Actual scores shown for all seven participants. Cut-off indicated by red line.

PI-ED	
Boys (Actual scores)	Girls (Actual scores)
	26
	21
17	
14	
9	
7	
0	

Table 5: Frequency of outcomes of measurement of emotional distress by HADS. Number of cases in each category shown. Cut-off shown by red line

HADS		
Grade	Anxiety (Number of cases)	Depression (Number of cases)
Severe	0	0
Moderate	4	0
Mild	2	1
None	6	13

Section 5: Messages From Narratives

PROBLEMS AND SUPPORT DURING PARTICIPATION

All but six participants required or opted for support from a parent (n=13), carer (n=1), or sibling (n=1) during completion of the questionnaires and the interview. Remarkably, in most cases patients were allowed to make their own responses without prompting despite considerable delay and difficulty. A blend of determined independence and ready recognition of the need for help was displayed. This mirrored findings of Livesley and Long (2014) regarding the participation of children in research. Indeed, several participants likened their situation to being like a child again in some ways and still needing their parents.

Of the six who were interviewed alone, three required some degree of assistance from the researcher, being either unable to read the materials or to write responses. Transcription was difficult in seven cases due to the speech problems experienced by the participants. The interviews lasted from 23 minutes to 45 minutes, and they were conducted after completion of the questionnaires. A number of Issues were identified from the interviews.

COMMON ISSUES FROM THE UNDER-18 YEARS GROUP

Unsurprisingly, given the age group under consideration, problems and concerns about school predominated in this age group. Other common issues related to recognition of, acceptance of, and resistance to, disability; denial and aspiration; the discovery of supportive services; and perceptions of quality of life and thoughts about the future.

School

Schools were often unprepared for the return of pupils who had missed whole years of study and who recommenced with significant cognitive and physical problems.

> *Getting to school was difficult because the school and all that was a massive debate or stuff. Because school didn't know. They hadn't done this before, so they'd no idea what they were supposed to do, how they should tackle it. So they were learning just as much as we were. (MB27)*

Over-compensation by school staff, while well-meant, could be counter-productive.

> *I think probably mollycoddled a little bit to the extent I think he got over-reliant on school staff as in dinner ladies, and he was sort of hanging on their coat tails at lunchtimes and break times. And I think from every slightest little thing they were on the phone or "can you come and get him, can you come…". (Father MB26)*
>
> *They were quite soft with him at times. The slightest little thing, like if he's coming down with a cold and he'll start in school, they'll phone up: "He doesn't feel too good, can you come and get him". And they wouldn't do that with anyone else. So that affects how he achieves at school then. (Mother MB27)*

Once patients appear externally well, recognition of needs ceased.

> *They were really good when he looked ill, but I suppose it was harder once he had finished the treatment and then his hair grew back and his lines came out, and I suppose then… It's only now looking back and seeing what he got in high school you realise he didn't get it there, in primary school. (Mother MB3)*
>
> *The treatment finishes and then suddenly it was like, and that was a relief. She's done the treatment, it's great. She'd gone from looking like she does now to looking like this shell of a child, didn't you? With no hair, in a wheelchair and eyes all over the place and not hearing. And then suddenly, it's all over now. People look at it and*

> *say "well what's your problem". She has lost two years of school, and everybody just thinks that you're well now. (Mother MB25)*
>
> *Yes. But they can't see that there are still loads of problems, and I can't do the same things as them sometimes. (MB25)*

The change from primary school to secondary school was a time of particular challenges.

> *In primary school, she was just poorly. So everybody was nice and everybody came to visit you, and then it was cards and everything. Then you go to secondary school and some of your friends were the same and there's a lot of new friends. But you're still in and out, in and out a lot, and it's so hard, I think, for you to fit in. (Mother MB25)*
>
> *And I think it's because I'm off for quite a lot and, not necessarily just appointments but with tiredness or ill or something. (MB25)*
>
> *The psychologist has helped him through. But the support side that way isn't great in the transition into secondary school from primary. The psychologist was fundamental in saying what he needed and what he needed to do and such like. (Mother MB26)*

Even the move from "infant school" to "junior school" brought problems.

> *He missed a huge chunk of what you would call the basics… He was at that age that progressed into, like, the junior school side of it from the infant side and it was like he missed probably what's the most important transition period in his primary school. (Father MB26)*
>
> *I feel he missed two years of his life. Social skills as well. He's missed that socialising aspect of school. (Mother MB26)*

Achieving and maintaining recognition by teachers and school peers was a constant source of concern.

> *Obviously there was going to be learning difficulties, growing, you know, adapting to general life, but try and get that through to the school is a total different thing because they were still giving him the work for a normal age child. We started telling them that he is not a normal aged child. It is hard getting it through to them. It is still a battle now. I mean, they send Maths homework home. Well, we can't do it. What is the chance of him doing it? So we tell school this, you know. Send him home with maths that he is capable of doing, because we are doing it for him, so he is not learning anything. Then they are saying "well, you shouldn't be doing it for him". And We are trying to tell them, "you need to sort a plan out and put it in for him, what he can do". We have sent letters in. We have rung them up. We have told them at parents evenings that the work is too hard for him and what he is like when he gets so wound up, to the brink where you want to cry, don't you, with the frustration. But then, also shouting at his mum: he feels the guilt then, and the remorse. (Father MB13)*
>
> *We've been in and out of school. There are hiccups. There's been a classic example today, really. She can't really walk in a straight line still and yet the PE teachers think they know best. So she's been torn off a strip by a PE teacher today. I think they think she is the sort of kid that's got a bit of a headache or a sore ankle and can't be bothered. So they've torn a strip off her today. Her self-esteem is shot to pieces. (Mother MB25)*
>
> *We get the impression with the school system that the elite students are looked after and then you've got kids like him that have statements. They segregate them away. So we found in GCSE year that because they're so results-driven they try to convince you not to do GCSEs it is going to show as a fail on their stats, to go to do something of a lesser standard that will get you a qualification which is worthless pretty much. They say, "actually, you can do an entry level, which will get you this NVQ" or whatever. It is in nothingness, it's a pointless exam. (Father MB26)*

While the focus on care was appreciated, there was concern that this was achieved at the expense of educational attainment.

> *I don't think they did a lot for his education. Education-wise, he was put back in just as a natural progression as to where he was, and they sort of muddled along, if you will, and it was, like, "Oh he'll pick it up". What we think should have happened is, they should have put him back. (Father MB26)*
>
> *He'd missed all the basic spellings, days of week, months, just basics. And he'd missed joined-up writing and he's never caught up, so his handwriting's poor, his reading skills aren't as good as probably they should be for his age group, and spelling is poor. So we think they should have either given him extra or just said, "look you've missed too much, we need to put you back a year". (Mother MB26)*

On occasion, although there were frustrating problems at school, there was evidence that teachers tried to help.

> *She has problems reading, concentrating, remembering the lessons. Sometimes, the teacher would explain the lesson, and then she forgets all about it, and she has to go through it again. I mean this year - she still deals with it right now. (Mother MB18)*
>
> *Sometimes they used to shout at me for not understanding as well. (MB18)*
>
> *To be fair to the school, they've done a really good job with her. Instead of having one class a week of art they've given her two just to make her feel better about herself because she was really good at that. And she achieved a lot. She had lots of support from teachers. "Where shall I go and what shall I do?" They've done this with her through secondary school. (Mother MB18)*

Recognition, Acceptance and Resistance to Disability

Social issues and loss of self-esteem were common problems.

> *She's always conscious of how she looks, and I have to do some special, you know, hair… like puff her hair up and try to make it as beautiful as possible. Plus she's always at the mirror, and she says "Why me?" She's got a younger sister: "Oh, I wish my hair was like my sister's." (Mother MB18)*
>
> *I have like my own friend, but I can't sit down and chat. Not really. Like at lunchtime and that, I just go by myself. It's like if I don't trust… I don't know if he's going to be my friend, he's going to leave me to go with another one so I'm not quite sure. I'm worried he might not understand me and that. (MB18)*
>
> *She has difficulties even socialising with other teenagers. She feels like she doesn't know how to cooperate with people her age, or sometimes she doesn't understand what people are saying. Last week we've been to a party and all the girls were dressed up. She was dressed up and she went and sat for a bit with the other girls and then came back and sat by herself. She said "take me home, I don't like it." (Mother MB18)*

Friendships, normally a vital part of children's lives were often lost, partly because of time spent in hospital or being too unwell at home to socialise, and partly because of the inability to join in with peer activities.

> *I've got a few friends. And I lost quite a lot of them because of my brain tumour. I didn't go to the same school, and I didn't get the chance to make the new friends that you would in a new school. But I still talk to them. Just walking places. I tire quickly, I've not got great stamina so my legs just start giving out because they get tired. Where you've got people going over to another district, and I can't do that, and that limits me to where I'm allowed to go. Just physically can't do it. It's like being a kid again. Like you've got boundaries on where you're allowed to go. (MB27)*

> *You just don't get out as much as the rest because they all moved on. That whole first year, obviously they were making new friends, getting used to everything. He wasn't even there for the year, not at home nor nothing, he was just always in the hospital so he never saw them. They didn't realise when he did come home that he was actually there, so totally lost contact. (Mother MB27)*

There is a general tendency for overt disability to be exaggerated in society, while the impact of less striking problems might be under-estimated and left unresolved.

