
INVESTIGATING THE
PSYCHOSOCIAL OUTCOMES
OF YOUNG ADULT
SURVIVORS OF CHILDHOOD
AND ADOLESCENT CANCER

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Abstract

While several studies report survivors of childhood and adolescent cancer to have affected outcomes in areas such as health-related quality of life, psychological health, education, employment and relationships, other studies report positive findings. Inconsistencies in the measures and methods used across studies hinder our ability to draw conclusions from the research and there is also a lack of measures which are designed specifically to capture the concerns of these survivors. In addition, survivors' subjective perceptions have been identified as potentially crucial risk factors for poorer psychosocial outcomes, but receive less attention than traditional risk factors involving disease and demographics.

The research for this thesis employed a mixed methods design, in the form of an exploratory sequential design, with the purpose of providing a comprehensive investigation of the psychosocial outcomes of young adult survivors of childhood and adolescent cancer. The first study aimed to qualitatively explore survivors' own perceptions of the impact of cancer and the influence it has had on their lives. The second study aimed to quantitatively investigate the outcomes and concepts identified in the qualitative study, and in a review of the literature, in a larger sample of survivors. In both studies, the survivors own views, experiences and concerns were of central importance.

Overall, survivors reported high levels of achievement and functioning. However, it was evident that a minority of survivors may benefit from further support and information with regards to fertility, education, employment, concerns about the impact of cancer and future health. Results of the questionnaire study indicate that survivors' views, as assessed by the Impact of Cancer for Childhood Cancer Survivors scale, may be associated with health-related quality of life and distress outcomes. Results suggest that overall the mixed methods study enabled a comprehensive investigation of psychosocial outcomes. The research indicates that health professionals should monitor the psychosocial health of even long-term survivors of childhood and adolescent cancer.

Keywords: childhood cancer; adolescent cancer; survivorship; psychosocial

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Abbreviations

BCCSS	British Childhood Cancer Survivor Study
BP	Bodily pain sub-scale
BSI-18	Brief Symptom Inventory-18
CCSS	Childhood Cancer Survivor Study
CNS	Central nervous system
CRUK	Cancer Research, UK
GH	General health sub-scale
GSI	Global severity index
HND	Higher National Diploma
HRQoL	Health-related quality of life
IOC-CS	Impact of Cancer for Childhood Cancer Survivors
IPA	Interpretative phenomenological analysis
LTFU	Long-term follow-up
MCS	Mental component summary
MH	Mental health sub-scale
NCI	National Cancer Institute
NCIN	National Cancer Intelligence Network
NRYPMDR	Northern Region Young Person's Malignant Disease Registry
PCS	Physical component summary
PF	Physical function sub-scale
PTG	Post traumatic growth
PTGI	Post Traumatic Growth Inventory
QoL	Quality of life
RE	Role-emotional sub-scale
RP	Role-physical sub-scale
RVI	Royal Victoria Infirmary
SF	Social functioning sub-scale
SD	Standard deviation
SF-36	Short Form-36
SWEMWBS	Shortened Warwick-Edinburgh Mental Well-being Scale
U.K.	United Kingdom
U.S.	United States
VT	Vitality sub-scale
WHO	World Health Organisation

Chapter overview

Chapter 1.

The opening chapter sets the context for the thesis. The main types of cancer diagnosed in childhood and adolescence, the therapies which aim to cure these cancers and the survival rates for these young patients are summarised. The issue that many survivors are at increased risk of physical and neurocognitive problems due to the cancer and its treatment is then introduced. This is followed by a brief introduction to psychosocial outcomes which are the psychological, emotional and social aspects of surviving childhood and adolescent cancer.

Chapter 2.

Chapter 2 critically appraises the available research for psychosocial outcomes in survivors of childhood and adolescent cancer. The chapter begins by reviewing the literature for the outcomes of health-related quality of life and psychological distress, as well as the growing evidence-base relating to positive psychological changes in this survivor group. The evidence for poorer social outcomes is then evaluated. The limitations of current research are then outlined before presenting why our understanding of psychosocial outcomes may be expanded by employing a mixed methodology study design. The chapter concludes with the aims and objectives of the thesis.

Chapter 3.

Chapter 3 presents the first step of the mixed methodology design. This qualitative study aimed to gain insight into the views of young adult survivors of childhood and adolescent cancer regarding the impact that cancer has had on them and their lives

and the issues they consider to be important. The chapter begins by stating the benefits of using qualitative methods before detailing the procedures of the study and justifying the chosen analytical method (thematic analysis). Findings from 12 survivors are illustrated by anonymised quotes taken directly from the participants. The chapter concludes with a discussion of the findings in relation to previous research and a consideration of the limitations and strengths of the study.

Chapter 4.

Chapter 4 presents a subsequent quantitative study. The qualitative study (chapter 3) and the literature review (chapter 2) helped to identify issues considered important both by survivors and researchers when investigating psychosocial outcomes in survivors, as well as identifying instruments which may help to further understand the topic. Using this information, a questionnaire was developed which was a combination of existing measures and new items designed specifically for this study. The methods used to develop the questionnaire are detailed in Appendix E. This quantitative study aimed to expand our understanding of psychosocial outcomes and was administered to young adult survivors of childhood and adolescent cancer. Chapter 4 presents the results of administering the questionnaire and also evaluates the feasibility and acceptability of the proposed research measures, methods and procedures. The results are then discussed in the context of previous research findings.

Chapter 5.

The final chapter briefly integrates findings from both studies undertaken as part of this thesis, and presents clinical implications and suggestions for future research.

Chapter 1 Introduction

1.1 Cancer in children and adolescents

Cancer in childhood and adolescence is rare, accounting for only 2% of all cancers in industrialised nations (Stiller & Shah, 2012). In the UK, there are approximately 1400 new cases of childhood cancer each year with 1 in 600 young people by the age of 14 years, and 1 in 300 by the age of 24 years, receiving a diagnosis (Bailey & Skinner, 2010). Cancer is a group of more than 100 distinct diseases which all result from the unregulated and abnormal proliferation of cells (Cooper, 2000).

The types of cancer which develop in children differ to those seen in adults. Although many adult cancers are strongly related to lifestyle factors, lifestyle is believed to have little influence on the development of cancers in children and adolescents. However, risk factors for childhood and adolescent cancers are not well understood. It is hypothesised that the development of cancer in adolescence and young adulthood is largely related to maturation, while cancer in the younger child is believed to be associated with a range of prenatal and perinatal risk factors (Bhattacharya *et al.* 2014). Certain infections, congenital anomalies and genetic disorders have also been suggested to be related to cancers in young people (Cancer Research UK [CRUK], 2015a).

Cancers in young people are generally grouped into three broad categories of haematological, central nervous system (CNS) and solid tumour malignancies. Leukaemia, a haematological malignancy, is the most common childhood cancer accounting for approximately 30% of diagnoses (Children with Cancer UK, 2016). However, the incidence of malignancies varies with age and in adolescence,

lymphomas, especially Hodgkin lymphoma, overtake leukaemia to be the main form of cancer (25%) (Stiller & Shah, 2012).

CNS tumours of the brain or spinal cord are the second most common form of cancer in both childhood (26%) and adolescence (14%) (Children with Cancer UK, 2016). There are many different types of CNS tumours, all differing in terms of prognosis. Low grade brain tumours are slow growing, are not likely to spread, may only require surgery and are less likely to reoccur if successfully removed (CRUK, 2015b). These may also be referred to as benign tumours, but although benign tumours are considered less serious in other parts of the body, in the brain these tumour types have the potential to cause serious damage and can be life threatening (Bailey & Skinner, 2010). High grade brain tumours develop faster, can metastasise, are more likely to return after removal and will require chemotherapy and radiotherapy as well as surgery (CRUK, 2015b).

Tumours in childhood may also develop in the cells of organs such as the ovaries, testes, kidney and liver; soft tissues which support and protect the bodily organs such as muscle or fat; or in any bones of the skeleton. These rarer forms of the disease account for a relatively small number of cases of childhood cancer.

1.2 Treatment of childhood and adolescent cancer

There have been significant advancements in the treatment of childhood and adolescent cancer over recent decades. This has largely been a consequence of the inclusion of many child and adolescent patients in national and international clinical trials, and the centralisation of care in the UK (Stiller *et al.* 2012). Now children and adolescents in the UK are treated in one of 21 specialist centres by multidisciplinary

and specialised staff, and one of these centres is located at the Royal Victoria Infirmary (RVI) in Newcastle upon Tyne.

The goal of treating cancer is to maximize survival, whilst minimising the potential for both short-term and long-term toxicities. The treatment regime will depend on many factors, including the type and location of the tumour as well as how advanced the cancer is. However, a combination of therapies will usually be used, with the main forms being chemotherapy, radiotherapy and surgery.

1.3 Survival from childhood and adolescent cancer

Improvements in the therapies available to treat childhood and adolescent cancer have led to a dramatic improvement in survival rates and a large and increasing population of survivors (Skinner *et al.* 2006). As demonstrated in Figure 1, five-year survival rates have doubled from less than 30% for children diagnosed in the late 1960s, to 77% for those diagnosed in the late 1990's (Stiller, 2007).

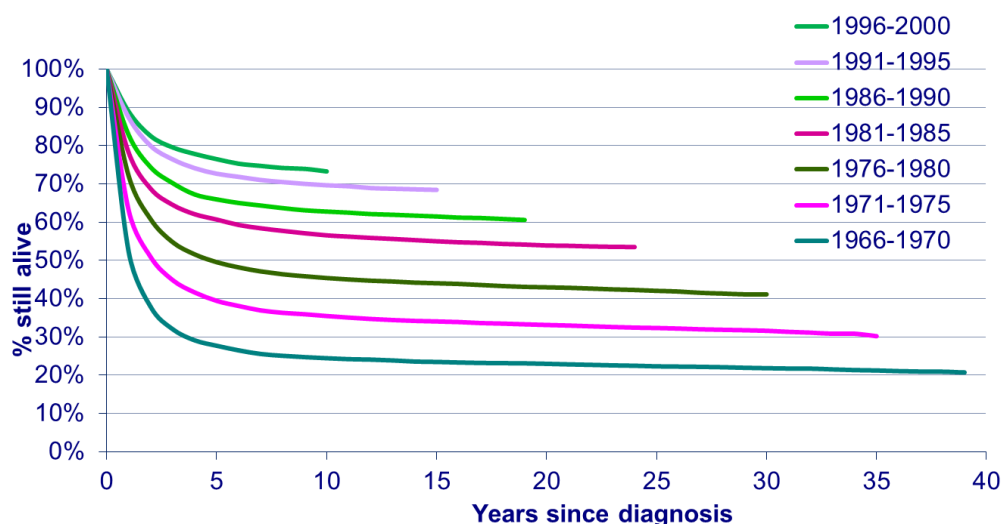


Figure 1 Survival of childhood cancer patients (aged 0-14 years), by period of diagnosis (1966-2000) (Stiller, 2007)

The most recent estimate of five-year survival in the UK is that 82% of children diagnosed aged 0-14 years will survive (National Cancer Intelligence Network [NCIN] 2012). Survival rates for the 15-24 year age group have also improved greatly with five-year survival also currently at 82% (NCIN, 2014). However, survival rates vary significantly across diagnoses with 57% of bone tumour patients surviving ten-years, compared to 99% of retinoblastoma patients (CRUK, 2015c). Therefore, encouraging overall survival rates can mask the issue that many children and adolescents do not survive, and for those who do, it is now known that survival often comes at a cost.

1.4 Late effects of treatment in survivors of childhood and adolescent cancer

The intense and invasive treatments which enable survival are now known to predispose the young person to a number of adverse health-related outcomes. While some ill-effects of the cancer and its therapy may occur at the time of treatment and persist through into adult life, other adverse effects may not surface until months, years or even decades after treatment has ceased (Landier *et al.* 2006). These have been termed as late adverse effects of treatment and long-term clinical follow-up (LTFU) of survivors aims to facilitate early detection or ideally prevention of these adverse effects (Skinner *et al.* 2006).

The burden of ill-health in childhood cancer survivors is reported to be substantial, even before they reach mid to late adulthood. A recent estimate is that 42% of 84,590 U.S. survivors of childhood and adolescent cancer aged 20-29 years have a mild or moderate chronic physical health condition, while 29% have a severe, disabling or life threatening condition (Phillips *et al.* 2015). Phillips' (2015) findings mirror those of well cited large cohort studies by Oeffinger (2006) and Geenen (2007) which also

report high levels of physical morbidity in relatively young survivors (with a mean age of 27 years and median age of 24 years respectively). The physical late adverse effects of cancer and its treatment are wide ranging and can affect any system or organ in the body. This results in a myriad of health problems such as secondary cancers; thyroid abnormalities; premature menopause; pulmonary complications; cardiovascular and cerebrovascular disease; neurological and neurosensory dysfunction; and obesity (Hudson *et al.* 2009; Skinner, 2012).

Survivors, and particularly survivors of CNS tumours or those who have undergone toxic treatment to the CNS, may also develop neurocognitive deficits affecting attention, memory and processing speed, visual perceptual skills and executive function (Askins & Moore, 2008; Nathan *et al.* 2007). It is estimated that neurocognitive deficits affects approximately 40% U.S. childhood cancer survivors aged between 20-29 years (Phillips *et al.* 2015). However, the prevalence may be much higher in CNS survivors (Castellino *et al.* 2014; Nathan *et al.* 2007). Neurocognitive dysfunction can affect several areas of survivors' lives including educational performance and achievement, the ability to live independently and a survivor's social functioning (Nathan *et al.* 2007).

1.5 Introducing psychosocial outcomes in survivors of childhood and adolescent cancer

In medicine, the term 'psychosocial' refers to the psychological, emotional and social aspects of a disease and its treatment (National Cancer Institute [NCI], 2015). Children and adolescents diagnosed with cancer will face many psychological, emotional and social challenges. For these young patients, the cancer occurs at a critical period of life where they are experiencing significant biopsychosocial

development. Although early childhood is characterised by high parental dependence, and adolescence involves emerging freedom and independence, the diagnosis of cancer in either developmental stage brings a time of great change in both physical appearance and physical energy, hospitalisation and health complications (Eiser & Kuperberg 2007). Cancer is a traumatic and unexpected event which will affect the young person's relationships with the world around them and will force a change on family relationships and friendships (Hokkanen *et al.* 2004).

With improved survival, the psychosocial outcomes of long-term survivors have emerged as an important area of survivorship research. In recognition of the need to understand the long-term outcomes of the growing population of childhood and adolescent cancer survivors who remain at risk of both physical and psychosocial sequelae, it has been stated that research should focus on survivors who have survived for five or more years from diagnosis (Hawkins *et al.* 2008; Surbone & Tralongo, 2016). However, the lack of research with these long-term survivors is a continuing gap in cancer research (Surbone & Tralongo, 2016).

Although it is often assumed that long-term survivors of childhood and adolescent cancer will demonstrate poor psychological health, this is not always evidenced in the literature. Reviews have surmised that most survivors are in relative good psychological health, but do highlight that certain sub-groups of survivors appear to be at risk for psychological distress or poor health-related quality of life outcomes (Bitsko *et al.* 2016; Klassen *et al.* 2011; McDougall & Tsonis, 2009). It has also been stated that those who experience cancer during childhood or adolescence may be less likely to achieve social outcomes which are considered by society to signify successful adaptation to adulthood (Stam *et al.* 2005) and lower levels of education,

employment (de Boer *et al.* 2006), marriage and parenthood (Pivetta *et al.* 2011) have been reported in survivors in comparison to similar-aged controls and population data.

Despite research focussing on the negative consequences of cancer, it has been acknowledged that survivors may report a positive psychological change or report benefits as a result of their cancer experience. This has been demonstrated through survey studies (Zebrack & Landier, 2011; Gianinazzi *et al.* 2016) and qualitative studies, with findings suggesting that most survivors simultaneously report both negative and positive consequences of cancer (Fauske *et al.* 2015; Lehmann *et al.* 2014).

Several medical and sociodemographic factors have been associated with poorer psychosocial outcomes in survivors, particularly having a cancer of, or receiving treatment to, the CNS (de Boer *et al.* 2006; Frobisher *et al.* 2007; Klassen *et al.* 2011; Lancashire *et al.* 2010; Zeltzer *et al.* 2009). However, it also recognised that how a survivor views the cancer experience and its impact on their health may be central to an enhanced understanding of psychosocial outcomes (Klassen *et al.* 2011).

It is well established that the beliefs that patients' hold about their medical condition and health threats are associated with a number of health-related outcomes, both physical and psychological. Although not always medically accurate, these health beliefs are rational and logical from the individual's subjective viewpoint and serve as an objective reality (Benyamini, 2011). In addition, for survivors long cured of the cancer, the beliefs they have in relation to themselves, their world and their future can provide an important avenue for understanding psychosocial outcomes and informing potential treatment approaches (Rourke *et al.* 2015).

Research has indicated that for survivors of childhood and adolescent cancer, their subjective beliefs and perceptions about the extent to which they perceive cancer to have impacted on their lives and their views of their present and future health may be stronger risk factors of psychological distress and anxiety disorders, perhaps more so than objective demographic, disease and treatment factors (Bitsko *et al.* 2016; Rourke *et al.* 2007; Zebrack & Landier, 2011). Survivors may also have beliefs, views or experiences which might impact on their progress in social outcomes of education, employment and relationships. Accessing these views may give a more detailed insight into why, and in what way, the experience of cancer as a child or teenager may affect a young adult's life and their psychosocial development.

Despite the potential importance of survivors' perceptions, studies tend to focus on disease, treatment and demographic risk factors for poor psychosocial outcomes and the use of generic quantitative measures which generally do not allow the survivors' subjective beliefs, views and additional experiences to be evaluated. Heterogeneity across studies in terms of the measures they use and the characteristics of the samples they recruit (e.g. diagnoses, age at diagnosis, age at study) have contributed to difficulties in reaching conclusions from the available evidence regarding long-term psychosocial outcomes in childhood and adolescent cancer survivors.

Therefore, the purpose of this thesis is to conduct an investigation of the psychosocial outcomes of survivors of childhood and adolescent cancer. Due to the many ways in which cancer may impact on a young person's life, the thesis will use methods which enable a comprehensive exploration by evaluating several psychosocial outcomes whilst also enabling survivors to share their own views on the psychosocial impact of their cancer experience. The thesis will focus on long-term survivors (≥ 5 years from

diagnosis) who are currently of young adult age and in particular, those who are in what has been termed as 'emerging adulthood'. Arnett (2000) proposes emerging adulthood as a distinct demographic period which begins at age 18 years and lasts until the late twenties, in which individuals seek to establish themselves in the world (Arnett, 2000). Emerging adulthood is stated to be a period of change in terms of love, work and views where different life directions will be explored and by the end of which, people in their late twenties will be likely to have made lasting life choices (Arnett, 2000). Therefore, by studying this age range survivors may be able to give a unique perspective into how cancer has influenced their life, themselves and their expectations for the future. It may be also be possible to identify age-specific issues which would otherwise be lost in studies involving survivors with a wide age range.

Chapter 2 presents a literature review of psychosocial outcomes in long-term survivors of childhood and adolescent cancer before presenting the specific aims and objectives of the thesis.

Chapter 2 Literature review of psychosocial outcomes in survivors of childhood and adolescent cancer

2.1 Introduction

This chapter will review the main findings to date concerning the psychosocial outcomes of long-term survivors of childhood and adolescent cancer. The chapter presents research findings regarding health-related quality of life, quality of life and the psychological and emotional impact of cancer before focusing on the social outcomes. In all sections, the main findings of both quantitative and qualitative research are summarised, as are the limitations of the current evidence-base. The chapter concludes by presenting the need for further research and the potential advantages that using a mixed methodology may bring to research, before presenting the aims and objectives of the thesis.

2.2 Health-related quality of life and quality of life outcomes

Survival rates tell us little of the quality of survivors' lives and whether they are endured or enjoyed (Zebrack & Zeltzer, 2003). Health-related quality of life (HRQoL) in childhood cancer survivors has been described as the physical, psychological, social and emotional impact of a disease on everyday life (Eiser, 2009; Stam *et al.* 2006). However, there is a lack of clarity regarding the definition of HRQoL. Bowling (2001) suggests that HRQoL should also involve survivors' perceptions of health, fitness, life-satisfaction, well-being and future prospects are also of importance.

The term HRQoL is also often used interchangeably with quality of life (QoL) (McDougall & Tsosis, 2009). However, QoL is stated to be a broader construct than HRQoL and includes areas of life which are not affected by health (Varni *et al.* 2007). The World Health Organization (WHO) state QoL is a concept which is affected by the

person's physical and psychological health, their level of independence, social relationships, personal beliefs and their relationship to salient features of their environment (WHO, 1997). This definition highlights the difficulty in distinguishing between those aspects of life that are affected by health and illness, and those aspects which are not.

However, the distinction between QoL and HRQoL may be demonstrated in a recent study by Gunn *et al.* (2015) involving 21 survivors of a childhood brain tumour (with a median age of 24 years). In response to the over reliance on quantitative measures to assess QoL and the recognition that cognitive limitations may impact on brain tumour survivors' participation in questionnaire-based studies, as well as the responses they give, the authors' utilised mixed methods to fully explore survivors the concept of QoL. Although survivors, many of whom had physical health problems, reported poorer scores on HRQoL measures compared to a general population control group in a number of functional areas (mobility, vision, hearing, eating, speech, mental function and sexual activity), in qualitative interviews all but two of the survivors described their QoL as being positive. Survivors appeared to value social aspects over and above physical function in their own evaluations of QoL. Therefore, while the generic HRQoL measures assess several areas of function and health which are theorized to impact on a person's HRQoL, they may do so at the expense of different aspects of life which survivors may consider to be more important.

Despite inconsistent and often contradictory findings across studies, narrative and systematic reviews have surmised that the majority of survivors report outcomes similar to population norms or aged-matched samples on quantitative measures of QoL and HRQoL (Cantrell *et al.* 2011; Langeveld *et al.* 2002; McDougall & Tsonis

2009). However, reviews have identified certain survivor groups at risk of poor HRQoL outcomes including CNS survivors, survivors who have received higher intensity treatment, survivors treated with cranial radiation (Klassen *et al.* 2011; McDougall & Tsonis, 2009) and survivors who report late adverse effects of treatment or health problems (Klassen *et al.* 2011). Socio-demographic factors such as being female, of older age at diagnosis, longer time since diagnosis and socio-economic status (represented by educational level, employment status and household income) have also been implicated in poorer HRQoL outcomes (Cantrell *et al.* 2011; Klassen *et al.* 2011; McDougall & Tsonis, 2009).

However, these reviews do acknowledge that their findings are constrained by the lack of agreement on what constitutes QoL and HRQoL and the large variety of measures utilised to measure these concepts, thus making comparability across studies problematic (Klassen *et al.* 2011; Langeveld *et al.* 2002; McDougall & Tsonis, 2009). These reviews also either include both childhood cancer survivors and patients still in treatment (Klassen *et al.* 2011) or childhood survivors of any age (Cantrell *et al.* 2010; McDougall & Tsonis, 2009) or are specific to young adult survivors of childhood cancer but are now dated (Langeveld *et al.* 2002).