> *I think the way they get treated when they're ill is different because people see that they're ill. And then you look at him now and he's just a normal teenager isn't he? And it's a fine line because I don't want him to think he's disabled in any way, but he does have other problems. It's difficult. And people can just think he's just a bit clumsy maybe, a little bit unbalanced; but he's not in a wheelchair or anything. People treat you differently.(Mother MB3)*

Sometimes it was simply too much to bear if others adopted a questioning approach when a patient was clearly unwell. It was less stressful just to pretend.

> *But it is really… It's a little bit like tears of a clown. Because she's got a great smile and a great personality, and people see that don't they? And sometimes inside they don't really know. (Mother MB25)*
>
> *And sometimes I do kind of have a smile even when I'm feeling a bit rubbish. I don't want to show it. Because otherwise people start asking questions. "Are you alright?" "Yes, yes. I'm fine". Because I can't be bothered having all those questions. (MB25)*

However, in some cases partial solutions were found to acknowledged disability.

> *I'm thinking normal kids don't want to play with him: because he's at a different level, he's like a different age. It's like he stayed at four where he was when the tumour was found. He's not so big as them, and he can't keep up. He does sometimes get angry watching other kids playing. His mum feels that he has a feeling that other kids are playing and doing things, and he is probably thinking "I can't do those things".* He can't do lots of things to play like other kids (such as) running and jumping, but he can do that [indicating console game] as well as anyone else. Then the disability is reduced. (Parents, MB12)

Denial and Aspiration

Although sometimes out of consciousness, realisation of their denial would come back to parents.

> *To be honest, as a parent, sometimes I'm in denial of so many things, you know. In my head I feel she's a normal girl. OK, so I'm in sort of a denial in some ways, you know, especially when she doesn't understand me sometimes, or I would say "why's she doing this?" And I forget that she's been through so much in the past. (Mother MB18)*

Despite the cognitive and physical problems experienced by some in the younger age group, they had aspiration for themselves and their future.

> *What comes next after college? Uni. Yeah. After that I may be a jewellery designer, yeah. I want to open my own business. (MB18)*
>
> *He's got massive people skills, he's such a pleasant person and everybody warms to him and he's got really good ideas on marketing and stuff. He's a bit of wheeler-dealer, without the practicality … so, you know, I'm still waiting for him to get it, whatever it is that he wants to do. (Mother MB3)*
>
> *Oh yes. I'll do a job. I just don't know what job to go for. Get a job. Get my own money in my bank. There is not much jobs around [county], though… (MB13)*

> *There is a lot of places where they want, you know, big superstores and that, take on… there is a lot of shops getting built. Semi-independence? Yeah. I would say so. I definitely think there would have to be somebody around him, or in the vicinity all the time… (Father MB13)*

For some patients, the reality of what would be possible was mixed in with their aspirations.

> *It's hard to see a future sometimes because, when it comes to school work and that I find it really hard. And quite pressurising. It's not too much of a problem being a bit behind, but taking new subjects a lot and remembering them all [is a problem]. I would like to be an actress or something and my memory is awful. I worry that my exams in the future are going to wreck that or my memory and stuff is not going to not help me in that sort of situation.*
>
> *I'm not exactly expecting much of myself because I just don't expect that I could do very well but I will try. Last year I thought I'd done really, really well and then I found that I'd done really bad at it. And really just felt disappointed with myself. So I'm really not kind of expecting much of myself, but I do try because I know I'm not exactly going to be exactly the same as everyone else, but it's the best that I'm going to do. (MB25)*

Discovering Services

Helpful services were found almost by accident, but being not specifically health-related, they were not well-advertised in clinic.

> *There must be other people that were like that, who don't want to go and play football where everybody's running 50 miles per hour and maybe seeing him as a hindrance. But there was nowhere really for him to go or to do, or if there was I never found it. And I did ask a few times. (Mother, MB3)*

For some families, a wealth of supportive services were available in the hospital or would be signposted clearly. However, this was not the case once they reverted to the care of their local district general hospital.

> *He took time to explain things and he would say, "Alright, you can get this, that and the other. Once you start looking for an iPod, you can get something that's in…we've got these magazines here. We can get you that. We can put you in touch with it. But then when we got transferred back to our local hospital, there's nothing. We go in and say, "have you got any catalogues that have implements that help hearing?" "Oh, no. We don't have anything like that." (Father MB26)*
>
> *I went to the fire brigade and asked if we could have a fire alarm at the top of the stairs, and one that we put under his pillow. If the fire alarm goes off it will shake and wake him. He said, "we don't usually do that for kids". So, we'll get that, we'll put it in. But we've had to do that ourselves by research and what have you, and now we need to start looking at different things for him, because his hearing isn't getting better; it's getting worse. (Mother MB26)*

Problems that occurred only episodically but repeatedly were seen to fall through the gap between services. A more timely multi-disciplinary review was sought rather than "take them to the GP if it happens again". The parents of one child were struggling to understand the reason for their son's intermittent irritability and angry outbursts.

> *The doctors did say that he won't be like normal kids, but I don't think that we knew just how that would turn out. Every six weeks or so…he's not himself, you know. The teachers as well, they don't know why he's like that. They thought it might be the shunt in his head, but we checked with the doctors and they said it can't be his shunt. He hits people at school and throws his shoes in the school bus. It still happens now. The doctor suggested that if it happens next time, see the GP: see if they can do some blood test and see if it's in his blood. So it's a bit of a mystery really. (Father, MB12)*

However, patients and parents also expressed a completely positive perspective on the response to reported problems.

> *If I had a problem I would just ring and speak to somebody. So it's not really been a problem. I've never felt I've got [only] five minutes and that's it. I've always felt that if we wanted to sit there for three hours I think we could've done. (Mother MB3)*

Perceptions of Quality of Life and Thoughts About the Future

Patients will often rate their quality of life much higher than their parents would report it. Given their often sustained dependence on their parents, this required tactful and insightful response in clinic.

> *I didn't want him to think that he could do nothing because he's had a tumour. I suppose I never talked about it when he was younger and then the longer you leave it I didn't know what to say. This is probably the most honest we've ever had a conversation about it. (Mother, MB3)*

While measured psychological quality of life seemed to be widely poor, some patients perceived an improvement over time.

> *She used to be very scared to get into public transport "Oh, people are looking at me", and stuff like that. She used to feel like that, but now you don't feel like that anymore do you? (Mother MB18)*
>
> *Yes, even though it's in the canteen in school, you know, people keep staring at me. I didn't like to go. But now I don't feel like that anymore. Yeah, things are improving, definitely, yeah. (MB18)*

Uncertainty about the future was a repeated concern.

> *To what extent do we assess… As he is growing up we're not sure then what to do then. What happens then? He's 11 now. And when he gets to 16, what happens then? That's what we're worried out. We're just thinking… is there not something that will trigger it so he will come out of that four year old stage. His mother is worried as well. We could do with him improving, like in his mind so that he moves on. We want him to make those big leaps, you know. I don't think he'll be able to be independent. I'm hoping that he will be, but you never know. (Father, MB12)*

This was not always the case, however.

> *And I think we were lucky having [Named oncologist] as well, because he obviously knew this was all going to be out there for us once she came through her treatment. He was very clever at drip-feeding information, so without wanting to sort of prepare you for terrible things that might not be, he had everything in place for when those sorts of things happened a bit. (Mother MB25)*
>
> *All the information we got from the hospitals and [Named oncologists], they all told us that his learning abilities will not be like a normal child. (Father MB13)*

COMMON ISSUES FROM THE 18-40 YEARS GROUP

In this group additional and different issues were raised. Problems had moved on to increased awareness of the lifelong impact of the tumour and treatment, with diminishing ability and the proliferation of problems, particularly psychological issues. Lack of transitional arrangements into adult life and a continual need to fight for services added to the burden. Both patients and parents experienced frustration with limitation and discrimination in occupation and work, while patients expressed wistful comparison with the lifestyles of others and persisting issues with social life.

Coming to terms with the lifelong impact of the tumour and treatment

The nature of enduring problems might change as patients moved into adult life.

> *Well, some things have carried on, but have decreased in how often. And then I've got side effects that have come from the treatment that I've been told will not really go, like headaches in the morning, sometimes vomiting in the morning. So that's something that I might just have to put up with. And the tiredness. Before, I couldn't concentrate for long enough to get back to studying. It's improved over time, you know. I can concentrate for quite a while now. I still have the problem of being really, really tired. (MB19)*

Patients were well aware of their cognitive and physical decline since diagnosis, usually accepting this with a degree of fatalism.

> *I haven't a clue any more. I was very bright when I was younger whereas now I am thick as two short planks. (MB15)*
>
> *I don't know how I feel about not being able to do things that I could do before. I used to say "I wish I could run" or I" wish I could ride my bike again". Can you picture it? With problems with balance and strength. I've got used to it. Adapted to it. (MB16)*
>
> *Some things are just not going to happen and you just shouldn't punish yourself about it. Will it come that I don't need the wheelchair? No, I don't think it will. I don't know. It might happen if by some miracle I don't have fatigue any more. But I do. So, I think, [it's been] 10 years. I've learnt to accept that I'm in a wheelchair now. Like, it might get better. I don't know. I take it as it is, but then if something gets better in some way, it's a bonus. (MB20)*
>
> *It's treating people like her normally, not feeling sorry and all this stuff. It's about this is the life you've got to live and I think that is a big thing, because some don't do that that and you know they mollycoddle them and spoil them and stuff like that. I think that's how she got on and for her own sake, you know, get on with life and stuff. (Mother MB7)*

Specific strategies were sometimes employed to overcome problems.

> *Balance was a major issue which affected me quite a bit. It upset me because before my treatment I was a gymnast. So I took dance classes and ballet to get my balance back. And my flexibility obviously wasn't brilliant, but it helped to get my balance back, especially the dancing. It made a difference socially because I became more and more normal. (MB19)*

Some problems were effectively concealed by patients as they learned to adapt to developing disabilities or to avoid the problem.

> *On top of that, they also diagnosed that his hearing was deteriorating but he wasn't telling anybody because he would eventually overcome it himself by the fact that he had learned to lip-read. He taught himself to lip-read. It was more in the home when everybody was talking and he couldn't see people that he wasn't responding, and it wasn't being noticed when he was talked to. Now because he is going deaf his speech is deteriorating and becoming indistinct. (Carer MB15)*

The effect on one respondent was that he could barely stand contact with others, feeling entirely unsociable, and finding nothing interesting any more.

> *In adult life I can't find anything that interests me. I'm quite bored and it takes a lot to interest me which is why I perhaps question a lot about what am I doing, what's this all about, where am I going with this, what's the point of me doing this. That's a problem sometimes now: I'm thinking about things too much.*
>
> *I'm self-employed now and it's the best thing that's ever happened to me because I couldn't stand the social environment that being employed brought for me. It's very difficult to relax when the phone rings, even if I put the phone down, or put the phone on silent I know I'm going to have to come back to it and deal with it. There's that apprehension there all the time. I've tried to get away from social environments but*

> *no matter what you do you've still got to interact with people in a way that I suppose in the long run seems to be making me uncomfortable.(MB9)*

Parents as well as patients noted the apparent proliferation of problems as adulthood changed the context of survival. MB10 and her mother listed the problems that had arisen during adulthood.

> *Her walking – help needed with that. Hearing – I need hearing aids now. You're too weak to lift pans for cooking. Fits. Tunnel vision – you can't cross the road on your own. (MB10 and Mother)*
>
> *I notice more of the change as he is getting into adulthood. I suppose you think he's going to be normal or you think him as normal, but then all these problems start hitting you with trying to get work. (Mother MB14)*
>
> *When you get to an age where the outside world starts impacting on him a bit more and he is trying to evolve and turn into a man himself, all the other issues of the world that he's been probably hidden from come into play. (Father MB14)*
>
> *And then my eyesight is now the one that is deteriorating. I started wearing glasses just over 12 months ago for television and computers, and 12 months down the line my sight has deteriorated, so I have to wear glasses all the time as I really can't see without them. And that is in the space of 12 months that my eyes have deteriorated. (MB15)*

Some problems came as a surprise to families. However, they acknowledged that they might have been warned of such late effects, but in the desperate hope first for their child's survival they may have either failed to register the information or have put it out of their minds.

> *We hadn't expected the cataracts and the hearing. It was more memory loss and learning difficulties, because they said it's going to destroy a lot of good cells. Maybe you find out things later on or maybe you're just mithered with what he's going through at the time. You're probably trying to take so much in and you're just focused 100% on his health, you don't care about anything else at the time. You'll worry about hearing later. But it seems like things have just happened one after another. (Mother MB14)*

Some of the problems that were experienced were described as hidden costs of survival. The likelihood of infertility is usually discussed, but one woman had become pregnant (thinking, at first, that this was impossible) only to suffer two miscarriages. She had never thought about this as a possible problem. She was determined to explain the distress that she had experienced.

> *I wanted to do this really because it's a good opportunity for somebody else who's in my position or who have had cancer and has been through this, for them to know what can happen. (MB7)*

The delayed onset of problems meant that some families had forgotten about their possible emergence. They wanted more timely reminders of problems to look out for at various points in the period of long-term follow-up, yet recognised the negative effect of continued reminders.

> *There is maybe an element that even when someone is going through or just coming out of the illness and treatment, if you hit them then with all these future problems, that's not going to always help as well, is it? Because you're going to think "Oh, my God". Then you're waiting for these things to happen that may not happen. I don't know whether… because things have caught us on a blindside that have happened as he gets older, we don't know whether there is anything else to come yet, you know? You wonder "do we know it all yet or is there anything else?". (Father MB14)*

Mental health suffered as a reaction to the multiple physical, psychological, cognitive and social problems encountered.

> *He has bouts of depression when he feels like he wants to die. He can't see the point of being here anymore. And then it goes off and he is fine. And then every now and again he get these bouts of depression again. (Carer MB15)*
>
> *The main things I suffer with are depression, and I have some problem with remembering words now and again, I just can't get the right word for things. It's getting strategies to cope with the problems, but the depression thing… I went off sick from work with depression. I was on medication and got sent back for a week and then got the sack for not having performed well enough: maybe because I'd got depression! (MB4)*

The frustration of being patient, and realising that this would continue potentially for the rest of her life prompted a rebellious reaction in one patient.

> *I'm meant to take my injections for the rest of my life and my tablets. I was just really sick of it all, so I just stopped taking it altogether. I just didn't want to be a patient anymore. I wanted to escape from the whole experience of needing help for everything. It felt like it's always hospital and I needed people helping me with this, that and the other, and that's the only thing I could control by throwing it away. I've said I am going to take them again, because I've realised how stupid it is not to take them. (MB21)*

Although not a clinical issue, one particular response, expressed in different ways by various patients or parents, was clearly unsettling.

> *It would be wonderful for somebody to say, 'Right, that's it now, you're fine, you're over it'? or even just to say, 'Right, that's it, now you know that you've got that, that and that against you on your tick sheet, those problems are always going to be there, but there aren't any more coming'. It feels like it's never-ending. (MB4)*

One particularly articulate young man repeatedly suggested that various problems that he had encountered might just be his personality. He was reluctant to ascribe problems to the tumour despite these being classical examples of late effects. He was not the only one at least to wonder how much to ascribe to the effects of the tumour.

> *It's difficult to tell what's normal and what's not normal. I'm not necessarily assuming that treatment or cancer that I've had is the cause of social or mental problems I might be facing now. It could just be my personality, the way I am. It's just me. (MB9)*
>
> *So it's always quite unsettling if you just feel a bit unwell and you think at the back of your mind, 'Is this related to the tumour or is it just me?' (MB4)*

Difficult transition into adult life

Although, the adult patients found that issues with benefits, education, social services, and disability support became far more important, health services were a part of the issue.

> *I think when they leave school the speech therapist signed her off, physio signed her off, occupational therapist signed her off because they said that the adult services is so different and you will not get the help so might as well just stop it now. (Mother MB16)*

While recognising cognitive problems in their children, parents might have to fight for an individualised appraisal of the individual's level of ability. Special schools, for example, were recognised as a major improvement after attendance at state school, but might mean being grouped with others of lower cognitive ability. This continued into the world of work and the attitudes of the staff in some agencies.

> *She went to a special needs course for independent living at college, but that didn't work out because she was put into the groups where they were mainly Down's syndrome pupils and autistic pupils, and a few with learning difficulties that were quite loud and aggressive. She got quite depressed. (Mother MB6)*

> *It was all sorts: a lot of the kids from all over the boroughs. There was all sorts, a lot of physical disabilities, a lot of mental disabilities and a lot of behavioural things… like a big melting pot. (Father MB14)*

> *She is just now having psychology reports. We're now just having social workers. All these people are now just becoming involved now because I've asked. She has always had a taxi to school and it carried on at college, even past 19. Then, one day, they suddenly said "she comes under adult services. You need to phone this number". I phoned them up, and it was, "it will take up to six months to get her referred". I just reached to the end of my tether. I just can't cope anymore. I've tried and tried, and everything she has needed was never there. You've always fought for it. (Mother MB16)*

The continual need to fight for health and social services was an experience shared by many, causing anger and often despair.

> *The times I've fought for everything for her from school, from Disability Living Allowance, everything. You've to fight for it, and you just feel so demoralised and so stressed that I can understand why people just think, oh I can't do it anymore. And they give up, and then the families suffer because they're not getting what should be on offer. (Mother MB6)*

Even before fighting to have services, parents had to know about their existence and find out how to make an application.

> *It wasn't that we said no to it. We were never offered anything like that. We had a talk to the social worker. She managed to help with the disability forms and things like that. (Mother MB14)*

> *All the services that was available when she was a child; they just all stopped completely. So I was unaware that she should have been transitioned over the adult services and she never was. And we're just dealing with it now. I thought it was all down to me. I didn't know all these other services what she was entitled to. People were saying to me, "why is she not getting this? Does she not get earning support allowance?" And I'm, like, "what's that?". I don't know because we've never had a social worker from becoming an adult. (Mother MB16)*

Although new cases receive the latest blend of services, existing patients may continue without such benefits – since these were not routine when they were younger. Their package might not catch up with recent service standard.

> *Sometimes when I've been here they've had like social workers. She's never seen a social worker, they've never been offered us, you know. And I just wondered whether a back-up thing would have been, you know, to give us where to go, what to do, help her to be independent. She's been coming here since she was four, and we've never been offered anything like that. Only just doctors. (Mother MB10)*

Limitation and discrimination in occupation and work

The need for occupation or work was expressed by most participants, whether as paid work, voluntary work, further educational study, or simply some activity to occupy the mind and perhaps feel valued. The work might be menial or low-skilled, but the activity of being.