The majority of research into QoL and HRQoL has been based on evidence from studies using a variety of quantitative measures, most of which are not disease specific to cancer (McDougall & Tsonis, 2009). Although the use of generic measures allows comparisons between cancer and non-cancer data, these measures do not address all issues important to young adult survivors of childhood cancer such as fertility and sexual health, resilience and body appearance (Nightingale *et al.* 2011) or relationships, perceived sense of self or parenthood (Quinn *et al.* 2012). In the

recent study by Gunn and colleagues (2015), survivors gave an insight into the aspects of life which they felt were important, such as experiencing positive psychological growth as a result of their cancer, the importance of social relationships, limitations in vocational opportunities and independent life, and having negative thoughts regarding their illness such as feelings of being different.

In recognition of the value of identifying the aspects survivors themselves consider to be central to their own QoL/HRQoL, and the lack of a measure specifically for young adult survivors of childhood cancer, Zebrack *et al.* (2009) conducted interviews with 64 survivors to inform the development of an age and disease specific measure. The resulting Impact of Cancer for Childhood Survivors scale (IOC-CS), assesses survivors' perceptions of how cancer has impacted on their lives in several QoL domains and covers issues relevant to young adult survivors which are absent from generic measures (Zebrack *et al.* 2010).

Like the studies outlined above, Zebrack and Landier (2011) reported that in multivariate models, socio-demographic factors (education, employment, relationship status) and the presence of physical health problems were associated with poorer HRQoL outcomes, as measured by the SF-36, a generic HRQoL measure used in some large cohort studies of adult survivors of childhood cancer. However, in the multivariate model, survivors' perceptions of the effect that cancer had on their life, as assessed by the IOC-CS, were also significantly associated with HRQoL and their addition substantially increased the variance (R^2) explained by the models. Therefore, these findings suggest that how the survivors perceive the impact of cancer may be a critical predictor of HRQoL (Zebrack *et al.* 2011).

To date, the IOC-CS is the only QoL self-report measure which has been developed specifically for young adult survivors of childhood and adolescent cancer. However, despite being informed by qualitative data from survivors, expert opinion and existing literature, all of which are essential to the development of QoL measures (Klassen *et al.* 2010), the IOC-CS has not been widely used and is still to benefit from further validation within further samples of young adult survivors (Zebrack *et al.* 2010).

2.3 Psychological and emotional outcomes

2.3.1 *Psychological distress*

Psychological distress is defined as a state of emotional suffering characterised by symptoms of depression (e.g. sadness, hopelessness) and anxiety (e.g. feeling tense, restlessness) which may also be accompanied by somatic symptoms (e.g. insomnia, headaches) (Mirowsky & Ross, 2002). Psychological distress in survivors is typically indicated by an elevated score on a psychological symptom questionnaire, such as the Brief Symptom Inventory (BSI) (Derogatis, 1993), or more commonly the shortened version, the BSI-18 (Derogatis, 2001). While the BSI assesses respondents across nine different scales (somatisation, obsessive-compulsive, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation and psychoticism) which are then summarised into a global severity index, the BSI-18 screens across three sub-scales of somatization, depression and anxiety, and an overall global severity index. These measures establish “caseness” in which respondents scoring above a certain value are identified as being at positive risk for distress.

In a report from the Childhood Cancer Survivor Study (CCSS; a large multi-institutional U.S. cohort study of childhood and adolescent cancer survivors ≥ 5 years from

diagnosis), analysis of data from over 9,500 survivors with a mean age of 27 years (range 18-48 years) found that after adjusting for age, sex and race, survivors were almost twice as likely (Odds Ratio [OR], 1.8; 95% Confidence Interval [CI], 1.6-2.1) to report clinically significant psychological distress in comparison to their siblings (Hudson *et al.* 2003). A later study from the CCSS cohort also confirmed that survivors reported higher levels of distress across the BSI-18 scales than their siblings, but lower levels compared to population norms (all $p < 0.003$) (Zeltzer *et al.* 2008).

Michel *et al.* (2010) reported that in a large Swiss population-based cohort of 987 childhood cancer survivors ≥ 5 years from diagnosis, adult survivors (mean age 28 years, range 20-49 years) scored lower than population norms on the somatisation, obsessive-compulsive, anxiety scales and the global severity index scales of the BSI (all p values < 0.001). A subsequent Swiss study reported that although overall long-term survivors (mean 20 years, range 16-19 years) demonstrated psychological distress levels comparable to siblings on the BSI-18 (all p values > 0.05), male survivors had higher levels of distress than male siblings on somatisation, depression and the global severity index (all p values < 0.03) (Gianinazzi *et al.* 2013). Therefore, although survivors appear to report higher distress levels than siblings, this seems to be similar or less than population norms.

However, it is highlighted by these large studies that although overall mean scores for distress may be lower than population norms, a significant minority of survivors are vulnerable to elevated distress (Michel *et al.* 2010; Zeltzer *et al.* 2009). Key risk factors are suggested to be female gender (Gianinazzi *et al.* 2013; Michel *et al.* 2010; Zeltzer *et al.* 2008; Zeltzer *et al.* 2009), older current age (Michel *et al.* 2010; Zeltzer *et al.* 2008), lower education, income and unemployment and being unmarried

(Zeltzer *et al.* 2008). The perceived presence of late effects has also been associated with increased distress. Michel *et al.* (2010) reported that in multivariate analyses, adult survivors who self-reported somatic problems had twice the odds (OR, 2.00; 95% CI, 1.29-3.11) and those who self-reported a psychological problem were almost at seven times an increased risk (OR, 6.74; 95% CI, 4.06-11.17) to be classed as a 'case' on the global severity index compared to survivors who reported no late effects. This finding was later supported by Gianinazzi and colleagues (2013) in a sample of adolescent survivors, although the risk of being a 'case' was much increased in those reporting psychological problems compared to survivors reporting no late effects (OR, 14.89; 95% CI, 4.72-46.99). Survivors with concerns with their physical appearance may also be at increased risk of distress when compared to those with no such concerns (OR, 5.48; 95% CI, 1.50-20.11) (Recklitis *et al.* 2003).

However, the evidence for the effect of objective cancer-related factors on psychological distress is mixed (Zebrack & Landier, 2011). While Zeltzer *et al.* (2008) report that having received cranial radiation was associated a slightly higher risk of depression (OR, 1.2; 95% CI, 1.0-1.5), the results from Recklitis and colleagues (2003) suggest these survivors are at five times the risk for screening positively for psychological distress (OR, 5.37; 95% CI, 1.63-17.70). However, this study was small in size (n=101) compared to Zeltzer's (2008) analysis of 7147 survivors.

Treatment received (chemotherapy, radiotherapy or surgery) was not found to be associated with 'caseness' for distress in Swiss survivors (Gianinazzi *et al.* 2013; Michel *et al.* 2003), and while Zeltzer and colleagues (2008) report that survivors of certain cancer diagnoses are more likely to be affected by distress (e.g. some types

of brain tumours, leukaemia and osteosarcoma), results from the Swiss survivor cohort did not (Michel *et al.* 2010; Gianinazzi *et al.* 2013).

Therefore, we do not fully understand the mechanisms underlying outcomes such as psychological distress which may be present many years after the cancer treatment has ended (Oancea *et al.* 2014). There is now recognition that as well as disease, treatment and demographic variables representing risk factors for poor psychological health, interpersonal factors such as survivor self-report of health problems and subjective health beliefs are also of importance, although these remain relatively understudied (Bitsko *et al.* 2016).

A recent review by the Children's Oncology Group, the world's largest organisation researching childhood and adolescent cancer, highlighted that survivor self-report measures may be the most accurate method of assessing physical functioning, the presence of late adverse effects of treatment and health beliefs (Bitsko *et al.* 2016). These self-reports are stated to be more strongly associated with psychosocial outcomes than provider reports (Hobbie *et al.* 2000; Rourke *et al.* 2007). Two studies which recruited young adult survivors of childhood cancer via the same cancer registry at a U.S. paediatric cancer centre, compared survivors with and without symptoms of post-traumatic stress. Both papers report that an individual's subjective appraisal of the intensity of the cancer treatment they received and the perceived past and current threat to their life were found to be associated with post-traumatic stress, more so than objective diagnostic and treatment variables (Hobbie *et al.* 2000; Rourke *et al.* 2007). In a longitudinal study of psychological distress involving 4569 survivors in the CCSS cohort, increased distress symptoms over time were associated with perceptions of worsening physical health (depression: OR, 3.3; 95% CI, 2.4–4.5;

anxiety: OR, 3.0; 95% CI, 2.2–4.0; somatisation: OR, 5.3; 95% CI, 3.9–7.4) (Brinkman *et al.* 2013). Zebrack and Landier (2011) also report that perceived impact of cancer, as assessed by the IOC-CS, is a critical risk factor for psychological distress ($p < 0.001$).

The importance of survivors' perceptions of their health to their psychosocial outcomes warrants conversations between the survivors and their healthcare provider to assess survivors' perceptions and understanding (Hobbie *et al.* 2000) and to educate survivors about aspects of the cancer or treatments which may cause them distress (Rourke *et al.* 2007). However, it is documented that survivors of childhood and adolescent cancer can often lack knowledge about the cancer they had, the treatment they received and the potential for late effects (Bashore *et al.* 2004; Hudson *et al.* 2003; Kadan-Lotticke *et al.* 2002; Knijnenburg *et al.* 2010; Syed *et al.* 2016). Survivors who do not have detailed knowledge of their treatment history and its possible effects on current and future health may not understand the importance of health behaviours and that risks such as future cardiovascular disease may be modified through healthy lifestyles. Equally, survivors who have negative perceptions or fears about their present or future health may develop symptoms of distress if these concerns are not addressed.

2.3.2 Positive psychological change

Although research has largely focussed on the potential for negative psychological consequences of cancer, there is increasing literature on the concepts of personal growth, benefit finding and post-traumatic growth in young adult survivors of childhood and adolescent cancer. Post-traumatic growth (PTG), stated to be a positive psychological change as a result of the struggle with a traumatic event such as cancer (Tedeschi & Calhoun, 1996) has been empirically investigated in adult

survivors of childhood cancer through the use of the self-report Post Traumatic Growth Inventory (PTGI) devised by Tedeschi and Calhoun (1996). The PTGI comprises of the five sub-scales of: relating to others; new possibilities, personal strength; spiritual change and appreciation of life. Gianinazzi *et al.* (2016) reported that only 1% of 309 Swiss childhood cancer survivors reported no PTG in any of the scales and that the highest growth was in 'relating to others' and 'new possibilities'.

Higher PTG scores have been associated with female gender, older age at diagnosis (Gianinazzi *et al.* 2016; Yi *et al.* 2015; Zebrack *et al.* 2011), less time since diagnosis (Zebrack *et al.* 2011), and longer duration of treatment (Gianinazzi *et al.* 2016). The association of higher PTG with older age at diagnosis may suggest that a certain level of cognitive capacity and development is needed to enable the child to acknowledge the severity of the illness and reflect on their experiences (Gianinazzi *et al.* 2016; Zebrack *et al.* 2011). Developmental issues need to be considered when applying PTG theory to young children, namely what is growth as opposed to normal maturation and the child's level of awareness and understanding of the traumatic event (Kilmer & Gil-Rivas, 2010).

Benefit finding after cancer has also been reported in survivors of adult cancer (Baker *et al.* 2014). For adult cancer survivors, the concept of biographical disruption (Bury, 1982) is stated to be useful for explaining how cancer may disrupt a person's anticipated life path and result in a transformation of life views and views of self (Hubbard & Forbat, 2012). Bury (1982) explains that biographical disruption occurs where a critical event such as a serious illness disrupts the structures of everyday life and the knowledge which underpins them. Pain, suffering and death, all previously distant prospects for the individual, are brought into the forefront of their

consciousness commanding that individuals re-examine plans and expectations for future life (Bury, 1982).

Compared to adults, children and adolescents have a relatively short biographical history prior to illness. In contrast to biographical disruption which attributes the change in views directly as a result of the cancer, Zebrack *et al.* (2012) propose that for those who experience cancer at a young age, the cancer becomes an organising principle for the person's sense of self and view of life (Boals & Schuettler, 2011). Similarly, the theory of cognitive adaptation (Taylor, 1983) proposes that adjustment to a threatening event, such as cancer, centres around a search for meaning in the experience, an attempt to regain mastery over the event and over one's life, and an effort to restore self-esteem through self-enhancing evaluations.

An alternative explanation for the positive outcomes seen in survivors, particularly with regards to self-report HRQOL/QoL measures, is a response-bias, in which positive aspects of life are exaggerated and the negative aspects minimised, a form of self-denial (O'Leary *et al.* 2007). An alternate form of cognitive adaptation may also be that the experience of cancer results in a 'response shift' in the individual's internal standards or conceptualisation of QoL which may lead to the individual adjusting to, and accommodating, any ill effects of cancer or its treatment (Sprangers & Schwartz, 1999).

As discussed above, the IOC-CS (Zebrack *et al.* 2010) may offer an opportunity to investigate the extent to which young adult childhood cancer survivors perceive cancer to have impacted several areas of their lives, both negatively and positively, and how this associates with psychosocial outcomes such as distress. However,

qualitative methods will also allow a deeper insight into how and in what way survivors perceive the cancer to have impacted on them and their lives.

A Swedish study reported that, through analysis of 59 telephone interviews, survivors of childhood cancer belonged primarily to one of three groups: 'feeling like anyone else' (n=29, 49%); 'feeling almost like others' (n=26, 44%) and 'feeling different' (n=4, 7%) (Doukkali *et al.* 2013). Across four additional categories of 'thoughts of having cancer', 'presence of complications in daily life', 'ability to handle complications' and 'view of life', these three groups described the varying ways in which cancer had affected their lives. A positive 'view of life' was reported across the three main groups, although participants in the 'feeling different' group also shared negative views such as being left with scarring, or feelings of grief regarding their cancer. The authors conclude that the majority of survivors appeared to cope well in life with a small minority experiencing complications such as negative thoughts, having physical and mental health complications which affected daily life and which they struggled with (Doukkali *et al.* 2013). However, although the sample was relatively large for qualitative research, there was a large range in ages at interview (12-22 years, median age of 17 years), with 81% still attending school. Therefore, this paper may be more relevant to the views of adolescents than survivors who are entering young adulthood, which the studies in this thesis aim to explore.

While the participants of Doukkali and colleagues' (2013) study were survivors diagnosed between 7-16 years old, another Swedish qualitative study presents data from seven survivors diagnosed at a younger age (<1-14 years) which aimed to describe how young adults experience being a survivor of childhood cancer (Enskar & Bertero, 2010). An overall theme was that negative experiences were described as

being compensated for with positive views and expectations in which survivors tried to live as normal a life as possible, whilst also struggling with bodily changes and negative emotions as a result of the cancer. In parallel with this, family relationships were reported to be stronger and survivors reported experiencing personal growth (Enskar & Bertero, 2010).

A recent paper reports results from a longitudinal telephone interview study with Swedish survivors of adolescent cancer ten years after diagnosis (aged 23-29 years at study) which concluded that the majority of young adults reported both negative and positive consequences (Lehmann *et al.* 2014). At ten years post-diagnosis, survivors reported additional negative and positive consequences of cancer: existential thoughts about loss and life, health worries, fertility concern, frustrations with health care were concerns not reported at earlier time points (three and four years after diagnosis), while compassion for others was a new gain. At all time-points, bodily concerns were the most salient theme for these survivors, the majority of which had experienced haematological cancers.

To summarise, HRQoL, psychological distress and psychological growth have been cited as important psychosocial outcomes in survivors. On the whole, survivors are stated to have HRQoL and distress outcomes comparable to peers or population norms, although sub-groups of survivors have been identified as being at increased risk of poorer outcomes, in particular CNS survivors (Bitsko *et al.* 2016; Klassen *et al.* 2011; McDougall & Tsonis, 2009). However, comparison and integration of much of the research findings continues to be problematic due to methodological heterogeneity across studies. Studies may use diverse samples in terms of their characteristics (e.g. diagnosis, age at diagnosis, current age) which limits the ability

of research to identify issues pertinent to certain groups of survivors. For instance, issues which influence HRQoL in young adults are likely to differ to those in older adults (Zeltzer *et al.* 2009). Further difficulties arise where studies continue to employ a variety of measures to evaluate outcomes such as HRQoL which are difficult to define (Bitsko *et al.* 2016).

These issues have been aided by the large cohort studies which have been established, such as the CCSS. These studies can lead to the identification of sub-groups of survivors who may be at greater risk of poor HRQoL or psychological outcomes. However, such studies rely on quantitative measures which limit the investigation of the survivors own views and beliefs and the potential influence that these subjective factors may have on survivors' HRQoL, psychological and emotional outcomes.

2.4 Social outcomes

Adaption to adult life is associated with achieving a number of developmental milestones including educational attainment; entering employment and developing a career; achieving independent living; forming intimate relationships; and marriage and parenthood. Difficulties in achieving these desired social outcomes may be exacerbated by both the experience of cancer and its treatment as well as physical and psychological late effects of treatment (Gurney *et al.* 2009). Thus, it is suggested that survivors may be less likely to achieve developmental milestones which are associated with successful adjustment to adult life (Stam *et al.* 2005).

2.4.1 Relationships

Finding a life partner has been stated to be central to life satisfaction (Syse *et al.* 2008) while achieving intimacy in a relationship is a crucial milestone to adulthood (Conger *et al.* 2000). Studies have often focussed on the attainment of relationship milestones, particularly marriage, as an indicator of young adult survivors' social functioning (Thompson *et al.* 2009). While several studies report that survivors of childhood and adolescent cancer are less likely to marry than siblings or the general population (Dieluweit *et al.* 2010; Felder-Puig *et al.* 1998; Frobisher *et al.* 2007; Hays *et al.* 1992; Janson *et al.* 2009; Langeveld *et al.* 2003; Pivetta *et al.* 2010; Rauck *et al.* 1999), others do not (Dolgin *et al.* 1999; Johannsdottir *et al.* 2010; Thompson *et al.* 2009).

However, several of the studies which report no differences have utilised small samples with a relatively young age (18-35 years) and excluded or had low numbers of CNS survivors (Dolgin *et al.* 1999; Thompson *et al.* 2009). In contrast, large cohort studies have allowed for the marriage rates across different age and cancer groups to be compared to general population data for British (Frobisher *et al.* 2007), American (Janson *et al.* 2009) and Italian childhood cancer survivors (Pivetta *et al.* 2010). All of these survivor cohorts are of significant size ranging from just over 6000 survivors (Pivetta *et al.* 2010) to almost 10,000 (Frobisher *et al.* 2007). However, these studies tend to count only legal marriage between those of the opposite sex (Frobisher *et al.* 2007; Pivetta *et al.* 2010), although, a recent paper found that survivors in a population-based Swiss cohort reported significantly lower rates of both marriage and life partnership than sibling controls (Wengenroth *et al.* 2014).

Several cancer-related factors have been associated with reduced odds of being married including being a survivor of CNS malignancy (Frobisher *et al.* 2007; Janson *et al.* 2009; Langeveld *et al.* 2003; Pivetta *et al.* 2010; Rauck *et al.* 1999; Syse *et al.* 2008), with British CNS survivors being 50% less likely to marry than British leukaemia survivors (OR, 0.5; 95% CI, 0.37-0.67). Younger age at diagnosis (Janson *et al.* 2009), and being treated with radiation (Dieluweit *et al.* 2010; Frobisher *et al.* 2007; Janson *et al.* 2009) are also associated with a reduced likelihood of marriage. Wengenroth *et al.* (2014) reported similar factors associated with lower rates of life partnership in survivors.

The presence of emotional distress or impaired social functioning (Frobisher *et al.* 2007; Janson *et al.* 2009) and higher levels of education have also been associated with lower rates of marriage (Frobisher *et al.* 2007). In Janson *et al.* (2009) univariate analyses suggested that survivors who perceived they had a fertility problem had slightly reduced odds of being married (OR, 0.91; 95% CI, 0.87-0.95) than those who did not. However, this was not included in a subsequent multivariate analyses due to the potential direction of the association as fertility status may not be determined until after marriage (Janson *et al.* 2009).

Overall, romantic relationships other than marriage have received much less attention. Despite reporting lower rates of marriage, some studies have simultaneously found no significant differences in the rates of long-term relationships (Dieluweit *et al.* 2010; Felder-Puig *et al.* 1998; Gerhardt *et al.* 2007). This may suggest that if survivors are in relationships at a similar rate to controls but are less often married, then there may be factors which are influencing their decision to not marry, or factors preventing them from marrying. Survivors may simply

experience 'lost years' which may result in them taking longer than peers to finalise their education, begin their career and enter committed relationships, or delays in their sexual development. Conversely survivors may have adverse health effects as a result of the cancer and its treatment which may impact on marital rates or harbour concerns which may interfere with relationships such as concerns about disclosing a history of cancer, having a negative body image, and worries about their fertility status (Thompson *et al.* 2013; Zebrack *et al.* 2004). However, survivors' views or concerns are not explored by these larger quantitative studies.

Gaining wider information about relationships other than marriage and exploring survivors' views on whether they feel their romantic relationships have been affected, and their plans for marriage may provide a fuller understanding of relationship outcomes in survivors. Dolgin *et al.* (1999) reported that in a sample of 64 childhood cancer survivors aged over 18 (mean age 24 years), 46% felt that cancer had impacted on their attainment of social and family goals. This was despite there being a tendency for more survivors being married or cohabiting with a partner (31%) compared to a controls (20%), although this difference was not statistically significant. Therefore, survivors may feel that cancer has affected their lives, even when there appears to be no differences in their outcomes compared to control groups. This underlines the importance of a survivor's own evaluation of their circumstances. Developmental milestones such as independent living are also important to research as they are the precursors to marriage (Gerhardt *et al.* 2007). Some research suggests that survivors may live longer with parents compared to controls (Dieluweit *et al.* 2010; Langeveld *et al.* 2003; Felder Puig *et al.* 1998) which may be linked to a continuing dependency on parents and a delay in achieving independence.

2.4.2 Parenthood

Although treatment options now exist to prevent infertility, it still remains a long-term risk for those who are treated for childhood cancer (Ginsberg *et al.* 2010) and is one of the most common and life-altering complications for survivors (Hudson, 2010). The risk to fertility is generally related to the organs involved in the cancer and the form and intensity of the treatment received, as well as the age at which this was experienced and the sex of the patient (Hudson, 2010; Metzger *et al.* 2013).

Several U.S. and European studies have reported that survivors of childhood and adolescent cancer are less likely to be parents than comparison groups (Dieluweit *et al.* 2010; Langeveld *et al.* 2003; Frobisher *et al.* 2007; Hohmann *et al.* 2011; Johannsdottir *et al.* 2010; Madanat *et al.* 2008; Pivetta *et al.* 2010; Reulen *et al.* 2009) with estimates from large scale studies suggesting that the probability of being a parent after childhood cancer reduces by around 50% (Reulen *et al.* 2009; Madanat *et al.* 2008).