> *I wanted to do hairdressing so that's what I did. Got somewhere close to home and I worked there, I think it was one day a week. And then when I finished school they kept me on, on a Saturday. And then I was there for I think about six months and then they had to let me go because they couldn't afford to keep me anymore. It was washing people's hair and making drinks and sweeping up. Then I did a hairdressing course and got a job at a hair studio. (MB7)*

> *He doesn't need a full-time job but he needs something, because if he didn't have these volunteering jobs he would be just up in that bedroom all day. Something for him to get involved with, just to take time up, to give him something to do, because if they haven't that you can see why just normal people that are unemployed get depressed. (Mother MB14)*
>
> *You don't want to be at home vegetating and getting even more depressed and watching life go by in front of you. People are nice, my nan is really good to me, but it's not the same as being with other people in your own age group, is it? Places don't do part time, and it's travelling to get there. It's just frustrating. (MB16)*

Many respondents expressed frustration with the inability to secure or retain paid work or even voluntary positions. This was combined with a lack of opportunities for general occupation.

> *She wants to be like everybody else. She wants to go out shopping by herself. She wants to do things by herself, but she's just too vulnerable, you know. She'd just be a target. And she doesn't have much understanding of money and danger, you know, things like that. One day she just said "I feel like I am that little mouse in a corner where it's a room full of elephants". (Mother MB6)*

The enduring limitations caused by the tumour and the treatment impacted directly on employment prospects, but this was compounded first by lack of appreciation of such limitations, and then by unhelpful attitudes towards families by some agencies and individuals.

> *Once the education came to its natural conclusion, you go into the job centre, and for two years we had a running battle with the authorities. It was only after two years when we finally got him to see a disability adviser. He should have had help straight away and there was nothing for him. He was just forgotten. And then they stopped his money because they were expecting him to go on to computer courses, to turn up at places asking for work, to write letters. They didn't know his limitations. (Father MB14)*
>
> *Because I was his appointee, I had to go with him to sign on, which I didn't know and I didn't get told. He'd been signing on for over a year on his own, and then when all the disability came out she said "Well, you're supposed to be with him". He doesn't want to take his mum to go and sign on, and it was really embarrassing. (Mother MB14)*
>
> MB20 explained off-tape about a problem with a benefits officer being curt and bullying on the phone because of lack of understanding of the problem. The patient found this frustrating since she was living independently and trying to be as self-caring as possible, but her speech difficulty provoked an irritable and unhelpful response. Telephone calls could be difficult.

One participant experienced termination of her opportunity to work in a nursery because of the employer's knowledge of her disability – despite a history of successful working in another child care nursery.

> *Someone said "I have got a nursery. Come and work with me. You are really good." So I did, and I thought, you know, this is great. But then within a few months they said "your disability is getting too obvious and too hard: you are going to have to go". It was really strange. I had been in other workplaces as well where you just do the same things as other staff but because they know, in the back of their mind, that you have got the word disability hanging over you, it is, kind of, you are on a double red line already, without starting.*
> *I also worked in a college as a support worker. I was really enjoying it, and everyone thought I was doing really well. Three or four months down the line, the management said I come under the DDA, because I had a childhood brain tumour: "It has come to*

> *our attention you have a disability and you didn't tell us this". Well, I did. It is on my CV. I told them at the interview. A manager asked me "how long have you known you are dyslexic?" I said "What? For about two seconds!" (MB11)*

Sometimes a situation of double jeopardy developed, with patients being restricted in employment because of disabilities being declared, yet also continuing to be criticised publicly because of the absence of immediately recognisable signs of disability.

> *She used to go to an under-fives unit to help out. She had to wash up and sharpen pencils, but wasn't gaining anything. And then they just said that it would be better for her to look for somewhere else to get more experience. But where do you start? She cannot pick the children up. If a child falls her reactions are not quick enough to catch them. She knows that. So she doesn't do it. She is not stupid…*
>
> *She sat in the disabled seats right near the bus driver, and somebody came and said, "get out of the seat: you shouldn't be there". Obviously there doesn't look anything wrong with her to look at. She just tried to get up and move. She would not argue with them. And then she fell over moving. (Mother MB16)*

The frustrated desire to engage in child care work was echoed by another participant.

> *You see now, with her having her balance and everything, I mean she loves children and she was going and helping in an after-school club, but I can't send her to after-school club if she is going to be falling all over. (Mother MB8)*
>
> *A child fell on his knee and I might lift it up. And I fall, you know. (MB8)*

Those patients who felt more positive about their life, often despite significant disabilities, were clear on the factors that had been important in achieving.

> *I was determined. I think that was just to prove that I could do it. I was motivated but I also had a lot of support around me family-wise, and educationally I had a lot of support. I had friends outside of school that knew me before I was ill, so I never really lost them, but they weren't really that local so I didn't see them. (MB19)*
>
> *I was an A* student. I was clever, but… Well the doctors said I wouldn't carry on with my education after I was 16, but I had strength and determination – I got an award for that - went to college and got two A levels and then went to uni and got a degree. So, I showed them. I'm working with young people. I'm a part-time youth worker at the minute. Yeah. I only do six hours because I've got fatigue now. Well eventually I want to get a more permanent job, because it's only temporary, this one. (MB20)*

Absence of social life

As patients moved into adult life the impact of the illness and treatment on social life revealed itself increasingly, with physical disability, sensory damage, and inability to keep up with peers exerting a particular impact. Often the problems with social life began while off school and intensified on return to school. Some patients felt that failure to regain social status and links at secondary school was the root of later problems.

> *I would say that about high school. Socially it was a disaster. (MB19)*
>
> *Socialising is a bit more of an issue. Some of the lads invited him down to the pub but by the time they had had two or three drinks they were standing outside smoking and chatting together and he would find himself at the table on his own. They would come and go but they are outside, while… (Carer MB15) …I'm inside. Billy No-mates. (MB15)*
>
> *My wish would be for her to be able to go out to either work voluntarily or paid in a job that she enjoys doing, that she's happy doing and have a bit of a social life, you know. I'm not saying going out partying every night, just doing something that she enjoys, with people that she can interact with. (Mother MB6)*

> *You see, that is another thing I get depressed with, I spend a lot of time with older people than myself, nothing for myself. It is good to do those things, but I need something with people my age. And it does depress me a lot, thinking about things, and I have to go and find something else to occupy myself with. (MB8)*
>
> *I did have friends but I used to hang around with the quieter bunch. It means less people. It was the better choice for me because if I go to the pub and there's a load of people I struggle to hear because there's too much noise for me, that's you know, just because I can't hear in this ear anymore. So that might be a contributory factor as to why I choose to be unsociable. That lack of wanting to be sociable might over time also have made me fear social environments. (MB9)*

Some parents expressed the need for direction and signposted opportunities.

> *There are clubs and volunteer things, but if it was put to him that you are going on this, that might be a good thing, if there was anything like that. Not so much a group where it would just be people with the same illnesses all moaning to each other, you know, not that sort of thing, but something with a positive spin on it. (Mother MB14)*
>
> *It's having stuff after it, something for him to do so they can engage in some sort of activity. To make them feel that it's not totally limiting them, because that's how it must feel for him when he sees what's going on in the world and what they can and can't do. (Father MB14)*
>
> *Things like social places for them to go, like, or even, not a workplace but, you know, something that helps them to do certain things that will get them out and into the world. That's what is needed. (Mother MB10)*

Comparison by patients of their situation with the lifestyles of others was unhelpful but unavoidable.

> *She wants to be like everybody else. She wants to go out shopping by herself. She wants to do things by herself, but she's just too vulnerable, you know. She'd just be a target. And she doesn't have much understanding of money and danger, you know, things like that. One day she just said "I feel like I am that little mouse in a corner where it's a room full of elephants". (Mother MB6)*
>
> *I did have a lovely head of hair and it was all down here. And now, seeing my cousins having a lovely head of hair and they can tie it up and everything, it really, really depresses me and I have to be hard on myself. See what I feel is, all the people I know are married, got a job and got a house and I am just stuck in a rut because I am disabled. (MB8)*
>
> *I get a bit upset sometimes because my brother and sister can do things on their own. I can't. I get a bit upset about that. (MB10)*

When friends were not lost, a more positive experience was described.

> *Yeah, well there was about five or six friends that were there all the time, and a few of them still are. I still see a friend who I've known since I was five and she's still there. I keep up with people through social networking, too. That was very important because otherwise it could be very isolating, couldn't it? (MB20)*

COMPARISON BETWEEN GROUPS

Comparing the interview findings of the under 18 years of age group and those aged 18-40 years highlighted both important common themes and also differences for survivorship issues that relate to increasing age from diagnosis and therapy. For both groups of survivors, social isolation could be a major survivorship issue, and the challenges encountered in terms of finding support from the health and social care sectors was highlighted by many respondents. In addition, the times of transition, for example from primary school to high school and the transition to adult life, were identified as being difficult and challenging by survivors of childhood medulloblastoma.

For both groups, the major ‘occupations’ of either school or work were central survivorship issues. At the school interface issues such as maintaining peer recognition and finding a balance between support and enablement were felt to be important, while for older survivors there was a perception of limitation and discrimination in the workplace.

The younger survivors described the evolving recognition, acceptance and resistance to disability and their mechanisms for dealing with these issues such as pretence. However, they still held aspirations for the future despite their physical and cognitive problems. In partial contrast to this, patients from the 18-40 years age group were, perhaps, more accepting of issues such as physical and cognitive decline over time, and the life-long impact of the tumour and treatment.