While some studies utilise rates of parenthood as an accurate proxy for fertility in survivors of childhood and adolescent cancer (Madanat *et al.* 2008), other studies such as Pivetta *et al.* (2010) use parenthood and marriage as indicators of the influence cancer has had on the social and behavioural choices of survivors. However, the wider issues for survivors of childhood and adolescent cancer not becoming parents have not been widely explored and few studies have asked survivors for their own views. In a study of over 2000 survivors with a mean age of 26 years (range 19-43), 70% stated that they were too young to start a family, 25% hadn't found the right partner and 14% stated it was due to either having no partner or for financial reasons. Only 4% had responded that 'getting pregnant has not worked out' (Hohmann *et al.*

2011). Survivors may also report that cancer has increased the importance they place on parenthood (Langeveld *et al.* 2002). Although survivors may report that their fertility is not an important issue to them at diagnosis, its importance can increase with age and more so when they became aware that their fertility may have been affected (Crawshaw & Sloper, 2006; Nieman *et al.* 2007).

2.4.3 Educational outcomes

Educational attainment is stated to be a basic determinant of quality of life, and a lack of education can limit an individual's access to good jobs and may increase the risk of social exclusion and poverty (Eurostat, 2013). Several studies have reported there to be no significant deficits in educational achievement of survivors of childhood and adolescent cancer above school age (Boman *et al.* 2010; Dolgin *et al.* 1999; Hays *et al.* 1992; Gray *et al.* 1992b; Kuehni *et al.* 2012a; Jonhansdottir *et al.* 2010). However, two of these studies excluded CNS survivors from their sample (Dolgin *et al.* 1999; Hays *et al.* 1992) and large national cohort studies have reported that CNS survivors have poorer educational outcomes than other diagnostic groups (Boman *et al.* 2010; Lancashire *et al.* 2010; Kuehni *et al.* 2012a). This is perhaps not unexpected as cranial radiation has been linked to neurocognitive deficits (Zeltzer *et al.* 2009) and physical and hearing disabilities (Lorenzi *et al.* 2009), while surgery to remove brain tumours can affect attention, processing speed, visual/perceptual skills and memory (Bruce *et al.* 2008). Other risk factors identified in the literature for poorer educational outcomes has been female gender, younger age at diagnosis and older age at study (Lancashire *et al.* 2011; Lorenzi *et al.* 2009).

In a cohort of over 10,000 British childhood cancer survivors, survivors were found to have lower levels of educational attainment as compared to the general population

(Lancashire *et al.* 2010). However, sub-analyses revealed that the differences were accounted for by CNS survivors and leukaemia survivors treated with cranial irradiation. In contrast, other sub-groups of survivors (bone sarcoma and retinoblastoma survivors) achieved higher in school examinations than the general population. However, the authors do not offer an explanation as to why this may be.

Survivors may experience several issues with regards to their schooling. First, although absence from school is greatest in the year after diagnosis, it is a problem at all stages of the illness (Moore *et al.* 2009). Survivors may also be more likely than siblings or peers to repeat school years (Gerhardt *et al.* 2007; Gray *et al.* 1992), require extra tutoring (Lähteenmäki *et al.* 2002), access learning disabilities programs or special education (Langeveld *et al.* 2003; Lorenzi *et al.* 2010) or experience bullying at school (Lähteenmäki *et al.* 2002). In 288 Swiss childhood cancer survivors, 30% had repeated a school year, 35% had received supportive tutoring, and 7% had attended a special school, although proportions differed across diagnoses with CNS survivors reporting higher numbers (Kuehni *et al.* 2012a).

There is however little information on how survivors view the impact of cancer to be on their education. In a qualitative study with 14 childhood brain tumour survivors (aged 17-29 years), all reported cognitive effects such as memory loss and problems with reading, writing and mathematics. Some also reported that they felt they 'appeared normal' to others which meant that their need for help with school work was not recognised (Boydell *et al.* 2008). Dumas *et al.* (2015) interviewed 80 childhood cancer survivors with a mean age of 38 years (range 27-53) and while 30% felt that their illness had influenced their choice of education and career, 70% (n=52) did not.

2.4.4 Employment

Employment is central to identity, social roles and social status, and is the most important means by which an individual will obtain the economic resources to be able to participate fully in society. Employment also meets an individual's important psychosocial needs in societies where being employed is the norm (Waddell & Burton, 2006). A systematic review which included meta-analyses involving 24 studies of long-term (≥ 5 years from diagnosis) adult survivors (aged ≥ 18 years) of childhood and adolescent cancer found that, overall, survivors were almost twice as likely to become unemployed than healthy controls (OR, 1.85; 95% CI, 1.27-2.69) (de Boer *et al.* 2006). However, the risk of unemployment was not evidenced equally across diagnostic groups in that while the CNS tumours were almost five times more likely than controls to be unemployed (OR, 4.74; 95% CI, 1.21-18.65), for survivors of blood and bone cancers the risk of unemployment was elevated, but not significantly so (OR, 1.4; 95% CI, 0.79-2.55). However, de Boer and colleagues (2006) acknowledge their results may be affected by heterogeneity and even separate analyses for the diagnostic groups resulted in considerable heterogeneity (I^2 of approximately 87%), suggesting that the studies in the meta-analysis varied substantially. In addition, de Boer *et al.* (2006) state that the quality of the studies included in the analyses varied extensively.

Further risk factors for unemployment in survivors of childhood and adolescent cancer have been younger age at diagnosis (de Boer *et al.* 2006; Pang *et al.* 2008; Holmqvist *et al.* 2010), having received cranial radiation (Pang *et al.* 2008; Holmqvist *et al.* 2010), being female, not finishing school (Pang *et al.* 2008), having neurocognitive defects (Dieluweit *et al.* 2011; Kirchhoff *et al.* 2011a); having a chronic medical condition (Pang *et al.* 2006) and poor physical health (Kirchhoff *et al.* 2011a).

Gurney *et al.* (2009), wrote that although employment status of survivors has been addressed in the literature, the quality and satisfaction of this employment has yet to be assessed. In a qualitative study with brain tumour survivors, although all were employed, not all were in their desired occupation with some reporting a lack of energy had affected their ability to do work (Carlson-Green *et al.* 2009). Survivors have also reported job discrimination (Langeveld *et al.* 2003), being denied entry to the armed forces (Dolgin *et al.* 1999; Hays *et al.* 1992), feelings of workplace rejection due to cancer history (Dolgin *et al.* 1999), and perceptions that cancer had affected their employment opportunities and possibilities (Dolgin *et al.* 1999; Dumas *et al.* 2015; Eiser *et al.* 1997; Felder-Puig *et al.* 1998).

From the results of their qualitative study with French survivors, Dumas and colleagues (2015) report that the perception of restricted employment choices may paradoxically result in a positive impact on occupational status as male survivors may pursue professional occupations, as opposed to those involving manual and physical work. In contrast, quantitative studies suggest that survivors of childhood cancer are less likely to be in a professional/white collar occupation than siblings (Ishida *et al.* 2011; Kirchoff *et al.* 2011b) and may be less likely to be in a physical occupation (Kirchoff *et al.* 2011b). Female survivors in particular have reported that the experience of cancer has orientated them towards careers in health or involving the care of others (Dumas *et al.* 2015; Eiser *et al.* 1997). In support of this, Ishida's study of Japanese survivors reported survivors were more likely to in medical jobs than their siblings (Ishida *et al.* 2011).

As the case with psychological outcomes, findings regarding the social outcomes of survivors are difficult to summarise due to methodological heterogeneity across

studies. There is evidence that cancer survivors may be more likely to never marry and experience unemployment. However, as outlined above, the perceptions of survivors are deemed to be important to their psychological outcomes, therefore, their views of how these social outcomes are affected may be insightful, but lacking from the current literature. Research also focuses on the endpoints of marriage, parenthood and employment and the attainment of these as an indicator of effective adjustment and of successfully reaching adulthood. However, in line with the theory of emerging adulthood, it may be insightful to gain the perspectives of young adult survivors of cancer who are in a period of life when they are exploring different directions and potentially making important decisions about their lives.

2.5 Rationale for the thesis

Research from large-scale cohort studies such as the British Childhood Cancer Survivor Study (BCCSS)(Frobisher *et al.* 2007; Hawkins *et al.* 2008; Lancashire *et al.* 2010), the CCSS (Janson *et al.* 2009; Zebrack *et al.* 2004; Zeltzer *et al.* 2009; Robison *et al.* 2002) and Swiss Childhood Cancer Survivor Study (SCCSS) (Gianinazzi *et al.* 2013; Gianinazzi *et al.* 2016; Kuehni *et al.* 2012a; Kuehni *et al.* 2012b; Michel *et al.* 2010) have been valuable as they have enabled the assessment of the prevalence of adverse psychosocial outcomes in survivors and the identification of subgroups of survivors who may fare less well.

However, we still lack a full understanding of the influences on psychosocial outcomes in survivors (McDougall & Tsonis, 2009; Maurice-Stam *et al.* 2009) and continued use of generic measures will not enable a comprehensive exploration of psychosocial outcomes in survivors. The views of the impact of cancer from the perspective of the survivors is lacking from the literature, despite survivors'

perceptions being identified as a potentially important determinant of psychological outcomes (Hobbie *et al.* 2000; Rourke *et al.* 2007; Zebrack & Landier, 2011). Accessing their views may also help our understanding of their progression in what are thought of as important developmental milestones which may aid our interpretation of the findings from quantitative studies.

While quantitative studies are typically concerned with experimental and objective testing of pre-defined variables to test cause and effect (Finlay, 2011), qualitative research aims to gain a subjective understanding from the participants' perspective, aiming to gain knowledge of meanings, experiences and processes (Willig, 2001). Each approach has its strengths and limitations and thus, by utilising both qualitative and quantitative techniques, we may develop a fuller picture and a more comprehensive understanding of the phenomena under investigation (Green, 2007; Johnson *et al.* 2007). This is of particular interest in health research due to the multi-faceted nature of health and illness (Morgan, 1998). Mixed methods are suitable when qualitative or quantitative methods alone cannot fully understand the problem (Cresswell & Plano Clark, 2011), and as outlined above, we do not currently have a full understanding of the psychosocial impact of cancer on survivors.

However, in practice, combining qualitative and quantitative methods has proved challenging (Morgan, 1998). Not only can it be technically challenging to do effectively (Morgan, 1998) but quantitative and qualitative research stem from different theoretical perspectives (Braun & Clarke, 2013). While, quantitative research is aligned with positivism which is concerned with discovering the one true reality (Braun & Clarke, 2013), qualitative research is associated with a constructivist paradigm which states that what we know of the world is socially constructed,

therefore, there is not one truth but multiple interpretations of reality (Johnson & Onwuegbuzie, 2004).

Purists from both sides of the argument view their paradigms as the ideal for research, with a belief that both methods should not be combined (Johnson & Onwuegbuzie, 2004). However, pragmatists suggest that research methods should not be dependent on paradigms, and methods should be combined if doing so provides the methodology most suited to answering the research question (Johnson & Onwuegbuzie, 2004). However, this approach is criticised for switching between paradigms (McEvoy & Richards, 2006).

Maxwell and Mittapalli (2010) argue that critical realism is a theoretical perspective which is compatible to mixed methods research. Critical realism rejects that we have an objective knowledge of the world, that reality exists independently of our perceptions and understandings and that there may be several alternative theories of the phenomena (Maxwell & Mittapalli, 2010). Therefore, critical realism, combines a realist perspective with a constructivist epistemology (Maxwell & Mittapalli, 2010). McEvoy and Richards (2006) state that the aim of a critical realist approach is not to identify generalizable laws as in positivism, or identify lived experience as in constructivism, but to develop deeper levels of understanding and explanation. Therefore, researchers choose the method which will most effectively answer their research question.

Authors have highlighted the importance of distinguishing the motivations researchers have for utilising mixed methods from the specific research designs which can then be used to meet these goals (Morgan, 1998). Researchers must first identify the purpose for combining methods before selecting the appropriate

methods which are linked to, and will ultimately serve, that purpose (Green, 2007). Green (2007) states that the overall purpose of combining methods is to 'better understand' the phenomenon in question, however, there are different forms of 'better understanding' within mixed methods. Green (2007) presents five basic purposes or forms of 'better understanding' in mixed methods: triangulation, complementary, development, initiation and expansion.

A complementary purpose for mixing methods seeks a 'broader, deeper and more comprehensive' understanding by using methods that will 'tap into different facets or dimensions of the same complex phenomenon' (Green, 2007, p.101), which in this thesis is the psychosocial impact of cancer. Using both qualitative and quantitative methods results in a more comprehensive and complete conclusion to the research (Green, 2007).

Although authors from across the disciplines have proposed typologies for mixed methods designs, these are many, are inconsistent, and use divergent terminology (Tashakkori & Teddlie, 2010). However, Cresswell & Plano Clark (2011) state there are four main types of mixed methods design: triangulation, embedded, explanatory and exploratory. An exploratory sequential design is most suited for qualitatively exploring a phenomenon from the participant's view before using the results to inform and identify important variables or instruments to study in a subsequent quantitative study in order to enhance the entire study and enable a more complete understanding of the topic (Cresswell & Plano Clark, 2011).

Therefore, a mixed methods approach using an exploratory sequential design was adopted for the current thesis (Figure 2). Chapter 3 presents a qualitative study which was first conducted with young adult survivors of childhood and adolescent cancer

to explore their perspectives regarding the long-term impact and influence cancer has had on their lives. The results from this qualitative study and the literature review then informed the important issues and variables to research in the subsequent quantitative study to ensure a comprehensive investigation of psychosocial outcomes in survivors was undertaken (chapter 4). This quantitative phase can also try to explore the broader implications, applicability and generalisation of qualitative findings gained from a small sample of survivors (Cresswell & Plano Clark, 2011). By combining methods, the complex long-term impact of the disease can be explored via the subjective perceptions of the survivors, as well as investigating the impact of cancer which may be reflected in objective outcomes such as educational or employment status.

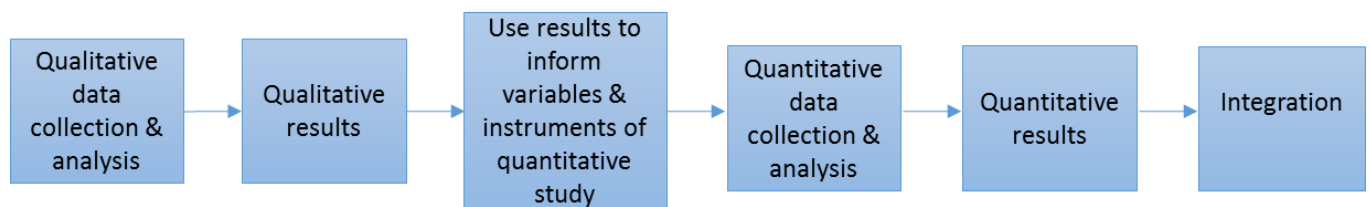


Figure 2 Exploratory sequential study design

2.6 Aims and objectives of the thesis

Aim

This mixed methods study will aim to conduct a comprehensive investigation of the long-term psychosocial impact of cancer in young adult survivors of childhood and adolescent cancer. This study will use an exploratory design in which qualitative and quantitative data will be collected in sequence and analysed separately, but with a final integration of the overall findings. The qualitative data phase will first explore the psychosocial impact of cancer from the perspective of young adult survivors. The results of which will then be used to inform a quantitative data phase which will be used to further explore the long-term psychosocial impact of childhood and adolescent cancer. By collecting both qualitative and quantitative data the study aims to provide a more complete understanding by exploring different dimensions of the psychosocial impact of cancer.

Objectives

- 1) To qualitatively explore the long-term psychosocial impact of cancer from the perspective of young adult survivors of childhood and adolescent cancer and to gain their views on how cancer has influenced their lives so far.
- 2) To conduct a literature review on the psychosocial outcomes in long-term survivors of childhood and adolescent cancer.
- 3) To use the results of the literature review and the qualitative study to inform a quantitative questionnaire which will be administered to a larger sample of young adult survivors of childhood and adolescent cancer. This questionnaire will further the investigation of the long-term psychosocial impact of cancer by evaluating a range of important psychosocial outcomes using a range of

measures and items. The comprehensiveness, feasibility and acceptability of the questionnaire will also be examined.

Chapter 3 A qualitative exploration of the psychosocial impact of childhood and adolescent cancer in young adult survivors

3.1 Preface

The author of the thesis led on all aspects of the study, under the direction of workplace supervisors. This included designing the study, the choice of analytical methods, gaining the necessary approvals, conducting the focus group and interviews, and analysis of the resulting data.

A fellow research assistant acted as a co-facilitator in the focus group and interview transcription was carried out by a university secretary.

The author was a co-applicant on a successful funding application to Newcastle Healthcare Charities for this study.

The results from this study were disseminated by:

Oral presentations

- Long-term psychosocial outcomes after childhood cancer at the 12th PanCare meeting, Amsterdam (2013)
- The long-term psychosocial impact of cancer - the views of young adult survivors of childhood cancer at the Epidemiology Group theme meeting, Institute of Health & Society, Newcastle University (2014)
- The long-term psychosocial impact of cancer - the views of young adult survivors of childhood and adolescent cancer at the British Psychosocial Oncology Society conference, Leeds (2015)

Poster presentations

- The long-term psychosocial impact of cancer - the views of young adult survivors of childhood cancer at the European Symposium on Late Complications after Childhood Cancer, Edinburgh (2014)

- The long-term psychosocial impact of cancer - the views of young adult survivors of childhood cancer at the Epidemiology Theme Research Day, Institute of Health & Society, Newcastle University (2014)

- The long-term psychosocial impact of cancer - the views of young adult survivors of childhood cancer at the Division of Health Psychology Annual Conference, York (2014)

Publication

- Brown, M. C., Pearce, M. S., Bailey, S., & Skinner, R. (2016). The long-term psychosocial impact of cancer: the views of young adult survivors of childhood cancer. *European Journal of Cancer Care*, 25 (3), 428-439.

3.2 Introduction

As the first phase in an exploratory sequential design, qualitative research can provide the means by which the phenomenon can be explored from the view point of the participants. This was deemed as a crucial stage in the present research due to the dominance of quantitative studies in psychosocial research. The studies most often utilise generic measures which do not cover issues pertinent to young adult survivors of childhood and adolescent cancer (Nightingale *et al.* 2011) and there is a lack of focus on survivors' views in the current literature. Therefore, to fulfil the aim of conducting a comprehensive investigation of long-term psychosocial outcomes it was essential to first fully explore the perspective of the survivor.

This chapter presents a qualitative study that was conducted with young adult survivors of childhood and adolescent cancer who were treated at the Royal Victoria Infirmary, Newcastle-upon Tyne.

3.3 Aims and objectives

- The aim of this first study was to explore the psychosocial impact of cancer from the perspective of young adult survivors of childhood cancer and to gain their views on how cancer has influenced their lives so far.

3.4 Methods

Qualitative methods allow us to gain a subjective understanding from the perspective of the survivors, enabling them to describe their own personal experiences, views and beliefs in their own words (Willig, 2001). By exploring young survivors' views and how they make sense of their cancer experience in their present lives as they move towards adulthood, it may permit a deeper exploration of the issues affecting their psychosocial outcomes. This may help to identify issues considered important to the survivors, as well as issues not yet addressed by current research.

3.4.1 Study population

The study population were survivors who had been treated at the RVI, Newcastle upon Tyne in the North East of England. The RVI is one of the 21 specialist treatment centres for childhood cancer in the UK and is the principal treatment centre for all of the Northern region of England (Figure 3).

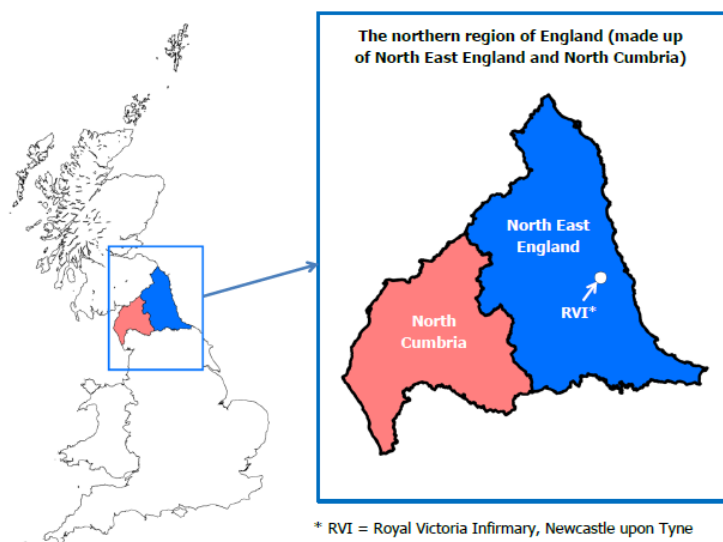


Figure 3 Map illustrating the geographic location of the Northern region of England

Reproduced with kind permission of K. Blakey, Newcastle University

Patients were identified via the Northern Region Young Person's Malignant Disease Registry (NRYPM DR). The NRYPM DR was established in 1968 and is a specialist registry of young people diagnosed with cancer under 25 years of age (Cotterill *et al.* 2000). The registry aims for complete coverage and so all individuals aged less than 25 years who have been diagnosed with a malignant (or benign CNS) tumour whilst resident in the northern region are eligible for registration.

Demographic details (including age, gender and residential address) as well as details of diagnosis and treatment, relapse and vital status are recorded in the NRYPM DR. Patient data are updated regularly by the registry secretary, with data for long-term survivors being updated approximately yearly. For long-term survivors who still attend the long-term follow-up (LTFU) clinics at the RVI, data is updated with information entered onto LTFU care plans by the LTFU nurse specialist. These care plans detail any emerging complications of treatment or health problems for the survivor. For survivors who are no longer in follow-up, either through non-attendance at appointments or because they have been discharged, contact is made with the named general practitioner and a request is made for an update on the survivor's health and any change in vital status.

By using the registry to identify eligible patients, this study aimed to include survivors who were both in LTFU care and those who were not.

3.4.2 Inclusion criteria

The primary inclusion criteria were that survivors had been diagnosed with a CNS tumour, other solid tumour or haematological malignancy before the age of 18 years; were aged 18-30 at the time of the study; were five or more years from diagnosis; were tumour/cancer free; and English speaking.

3.4.3 Procedure

Ethical approval

A favourable opinion was given by Newcastle and North Tyneside 2 Research Ethics Committee (Appendix A).

Sampling

Purposive sampling was employed in which the details of potentially eligible survivors who met the primary inclusion criteria were extracted from the NRYPMDB database. Paediatric oncology and haematology consultants based at the RVI were then asked to review the details of survivors who had been, or were still in their care, to confirm their eligibility. They were also asked to consider the survivors suitability for the study to ensure that vulnerable individuals and those who may lack the capacity to give informed consent were not approached. Therefore, additional inclusion criteria were the absence of severe or life threatening late effects (defined as grade 3 or above according to the NCI's Common Terminology Criteria for Adverse Effects v3.0) (NCI, 2009), and the absence of severe learning disabilities.

Recruitment

A process of rolling recruitment was used in that consultants were asked to identify between 5-10 eligible survivors at a time. Study invitation packs were then sent to these survivors via the consultant's secretary. Each pack contained an information leaflet detailing the study background and an invitation letter personally signed by their consultant (Appendix B). On receiving the invitation, survivors were asked to contact their consultant, or the researcher, if they were interested in taking part in the study or wished more information.

Heterogeneity and diversity in the sample was sought to enable the elicitation of a broad range of experiences and views. Therefore, key characteristics (diagnosis and current age) of those recruited to the study were reviewed in order to inform the purposive sampling and the identification of survivors with the required characteristics. Recruitment was conducted alongside data collection.

Data collection

Survivors were initially invited to take part in focus groups, with at least one focus group covering each of the three diagnostic groups (CNS, other solid tumours and haematological malignancies). Focus groups had been the initial method for data collection as participants are able to explore the issues which are of importance to them which may take the research into new and unexpected directions (Kitzinger, 1995). This was deemed particularly useful due to the exploratory nature of the study. Focus groups are also stated to be useful in encouraging those who may otherwise be intimidated by interviews and are well suited for exploring sensitive issues (Kitzinger, 1995; Wilkinson, 2004). In particular, diagnostic specific focus groups were chosen with the view that survivors may feel more comfortable disclosing their views with individuals with a similar diagnosis and illness experience.

However, response rates to the study invitations were disappointing. In addition, of those survivors who registered their interest in the study, organising focus groups proved to be problematic due to the range in survivors' geographical locations and their study or work commitments. Therefore, after holding one focus group, the design of the study was amended to allow one-to-one telephone interviews. The potential implications of this change of design on the study are outlined in the discussion.

The focus group took place at a university research department and was moderated by the researcher with the assistance of a co-facilitator who took detailed field notes of verbal and non-verbal cues throughout. Telephone interviews were conducted at a date and time chosen by the participant.

Recommended procedures for focus groups (Krueger & Casey, 2009; Wilkinson, 2004; Wilkinson, Joffe & Yardley, 2004) and telephone interviews (Burke & Miller, 2001) were followed in terms of planning and conduct. Prior to participation, all participants provided informed consent. This was written consent for the focus group and verbal consent audio-recorded for the telephone interviews (Appendix C).

Interview guide

Both the focus group and telephone interviews were guided by a semi-structured interview guide (Appendix D). The questions were open-ended and neutral to encourage the participant to respond without leading their answers (Smith, Flowers & Larkin, 2009). The technique of funnelling was utilised in that participants were initially asked the broad question of “How do you feel cancer has impacted on your life, if at all?” followed by questions for their views on more specific areas of their life including education, careers, relationships and health, with the more sensitive and personal questions towards the end. However, the responses of the participant influenced the exact ordering of the questions with care being taken that all topics had been covered. Follow-up questions and prompts were included to encourage the participant to expand on their answer. These prompts varied between interviews as they were dependent on what each participant said (Wilkinson *et al.* 2004). At the conclusion of both the focus group and the telephone interviews, participants were invited to raise any additional issues which were important to them. Participants

were also asked to contact their consultant if taking part in the study had raised any questions or concerns.

3.4.4 Data analysis

Analytical approach

Thematic analysis was chosen as the most appropriate method of qualitative data analysis for the study. Braun and Clarke (2006) define thematic analysis as a method for identifying themes and patterns of meaning across a data set. Although a popular method, thematic analysis is often poorly described in research. Additionally, as the use of thematic coding is a method common across many forms of qualitative analysis there exists the notion that thematic analysis is not a method in its own right (Braun & Clarke, 2013). To address this, Braun and Clarke (2006) outline clear and systematic steps for carrying out thematic analysis, successfully presenting the case for it to be considered a valued analytical method for qualitative research.

Thematic analysis is often considered less sophisticated in comparison to other qualitative methods such as interpretative phenomenological analysis (IPA). However, many IPA studies actually fail to be interpretative and do not go beyond initial descriptive analyses (Hefferon & Gil-Rodrigues, 2011). A stand-out feature of IPA is its explicitness in acknowledging the role of the researcher in interpreting the data. However, Braun and Clarke (2006) underline the active role of the researcher in thematic analysis and dismiss the concept of themes 'emerging' from the data as if the researcher is passive in the analytical process. In addition, by adopting a reflexive approach a researcher is able to consider their role in the construction of the knowledge resulting from the data (Willig, 2001). Another strength of thematic

analysis is that it is a flexible method in that it is not tied to a particular theoretical basis as is the case with IPA (Braun & Clarke, 2013).

Analytical procedure

Both the focus group and the telephone interviews were audio recorded and transcribed verbatim. The resulting data were analysed using thematic analysis using procedures described by Braun and Clarke (2006). Analysis began with familiarisation and active engagement with the data through repeated listening to the recordings and reading of the transcripts. This was followed by a thorough and systematic coding of the data relevant to the research question, taking care to note instances where participant's views were different or in conflict with the consensus. Coding focussed on the semantic meanings within the data, thus what was explicitly stated in the words used by participants. Identified codes were then reviewed and collated into themes. Coded extracts within each theme were then assessed for coherence and representativeness of the whole data set, before refining themes and creating clear definitions and names for each. The resulting themes are presented below and are illustrated through quotes from the participants.

As the aim of the current study was to explore the participants' personal views, the analysis was inductive. By using this approach, it is the data as opposed to the researcher's theoretical interest which drive the analysis. In doing so, the resulting themes may differ greatly from the questions that were initially asked of the participants (Braun & Clarke, 2006). Care was then taken to ensure that the analysis moved beyond description to interpretation by discussing findings in relation to theory and previous literature.

Braun & Clarke (2006) provide a 15-point checklist for achieving good thematic analysis which stipulates clear criteria throughout the analytical process from transcription to producing the written report (Table 1).

Table 1 Braun & Clarke's (2006) criteria for good thematic analysis

Process	No	Criteria
Transcription	1	Data transcribed to an appropriate level of detail. Transcripts checked against recordings for accuracy.
Coding	2	Each data item given equal attention in coding process.
	3	Coding process is thorough, inclusive and comprehensive. Themes not generated from a few vivid examples.
	4	All relevant extracts for each theme have been collated.
	5	Themes checked against each other and with back to the original data set.
	6	Themes internally coherent, consistent and distinctive.
Analysis	7	Data analysed and interpreted rather than just paraphrased or described.
	8	Analysis and data match each other – the extracts illustrate analytic claims.
	9	Analysis tells a convincing and well-organised story about the data and topic.
	10	There is a good balance between analytic narrative and illustrative extracts.
Overall	11	Enough time has been allocated to complete all phases of analysis adequately.
Written report	12	The assumptions about, and specific approach, to thematic analysis are clearly explained.
	13	A good fit between what is claimed to have been done and what is shown to be done.
	14	Language and concepts used in the report are consistent with the epistemological position of the analysis.
	15	The researcher is active in the research process: themes do not just emerge.

Care was taken to conduct the study in accordance with these criteria and general criteria for achieving good quality research provided by Yardley (2000). Yardley's guidance centres on the researcher being sensitive to the context of the study; being committed and thorough in the conduct of the study; being transparent and coherent in what was done; reflecting on factors which may have affected the findings; and producing research which has the potential to influence the beliefs or actions of others.

3.5 Results

A total of 122 survivors (69 solid tumour; 28 CNS; 25 haematological) were invited to take part in the study. At the end of the study 12 survivors had taken part in either a focus group (n=4) or a telephone interview (n=8). This final response rate was 10%. The characteristics of the participants are shown in Table 2. The focus group lasted 75 minutes and the interviews lasted between 26 and 95 (median 48) minutes.

Table 2 Participant characteristics

Participant characteristics	
Gender (n)	
<i>Female</i>	8
<i>Male</i>	4
Diagnosis (n)	
<i>Haematological</i>	
<i>Leukaemia</i>	3
<i>Hodgkin lymphoma</i>	1
<i>Solid tumour</i>	
<i>Rhabdomyosarcoma</i>	2
<i>Fibrosarcoma</i>	1
<i>Ewing's sarcoma</i>	1
<i>Germ cell tumour</i>	2
<i>Central Nervous System tumour (CNS)</i>	2
Employment (n)	
<i>Student</i>	6
<i>Employed</i>	5
<i>Unemployed</i>	1
Age at study (years), <i>Median (range)</i>	23 (18-30)
Age at diagnosis (years), <i>Median (range)</i>	7 (<1-16)
0-5	5
6-11	3
12 and over	4

Six participants were survivors of solid tumours, four of haematological malignancies and two of central nervous system tumours. There were more females (n=8) compared to males (n=4). The median age of participants at the time of diagnosis was seven years (range <1-16 years) and the median age at the time of the study was 23

years (range 18-30 years). All but one survivor were in further education or employment.

Through the narratives of the survivors, it was clear that there were differing views regarding the extent to which cancer had impacted on their present lives and this did not seem to be related to age at diagnosis, or indeed diagnosis. While four participants felt strongly that being a survivor was a continuing influence on how they led their lives, for a few their illness was firmly in the past. In general, it was commented that the impact of cancer on them, and their subsequent lives, was a difficult concept to consider due to their young age at diagnosis and hence their lack of self-awareness at the time. However, several survivors stated that every so often they gave thought to where they would be in life if they had not experienced the illness. No survivor felt that they were in a worse off place because of their past illness.

However, it was apparent that many survivors were still experiencing negative issues in their lives which were clearly connected to their past illness. The analysis of the data identified three main themes of *altered life perspectives*, *perceptions of self* and *lasting effects on relationships* through which the survivors described the effect that cancer was perceived to have on their present lives.

3.5.1 Altered life perspectives

This theme encompasses the many ways in which survivors described the experience of cancer to have influenced their current perspectives on life. These altered viewpoints and approaches to life were largely attributed to an explicit awareness of the uncertainty and unpredictability of life and health.

You can't plan

Survivors were acutely aware of the changeable nature of life and the concepts of unpredictability and uncertainty had evidently been assimilated into their life views. As described by Stephen, there was a strong rational feeling that no-one knows what the future holds:

"I can't... you can't plan. I get annoyed with people who plan in, you know, 'what if, what if, what if?'...it does my head in. You can't plan." (Stephen, age 24 years, leukaemia)

All survivors discussed uncertainty in the context of future health. The possibility of "it" - the original cancer returning, or of developing a different form of cancer in later life was acknowledged. Some survivors reflected on the treatment they had received and its potential for long-term adverse effects on their health, although there was no certainty among the survivors of what these effects could actually be:

"Em...I think I don't still fully know em I think I had a lot of x-rays when I was young which has made me quite worried about the risks of those. Em ..that's probably the main thing I worry about now to be honest. I think that will have some kind of long-term

*affect whatever that might be.” (Chloe, age 25 years,
rhabdomyosarcoma)*

Uncertainty was linked to a lack of knowledge and understanding about the potential effects on health which were in part connected to a young age at diagnosis. However, most survivors commented that although there was an awareness of the uncertainty of health, it was something that was at the ‘back of their minds’ which did not dominate their thoughts and was not reported to cause significant distress:

“I don’t ever worry that you know something like this would happen again, I don’t worry about the cancer coming back or yeah I don’t. I mean I think probably always there in the back of my mind that I know it’s possible but it’s not something that worries me.” (Joanna, age 20 years, fibrosarcoma)

“Might get a different form of it. I don’t know. Em it’s a passing thought it’s not something that stays it’s just a, “I wonder if”, because, I heard it when I was a child that they say that you’re more likely to get it...oh I don’t know” (Katie, 25 years, leukaemia)

To cope with life’s uncertainties, survivors held the view that worrying about what may or may not happen, or things you cannot control was counterproductive; it was more constructive for the survivors to “live each day as it is” and to “look forward” as opposed to dwelling on the past. A few survivors shared the belief that they would be able to deal with bad situations “as and when they come along”, with one male survivor of leukaemia, Daniel, commenting “I beat it before, I’ll beat it again” and

“everything will be fine in the end”. However, a few survivors appeared to have adopted a *“whatever happens, happens”* approach to future health:

“I’d be scared if I got it again I think but I don’t think about it as an ongoing, I’m not a worrier about it on a day-to-day basis, oh I’m going to get it again. If it happens it happens.” (Stephen, 24 years old, leukaemia)

“I don’t really think about... I try not to think about what could happen, just let it happen sort of thing so... I just think to myself, well, it’s got to happen at some point sort of thing. So I might as well just get along with it”. (Craig, 22 years old, central nervous system tumour)

Although a strong desire to have a family was evident in most survivors, feelings of uncertainty surrounding their fertility status were discussed by most. Although this was acknowledged as a concern, for those who were not ready to start a family it was stated that this was not the right time for them to have confirmation of their fertility status. There was a feeling that having confirmation of whether they could have children or not, could be more detrimental than not knowing. For these survivors, they preferred to address this when they and their partners had started to consider a family:

“I don’t really want to think about that while I’m at uni because I want to enjoy uni for what it is em but I imagine when it comes to that time em when I’m looking to have children whatever I’ll probably just have a sit down with whoever I’m with and say look

this is a joint decision now so would you like to know or would you like to just go with it or but I've I'll take that when it comes."

(William, 21 years old, rhabdomyosarcoma)

"I think if I find out now, that I'm not [fertile], that would have bigger implications of just not really knowing... because no one really knows."(Chloe, 25 years old, rhabdomyosarcoma)

The two female survivors who were married had reached the stage of life where fertility was a central concern. For one who was diagnosed at age 14, she reflected that for her, her ability to have children had been paramount from the beginning, *"it was the first thing I asked. The first thing I said"*. Despite being told there was a *"slim"* chance she could have a family, to her surprise, she had gone on to have two children naturally:

"I just had to kind of wait and see. It was one of those things that was out of my hands. I mean I had convinced myself it [getting pregnant] was going to take a long time. I was probably going to have to be backwards and forwards to try and get help, and luckily I didn't have to". (Emily, 30 years old, germ cell tumour)

However, for the other married survivor, despite having an ovary removed after being diagnosed at 16 she recalled *"the exact words that they [the consultant] used was that if you were able to conceive before, you should be able to conceive now"*. She was actively trying for a family and had so far had been unsuccessful. The uncertainty surrounding her fertility status had caused a strain within her

relationship. She felt a huge pressure to be able to have children. Like the other survivors, and evidenced in the quotes above, it was felt that the only way to be certain about your fertility status was to become pregnant:

“It’s always been something sort of hanging over my head. I think that’s probably the best way to describe it. It’s always kind of in the background. It just niggles away em... I suppose until you find out.” (Amy, 24 years old, germ cell tumour)

Making the most of it

The unpredictability of life highlighted to these young adults the importance of taking opportunities and being thankful and appreciative for what they had. Some felt that overcoming the illness had given them drive to make the most of their life:

“Had I not had cancer I think I would have been a little bit more of a home-bird and a little less kind of get out there and go. Whereas having cancer has just made me want to make the most of life cos I know that I could turn round tomorrow and I could have cancer. I could turn round tomorrow and like get hit by a bus.” (Laura, 24 years old, Ewing’s sarcoma)

Yeah I think there’s definitely the whole thing where a lot of my view is life’s too short to wait on things and you don’t know what’s going to happen and you know just go for it while you can, basically. (Amy, 24 years, germ cell tumour)

It was important for the survivors' to have sense of purpose in their life. Some described themselves as being "lucky" to have survived. In particular, survivors discussed how employment was an important life domain in which purpose could be found. For most it was conveyed that it was seen to be valuable to have a job which was meaningful and fulfilling:

"It's really doing something it doesn't matter what it is. Just as long as it's doing something that's, that's useful, that has a meaning to it I guess. I don't know whether... I couldn't tell obviously whether it would have been...I imagine it would have been different if I hadn't been ill." (Stephen, 24 years old, leukaemia)

However, for Craig who was currently receiving disability allowance, and in the past had unsuccessfully applied for a range of positions, a sense of purpose was to be found in any job:

"Just anything really. Just applied for anything I thought I could do. I wasn't really bothered what I was doing, as long as I was doing something..." (Craig, 22 years old, central nervous system tumour)

Helping others

In general, many survivors felt that their experiences had led them to have greater empathy and consideration in how they treated or viewed others. Half of the survivors specifically stated they had given thought to how they could use their illness experience to help others. Their illness had also exposed them to new experiences

such as a hospital environment and being cared for, with several stating that this had led them to consider a career in health and social care.

“helping the poor, helping the sick, helping people you know with the strength that I have, with the support that I had with my experiences. Helping others... it’s probably my biggest goal in life.”
(Joanna, 20 years old, fibrosarcoma)

However, for one survivor who went on to study medicine, she was adamant that cancer had no bearing on her present life or on her choice of career:

“Yeah I don’t think it has, no I think people often assume it would have done but I really don’t think it has it was so long ago.. em if anything em for quite a while I never wanted to go into hospital again.. em so yeah I don’t think it had any bearing.” *(Chloe, 25 years old, rhabdomyosarcoma)*

3.5.2 Perceptions of self

This theme presents findings relating to how survivors felt that surviving cancer had influenced how they perceived themselves and how others may also perceive them. Many survivors described adaptation to, and acceptance of, the changes to their physical appearance, whilst other survivors described persisting issues with their changed appearance which they felt now set them apart from others. However, surviving an ordeal such as cancer also made survivors aware of their own strengths as well as potential limitations.

It's just who I am

The majority of survivors reflected on how the illness experience had influenced their perceptions of themselves. Adapting to a changed appearance was particularly salient and this was across all diagnoses. For those who had received surgery as a young child, they had learned to adapt to their physical changes: it had become accepted as part of who they were:

"If it was happening now, God forbid, I imagine it would be different. I imagine I would feel more self-conscious. But because I've grown up with it from such a young age, it's just who I am. So it's never really, never really bothered me" (William, 21 years old, rhabdomyosarcoma)

"I count my scars and when people are like I've got this scar and I'm like you haven't got a scar I've got the scars. I can win, I win every time." (Laura, 24 years old, Ewings sarcoma)

For those who were diagnosed as teenagers, they reflected on their initial self-consciousness as a result of their treatment. However, this was reported to have lessened with age. Acceptance of a physical appearance altered by surgery was said to be made easier by the passing of time, and the fading and the neatness of the scars that remained. Most survivors who had stated they had undergone surgery stated that there was no use dwelling on what could not be changed:

"Initially I was quite self-conscious about it. I wouldn't ever, you know, wear bikinis or that kind of thing em... but I think I'm getting older and the scar's fading and looking a bit neater and just all

that kind of thing. It becomes less of a worry and I don't, I don't tend to dwell on it." (Joanna, 20 years old, fibrosarcoma)

Different to others

Physical appearance was largely discussed in relation to how survivors felt they were perceived by others. Although the above survivors seemed to have become comfortable in themselves and adapted to their changed appearance due to surgery, it was clear that overall, the location and visibility of the scars was an important factor to survivors in considering whether the scarring had left an impact on them or not. For those who had scars which were hidden under clothing, these scars could be revealed to trusted others as and when they felt comfortable, as demonstrated by William who had a testicle removed as a child:

"Before things get too serious I just like to say "look, just to warn you"... em but I mean that's never been a problem relationship wise" (William, 21 years, rhabdomyosarcoma)

However, it appeared that for survivors who felt that their physical consequences of their past illness were more obvious, for instance due to scarring to the face, or from poor hair re-growth after chemotherapy, they were not able to hide this from others. They reported feelings of anxiety and a loss of confidence as a result of this:

I don't think I'm, I'm just not a very confident person at all really I don't think. Em even if, even if it's just going into town for shopping or going to the library to do work I'm always conscious about people looking at me, thinking about what I look like and I'm worried about people what, what, what they going to say

about me or if they're going to say anything to my face or if they're going to say it behind my back. Just, it's just always a worry about what, I think it's just an image conscious thing in my mind and I'm probably over paranoid about it to be honest because I do panic about it a lot. (Claire, 21 years, central nervous system tumour)

"I think the only reason it has an ongoing kind of, the only reason I ever think about it or the fact that it has any bearing on me is because the surgery was on my face and I think that if it was somewhere no one could see it, I wouldn't think about it ever."
(Chloe, 25 years, rhabdomyosarcoma)

However, there was also evidence that even in the absence of visible physical signs, survivors may still struggle with how they appear to others. Lucy's struggle with her appearance during her illness had persisted into young adulthood and she stated that she was aware she still possessed a distorted view of how she looked to others:

"The one thing that really bugged me was just my appearance all the way through. This year I like got to the point where I was saying 'oh God' and I've had counselling all this year and I'm still having counselling now. I struggle with that aspect... I still think people see us looking the same as when I did when I was ill" (Lucy, 18 years old, Hodgkin lymphoma)

An awareness of own strengths and limitations

The majority of the participants gave insight into how the experience of cancer had influenced their perceptions of their own ability. Being determined, motivated and realising your own strength were discussed. For some, the experience was said to have given them the strength to deal with situations they found challenging:

“because I kind of learned to push myself to get through the illness and to get through the new school transition, that sort of thing but that seems to be the mentality. That even if something seems to be in the way or stopping me, that it doesn’t stop me. That it just kind of gives me the momentum or the motivation to kind of push through it.” (Joanna, 20 years old, fibrosarcoma)

These views were shared particularly in relation to education. Despite recalling factors which could have impacted on their education such as missing school due to ill health and treatment and struggling with being the child with cancer, several stated they had been determined to do well academically. For a few this meant that they had gone on to exceed their own, and others, expectations in terms of their educational achievement:

“I didn’t really expect that I would go on to do a degree and nobody else expected that I would get that far and I did...and I passed... and I got a first” (Katie, Leukaemia)

“When I went to school I actually did a lot better and I’m not entirely sure why... I was more driven to do things and I achieved higher than I did before” (Stephen, 25 years old, leukaemia)

I used to miss physics on a Thursday every week but em er I ended up like at the end of the year getting somethin like 89% physics. My mum was just like 'well how have you done that?' and like yeah I remember that and just thinking well it obviously is not affecting my school (Daniel, 19 years old, leukaemia)

Conversely, for a few survivors there was an awareness of their limitations. For two survivors who been left with either a physical or a visual impairment from a young age, they reported that they had learnt to adapt to these limitations and to compensate where needed:

"my tendons for my leg are shorter but I just compensate for it...em and like I've taught myself to compensate exactly the same and I've just gotten on with it". (Laura, 24 years old, Ewings sarcoma)

"Em because it, because I've had it that long I just kind of adapted to it and I automatically do things automatically myself without even realising. (Claire, 21 years old, central nervous system tumour)

However, for Claire although she didn't perceive her impaired vision to be a hindrance to herself in the workplace, it did impact on her confidence to take part in physical activities. A few survivors also commented on problems with tiredness, concentration or memory which they perceived to impact on their ability to undertake writing tasks for work or university. Craig, who had been diagnosed with

short-term memory loss, was acutely aware that he was unable to perform at the level of his peers, even in basic tasks. He had since struggled with his memory in college:

“I knew for a fact before I left school there was certain, certain jobs that I couldn’t do, and some of them were jobs that I thought were quite basic like working at McDonalds or Burger King. But I wouldn’t be able to do that with my memory problems...wouldn’t be able to keep up”. (Craig, 22 years old, central nervous system tumour)

3.5.3 Lasting effects on relationships

The final theme presents data on how survivors described their cancer experience to have influenced their relationships with their parents, siblings, as well as romantic relationships and friendships.