In summary, for both patients groups a major concern was the availability of, and signposting to, appropriate services for support such as psychological well-being and social services to which they were entitled but about which they were often ignorant. The findings indicate the broad range of survivorship issues that could be supported with appropriate resources.

Section 6: Outcomes of Text Mining

CLINICAL CONCEPTS

Using the results from the text mining techniques, the data was analysed by each individual transition group as well as an overall aggregated cohort analysis of common concepts that appeared in respective datasets: patient interview transcripts (PN) and clinical (CN) narratives. In particular, the analysis considered common clinical concepts versus HrQoL concepts (Table 6).

Table 6: Common clinical concepts and HrQoL concepts

Common clinical concepts	HrQoL concepts
Physical functioning	Oncology diagnosis
Emotional functioning	Oncology investigation
Social functioning	Oncology treatment
Cognitive functioning	Endocrinology diagnosis
School functioning	Endocrinology investigation
Sensory and pain	Endocrinology treatment
Other wellbeing	
Activity	
Home and family	

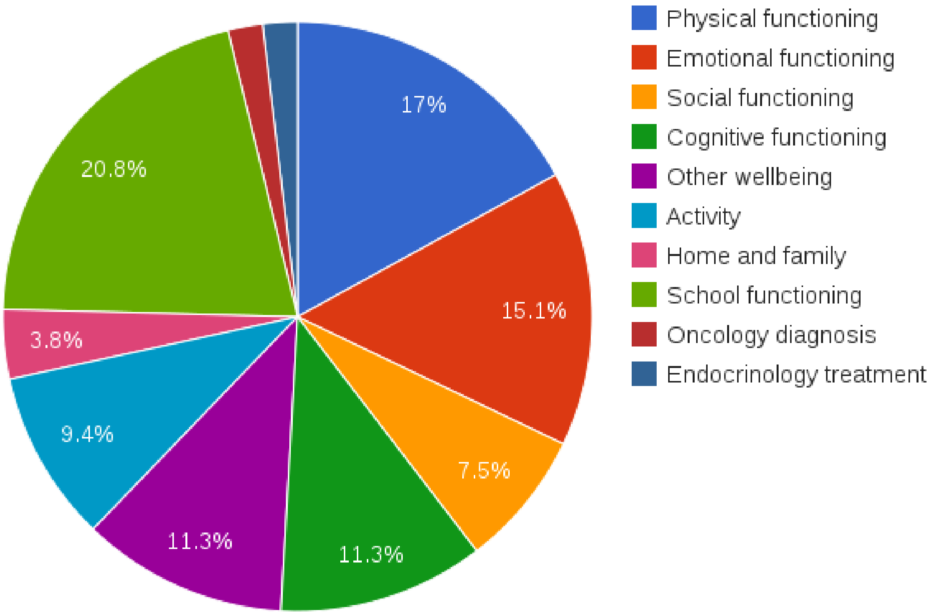
TRANSITION GROUP: 11-16

While concepts mentioned in patient (PN) and clinical (CN) narratives in this transition group seem to have notable overlap, it is obvious that there is a clear bias between health concepts in corresponding narrative types. For example, the top concept types in PN were *School functioning*, *Physical functioning*, and *Emotional functioning*. On the other hand, *Endocrinology investigation*, *Oncology treatment* and *Other wellbeing* concepts appeared most frequently in the CNs (Table 7).

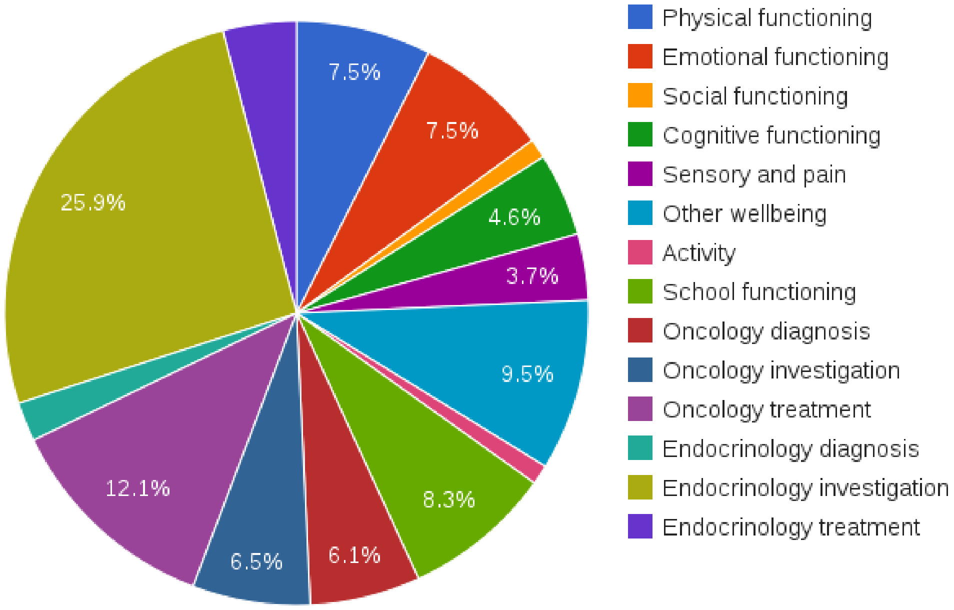
Table 7: Young transition group: concept distribution

Concept type	Patient narrative		Clinical narrative	
	Frequency	Proportion	Frequency	Proportion
Physical functioning	9	17.0%	55	7.5%
Emotional functioning	8	15.1%	55	7.5%
Cognitive functioning	6	11.3%		
Other wellbeing	6	11.3%	70	9.5%
School functioning	11	20.8%	61	8.3%
Oncology treatment			89	12.1%
Endocrinology investigation			191	25.9%

Moreover, despite the evident bias with regard to the narrative type, it is notable that PNs contain many of the concepts prevalent in CNs (Figure 5). However, this is not true in reverse. For example, while the top three concepts in PNs appear in CNs, only one out of the top three concepts in CNs appear in PNs. While a bias was observed in terms of proportion of concept types between PNs and CNs, the CNs seemed to be more inclusive of the breadth of concepts.



a. Proportion of concepts found in patient narrative



b. Proportion of concepts found in clinical narrative

Figure 5: Comparison of proportion of concepts identified in PNs and CNs for the young transition group

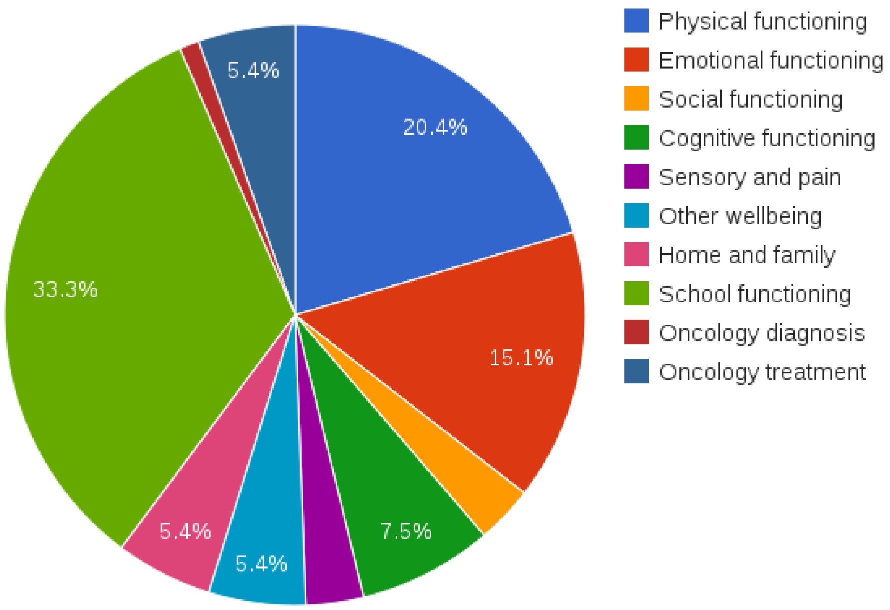
TRANSITION GROUP: ADULT

The adult transition group generally reflects the results from the young transition group. The top three mentioned concepts in PNs are *School functioning*, *Physical functioning* and *Emotional functioning*. On the other hand, the most common concepts in CNs include *Endocrinology investigation*, *Oncology treatment*, and *Emotional functioning* (Table 8). Again, a clear contrast was observed in the type of concepts common in respective narrative types.