Family

Parents were stated to be an important source of continuing support and encouragement and it was clear that survivors had close bonds with their parents. Some survivors felt that in retrospect, due to their young age at treatment, their illness experience had affected their parents more than it had them. Many survivors referred to the protectiveness of their parents as they were growing up, which, in some cases, was seen to restrict their ability to have experiences and achieve independence and self-confidence. Survivors stated that even as young adults, their parents continued to find it hard to let go, particularly with respect to them leaving the family home:

“my mum’s like “are you sure you want to do that?” and more “stay here”. And with everyone else’s parents it’s like “oh yeah, just go, want you out, get rid of you” (laughs). (Katie, 25 years old, leukaemia)

However, for two participants, emotional issues were said to be affecting their communication and closeness with family, and particularly their mothers:

“She thinks that my emotions get out of control sometimes when something happens. If it causes an argument the argument will be blown all out of proportion and she thinks that’s to do with my

emotions just getting in the way completely.” (Claire, 21 years old, central nervous system tumour)

Two survivors also reported persisting feelings of resentment from siblings, with one commenting that *“the resentment has just grown with him as he’s grown up”*. This was stated to originate from feelings of neglect in favour of their sick sibling and at being forced to miss out on time with their parents. These survivors stated that they felt they had gone on to achieve more than their siblings:

“she’s more em... close to mum because of it because she didn’t get to spend the time with her when I was poorly...so it affected my family quite a lot as well. Quite a bit more than me, ‘cause I seem to have done a lot better than what my sister’s been able to. Because she, I don’t know, it affected her later on” (Katie, 25 years old, leukaemia)

A family of their own

Although only two of the participants were married, most reported to be in a relationship and did not report that their cancer had impacted on their ability to meet a partner. All but one survivor reported that they had shared their illness history with their partners. Only one survivor was a parent, and although marriage and children were desired in most cases, it was acknowledged by the majority that this was not a consideration at the present time. There was a wish to finish their education and be settled before having a family. Most survivors considered themselves too young at present:

“I’d obviously like to get university out of the way, get my job established first and me and my partner have been talking about finding a house together when I’ve finished university. I think it’s just getting established first and finding the right time.” (Claire, 21 years old, central nervous system tumour)

Conversely, one married survivor who was actively trying for a family felt that starting a family *“would have been a bit further down the line”* if she had not experienced cancer. Her illness experience had led her to place a greater value on having a family. Other survivors similarly stated that as a result of their experience they were motivated to have children to be able to care for another:

“I want to give back what mum gave to me” (Katie, 25 years old, leukaemia)

“to care for someone younger than you and make sure they get the best chance in life” (Stephen, 24 years old, leukaemia)

Friendships

Most survivors did not report there to be lasting effects on their friendships. For some, friends were stated to be a good source of support whilst ill, and for most whose friendships suffered, their relationships were said to recover in time. However, a few survivors commented that they were aware they now had few close friends. Experiencing a loss of friends through cancer at a young age, not wanting to cause anyone concern, worrying about people’s reactions to a disclosure of cancer history, anxiety at being viewed as “different” to others, missing out on the stage of life when

other friends were making close bonds and consequently feeling you had little in common with your peers were reported as negative influences on friendships:

"I don't really have close friends now... em I used to before. But I think... I always remember telling one of my friends when, when I'd been diagnosed with depression, she said "how can you have depression, you're too young", and that kind of said it all for me. It was just a bit too much em but again I think it's the whole thing well why waste your time with people where you're unhappy you know where, where you're not getting on with people, where you're unhappy or where you know where you might go out somewhere and kind of grin and bear it you know, just so you're out and about... Whereas now I'm just a bit more the opposite well if I'm not happy doing it then I don't really want to go, I'm not going." (Amy, 24 years old, germ cell tumour)

"I make friends very easily em I'm very kind of I'm very open and social but I don't like making close friends. Em it's not that I don't like I just I think I get to a point where I stop myself em because well first of all I lost friends and then second of all I don't like people suffering because of me. That's a really odd thing to say..."
(Laura, 24 years old, Ewing's sarcoma)

One survivor who stated that although he had never been that sociable, he felt this was more so since his treatment at age 11. He found it difficult to talk to new people.

He preferred to stay at home or use social media. He also preferred to spend time with people older than himself, as opposed to people his own age:

"I don't know I've just been like that since, since I was like the week after all my treatment and everything started getting very cautious around people getting nervous when speaking to them."

(Craig, 22 years old, central nervous system tumour)

Conversely, Claire who was diagnosed at a very young age, felt her experience had led to her valuing friendships more than she would have otherwise. However, although this was a positive consequence of the cancer, Claire felt that it had the potential to affect friendships:

Em (pause) I am very, I think I'm very close to people when, like when I meet new people I think I'm very, I think I value friendship a lot more and value my family a lot more because of what I've been through. Em and sometimes I think people find that a bit strange because of how close I get and how quickly I become close to them. (Claire, 21 years old, central nervous system tumour)

3.6 Discussion

This study aimed to explore the long-term impact of cancer from the perspective of the survivor as a young adult and gain an insight into how they perceived cancer to have influenced their life paths, if at all. Although there were mixed opinions as to whether cancer had indeed left a long-term impact, all participants went on to share physical, emotional and social consequences of their experience which had continued into young adulthood. The most salient theme was that survivors perceived their experience to have influenced their present outlook on life. Although this was voiced in a positive way through a greater appreciation of life and of others, this was due to an awareness that life can be uncertain and short. Survivors also reflected on how they felt the cancer had impacted on how they, and others, viewed themselves. This was from both a physical and a psychological perspective. Their views with regards to how relationships were affected were also explored and in doing so, both positive and negative effects on relationships were found.

3.6.1 Altered life perspectives

Uncertainty is a factor in all our lives, however, similar to the findings of Parry (2003) and more recently Lehmann *et al.* (2014), survivors possessed a heightened awareness of the potential uncertainty of health, fertility and life as a result of their illness experience. Mishel (1988) states uncertainty to be the inability to determine the meaning of illness-related events, which can occur when the individual is unable to predict the outcome due to a lack of sufficient cues. Uncertainty is a central theme for children diagnosed with cancer and their families due to entering the unknown (Woodgate & Denger, 2002). However, less is known about how long-term survivors conceptualise uncertainty (Parry, 2003).

Uncertainty is stated to be a theme in childhood and adolescent cancer survivorship due to the risk of the development of cancer or late adverse effects of treatment (Santacroce & Lee, 2006). Similarly, survivors in the present study experienced uncertainty through the belief that they were potentially at risk of a relapse or of developing a secondary cancer in future, with most acknowledging they lacked information about these, and other potential risks. However, few survivors acknowledged the potential for the development late adverse effects of treatment, and of those who did, the majority were uncertain about what the potential late adverse effects could be and were aware that their knowledge of late effects was lacking, which has been previously reported (e.g. Bashore *et al.* 2004).

Survivors were generally unsure of their ability to have children, which although presented uncertainty, for most it was not referred to as a source of distress. While some survivors may want to confront their fears about their reproductive health, as found by Thompson *et al.* (2013), the majority of survivors in this study stated their fertility status was not something they wanted to address at the present time. Rather, their focus in life was on their education and enjoying young adulthood. Although the majority of participants in the present study were female, a similar finding was reported by Green *et al.* (2003) in young male survivors of childhood cancer.

Parry (2003) has previously reported that uncertainty about fertility was not perceived by survivors as distressing due to their views that adoption was a possible alternative. However, it is noted that survivors both in Parry's (2003) study and in the present study were relatively young, with average age of 22 and 23 years respectively. Therefore, it is not suggested that fertility issues are not a cause of concern for survivors, but that it may be dependent on life stage and the priorities of

the person at that time. As noted by Woodgate and Denger (2002), uncertainty differs across contexts and the presence of it should not always be presumed to be something negative. Survivors may receive information about their fertility at a young age when they are unable to see the relevance it will have on their lives, and some survivors may prefer to 'partially process' the information and return to the issue at a time of their choosing (Green *et al.* 2003).

Uncertainty was discussed as being the catalyst for an altered life view, which was more positive in relation to life and feelings towards others, and also prompted the need for life to have a deep sense of purpose. Improved views of life and of oneself have been previously described (Doukkali *et al.* 2013; Enskar & Bertero, 2010; Parry & Chesler, 2005). This was even evidenced in several survivors who were diagnosed in adolescence and even in early childhood. These findings may be in contrast to the findings of studies which utilise the PTGI and find that higher positive impact of cancer is reported by older survivors (Gianinazzi *et al.* 2016; Zebrack *et al.* 2012). PTG may not be applicable to survivors who experienced their illness at a very young age and have few memories of the event. It may be more suitable to conceptualise the cancer as an organising principle in life, as suggested by (Zebrack *et al.* 2012).

As reported by Lehmann *et al.* (2014), survivors simultaneously reported both positive and negative outcomes of their illness experience. As reported by previous studies (Dumas *et al.* 2015; Eiser 1997), cancer appears to have influenced their career choice of some survivors towards health and several were either employed in, or studying towards, a career in a caring profession. In the present study, the change in life views was stated to influence career choices with many being motivated to help others. In addition, being ill as a child gave them experience and knowledge

which they felt they could use to help others. For most, there was also a need to have a job which was meaningful. Having a career and employment was conveyed as an important goal by all and it has previously been reported that survivors are more likely to report work as being the main concern in their life as compared to aged-matched controls (Badell *et al.* 1998). As Schwarz *et al.* (2012) discuss, employment and occupational choice are important factors in the formation of personal identity and in achieving full adulthood.

Survivors gave insight into how they incorporated uncertainty into their lives, such as living each day at a time. However, a 'whatever happen, happens' view to future health was also stated by some. Although this may be a successful way to cope with uncertainty, it may hint that some survivors may feel that they have limited control over their future health and could potentially adopt a fatalist coping mechanism, as also noted by Parry (2003).

3.6.2 Perceptions of self

Given the importance of body image in adolescence, the body-altering effects of treatment were reported to be distressing during this life-stage. However, the majority of survivors stated that over time they had grown to accept physical changes and scarring as part of who they were. Recently, Lehmann *et al.* (2016) reported that long-term survivors of childhood cancer had comparable levels of body image with healthy controls, although it is noted that CNS survivors were not included in this study.

Although they felt the need to pre-warn partners and explain their scars prior to intimate relationships, the visible consequences of surgery were not reported to have an impact on relationships. Most of these survivors had received surgery to their

abdomen and limbs which could be covered and revealed when they wished. However, it was evident that for some survivors, the loss of control over the visibility and appearance of scars to others seemed to be the cause of angst, anxiety and loss of confidence. While scarring has been associated with psychological distress, this has recently been found to be more so for head and neck scarring and survivors with persistent hair loss, suggesting that outwardly visible physical appearance may be key in emotional adjustment (Kinahan *et al.* 2012).

As found in previous studies, survivors described their feelings of strength and determination as a result of their experience (Parry & Chesler, 2005; Zebrack *et al.* 2012). In particular, survivors often attributed their educational achievements to a new found drive stemming from their illness. Similarly, Lewis *et al.* (2013) reported that survivors attributed greater importance to their education after their illness, and determination and motivation to achieve future goals has been demonstrated in focus groups with young adult survivors of childhood brain tumours (Boydell *et al.* 2008). On the whole, survivors did not feel that their education had been adversely affected. The majority of survivors in the study had completed, or were currently in, higher education. This could in part, help to explain the findings of Lancashire *et al.* (2010) who reported that while central nervous system tumour survivors and survivors treated with cranial radiation had educational deficits in comparison to the general population, some sub-groups (bone sarcomas and retinoblastoma) achieved at a higher level in school examinations than the controls (Lancashire *et al.* 2010).

3.6.3 Lasting effects on relationships

Although marriage is used as an indicator of social adjustment and parenthood is often used as a measure of fertility (Madanat *et al.* 2008), there are many factors

which may influence whether a person marries or has a child or not. As previously reported by Hohmann *et al.* (2011), the majority of survivors considered themselves too young to be parents, wanting to continue with their education and careers before settling down. However, many were in committed relationships and stated that marriage and children were goals in later life. It also appeared that the cancer experience had enhanced the value some survivors placed on having their own family (Langeveld *et al.* 2002). These findings suggest underline the importance of considering relationships other than marriage as a research outcome. However, this is not to say that survivors' relationships are not affected, only that the study is limited by the young age of the sample and further outcomes such as sexual functioning were not explored.

The long-term effects of childhood cancer on families are not well known as research has tended to focus on the family of the newly diagnosed patient (Hoven *et al.* 2013). Previously it has been reported that survivors may perceive the illness experience to have changed their family for the better (Gray *et al.* 1992) whilst others may still experience feelings of guilt at what their families had to deal with as a result of their cancer (Carlson-Green 2009). No feelings of guilt were reported here, however, there was a sentiment from some that the illness had affected their families more than it had them. Siblings experience a range of feelings and unmet needs throughout the patient's illness trajectory (Wilkins & Woodgate 2005) and there was evidence that this can still affect relationships between siblings into young adulthood with suggestions that some siblings may in fact do less well than the survivor.

On the whole, most survivors were positive about their friendships. However, changes in relationships with peers may persist over time resulting in survivors

perceiving themselves to have few close friendships. Barrera *et al.* (2005) previously found that, as reported by parents, survivors were more likely than controls to have no close friends. A range of factors which were perceived to affect friendships were given by survivors in this study, however, survivors did not report to be distressed by this.

3.6.4 Strengths and limitations of the study

The qualitative approach of this study enabled survivors to share their views and for us to gain potentially new insights into how survivors consider the cancer experience to have influenced their lives. A holistic approach was adopted by the study in that several outcomes were investigated at once. In this way, the researcher gained an understanding of the relationships between outcomes and how several areas of a survivor's life may be connected, for instance how a change in life views may influence other areas of life such as career goals and relationships.

Although the value of generalisability in qualitative research is debated, it is believed that the results of this study illustrate viewpoints which may be relevant to other childhood cancer survivors with similar characteristics. The study included long-term survivors of a range of childhood cancers diagnosed under the age of 18 and aged between 18-30 at time of study with themes being present across all diagnostic and age groups. However, it is acknowledged that this study had a low response rate and that the resulting sample had a higher proportion of females than males which means a cautious approach must be taken when interpreting any such findings. As with much previous research, the participants self-selected and the study may have attracted survivors who were higher functioning or who were more motivated to relate their experiences. However, qualitative studies of young cancer survivors often

only approach survivors who are in LTFU care (Enskar & Bertero *et al.* 2010; Zebrack *et al.* 2004). By identifying survivors from a registry it was possible to invite those who had been discharged or did not attend LTFU appointments, as well as those still in follow-up.

Although the study had initially aimed to utilise focus groups, recruitment difficulties meant that the majority of data were instead collected via telephone interviews. Although the data collected via the focus group did not appear to differ from subsequent interview data in scope, it is probable that the interviews enabled a deeper exploration of the individual's own personal views. While focus groups explore collective views and generate data that is a result of an interaction between group members (Wilkinson, 2004), semi-structured interviews are stated to be most suitable for research questions which enquire about the experiences or explore the perceptions of individuals (Braun & Clarke, 2013). Therefore, on reflection, semi-structured interviews may have been more appropriate method of data collection for this study from the outset.

Although telephone interviews are more associated with the collection of survey data as opposed to in-depth qualitative data, a review of telephone interview studies by Novick (2008) found the resulting data had been rich and of high quality. Telephone interviews may allow the participant to take part whilst in their own environment with anonymity and privacy, therefore, participants potentially may be more comfortable and relaxed than when in a face-to-face situation.

A low response rate, as seen in this study, is a problem evident in previous qualitative work with CCS (Earle *et al.* 2005) and recruitment is a recognised challenge for studies involving young adult survivors of cancer (Aubin, 2011). Participant rates may have

been improved by recruiting survivors from the LTFU clinic, as opposed to recruiting by mail. However, recruiting via clinics would exclude survivors who had been discharged or did not attend follow-up.

3.6.5 Conclusion

Young adulthood is stated to be a time where independent living, intimate relationships and vocational goals are pursued (Henderson *et al.* 2010). This study was able to explore whether an experience of cancer at an early age is perceived by survivors to have impacted on, and influenced, several areas of their lives. The use of qualitative methods enabled survivors to communicate their views and beliefs in their own words. By investigating the general impact of cancer as opposed to focusing on one particular area of life (e.g. relationships) it was possible to achieve an insight into the inter-relatedness of psychosocial outcomes in childhood and adolescent cancer survivors.

The review of previous research (Chapter 2) and the findings of this qualitative study were then used to inform the content of a quantitative questionnaire study to further investigate psychosocial outcomes in survivors. This is now discussed in chapter 4.

Chapter 4 A questionnaire study to investigate psychosocial outcomes in young adult survivors of childhood and adolescent cancer

4.1 Preface

The author of the thesis led the design of this study to administer a questionnaire to further the investigation of psychosocial outcomes in young adult survivors of childhood and adolescent cancer. This study was successful in obtaining a project grant from Children with Cancer, UK, for which the author was a named co-applicant.

The author developed the questionnaire booklet with advice from workplace supervisors, was responsible for developing the protocol for the study and for obtaining NHS approvals, co-ordinating its distribution to survivors and for analysing the resulting data.

The work produced by this study has been disseminated by:

Oral presentation

- The long-term psychosocial impact of cancer - the views of young adult survivors of childhood cancer at the Epidemiology Group theme meeting, Institute of Health & Society, Newcastle University (2014)

Poster presentation

- Developing and piloting a questionnaire to investigate important psychosocial outcomes in survivors of childhood and adolescent cancer at the British Psychosocial Oncology Society conference, Leeds (2015)

4.2 Introduction

The aim of this second phase of the exploratory sequential design was to combine the knowledge presented in previous chapters, to inform a complementary quantitative study. The literature review (chapter 2) identified outcomes considered central to psychosocial outcomes of young adult survivors of childhood and adolescent cancer, namely health-related quality of life, psychological distress and psychological growth. In addition, educational, employment and relationship outcomes were prominent. The importance of the subjective views of survivors regarding the impact of cancer on their lives and the effect that that these views may have on psychosocial outcomes were also highlighted. In phase one of the study, the qualitative study (chapter 3) explored the subjective views of survivors on the impact that cancer has had on their lives and the way in which this illness experience may have influenced their lives so far

The present study involved administration of a self-report questionnaire to survivors with the aim of furthering the investigation of psychosocial outcomes in young adult survivors of childhood and adolescent cancer. The questionnaire encompassed items to extend the investigation of variables identified in the qualitative phase and explore the generalisation of these issues in a larger sample of survivors. The quantitative phase also aimed to be complementary by gaining information on issues identified in the qualitative phase and the literature search in order to evaluate the many different dimensions of psychosocial outcomes in survivors, for instance survivors subjective views of the impact that cancer has had on their lives and the life-long impact of the disease which may be reflected in objective outcomes such as educational, educational and relationship outcomes. It was hoped that the questionnaire would increase the knowledge of the psychosocial outcomes of survivors who have been

treated at the RVI, Newcastle upon Tyne, as well as leading to a greater understanding of the factors which are associated with, and may determine, poorer psychosocial outcomes in this group.

As the questionnaire was envisaged to be detailed, contain new items and was a new method of data collection in the childhood and adolescent cancer survivor population at the RVI, piloting and assessing the acceptability and feasibility of such a questionnaire was deemed necessary. Testing methods, procedures and data collection measures prior to a full scale study is considered good study design (van Teijlingen & Hundley, 2001).

4.3 Aims and objectives

Aim

To undertake a quantitative questionnaire study to further investigate long-term psychosocial outcomes in young adult survivors of childhood and adolescent cancer.

Objectives

- To use the results of the literature review and the qualitative study to inform the content of a questionnaire by identifying important variables and instruments for inclusion.
- To administer the final questionnaire to a sample of young adult survivors of childhood and adolescent cancer.
- To evaluate the feasibility and acceptability of the questionnaire to inform future administration.

4.4 Methods

Questionnaires provide a method of eliciting information from people about their knowledge, attributes, emotions, behaviour, beliefs, and attitudes (Rattray & Jones, 2005). The aim is that by gaining statistical information on the characteristics of the subset of people who respond to the questionnaire, inferences can be made to the target population they represent (Fowler, 2009).

This questionnaire study was conducted in two stages. The first stage aimed to develop the questionnaire content and an overview of this process, an overview of the questionnaire, as well as the final questionnaire, is located in Appendix E. The second stage then administered the questionnaire in a sample of young adult survivors of childhood and adolescent cancer and this is presented below.

4.4.1 Administration of the questionnaire

A pilot study is a smaller version of the proposed research and is conducted prior to the main study (National Institute for Health Research, 2016). Pilot studies are central to good study design (van Teijlingen & Hundley, 2001), and can be used to test the feasibility and acceptability of the proposed research measures, methods and procedures of a study (Hertzog, 2007; Thabane *et al.* 2010). For this piloting the questionnaire, the issues relating to feasibility were the proposed procedures for identifying and recruiting eligible participants, the response rate from survivors, the completeness of the resulting data, and the adequacy of the research measures and newly developed items. Pilot studies can also be useful by identifying possible effects and associations which could be investigated further in a larger study, as well as providing better estimates of likely statistical power (Everitt, 2006). Acceptability of

the questionnaire to survivors was indicated via the participant response rate and participant feedback.

4.4.2 Procedure

Ethical approval

The final questionnaire and patient information documents were approved by Newcastle upon Tyne Hospitals NHS Foundation Trust Research & Development before a favourable ethical opinion was granted by the North Tyneside and Newcastle 2 NHS Research Ethics Committee (Appendix F).

Study population

The inclusion criteria for eligible participants matched those used in the qualitative study presented in Chapter 3. Eligible participants were young adults (18-30 years old at time of study) diagnosed with cancer at ≤ 18 years old, who were diagnosed ≥ 5 years ago and who were English speaking. As before, survivors were identified via the Northern Region Young Persons' Malignant Disease Registry (NRYPMDR).

Recruitment

As in the qualitative study, details of potentially eligible survivors were extracted from the NRYPMDR database. Survivors were then screened by their consultants to confirm eligibility and that it was appropriate to contact them. Eligible patients were sent a questionnaire pack via the consultant's secretary which contained: 1) an information sheet about the study; 2) a consent form; 3) a questionnaire booklet with an evaluation form; 4) a refusal form for those not wanting to take part to indicate reasons why; and 5) a pre-paid addressed envelope. (Appendices G, H & I). Each questionnaire pack was assigned a unique study ID.

If survivors had not responded within approximately 3 weeks of receiving the questionnaire, a reminder letter was sent via the consultant's secretary. On receipt of the questionnaire, respondents were sent a thank you letter. Only after this point was the researcher able to access the survivor's personal details, for those who had given consent, from the NRYPMR including date of birth, diagnosis, date of diagnosis, and age at diagnosis.

4.4.3 Measures

The aim of the questionnaire was to investigate further issues, concepts and variables which had been uncovered in the qualitative study with survivors, as well as in the literature review. The aim was to administer a questionnaire which would be comprehensive and would enable an evaluation of the complex interaction of multiple variables (physical, psychological and social) which may influence long-term survivors' psychosocial outcomes. The questionnaire would be cross-sectional which would obtain data on the survivors' current status as well as retrospective information.

The subjective views of the survivor as to how the cancer has impacted or influenced their life, concerns they have about their health and their future, how they view themselves, how others view them and the impact of cancer on their relationships were important themes in chapter 3. The literature search and the qualitative study highlighted that survivors' perceptions and subjective appraisals of the impact of cancer are important factors to consider and have been found to be associated with psychological outcomes (Hobbie et al. 2000; Rourke et al. 2007; Zebrack & Landier, 2011).

The Impact of Cancer Scale for Childhood Cancer Survivors (IOC-CS) is a measure of survivors' perceptions of the impact of cancer on their lives over several areas including life views, how they view themselves and relationships (Zebrack *et al.* 2010). Therefore this measure was considered of great importance to the quantitative study. The IOC-CS and other measures included in the questionnaire are discussed below.

The Impact of Cancer for Childhood Cancer Survivors (IOC-CS)

The IOC-CS aims to assess survivors' perceptions of how cancer has affected their lives across several quality of life domains and its development was informed by interviews with 64 young adult cancer survivors aged 18-35 years (Zebrack *et al.* 2009).

The original version of the IOC-CS contains 73 items which each relate to either a positive or negative outcome of cancer. Items are scored on a five-point Likert scale ranging from *Not at all (1)* to *Very much (5)*. A psychometric evaluation of the IOC-CS has suggested that 45 of the items contributed to eight specific subscales: life challenges; body/health; talking with parents; personal growth; thinking/memory problems; health literacy; socialising and financial problems (Zebrack *et al.* 2010). Each subscale is suggestive of either a positive or negative outcome and the score of each is calculated by summing the responses to the relevant items and dividing by the number of items to give the mean.

Zebrack and Landier (2011) propose that as well as the subscales, overall positive and negative impact scores can be calculated by calculating the mean of the 25 items suggestive of positive outcomes (items in the subscales of body/health; talking with parents; personal growth; health literacy; and socialising subscales) and the 20 items

relating to negative outcomes (items in the subscales of life challenges; thinking/memory problems; and financial problems subscales). Further items in the IOC-CS, which are not included in the impact scales or the above subscales, ask survivors about concerns with siblings and concerns about fertility and intimate relationships. Zebrack *et al.* (2010) suggest these items contribute to three additional scales of: sibling concerns, relationship concerns (non-partnered); and relationship concerns (partnered). Higher scores on all the subscales and the impact scores indicate a greater perceived impact of cancer, whether that be positive or negative.

Therefore, the IOC-CS measures aspects of long-term survivorship currently not assessed by existing tools used in childhood and adolescent cancer survivors (Zebrack *et al.* 2009). Although this measure was specifically designed for use in young adult childhood cancer survivors, it has not previously been used in a British survivor population. The authors recommend that it be used in combination with other measures of HRQoL, such as the SF-36 (Zebrack *et al.* 2010).

SF-36 Version 2

As discussed in the literature review, HRQoL is considered an important outcome in survivors, however, a disease-specific validated measure of HRQoL does not currently exist for young adult survivors of childhood cancer. The SF-36 Version 2 (SF-36v2) provides a broad overview of a patient's health status and its effect on his or her functioning (Ware *et al.* 1996). Although a generic measure, the earlier version of the SF-36 was validated within a British sample of adult survivors of childhood cancer (Reulen *et al.* 2006). However, the SF-36v2 is recommended for research as it contains minor modifications to the wording of six items which makes it more acceptable in the British context (Jenkinson *et al.* 1999).

The SF-36v2 gives an overall health profile by measuring eight dimensions of physical functioning (PF), role-physical (RP), bodily pain (BP), general health (GH), vitality (VT), social functioning (SF), role-emotional (RE), and mental health (MH). In addition, two psychometrically based summary measures are calculated, the physical component summary (PCS) and the mental component summary (MCS). Higher scores on the health domain scales and the component summary measures indicate better health.

Due to the population norms for the U.K. dating from 1999 (Jenkinson *et al.* 1999), it is advised to use the 2009 U.S. norms (personal communication, Jenkinson 29th Feb 2016). In support of this, the U.S. and U.K. scoring algorithms have been reported to produce summary measures scores which are very highly correlated (0.997 for the PCS summary scale and 0.995 for the MCS summary scale) (Ware *et al.* 1998).

The raw scores of the SF-36v2 are transformed to norm-based T-scores with a general population mean of 50 and standard deviation of 10. This procedure is recommended to enable the health domain scale results to be meaningfully compared with one another and to simplify interpretation of the results based on 2009 U.S. general population normative data scores (Maruish, 2011). For analysing group level data, T-scores of 47-53 are considered within the normal range. Scores below this may signify impairment, whilst scores above this range may indicate above-average functioning (Maruish, 2011).

The Brief Symptom Inventory-18 (BSI-18)

Psychological distress is considered an important outcome in survivors as evidenced in the literature review. The BSI-18 is a brief self-report measure with 18 items designed to screen for psychological distress symptoms experienced over the previous seven days (Derogatis, 2001). By using the BSI-18 it will be possible to

identify risk factors which may be associated with increased distress in survivors such as the presence of scarring as was suggested in the qualitative study. The BSI-18 has been previously validated in adult survivors of childhood cancer (Recklitis & Rodrigues, 2007). Items are rated on a five-point Likert scale ranging from 'Not at all' (0) to 'Extremely' (4) and contribute to three symptom dimensions of somatisation, depression and anxiety and a global severity index (GSI) which represents the respondents' overall level of psychological distress. Raw scores for each of the three subscales and the GSI are converted to gender specific standardised T-scores using adult community based norms. The standardized T-scores for BSI-18 scales have a mean of 50 and a standard deviation of 10. Higher scores are associated with higher levels of distress. A T-score of ≥ 63 on the GSI or any two of the three subscales suggests 'caseness' which suggests respondents are testing positive for distress and require further assessment (Derogatis, 2001).

The Shortened Warwick-Edinburgh Mental Well-being Scale (SWEMWBS)

In addition to measuring psychological distress, and related to the findings that survivors often report good levels of wellbeing, a measure of mental well-being was included. The seven-item SWEMWBS which has been developed and validated in the UK was chosen (Tennant *et al.* 2007). The positively worded items cover the feeling and functioning aspects of mental wellbeing and response categories range from *None of the time (1)* to *All of the time (5)*. Higher scores are indicative of better mental wellbeing, however, the SWEMWBS does not state a cut-off point indicating mental wellbeing or otherwise. Therefore, it is stated that the SWEMBWS is not a clinical tool, but may be of use in investigating the determinants of mental wellbeing

(Warwick Medical School, 2014). Normative data are available via the Health Survey for England (2011) (Warwick Medical School, 2014).

As outlined in Appendix E, items to assess social outcomes of education, employment, income and relationship status were adapted from existing surveys including the Health Survey for England 2012 (Health and Social Care Information Centre, 2012), the British Childhood Cancer Survivor Study (Hawkins, 1999), and the Childhood Cancer Survivor Study (Robison *et al.* 2002).

Additional items were also developed which aimed to capture detailed information on the survivors' experiences in, and views of, education, employment and relationships, for example: their level of satisfaction with educational attainment; experiences relating to education (e.g. time missed at school due to illness, educational support received); satisfaction with current job; problems encountered in gaining or keeping employment; perceived reasons for not being married or being a parent and level of desire to marry and to have children. To collect additional information, open questions were included in which the participants' were invited to expand on their answer to a previous question (e.g. if you have never been married, why do you think this is?). Open questions allow respondents freedom to answer as they wish (Oppenheim, 2005).

To assess survivors beliefs regarding their future health and the perceived likelihood of developing ill health, survivors were asked to indicate on a 5-point Likert scale the extent to which they felt it was *very unlikely (1)* or *very likely (5)* that they would develop each of 21 health issues in the future. These 21 listed health issues are well known late adverse effects of treatment in survivors (e.g. heart problems, diabetes, poor hearing, anxiety, depression). If respondents felt they had already developed

this health issue, they were asked to tick '*I already have this problem*'. To detect the presence of scarring, which was identified as being an important issue in the qualitative study, single items asked if survivors had scarring on three different areas of the body (face or neck; chest or stomach; legs or arms) and required a *yes/no* response.

4.4.4 Data processing and analysis

The primary aim of the study was to pilot the questionnaire and to test the overall feasibility and acceptability of the proposed research measures, methods and procedures of the study. The questionnaire was piloted to assess whether included measures would provide a valid, reliable and insightful assessment of psychosocial outcomes in survivors. The questionnaire was also to be evaluated as to whether it would allow an investigation into a range of factors which may be associated with poorer psychosocial outcomes and to identify possible associations which may be investigated in a larger study. Acceptability of the questionnaire to survivors was also of importance. Therefore, analysis at this stage was primarily descriptive.

Data quality was assessed for each measure by calculating the percentage of items with valid responses (Maruish, 2011). A large number of missing items would suggest items, or the measures, are unacceptable or unclear to respondents (Streiner & Norman, 2008). Cronbach's alpha was utilised to assess the internal consistency of scales in each measure. An acceptable value for Cronbach's alpha is ≥ 0.70 which suggests that the items within the scale are measuring the same underlying construct and is a measure of scale reliability (Maruish, 2011; Pevalin & Robson, 2011).

Prior to analyses, a Shapiro Wilk test was used to test normality of data. Results indicated that the subscales of the measures were not normally distributed. Despite the subscales of the SF-36 rarely being normally distributed, parametric methods are generally used (Torrance *et al.* 2009). It is also standard practice to report the mean for the SF-36 scores (Bowling, 1999), despite this being statistically incorrect to do so with non-normal data. The authors of the IOC-CS also state that mean scores should be utilised for scoring and analyses (Zebrack *et al.* 2010). Both parametric (t-test) and non-parametric (Man-Whitney *U*-test) tests were performed and were found to give similar results in terms of statistical significance. Therefore, it was decided to present the results of the parametric tests as doing so may help compare results to existing literature (Torrance *et al.* 2009).

One sample t-tests were used to test the samples mean score to expected norms for the SF-36v2, BSI-18, and SWEMWBS measures. Bivariate analyses were utilised to compare the mean scores of the SF-36 v2, BSI-18 and IOC-CS scales across demographic and disease and treatment variables. Independent t-tests were used to compare the mean scores between two variables (e.g. gender, relationship status) and one-way analysis of variance (ANOVA) tests were used to compare the means across three (e.g. cancer diagnosis, age at diagnosis). Where ANOVA results provided a significant p-value, a Scheffé multiple comparison test was used to detect between which groups the difference existed. The Scheffé test is stated to be a cautious post-hoc test which reduces the risk of a Type 1 error, although due to low power it may be less likely to detect a difference between the groups (Pallant, 2007).

To enable analysis, a number of variables were collapsed. Survivors were categorised by their age at diagnosis (0-5; 6-11; 12-18 years) to reflect being diagnosed as a young

child, older child and teenager; age at study (18-23; 24-29 years) and time since treatment (5-10; 11-15; 16-24 years) to capture any differences which may increase or lessen with age or over time since diagnosis. Analyses involving educational attainment included only survivors aged 21 years or above to better assess final educational outcomes and educational level was categorised as being 'degree level' or lower ('higher education certificate other than degree [e.g. Higher National Diploma] and A-level or GCSE). Categorisation of employment outcomes followed that of Zebrack & Landier (2011) in that survivors who were employed full-time or part-time, cared for home or family or were students were classed as being 'occupied'. Survivors who were unemployed and looking for work or unable to work due to illness or disability, were categorised as being 'unoccupied'. Income was dichotomised to a personal income of less than £200 and more than £201 a week. This cut-off point for collapsing income was for pragmatic reasons to produce two groups with roughly equal numbers of survivors. Survivors who were married, living as married, or in a significant committed relationship were categorised as being in a relationship, with remaining survivors being categorised as being single.

The list of 21 known late effects of childhood cancer were grouped as either somatic (e.g. hearing problems, dental damage, heart problems, hormone problems) or psychological late effects (e.g. depression, anxiety, mood swings, difficulty with learning and memory). Respondents who indicated they already had one or more of these health issues were categorised into a yes/no variable for the relevant late effects group (somatic or psychological). Surgery, chemotherapy, radiotherapy and stem cell transplant were also treated as categorical yes/no variables as was the self-report of scarring.

Correlation was used to assess concurrent validity of the IOC-CS in comparison to the scales of the SF-36 and BSI-18 as well as the SWEMWBS measure to the SF-36v2. Due to all not being normally distributed, the non-parametric Spearman's Rank Order Correlation was utilised.

Due to the limited data in the free text answers, they were analysed using basic content analysis, a systematic and objective process which enables the reduction of data by establishing categories and recording the frequency at which they are present in the data (Joffe & Yardley, 2004).

All data were first entered into a Microsoft Access database before being imported into the statistical software package Stata, version 12 (Statacorp, College Station, TX), for data cleaning and analyses.

4.5 Results

Recruitment to the study ran from March to December 2015. A total of 217 eligible survivors were posted a questionnaire (Figure 4), of which 213 questionnaires were understood to have been received by survivors. Three non-responder feedback sheets were returned: two survivors replied that they did not have the time to complete the questionnaire (0.9%) and one parent responded that their child would not be able to take part due to their special needs. A total of 94 completed questionnaires were returned giving a response rate of 44%. However, one participant was found to only be 3.7 years from diagnosis and, therefore, did not meet the eligibility criteria so was excluded from the analyses. Therefore, the analysis is based on 93/213 (44%) of questionnaires returned by young adult survivors of childhood cancer.

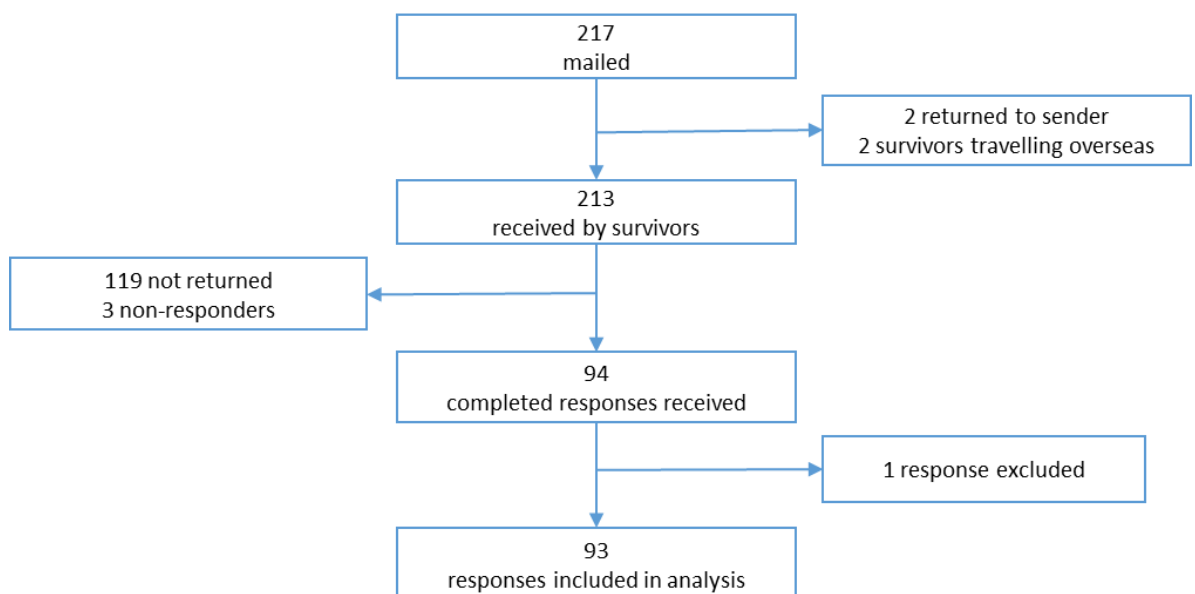


Figure 4 Flowchart detailing response to the questionnaire

4.5.1 Patient characteristics

Of the 93 respondents, 52 (55%) were female and almost all were white/European (Table 3). Median age at the time of study was 22.9 years (range 18-29 years). Median age at diagnosis was 9 years (range 0-18 years) with a median time since diagnosis of 13 years (range 4.6-24.3 years).

The most represented cancer group was haematological malignancies (n=36, 38.7%). The other cancer groups represented were CNS tumours (n=34, 36.6%) and other solid tumours (n=23, 24.7%). Patients reported they had received chemotherapy (n=59, 63.4%), radiotherapy (n=37, 39.8%), surgery (n=69, 74.2%), and stem cell transplant (n=13, 16.7%).

It was not possible to compare the characteristics of responders to non-responders due to the ethical constraints of the disease registry which meant that the researcher was not able to access personal data without patient consent.

Table 3 Characteristics of survivors who responded to questionnaire

Variable	Survivors n (%)
Gender (n=93)	
<i>Male</i>	41 (45)
<i>Female</i>	52 (55)
Ethnicity (n=93)	
<i>White European</i>	89 (95)
<i>Other</i>	4 (5)
Diagnosis (n=93)	
<i>Haematological</i>	36 (38.7)
<i>Central nervous system tumour</i>	34 (36.6)
<i>Other solid tumour</i>	23 (24.7)
Treatment (n=93)	
<i>Surgery</i>	69 (74.2)
<i>Chemotherapy</i>	59 (63.4)
<i>Radiotherapy</i>	37 (39.8)
<i>Stem cell transplant</i>	13 (16.7)
Age at study (years) (n=93)	
Mean (range)	22.9 (18-29)
<i>18-23</i>	53 (57.00)
<i>24-29</i>	40 (43.00)
Age at diagnosis (years) (n=93)	
Mean (range)	9 (0-18)
<i>0-5</i>	28 (30.1)
<i>6-11</i>	29 (31.2)
<i>12-18</i>	36 (38.7)
Time since diagnosis (years) (n=93)	
Mean (range)	13 (4.6-24.3)
<i>5-10</i>	35 (37.6)
<i>11-15</i>	31 (33.3)
<i>16-24</i>	27 (29.0)
Relationship status (n=92)	
<i>Married</i>	1 (1.1)
<i>Recognised civil partnership</i>	1 (1.1)
<i>Divorced</i>	1 (1.1)
<i>In long-term relationship (cohabiting)</i>	21 (22.8)
<i>In long-term relationship (not cohabiting)</i>	16 (17.4)
<i>In casual relationship</i>	4 (4.3)
<i>Single</i>	48 (52.2)

4.5.2 Social outcomes

To help put the study findings in context, the results regarding the social outcomes of the survivors will be presented prior to the psychological outcome data.

Marriage, relationships and parenthood

Only two survivors (2%) were married or in a civil partnership (Table 3). However, over 40% indicated that they were in a relationship, with 21 (22.8%) stating they co-habited with a long-term partner. The majority of unmarried survivors (66.7%) reported a high desire to marry in the future (Table 4). Common reasons for not being married at present were being too young/not the right time (n=41/68, 60.3%) and not having met the right person (n=17, 25.0%). Less common reasons were not wanting to get married (n=5, 7.4%), having confidence issues (n=3, 4.4%), having no desire for relationship (n=2, 2.9%), and not seeing marriage as a necessity (n=2, 2.9%).

Fifty (94.3%) of the single survivors completed a section about relationship concerns. Over half (56%) worried to some extent about not having a partner/spouse/boyfriend/girlfriend. To a lesser extent, worries were evident about disclosing a cancer history (34.0%), having sex (40.0%), and telling a potential partner that they may not be able to have children (40.0%).

Thirty-nine (93%) of the survivors currently in a relationship completed questions about relationship concerns. Conversely, these survivors reported few relationship worries. Only (5%) stated they were not comfortable talking to their partner about their health problems or worried about having sex with their partner. However, approximately quarter (25.6%) had some concerns that their partner would leave if they were to get cancer again.

Table 4 Survivors' views and concerns about romantic relationships

Item	Survivors n (%)
Desire to marry in unmarried respondents (n=90)	
<i>Quite a bit/very much</i>	60 (66.7)
<i>A little/somewhat</i>	20 (22.2)
<i>Not at all</i>	10 (11.1)
Relationship concerns for those not in significant relationship (n=50)	
Worry about not having a partner	
<i>Quite a bit/very much</i>	15 (30.0)
<i>A little/somewhat</i>	13 (26.0)
<i>Not at all</i>	22 (44.0)
Worry about telling a potential partner about cancer history	
<i>Quite a bit/very much</i>	6 (12.0)
<i>A little/somewhat</i>	11 (22.0)
<i>Not at all</i>	33 (66.0)
Worry about telling a potential partner may not have able to have children	
<i>Quite a bit/very much</i>	10 (20.0)
<i>A little/somewhat</i>	10 (20.0)
<i>Not at all</i>	30 (60.0)
Worry about having sex	
<i>Quite a bit/very much</i>	5 (10.0)
<i>A little/somewhat</i>	15 (30.0)
<i>Not at all</i>	30 (60.0)
Relationship concerns for those in significant relationship (n=39)	
Comfortable in taking to partner about health problem	
<i>Quite a bit/very much</i>	31 (79.5)
<i>A little/somewhat</i>	6 (15.4)
<i>Not at all</i>	2 (5.1)
Worry about my partner leaving me if I get cancer again	
<i>Quite a bit/very much</i>	2 (5.1)
<i>A little/somewhat</i>	8 (20.5)
<i>Not at all</i>	29 (74.4)
Worry about having sex with partner	
<i>Quite a bit/very much</i>	0
<i>A little/somewhat</i>	2 (5.1)
<i>Not at all</i>	37 (94.9)

Twenty-two (24.2%) survivors reported that they had been told that it would be unlikely they would ever be pregnant or father a child (Table 5). Only seven (7.6%) survivors (4 males) reported they had biological children. Almost three-quarters of childless survivors reported a high desire to have children in the future (71.6%). However, 23 (27.1%) survivors reported a high level of concern that they would not be able to have children. The most common reason for not yet having children was being too young (n=36/79; 45.6%) and not having found the right partner (n=14/79; 17.7%). Almost a third of survivors (n=28, 30.8%) stated that they were very concerned their children or future children may get cancer.

Table 5 Survivors' views and concerns about parenthood

Item	Survivors n (%)
Been told it will be unlikely to be pregnant or father a child (n=91) <i>Yes</i> <i>No</i> <i>Don't know</i>	 22 (24.2) 62 (68.1) 7 (7.7)
Have biological children (n=92) <i>Yes</i> <i>No</i>	 7 (7.6) 85 (92.4)
If no children, desire to have children in the future (n=81) <i>Quite a bit/very much</i> <i>A little/somewhat</i> <i>Not at all</i>	 58 (71.6) 18 (22.2) 5 (6.2)
If no children, concerned may not be able to have children (n=85) <i>Quite a bit/very much</i> <i>A little/somewhat</i> <i>Not at all</i>	 23 (27.1) 32 (37.7) 30 (35.3)
Concerned children/future children may get cancer (n=91) <i>Quite a bit/very much</i> <i>A little/somewhat</i> <i>Not at all</i>	 28 (30.8) 31 (34.0) 32 (35.2)

Education and employment

Almost a third of the sample reported a degree level education (Table 6). However, when restricted to survivors aged 21 years or above, approximately 40% had achieved a university education (n=28/68; 41.2%) and 12 (17.7%) had received a higher education certificate such as an HND or BTEC, 13 (19.4%) A-levels, 14 (14.93%) GCSE level. Sixteen (n=16/91, 17.6%) respondents of the overall sample were still in education (Table 7).