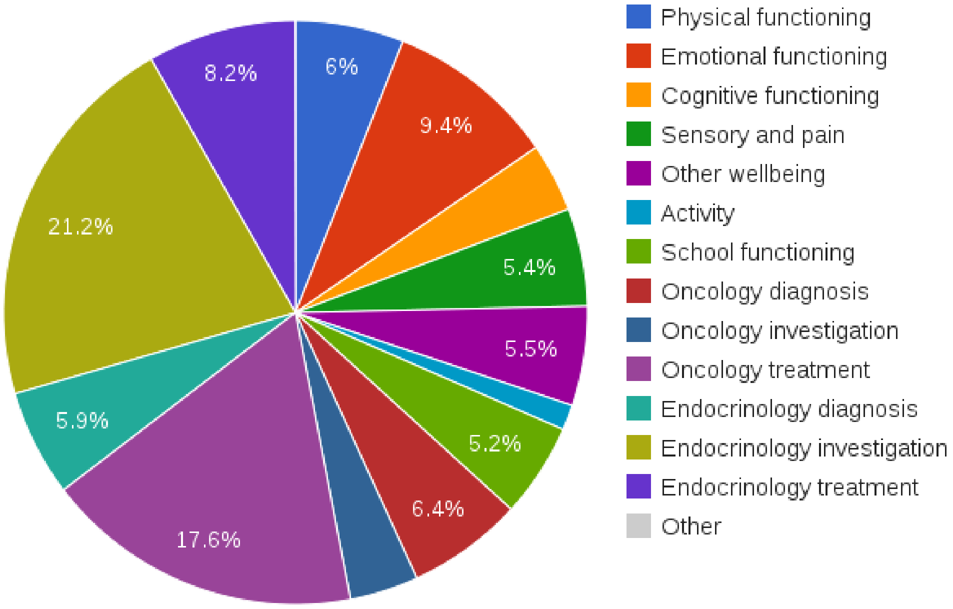
Table 8: Adult transition group: concept distribution

Concept type	Patient narrative		Clinical narrative	
	Frequency	Proportion	Frequency	Proportion
Physical functioning	19	20.4%		
Emotional functioning	14	15.1%	151	9.4%
Cognitive functioning	7	7.5%		
Home and family	5	5.4%		
School functioning	31	33.3%		
Oncology diagnosis			103	6.4%
Oncology treatment	5	5.4%	283	17.6%
Endocrinology investigation			340	21.2%
Endocrinology treatment			131	8.2%

Moreover, similar to the young transition group, CNs tended to include a broader range of concept types, while PNs were largely focused on HrQoL concepts. This conclusion is made more evident in Figure 6.



a. Proportion of concepts found in patient narrative



b. Proportion of concepts found in clinical narratives

Figure 6: Comparison of proportion of concepts identified in PNs and CNs for the adult transition group

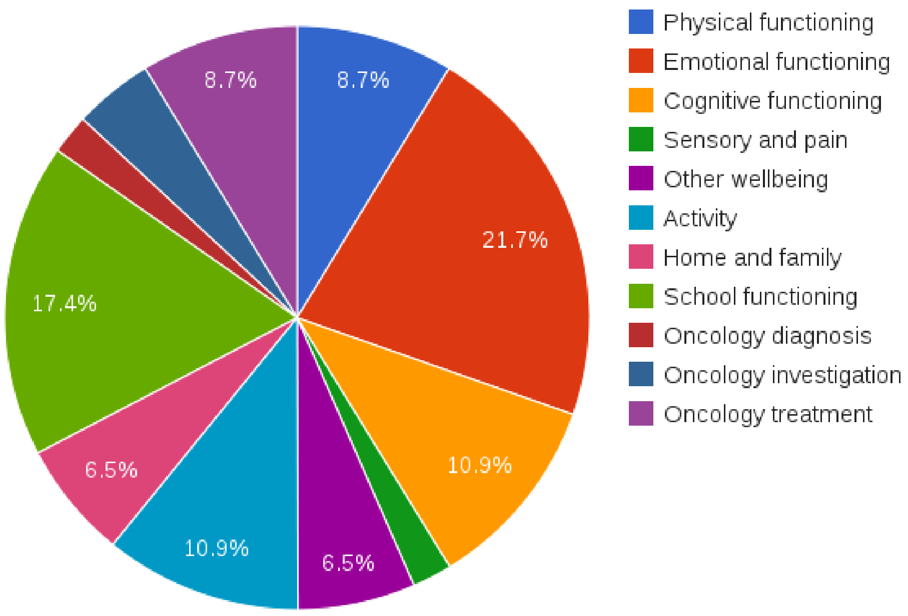
ADULT PATIENTS OLDER THAN THE ADULT TRANSITION GROUP

For this group there was a similar bias to that in the transition groups in terms of common concepts mentioned in different narrative types. For example, as shown in Table 9, *Emotional functioning*, *School functioning*, *Cognitive functioning*, and *Activity* were the most mentioned concepts in PNs. In contrast, CNs contains mostly mentions of *Endocrinology investigation*, *Oncology treatment*, and *Other wellbeing* concepts.

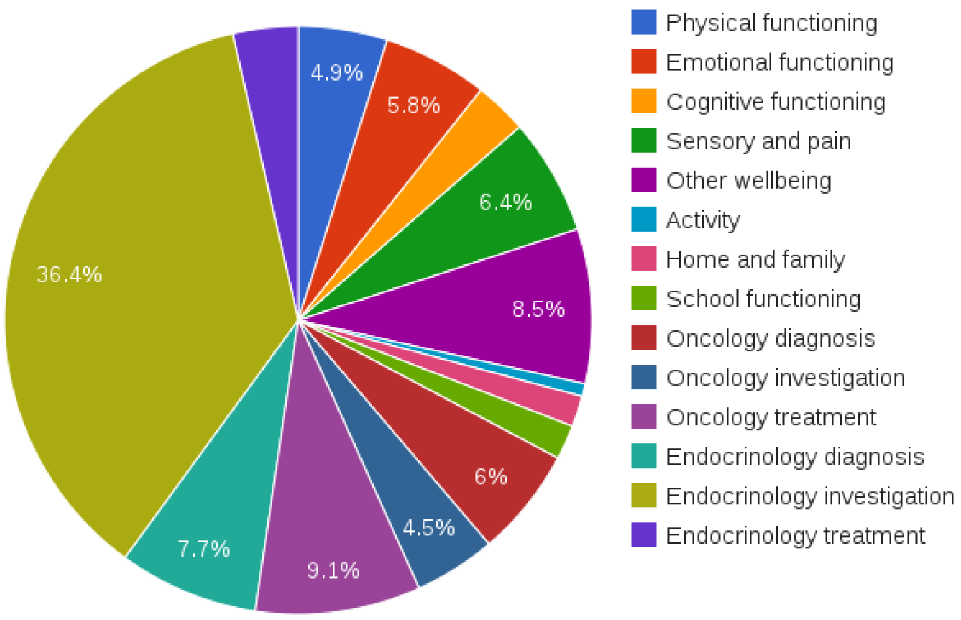
Table 9: Patients older than the adult transition group: concept distribution

Concept type	Patient narrative		Clinical narrative	
	Frequency	Proportion	Frequency	Proportion
Emotional functioning	10	21.7%		
Cognitive functioning	5	10.9%		
Sensory and pain			64	6.4%
Other wellbeing			85	8.5%
Activity	5	10.9%		
School functioning	8	17.4%		
Oncology treatment	4	8.7%	91	9.1%
Endocrinology diagnosis	-	-	77	7.7%
Endocrinology investigation	-	-	364	36.4%

Figure 7 shows a similar trend to previous groups in which HrQoL concepts tend to be more prevalent in PNs, while CNs include broader cover of mentions of concepts.



a. Proportion of concepts found in patient narratives



b. Proportion of concepts found in clinical narratives

Figure 7: **Comparison of proportion of concepts identified in PNs and CNs for patients older than the adult transition group**

OVERALL AGGREGATED COMPARISON

The aggregated concept analysis largely reflects the findings from the individual transition groups. The analysis of the whole cohort included a set of twenty clinical narratives or CNs (ie: hospital records) and corresponding patient narratives or PNs (ie: interview transcripts). The extracted data, using various NERs (clinical events and HrQoL mentions), were normalised using the proportion-based statistic of the frequency of mentions. The pie charts shown in Figure 8 and Figure 9 show a notable difference in concepts found in PNs versus CNs, respectively. For example, the most common concepts found in PNs are School functioning (24.5%), Emotional functioning (20.2%), Physical functioning (11.4%), and Cognitive functioning. On the other hand, Endocrinology investigation (22.7%), Oncology treatment (15.8%), Endocrinology treatment (8.2%) and Physical functioning (7.3%) are proportionally the most common concept types appearing in CNs.