A large majority of survivors (87%) had missed time at school due to their illness, with almost a third missing over 12 months (29%) (Table 6). However, of these survivors only six reported repeating a school year (7.6%). Almost half of the survivors (46.2%) had received extra tutoring whilst on treatment, while approximately 40% reported receiving extra educational support at school. Approximately a quarter of survivors (28.4%) stated they were bullied or teased at school due to their illness.

Overall, the majority stated they were satisfied with their educational attainment (n=59, 63%). However, 64 (68.8%) felt that their past illness had affected their educational achievements to some extent. Fifty-six survivors gave their views on how their illness had specifically affected their education. Reasons were: missed school (n=32, 53.3%); poor memory and concentration (n=9/60, 15%); decreased ability to learn (n=9/60; 15%); and fatigue (n=4, 6.7%). However, six (10.0%) stated a positive impact such as they learnt better, were more focused and achieved higher than before they were ill.

Table 6 Survivors' outcomes, experiences and views of education

Item	Survivors n (%)
Highest educational level (n=92)	
<i>University degree</i>	29 (31.5)
<i>Higher education certificate</i>	13 (14.1)
<i>A-level</i>	28 (30.4)
<i>GCSE</i>	20 (21.7)
<i>Don't know</i>	2 (2.18)
Time missed at school (n=93)	
<i>No</i>	12 (13.0)
<i>Yes, less than a month</i>	5 (5.4)
<i>Yes, between 1-6 months</i>	27 (29.0)
<i>Yes, 6-12 months</i>	22 (23.7)
<i>Yes, more than 12 months</i>	27 (29.0)
Repeated year at school (n=79)	
<i>No</i>	73 (92.4)
<i>Yes, 1 year</i>	5 (6.3)
<i>Yes, 2 years</i>	1 (1.3)
<i>Yes, more than 2 years</i>	0 (0)
Educational support	
<i>Received extra tutoring whilst on treatment (n=92)</i>	43 (46.7)
<i>Received extra educational support at school (n=91)</i>	36 (39.6)
Reasons for extra educational support (n=34)	
<i>Missed school</i>	25 (73.5)
<i>Problems learning or concentrating</i>	12 (35.3)
<i>Emotional or behavioural problems</i>	3 (8.8)
<i>Low scores on tests</i>	2 (5.9)
Satisfied with educational achievement (n=93)	
<i>Very much/Quite a bit</i>	59 (63.4)
<i>A little bit/Somewhat</i>	19 (20.4)
<i>Not at all</i>	15 (13.1)
Feel illness affected educational achievement (n=93)	
<i>Very much/Quite a bit</i>	31 (33.3)
<i>A little bit/Somewhat</i>	33 (35.5)
<i>Not at all</i>	29 (31.2)
Experienced bullying at school due to illness (n=88)	
<i>Yes</i>	25 (28.4)
<i>No</i>	63 (71.6)

At the time of study, almost half of the survivors (46.2%) reported to be in full-time employment (Table 7). Eight (8.8%) stated to be unemployed and looking for work and eight (8.8%) were unable to work due to illness or disability.

Table 7 Survivors' outcomes, experiences and views of employment

Item	Survivors n (%)
Employment status (n=92)	
Employed full-time	42 (46.2)
Employed part-time	16 (17.6)
Care for home/family	1 (1.1)
Unemployed & looking for work	8 (8.8)
Unable to work due to illness/disability	8 (8.8)
Student	16 (17.6)
Satisfied with current job (full-time employment) (n=42)	
<i>Very much/Quite a bit</i>	29 (69.0)
<i>A little bit/Somewhat</i>	10 (23.8)
<i>Not at all</i>	3 (7.1)
Having had cancer limits my ability to work (n=91)	
<i>Very much/Quite a bit</i>	16 (17.6)
<i>A little bit/Somewhat</i>	16 (17.6)
<i>Not at all</i>	59 (64.8)
Had problems getting or keeping employment due to cancer history (n=92)	
<i>Yes</i>	9 (9.8)
<i>No</i>	74 (80.4)
<i>Don't know</i>	4 (4.4)
<i>Never tried to get employment</i>	5 (5.4)
Cancer history prevented you from pursuing a career/occupation? (n=92)	
<i>Yes</i>	16 (17.4)
<i>No</i>	71 (77.2)
<i>Don't know</i>	2 (2.2)
<i>Never tried to get employment</i>	3 (3.3)

Of the full-time workers, the majority (69%) were highly satisfied with their current job. Approximately a third of respondents felt that their cancer history limited their ability to work to some extent (35.2%). However, only nine (9.8%) survivors stated they had experienced problems getting or keeping employment because of their

cancer history. Perceived reasons for this were discrimination (n=1), health and mobility issues (n=3), issues due to general cancer history (n=2), tiredness and irritability (n=1) and having poor vision (n=1).

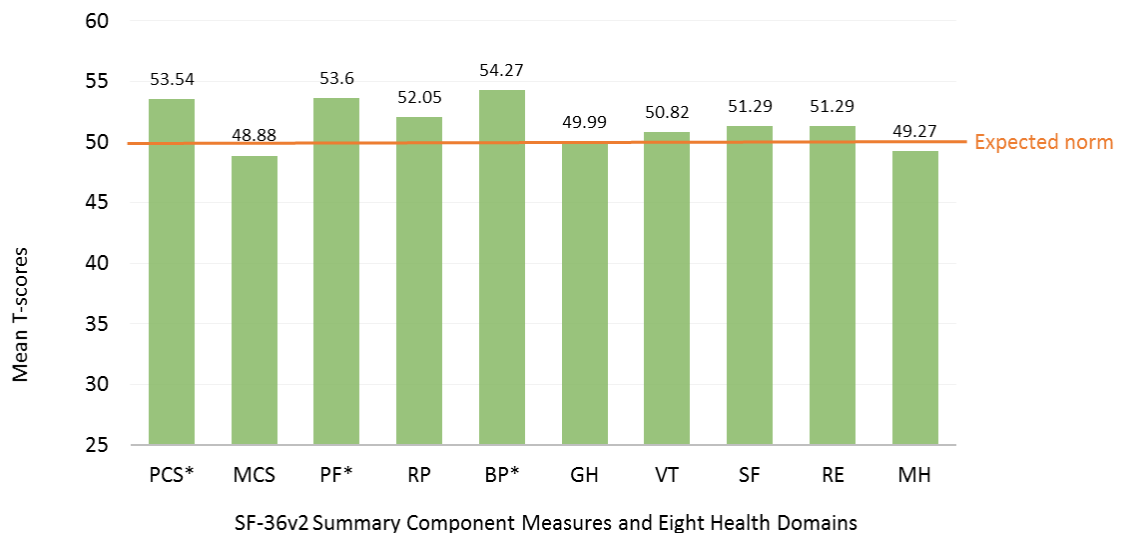
Sixteen (17.4%) survivors stated that their cancer history had prevented them from pursuing a career they wanted to do, including the military (n=3), emergency services (n=1), veterinary surgeon/zoo keeper (n=2), nursing (n=2), childcare (n=1), social work (n=1), career requiring university education (n=2) and working on a cruise ship (n=1). Reasons given for this were their cancer history in general (n=6), physical disability (n=5), learning difficulties (n=3), mental health issues (n=2), fatigue (n=3), self-esteem and confidence (n=2), and lack of education (n=2).

4.5.3 Psychological outcomes

Health related quality of life

Data completeness for the SF-36v2 was 99.5%. Cronbach's alpha for the eight health domain scores ranged from 0.84 to 0.96, and were 0.94 for the physical component summary (PCS) score and 0.93 for the mental health component summary (MCS) score, suggesting good to excellent internal consistency.

Survivors demonstrated scores within the expected normal range of 47-53 for the MCS and the subscales of mental health (MH), role-emotional (RE), social functioning (SF), vitality (VT), general health (GH) and role-physical (RP) (Figure 5). However, scores were significantly above the expected range for the PCS score and physical function (PF) and bodily pain (BP), suggesting good health status, particularly for aspects of physical function.



*One sample t-test to compare the means for the sample to standardised norms and significant at $p < 0.001$

Figure 5 The comparison of standardised mean T-scores for the SF-36v2 to expected norms

Ceiling effects, the proportion of respondents who score the maximum possible T-score (indicating good health status in terms of the SF-36v2), ranged from 3.5% for the vitality scale to 69.0% for the role-emotional scale (Table 8). Floor effects (respondents who score the lowest possible T-scores, indicating poor health status) ranged from 0% for vitality to 2.3% for social functioning.

Table 8 Ceiling and floor effects for SF26v2 subscales

	PF	RP	BP	GH	VT	SF	RE	MH
Ceiling effect %	58.6	59.8	51.7	12.6	3.5	57.5	69.0	5.8
Floor effect %	0	1.2	0	0	0	2.3	1.2	0

As shown in Table 9, female survivors (Mean =51.72, SD =11.43) reported lower PCS scores than male survivors (Mean =55.89, SD =6.96, $t(85) = 1.98$, $p=0.05$, two tailed). Survivors with a university degree reported higher PCS scores than survivors who had a higher education certificate, A-levels or GCSEs (Mean =56.85, SD 5.89 vs Mean =50.18, SD =11.49, $t(61) = 1.26$, $p=0.009$, two tailed). Survivors who were classed as being occupied, demonstrated higher PCS scores (Mean =55.91, SD =7.21) than survivors who were not (Mean =42.57, SD =13.49, $t(83) =5.36$, $p=0.00$, two tailed). Survivors with a higher personal income reported higher MCS scores (Mean =45.33, SD =13.70 vs Mean =51.10, SD =8.74, $t(83) =-2.29$, $p=0.02$, two tailed). No significant differences were demonstrated for the MCS and PCS scores for across the age at study groups and for relationship status (all p -values >0.05).

Table 9 Bivariate comparisons of the component summary scores of the SF36v2 to sociodemographic and medical variables

	PCS		MCS	
	Mean (SD)	P value	Mean (SD)	P value
Gender				
Male (n=38)	55.89 (6.96)	0.05	49.42 (10.66)	0.70
Female (n=49)	51.72 (11.43)		48.46 (12.08)	
Education¹				
University degree (n=26)	56.84 (5.89)	0.009	50.34 (6.78)	0.21
Higher education certificate or lower (n=37)	50.18 (11.49)		46.75 (13.33)	
Employment				
Occupied ² (n=71)	55.91 (7.21)	<0.001	49.56 (10.68)	0.15
Unoccupied ³ (n=14)	42.57 (13.49)		44.69 (14.96)	
Income				
≤£200 a week (n=38)	52.70 (10.00)	0.56	45.33 (13.70)	0.02
≥201 a week (n=43)	54.04 (10.43)		51.10 (8.74)	
Relationship status				
Married/in relationship (n=37)	54.26 (9.67)	0.56	50.12 (10.13)	0.39
Single (n=50)	53.00 (10.13)		47.96 (12.33)	
Age at study				
18-23 (n=51)	53.62 (9.76)	0.92	49.14 (12.09)	0.80
24-29 (n=36)	53.42 (10.23)		48.51 (10.57)	
Diagnosis group				
Haematological (n=28)	57.08 (7.61)	0.28	50.57 (9.33)	0.52
CNS (n=28)	52.97 (10.61)		51.83 (8.12)	
Solid tumour (n=31)	51.22 (10.47)		44.69 (14.42)	
Chemotherapy				
Yes (n=54)	53.54 (10.40)	0.94	49.31 (10.78)	0.67
No (n=32)	53.36 (9.27)		48.23 (12.76)	
Radiotherapy				
Yes (N=35)	50.59 (10.71)	0.04	50.10 (11.46)	0.29
No (N=46)	55.32 (9.33)		47.32 (11.84)	
Surgery				
Yes (n=63)	53.29 (9.78)	0.93	48.24 (11.75)	0.64
No (n=19)	53.54 (11.34)		49.70 (11.64)	
Age at diagnosis				
0-5 (n=28)	57.08 (7.60)	0.06	50.57 (9.33)	0.03
6-11 (n=28)	52.57 (10.61)		51.83 (8.12)	
12-18 (n=31)	51.22 (10.47)		44.69 (14.42)	
Time since diagnosis (years)				
5-10 (n=32)	50.88 (11.97)	0.06	46.61 (14.34)	0.36
11-15 (n=29)	53.27 (90.02)		50.56 (9.43)	
16-24 (n=26)	57.10 (6.79)		49.79 (9.18)	

¹ Survivors aged 21 or over only included in analyses; ² Includes full time employment, part-time employment, caring for home or family, student; ³ Includes unemployed and looking for work and unable to work due to illness or disability; **Bold** indicates results significant at p<0.05

Survivors treated with radiotherapy reported significantly lower PCS scores than those who had not (Mean =50.59, SD =10.71 vs Mean =55.32 (9.33), $t(79) = -2.12$,

p=0.04). There was a trend for those diagnosed at a younger age to demonstrate higher MCS scores suggesting better mental health ($F(2,84) = 3.52, p=0.03$). A Scheffé post hoc test indicated that the difference was significant for the survivors diagnosed at ages 12-18 years (Mean =44.69, SD =14.42) compared to those diagnosed aged 6-11 years old (Mean =51.83, SD =8.12, $p=0.05$). There was no significant difference in the PCS or the MCS scores across diagnosis, age at study or time since diagnosis groups (all $p>0.05$).

As seen in Table 10, survivors who reported somatic late-effects (Mean =50.68, SD =11.15) reported lower PCS T-scores than those who did not (Mean =58.70, SD =3.19, $t(85), p=0.0002$, two tailed). Similarly, survivors who reported psychological late-effects reported lower MCS T-scores (Mean =42.71, SD =13.93) than those who did not (Mean =53.44, SD =6.06, $t(85), p <0.001$, two tailed). There were no significant differences between survivors who reported scarring and those who did not (all p values >0.05).

Table 10 Bivariate comparisons of the component summary scores of the SF36v2 to self-reported late effects and scarring

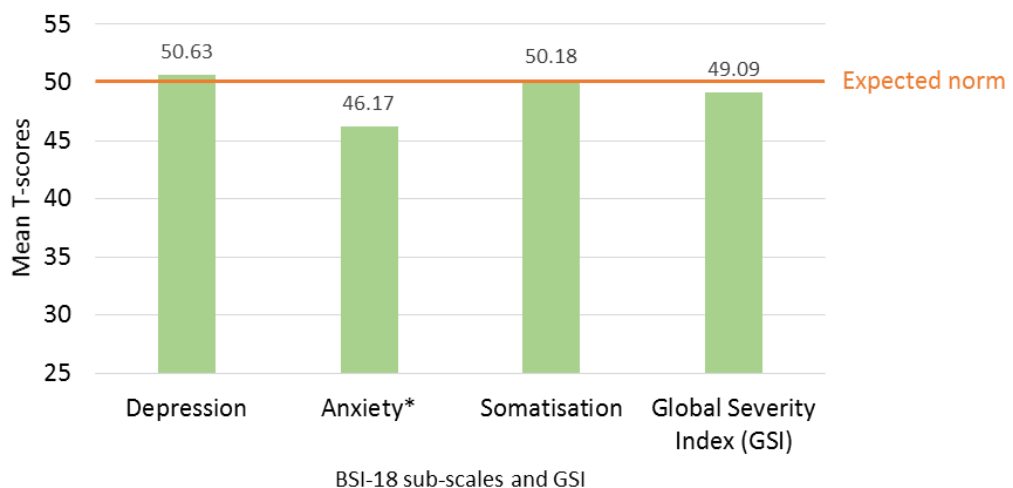
	PCS		MCS	
	Mean (SD)	P value	Mean (SD)	P value
Somatic late effects				
Yes (n=56)	50.68 (11.15)	<0.001	47.71 (11.84)	0.20
No (n=31)	58.70 (3.19)		50.98 (10.51)	
Psychological late effects				
Yes (n=37)	51.69 (10.67)	0.13	42.71 (13.93)	<0.001
No (n=50)	54.91 (9.16)		53.44 (6.06)	
Scarring to face/head/neck				
Yes (n=53)	52.89 (9.80)	0.48	47.81 (11.73)	0.27
No (n=33)	54.47 (10.27)		50.66 (11.05)	
Scarring to chest/stomach				
Yes (n=58)	54.33 (9.23)	0.26	49.76 (11.07)	0.33
No (n=28)	51.75 (11.28)		47.14 (12.35)	
Scarring to legs/arms				
Yes (n=24)	51.70 (12.11)	0.30	49.27 (12.56)	0.85
No (n=62)	54.19 (8.99)		48.76 (11.16)	

Bold indicates results significant at $p<0.05$;

Psychological distress

Data completeness for the BSI-18 was 98.7%. One survivor chose not to complete the measure. Three missing values were imputed as directed by Derogatis (2001). Ceiling effects were 0% for the subscales and 1.0% for the global severity index (GSI). Floor effects ranged from 21.7% to 48.9%, indicating low levels of distress symptoms. Cronbach's alpha for BSI-18 subscales were: depression (0.93); anxiety (0.81); somatisation (0.66); global severity index (0.91).

Overall, the majority of survivors in this study demonstrated few symptoms of psychological distress (Figure 6). Survivors mean scores did not differ significantly from standardised norms on the depression, somatisation or global severity index (GSI) scale. Survivors did, however, report significantly lower anxiety scores suggesting lower levels of anxiety symptoms than would be expected in general population.



*One sample t-test to compare the means for the sample to standardised norms and significant at $p < 0.01$

Figure 6 *The comparison of standardised mean T-scores for the BSI-18 to expected norms*

Thirteen (14.13%) survivors had T-scores above the threshold of ≥ 63 on the GSI or two of the subscales and were classified as 'cases' at positive risk of distress. Survivors classed as cases had poorer outcomes on the MCS scores (Mean =30.68, SD =14.22) than those who were not (mean =51.99, SD =7.18, $t(84) = 8.28$, $p < 0.001$, two tailed). There was no significant difference in the PCS scores of cases and non-cases ($p = 0.23$).

As shown in Table 11, survivors who were married or in a significant relationship reported significantly lower scores on the depression subscale (Mean =45.93, SD =8.83) compared to survivors who were single (Mean 54.25, SD 14.10, $t(90) = -3.27$, $p = 0.002$, two tailed) as well as lower GSI scores (Mean =46.20, SD =9.45 vs Mean =51.31, SD =12.48, $t(90) = -2.15$, $p = 0.03$, two tailed). Significantly lower scores were also reported by survivors with a higher personal income for depression (Mean =54.53, SD =14.21 v Mean =48.17, SD =11.04, $t(84) = 2.33$, $p = 0.02$, two tailed), anxiety (Mean =48.98, SD =11.52 vs Mean =44.39, SD =7.43, $t(84) = 2.22$, $p = 0.03$, two tailed) and the GSI (Mean =52.59, =SD 12.23 vs Mean =46.85, SD =10.15, $t(84) = 2.53$, $p = 0.01$, two tailed).

There were no significant differences in the subscales and GSI scores of the BSI-18 across diagnoses or by treatment (all p values > 0.05). However, GSI scores differed across the age at diagnosis groups ($F(2,89) = 4.75$, $p = 0.01$) with a Scheffé post hoc test suggesting that survivors diagnosed as teenagers reported higher GSI scores and overall psychological distress than survivors diagnosed at 0-5 years ($p = 0.05$) and 6-11 years old ($p = 0.03$). There was a significant different difference in somatisation scores by time since diagnosis ($F(2,89) = 3.18$, $p = 0.05$), with survivors 5-10 years from diagnosis reporting higher scores and more somatic symptoms than survivors 11-15 years from diagnosis ($p = 0.05$).

Table 11 Bivariate comparisons of the BSI-18 to sociodemographic and medical variables

	Depression Mean (SD)	Anxiety Mean (SD)	Somatisation Mean (SD)	GSI Mean (SD)
Gender				
Male (n=40)	50.48 (13.17)	46.63 (8.48)	48.3 (7.24)	49.00 (11.06)
Female (n=52)	50.75 (12.51)	45.83 (10.58)	51.63 (9.10)	49.15 (11.93)
Education¹				
University degree (n=28)	47.96 (9.77)	44.96 (5.28)	49.64 (6.93)	47.86 (8.27)
Higher education certificate or lower (n=41)	52.20 (13.9)	47.56 (11.09)	50.80(9.21)	50.21 (12.94)
Employment				
Occupied ² (n=74)	50.05 (12.39)	45.74 (9.33)	49.42 (8.08)	48.26 (11.22)
Unoccupied ³ (n=16)	53.19 (14.52)	48.31 (11.29)	53.19 (8.83)	53.06 (11.74)
Income				
≤£200 a week (n=40)	54.53 (14.21)	48.98 (11.52)	52.05 (9.43)	52.95 (12.23)
≥201a week (n=46)	48.17 (11.04)	44.39 (7.43)	49.34 (7.59)	46.85 (10.15)
Relationship status				
Married/in relationship (n=40)	45.93 (8.83) 54.25 (14.10)	44.30 (8.26) 47.62 (10.44)	51.03 (7.86) 49.54 (8.93)	46.20 (9.45) 51.31 (12.48)
Single (n=52)				
Diagnosis				
Haematological (n=36)	48.89 (11.64)	46.13 (9.30)	49.14 (7.13)	47.81 (10.75)
CNS (n=34)	52.64 (14.33)	47.44 (9.92)	51.21 (9.86)	51.00 (12.32)
Other solid tumour (n=22)	50.36 (11.91)	44.27 (9.90)	50.32 (8.35)	48.23 (11.50)
Age at study				
18-23 (n=52)	49.79 (12.51)	45.83 (10.14)	49.71 (9.40)	47.79 (12.16)
24-29 (n=40)	51.73 (13.10)	46.63 (9.06)	50.80 (7.14)	50.78 (10.48)
Chemotherapy				
Yes (n=59)	49.10 (11.34)	45.69 (9.25)	50.15 (8.01)	47.98 (10.89)
No (n=32)	53.00 (14.78)	46.88 (1.86)	50.31 (9.50)	50.48 (12.56)
Radiotherapy				
Yes (n=37)	51.32 (13.02)	47.25 (9.20)	51.27 (9.28)	50.57 (10.82)
No (n=49)	50.51 (12.84)	45.41 (10.24)	49.65 (7.97)	48.20 (12.27)
Surgery				
Yes (n=68)	51.31 (13.08)	46.22 (9.82)	50.37 (8.71)	49.41 (11.79)
No (n=19)	49.47 (2.93)	47.21 (9.97)	49.58 (8.43)	49.06 (11.65)
Age at diagnosis				
0-5 (n=28)	49.71 (12.68)	44.93 (9.29)	48.04 (7.66)	46.61 (11.72)
6-11 (n=28)	46.79 (10.21)	44.14 (7.17)	49.39 (8.51)	45.89 (9.83)
12-18 (n=31)	54.33 (13.80)	48.72 (11.16)	52.47 (8.70)	53.50 (11.40)
Time since diagnosis				
5-10 (n=35)	53.57 (14.08)	47.80 (11.17)	52.74 (9.61)	52.03 (12.60)
11-15 (n=30)	48.23 (11.22)	45.10 (8.18)	47.60 (7.25)	47.03 (10.09)
16-24 (n=27)	49.48 (12.17)	45.26 (9.05)	49.74 (7.40)	47.56 (11.07)

¹ Survivors aged 21 or over only included in analyses; ² Includes full time employment, part-time employment, caring for home or family, student; ³ Includes unemployed and looking for work and unable to work due to illness or disability; **Bold** indicates results significant at p<0.05

As shown in Table 12, survivors who reported somatic late effects scored significantly higher on the depression (Mean =53.07, SD =13.45 vs Mean =46.06, SD = 9.93, $t(90)=2.59$, $p=0.01$), somatisation (Mean =51.62, SD =8.80 vs Mean =47.50, SD =7.19, $t(90)=2.27$, $p=0.03$) as well as the GSI scale (Mean =51.62, SD =11.14 vs Mean =44.34 SD =10.78, $t(90)=3.02$, $p=0.003$).