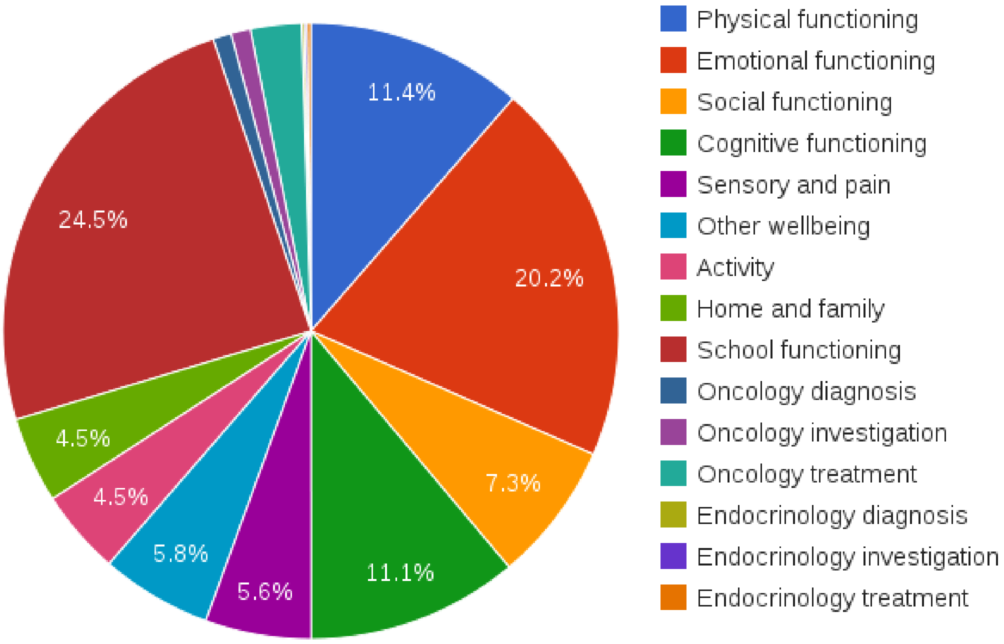


Figure 8: Proportion of concepts found in patient narratives

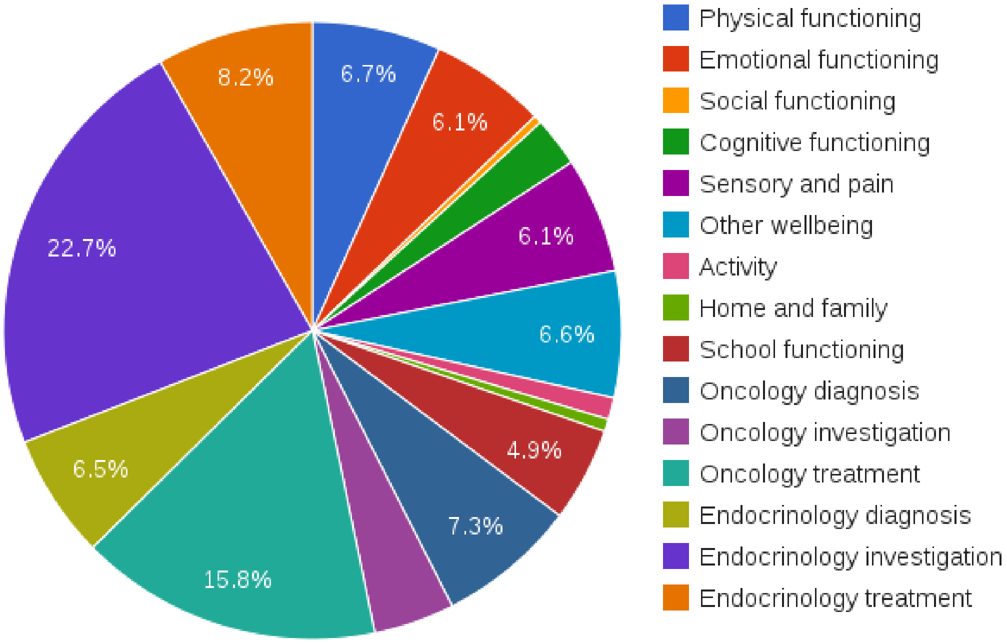


Figure 9: Proportion of concepts found in clinical narratives

COMPARISON BY BUBBLE CHART

An alternative data view is shown using a bubble chart. Figure 10 shows a clear difference in concept between PNs versus CNs. For example, traditional clinical concepts such as diagnosis, investigation and treatments are more prevalent in CNs. In contrast, HrQoL concepts are more common in PNs. While CNs contains all HrQoL concepts investigated, they occur proportionally less than in the PNs.

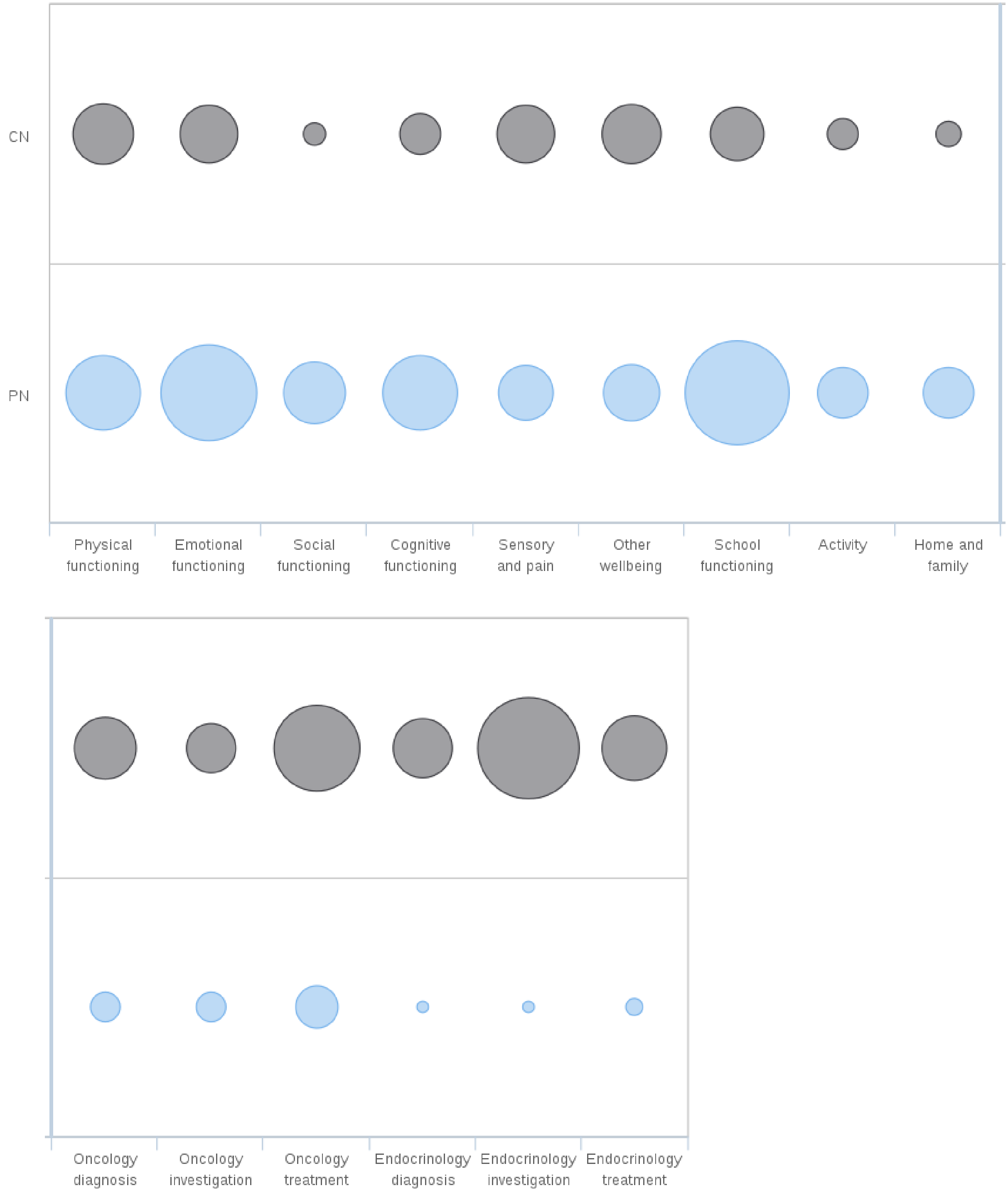


Figure 10: Concept analysis between patient and clinical narratives – whole cohort

The comparison between the focus of health professionals and patients in each of the three groups is illustrated below in examples of cases.

Figure 11: Example of under-18 group: MB03

There was significant symmetry between clinicians and patient in this case other than a distinct lack of concern on the patient’s part about ongoing monitoring for signs of further tumour or endocrine problems. More concern was shown by the patient about social functioning, activity and home life.

Figure 12: Example of 18-40 group (adult transition): MB20

It is clear that for the adult transition patient, physical functioning, particularly mobility, was a major issue, as were emotional problems and continuing frustrations about continuing with education. The medical concern was spread more evenly over a breadth of actual or potential problems that required monitoring, investigation, or treatment. This contrasts with the older group of adult patients for which function in a variety of aspects was most important and there was little correlation with health professional concerns. That school should remain a major issue is a matter mostly of patients recognising the enduring impact of missed time at school and associated lack of achievement.

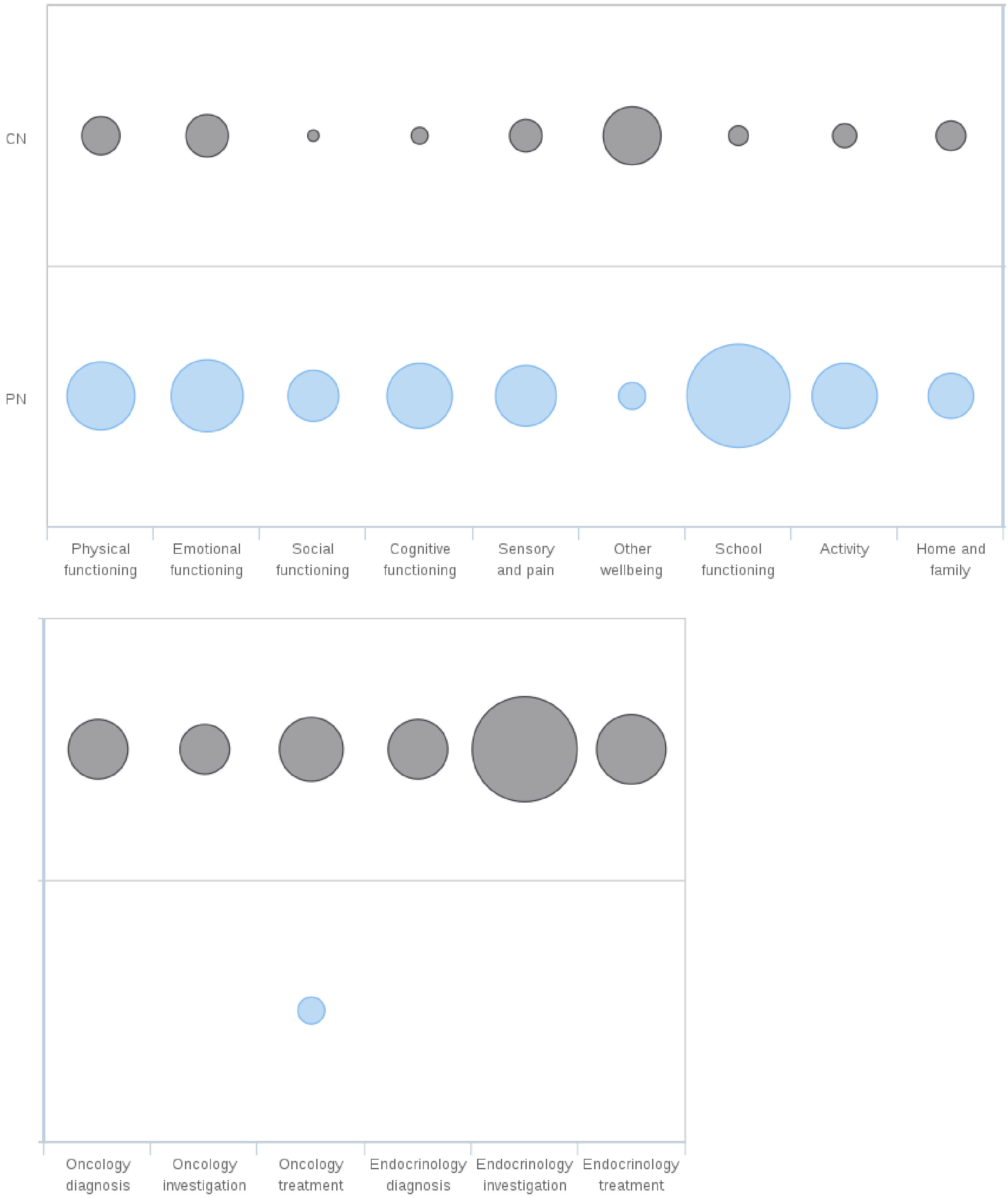
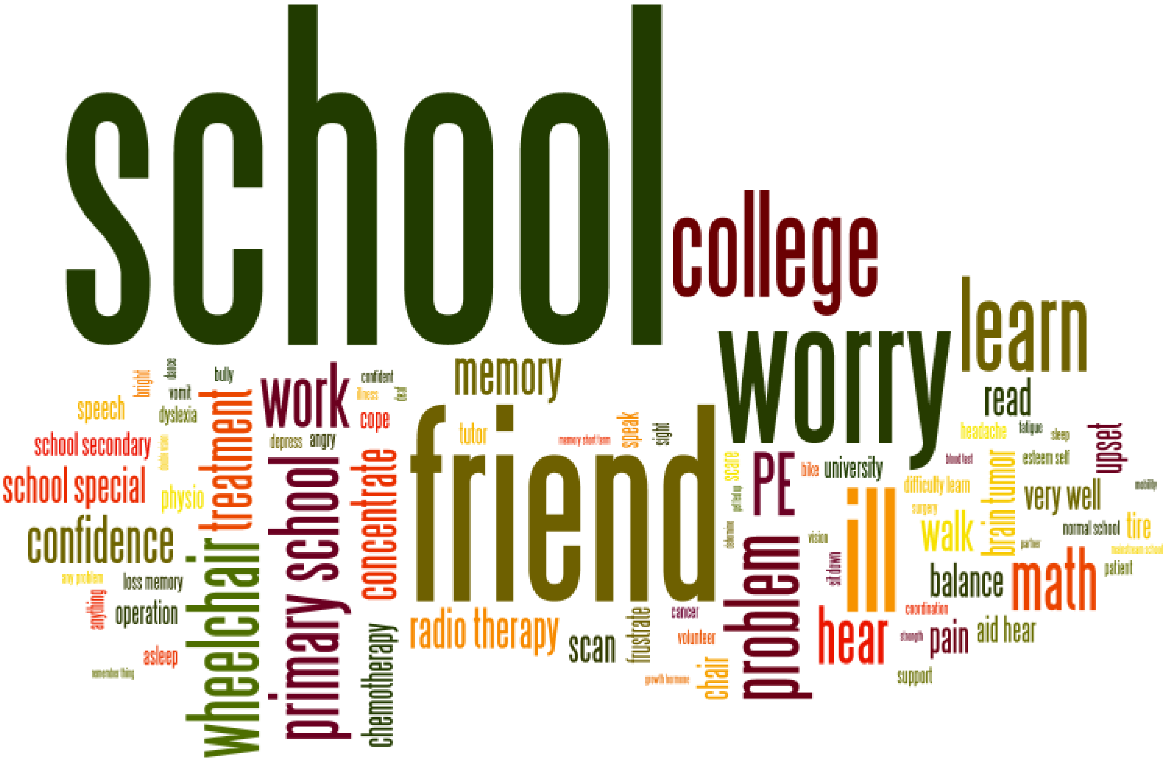


Figure 13: Example of 18-40 group (older than transition group: MB10

LEXICAL ANALYSIS – WHOLE COHORT

Figure 14 illustrates a lexical analysis (of the whole cohort) of frequent terms found in corresponding PNs and CNs. The word clouds use term frequency to set font size and show the most frequent terms in clinical (a) and patient (b) narratives. The size of the terms in the each word cloud is proportionally related to their frequency in each corresponding dataset. Further, a clear contrast in terminology is apparent: PNs contain typical HrQoL mentions while CNs contains common clinical concept.

(a) PNs: word cloud of the most common terms



(b) CNs: word cloud of the most common terms

Figure 14: Lexical analysis using word clouds

Section 7: Conclusions

LEARNING FROM THE STUDY

The study resulted in greater understanding of this patient group, its recognised and hidden problems, and something of the complexity of the interaction between health status, quality of life and psychological wellbeing as patients move on from the diagnosis through survivorship. Steps have also been made to move on with the use of clinical text mining to assist in the management and interpretation of complex clinical data.

HEALTH

Measures for most patients showed severely impaired health, particularly so for the under-18 group but only moderately less so for the adult group. A pattern of evolving problems over time was seen, characterised by an increasing burden in the number and severity of disabling health problems. Despite this, most patients reported that their overall health status was at least good, and often very good.

QUALITY OF LIFE

Diminished quality of life in most functions was evident for most patients in the under-18 group, and issues of psychosocial health were a greater problem than issues of physical functioning. Social and school functioning were constituent parts of this. Deficits in cognitive functioning exerted a serious impact on quality of life, as did pain. Family functioning was negatively affected in almost all cases, causing substantial reduction in parental and family quality of life. Measures of quality of life in the adult patients showed less pronounced impact: motor dysfunction and communication deficit caused significant reduction in quality of life for about a third of patients.

PSYCHOLOGICAL WELLBEING

Much greater problems with emotional distress were recorded for the under-18s than for the older patients. Adult patients denied problems with depression and a minority reported moderate problems with anxiety. This was not reflected in the interview data, however, and some patients identified additional struggling with emotional distress that was not captured by the instrument.

PATIENTS' DESCRIPTIONS OF THEIR PROBLEMS

For the under-18s, issues about school predominated, followed by notions of self-consciousness about and resignation to disability. Despite multiple problems, including cognitive deficits, patients retained aspiration to achieve more in life, and they sought a more positive response from schools and services. For the adult group, reflections on coming to terms with the lifelong impact of the tumour and associated treatment predominated, followed by the difficulties of transitioning into adult life and the limited opportunities to secure work (often linked to outright discrimination). Absence of a social life was increasingly burdensome. For both groups, the availability and signposting of health, social, and education services was problematic, and families often had to discover supportive services for themselves. Overall, a broad range of survivorship issues could be addressed with the provision of appropriate resources.

OUTCOMES OF TEXT MINING

It was clear that while there was crossover between the concepts identified by patients and by health professionals, the emphasis and priority placed on these differed – sometimes markedly – between them. Patients tended to prioritise school, emotional functioning, cognitive functioning, and physical functioning, while the medical record was focused more on oncological and endocrinological treatment, investigation, and physical functioning. In some ways, these might be seen as a function of the relevant roles of doctors and patients in that patients rely on doctors to deal with treatment and monitoring aspects and do not expect them to address issues about school, social life or the aftermath for family life. With current resources, the medical team may be able to do very little about problems relating to employment, social life or school.

LESSONS FOR FURTHER RESEARCH

The study process of measurement, interviewing, and review of medical records provided insights into some of the strengths and weaknesses of the adopted approach, overcoming some problems – such as the anonymisation and preparation of medical record data for analysis with text mining - and offering learning for the further pursuit of harmonisation of patients' and health professionals' concerns and endeavours.

In particular, it will be important to ensure that the same question is asked of patients and health professionals (the former directly through measurement of key concepts and through individual or family interviews; the latter through the proxy of medical records). The groupings, including the transition groups, appeared to help to tease out differences in concern over time. The selection of research instruments for single use, and to allow for more direct comparison between child and adult patients, would bear more consideration.

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Comparison of the expressed experiences of survivors of childhood medulloblastoma with measures of health and quality of life, and with issues identified in consultations

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This report can be referenced as

Long T, Estlin E, Keane J, Gattamaneni R, McCabe M, Kamaly-Asl I, Nenadic G, Dehghan A (2015) Comparison of the expressed experiences of survivors of childhood medulloblastoma with measures of health and quality of life, and with issues identified in consultations. Manchester: The Christie NHS Foundation Trust.

ISBN: 978-1-907842-74-0

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http://gnode1.mib.man.ac.uk/hecta.html