Survivors who reported psychological late effects scored significantly higher on the depression (Mean =57.76, SD =14.06 vs Mean =45.61, SD =8.85 $t(90)=5.09$, $p<0.001$), anxiety (Mean =51.45, SD =11.57 vs Mean =42.46, SD =5.69, $t(90)=4.93$, $p<0.001$), somatisation (Mean =52.89, SD =8.87 vs Mean =48.28, SD =7.69, $t(90)=2.66$, $p=0.009$) and the GSI scale (Mean =55.72, SD =11.59 vs Mean =44.43, SD =8.92, $t(90)=5.28$, $p<0.001$).

Table 12 Bivariate comparisons of the BSI-18 to self-reported late effects and scarring

	Depression Mean (SD)	Anxiety Mean (SD)	Somatisation Mean (SD)	GSI Mean (SD)
Somatic late effects				
Yes (n=60)	53.07 (13.45)	47.50 (9.70)	51.62 (8.80)	51.62 (11.14)
No (n=32)	46.06 (9.93)	43.69 (9.17)	47.50 (7.19)	44.34 (10.78)
Psychological late effects				
Yes (n=38)	57.76 (14.06)	51.45 (11.57)	52.89 (8.87)	55.72 (11.59)
No (n=54)	45.61 (8.85)	42.46 (5.69)	48.28 (7.69)	44.43 (8.92)
Scaring to face/head/neck				
Yes (n=58)	52.03 (12.94)	47.91 (10.00)	50.81 (8.99)	50.76 (11.61)
No (n=33)	48.48 (12.28)	43.36 (8.40)	49.36 (7.47)	46.64 (10.78)
Scaring to chest/stomach				
Yes (n=61)	49.74 (12.0)	45.62 (9.49)	50.49 (8.16)	48.46 (11.27)
No (n=30)	52.80 (14.17)	47.57 (10.02)	49.87 (9.17)	50.90 (11.77)
Scaring to legs/arms				
Yes (n=26)	57.85 (10.07)	47.96 (10.28)	52.81 (9.61)	51.42 (11.84)
No (n=65)	50.31 (12.70)	45.58 (9.40)	49.28 (7.80)	48.40 (11.24)

Bold indicates results significant at $p<0.05$

Although survivors with scaring to either their face, head or neck reported higher distress levels for all BSI-18 subscales, this was only significant for the anxiety subscale (Mean =47.91, SD =10.00 vs Mean =43.36, SD =8.40, $t(90)=2.21$, $p=0.03$).

Survivors with scarring to the chest or limbs did not report significantly different levels to survivors without (all p values >0.05).

Mental wellbeing

Data completion for the SWEMWBS was 100% and Cronbach's alpha was 0.91, suggesting excellent internal consistency of the scale items. The ceiling effect was 4% and the floor effect 1%. Survivors' mean scores for mental wellbeing as measured by the SWEMWBS were 23.25 (SD =5.14) with no difference compared to the population norm (Mean =23.6093, SD =3.90), $t(92) = -0.86$, $p > 0.05$ (two-tailed).

The relationship between mental wellbeing as measured by the SWEMWBS and HRQoL as measured by the scales and summary scores of the SF-36v2 were assessed (Table 13).

Table 13 Correlation between the SWEMWBS scores and the scales of the SF36v2

	PCS	MCS	PF	RP	BP	GH	VT	SF	RE	MH
SWEMWBS	0.15	0.68	0.23	0.35	0.32	0.50	0.55	0.52	0.46	0.71

SWEMWBS scores were weakly related to the PCS summary score and scales which mainly contribute to it (PF, RP and BP scales, all p values <0.01). However, there was a strong positive correlation between the SWEMWBS scores and the scales relating to mental health (all p values <0.001), and in particular the MCS summary score and the subscale of mental health. Therefore, indicating measurement of a similar construct and indicating concurrent validation for the SWEMWBS against the SF36v2, an established measure of mental health functioning. However, by doing so, this suggests that the SF-36v2 may be the most insightful measure as it provides health status across various domains as opposed to just mental wellbeing.

Perceptions of the impact of cancer

Data completeness for the IOC-CS was approximately 99%. Cronbach's alpha was calculated for each of the eight subscales of the IOC-CS, and were: life challenges (0.80); thinking and memory problems (0.78); financial problems (0.58) body and health (0.86); talking with parents (0.90); personal growth (0.75); health literacy (0.78) and socialising (0.77). Therefore, suggesting good internal consistency for all subscales, except the financial problems scale. The Cronbach's alpha was 0.87 and 0.84 for the overall positive and negative impact scales respectively.

Results from the IOC-CS are shown in Table 14. Survivors with a university degree scored both a higher positive impact of cancer (Mean =3.66, SD =0.55 vs Mean =3.33, SD =0.52; $t(54) = 1.85$, $p=0.02$, two tailed) and a lower negative impact of cancer (Mean =1.90, SD =0.35 vs Mean =2.27, SD =0.71, $t(57) = 2.24$, $p=0.03$, two tailed) than survivors without a degree level education. Survivors who were classed as occupied reported a lower negative impact of cancer than survivors who were unemployed or unable to work (Mean =1.96, SD =0.58 vs Mean =2.36, SD =0.70; $t(73) = -2.24$, $p=0.03$). Survivors who were single reported a higher negative impact of cancer than survivors who were married or in a committed relationship (Mean =2.19, SD =0.71 vs Mean = 1.85, SD =0.43, $t(75) = -2.50$, $p=0.01$).

A significant association was found between the negative impact scores and time since diagnosis ($F(2, 74) = 3.91$, $p=0.02$). A Scheffé post hoc test indicated that survivors within 5-10 years of diagnosis, reported a significantly higher negative impact (Mean =2.28, SD =0.67) than those 11-15 years from diagnosis (Mean =1.87, SD =0.56, $p=0.05$). There was a trend for survivors who were diagnosed at a younger age to have higher scores for the positive impact of cancer, although this was not

significant. However, survivors diagnosed older at age 12-18 reported significantly higher negative impact scores than the younger age groups ($F(2, 74) = 7.73$, $p = 0.0009$).

There were no significant associations for gender, education, income, age at study and diagnosis for the IOCS-CS impact scores (all p -values > 0.05).

Table 14 Bivariate comparisons of the IOC-CS scores to sociodemographic and medical variables

	IOC-CS Impact of cancer scores (range from 1-5)			
	Positive		Negative	
	Mean (SD)	P-value	Mean (SD)	P-value
Gender^a Male (n=37) Female (n=40)	3.57 (0.61) 3.44 (0.58)	0.31	2.04 (0.65) 2.03 (0.61)	0.94
Education¹ University degree (n=24) Higher education certificate or lower (n=32)	3.66 (0.55) 3.33 (0.52)	0.02	1.90 (0.35) 2.27 (0.71)	0.03
Employment Occupied ² (n=62) Unoccupied ³ (n=14)	3.57 (0.59) 3.23 (0.57)	0.06	1.96 (0.58) 2.36 (0.70)	0.03
Income ≤£200 a week (n=30) ≥201a week (n=42)	3.44 (0.57) 3.46 (0.58)	0.87	2.14 (0.71) 1.97 (0.56)	0.26
Relationship status Married/in relationship (n=34) Single (n=44)	3.61 (0.59) 3.42 (0.59)	0.16	1.85 (0.43) 2.19 (0.71)	0.01
Age at study 18-23 (n=44) 24-29 (n=34)	3.55 (0.64) 3.44 (0.54)	0.44	1.95 (0.61) 2.15 (0.63)	0.16
Diagnosis Haematological (n=29) CNS tumour (n=28) Solid tumour (n=20)	3.67 (0.57) 3.34 (0.61) 3.47 (0.57)	0.11	2.03 (0.62) 2.20 (0.65) 1.83 (0.55)	0.13
Age at diagnosis 0-5 (n=25) 6-11 (n=25) 12-18 (n=28)	3.62 (0.42) 3.60 (0.62) 3.30 (0.67)	0.09	1.75 (0.56) 1.93 (0.45) 2.36 (0.67)	0.0009
Time since diagnosis 5-10 years (n=28) 11-15 years (n=25) 16-25 years (n=25)	3.41 (0.72) 3.55 (0.59) 3.55 (0.44)	0.59	2.28 (0.67) 1.87 (0.56) 1.91 (0.55)	0.02

¹ Survivors aged 21 or over only included in analyses; ² Includes full time employment, part-time employment, caring for home or family, student; ³ Includes unemployed and looking for work and unable to work due to illness or disability; **Bold** indicates results significant at $p < 0.05$

As shown in Table 15, survivors with somatic late effects reported a higher negative impact of cancer (Mean =2.21, SD =0.65) than survivors who did not (Mean = 1.73, SD =0.44, $t(75) = 3.48$, $p=0.0009$).

Similarly, survivors who self-reported psychological late effects were much more likely to report both a lower perceived positive impact (Mean =3.22, SD =0.61 vs Mean =3.70, SD =0.51, $t(76) = -3.76$) and a higher perceived negative impact of cancer (Mean =2.45, SD =0.62 vs Mean =1.74, SD =0.43, $t(75) = 5.95$, $p<0.0001$).

Table 15 Bivariate comparisons of the IOC-CS to self-reported late effects, scarring and caseness on the BSI-18

	IOC-CS Impact of cancer scores (range from 1-5)			
	Positive		Negative	
	Mean (SD)	P-value	Mean (SD)	P-value
Somatic late effects Yes (n=50) No (n=28)	3.42 (0.57) 3.65 (0.622)	0.09	2.21 (0.65) 1.73 (0.44)	0.0009
Psychological late effects Yes (n=32) No (n=46)	3.22 (0.61) 3.70 (0.51)	0.0003	2.45 (0.62) 1.74 (0.43)	0.0000
Scarring to face/head/neck Yes (n=51) No (n=27)	3.45 (0.63) 3.59 (0.52)	0.31	2.15 (0.65) 1.83 (0.53)	0.03
Scarring to chest/stomach Yes (n=51) No (n=25)	3.59 (0.61) 3.34 (0.53)	0.08	2.04 (0.64) 2.04 (0.61)	0.98
Scarring to legs/arms Yes (n=18) No (n=60)	3.56 (0.73) 3.48 (0.55)	0.64	2.06 (0.70) 2.03 (0.60)	0.86
Caseness on BSI-18 Yes (n=11) No (n=66)	3.03 (0.52) 3.59 (0.57)	0.0036	2.80 (0.69) 1.90 (0.51)	0.0000

Survivors who were classed as cases on the BSI-18 also reported less positive impact (Mean =3.03, SD =0.52 vs Mean =3.59, SD =0.57, $t(75) =3.00$, $p=0.004$) and more

negative impact (Mean =2.80, SD =0.51 vs Mean =1.90, SD =0.69, $t(74) = -5.31$, $p < 0.001$) than those where not.

Association of IOC-CS with existing measures of HRQoL and psychological distress

To evaluate concurrent validity, the subscales and impact scores of the IOC-CS were correlated with those of the SF36v2 and the BSI-18 which are existing and validated measures of HRQoL and psychological distress (Table 16). Across most IOC-CS subscales, medium to strong relationships were observed with the scales of the SF36v2. In particular, the body/health scale and the socialising subscales of the IOC-CS demonstrated medium to large positive relationships with several scales of the SF36v2, specifically those which contribute to mental health. In general, the IOC-CS subscales and impact scores had stronger associations with the MCS, than the PCS scores.

The IOC-CS subscales were also moderately correlated with the depression, anxiety and global scale of the BSI-18. This was particularly the case for the life challenges scale, body and health, socialising and relationship concerns for those with no partner. Scales of personal growth, finance problems, sibling concerns and relationship concerns for those with a partner displayed weaker correlations across both the SF-36 and the BSI-18 scales.

Overall, the correlations between the IOC-CS and the SF36v2 were in the expected direction. The positive scales of the IOC-CS were positively correlated with SF36v2 subscales, and negatively associated with the BSI-18 subscales. A similar pattern was demonstrated for the negative IOC-CS but in the opposite direction (negatively related to SF36v2 and positively associated with BSI-18).

Table 16 Correlation between the IOC-CS scales with those of the SF36v2 and the BSI-18

	Subscales on IOC-CS										
	Life Challenges -	Body/health +	Talking with parents +	Personal growth +	Thinking/memory problems -	Health literacy +	Socialising +	Finance problems -	Sibling concerns -	Relationship concerns (no partner) -	Relationship concerns (partnered) -
SF-36v2											
Physical Function	-0.22	0.38	0.30	-0.39	-0.18	0.15	0.32	-0.11	-0.33	-0.31	0.11
Role-physical	-0.37	0.54	0.39	-0.27	-0.33	0.17	0.47	-0.08	-0.27	-0.26	0.14
Bodily pain	-0.39	0.34	0.34	-0.23	-0.40	0.36	0.31	-0.22	-0.35	-0.27	-0.07
General health	-0.49	0.66	0.32	-0.08	-0.40	0.35	0.50	-0.34	-0.21	-0.47	0.03
Vitality	-0.36	0.69	0.28	-0.23	-0.48	0.38	0.59	-0.31	-0.35	-0.41	0.11
Social functioning	-0.42	0.53	0.35	-0.15	-0.44	0.27	0.53	-0.11	-0.22	-0.35	0.16
Role-emotional	-0.38	0.46	0.38	-0.25	-0.22	0.28	0.49	-0.12	-0.14	-0.48	-0.10
Mental health	-0.48	0.66	0.35	-0.11	-0.38	0.48	0.57	-0.22	-0.22	-0.44	-0.05
PCS	-0.21	0.32	0.26	-0.16	-0.27	0.16	0.24	-0.19	-0.25	-0.09	0.06
MCS	-0.44	0.63	0.36	-0.10	-0.39	0.45	0.61	-0.24	-0.21	-0.41	0.03
BSI-18											
Depression	0.48	-0.54	-0.32	0.16	0.38	-0.31	-0.50	0.33	0.17	0.58	0.05
Anxiety	0.48	-0.46	-0.27	0.26	0.32	-0.30	-0.32	0.34	0.20	0.59	0.10
Somatisation	0.31	-0.33	-0.23	0.29	0.26	-0.19	-0.27	0.22	0.42	0.39	0.07
Global (GSI)	0.50	-0.57	-0.34	0.27	0.43	-0.29	-0.49	0.34	0.30	0.65	0.08
MHLC											
Internal	-0.27	0.51	0.01	0.11	-0.25	0.30	0.19	-0.30	0.02	-0.07	0.19
Chance	0.27	-0.22	-0.10	0.05	0.02	0.01	0.11	0.19	0.17	0.14	0.13
Powerful others	0.15	-0.21	-0.07	0.23	-0.05	0.22	0.07	-0.01	-0.03	0.20	-0.20

-/+ indicates whether the subscale is suggestive of negative or positive outcomes; **Bold** indicates correlation ≥ 0.50

4.5.4 Acceptability and feasibility of questionnaire

Respondent feedback was generally positive (Table 17). The majority indicated that they found the questionnaire interesting and easy to follow. However, about quarter of the survivors indicated that the questionnaire was difficult to fill in to some extent, although it is not known if this was due to it being emotionally and/or cognitively challenging. Approximately 40% agreed to some extent that the questionnaire was too long, although only 17.4% stated that this was moderate or strong agreement.

Table 17 Survivors' evaluations of questionnaire

Evaluation question	Survivor n (%)
Questionnaire was interesting (n=87)	
<i>Strongly agree</i>	20 (23.0)
<i>Moderately agree</i>	29 (33.3)
<i>Slightly agree</i>	32 (36.7)
<i>Slightly disagree</i>	2 (2.3)
<i>Moderately disagree</i>	3 (3.5)
<i>Strongly disagree</i>	1 (1.2)
Filling in the questionnaire was difficult (n=86)	
<i>Strongly agree</i>	0
<i>Moderately agree</i>	6 (6.98)
<i>Slightly agree</i>	16 (18.6)
<i>Slightly disagree</i>	7 (8.1)
<i>Moderately disagree</i>	19 (22.1)
<i>Strongly disagree</i>	38 (44.2)
The questionnaire is too long (n=86)	
<i>Strongly agree</i>	5 (5.8)
<i>Moderately agree</i>	10 (11.6)
<i>Slightly agree</i>	20 (23.3)
<i>Slightly disagree</i>	18 (20.9)
<i>Moderately disagree</i>	19 (19.8)
<i>Strongly disagree</i>	16 (18.6)
Layout of questionnaire is easy to follow (n=87)	
<i>Strongly agree</i>	50 (57.5)
<i>Moderately agree</i>	21 (24.1)
<i>Slightly agree</i>	12 (13.8)
<i>Slightly disagree</i>	3 (3.5)
<i>Moderately disagree</i>	1 (1.2)
<i>Strongly disagree</i>	0

Respondents were asked to specify any questions which they found difficult to complete. One survivor stated that a section within the IOC-CS entitled 'talking and thinking about cancer' appeared irrelevant as they were so young at diagnosis that they could not remember details. A brain tumour survivor indicated that all questions were difficult as they did not have cancer. Therefore, the wording of the questions may have to be amended to 'my cancer/tumour.' One respondent stated that the questions about income were hard to answer as their partner deals with the financial issues due to her bad memory and difficulty in making decisions. Another stated that the section on romantic relationships/marriage and parenthood were perhaps not relevant for people of such a young age and possibly too personal. One participant queried whether the question about scarring included scarring from spots and another commented that the scarring questions should allow greater detail as opposed to just multiple choice.

In terms of missing questions or topics, a section on social development was suggested as a useful addition (n=2) and a question about whether they felt they received the correct care (n=1). A general comment from one survivor was that the design of questionnaire was too childish for ages 18-30, and that they would also have preferred receiving an invite to register their interest in the study, prior to receiving the questionnaire through the post.

Through the process of data entry and analysis, some of items were found to need rephrasing or were considered redundant. As already discussed, the scarring question is one such item which will need rephrasing. Three questions on education were also not suitable for analysis. A question requesting the school year the survivor

was in when they left school got mixed responses with 31/93 giving the year (e.g. 2010) as opposed to the school year (e.g. year 13). In addition, the questions regarding the delay of exams were also not seen to provide useful information due to the low number of survivors who answered yes and the high number of 'I don't know' responses.

4.6 Discussion

4.6.1 *Summary of findings*

This chapter reports on the development and piloting of a questionnaire which aimed to assess psychosocial outcomes in young adult survivors of childhood and adolescent cancer and to investigate factors associated with less favourable psychosocial outcomes. In particular, the questionnaire aimed to include measures and items which would enable inclusion of survivors' subjective views as to what the impact of cancer had been for them. The specific aims of the pilot study were to pre-test the measures and items to identify any issues with their use and assess their suitability in this survivor population, as well as investigate associations which may be of interest to examine in a subsequent and larger study. The acceptability and feasibility of administering the questionnaire in a young survivor population was also evaluated.

In terms of the assessment of social outcomes, although few were married, many survivors were in long-term relationships with almost a quarter co-habiting with their partner. This may be considered a precursor for marriage and indeed marriage was desirable for the majority. In this sample of survivors with a mean age of 23 years, most stated that they were too young for marriage at present. In support of these views, the current age of first marriage in the UK is 36.5 years for males and 34.0 years for females (Office of National Statistics, 2012). Although romantic success in young people has been previously defined as establishing a committed relationship by the age of 26 years (Schulenberg *et al.* 2004), this may no longer apply to current young adults due to the rising age of first marriage. Indeed, Arnett's (2000) theory of

emerging adulthood marks the twenties as a period of instability and exploration in relationships.

However, for those not in long-term relationships, there was evidence of concerns which may impact on them psychologically and may affect their desire or ability to meet a partner, as reported by Thompson *et al.* (2013). Such worries included disclosure of history of cancer or potential infertility, having a sexual relationship and ultimately not finding a partner. The relationship concerns (no partner) subscale of the IOC-CS demonstrated moderate negative relationship with several of the mental health domains of the SF36v2, and a stronger positive relationship with depression, anxiety and overall distress. Similar findings were reported by Zebrack *et al.* (2010). In contrast, there was a weak correlation between relationship concerns of those who were in relationships and HRQoL and distress outcomes. This may have been due to the low level of relationship concerns reported by partnered survivors.

As reported in previous studies, desire to have a family was high (Zebrack *et al.* 2004), but as in the qualitative study, several were uncertain about their fertility status. A quarter of the sample also reported concern at not being able to become pregnant/father a child. Although not specifically explored in this study, fertility concerns in female survivors have been linked with depression (Gorman *et al.* 2015). Gorman's sample was, however, older (mean age of 28 years) with many having been diagnosed in young adulthood as opposed to childhood. In contrast, survivors in the qualitative study did not cite uncertainty about fertility as a source of distress as they were not ready to start a family, as also found by Nilsson *et al.* (2014). Being too

young or not yet finding the right partner were the most common reasons for not having children as in Hohmann *et al.* (2011).

Attainment of a degree level education was higher (40%) than that previously reported in a population based cohort of British childhood cancer survivors (17.9%) (Lancashire *et al.* 2010). However, the cohort used in Lancashire's (2010) study were survivors diagnosed between 1940 and 1991, whereas survivors in the current study were diagnosed in the period 1991-2011. During this time the therapies used to treat childhood cancer have changed to become more targeted and less toxic, for instance cranial irradiation is no longer used to treat leukaemia (Jenney, 2005). In addition, CNS survivors are the main group to demonstrate deficits in education (Boman *et al.* 2010; Lancashire *et al.* 2010; Kuehni *et al.* 2012a) and on closer inspection of the specific diagnoses of the CNS respondents in the present study, it was found that the majority were low grade brain tumours, therefore, presenting a biased representation of CNS survivors which explains the apparent good outcomes of this group. Participation in higher education in the U.K. has also increased by more than ten-fold since the 1950s (Bolton, 2012), with about 49% of 25-34 year olds now obtaining tertiary education (OECD, 2016).

Previous studies have stated that between 9% (Lähteenmäki *et al.* 2002) and 30% of survivors repeat a school year (Kuehni *et al.* 2012a). In this study the figure was only 7.6% of survivors. However, similar rates of extra tutoring and bullying were found (Kuehni *et al.* 2012a; Lähteenmäki *et al.* 2002). Despite the majority feeling that their illness had impacted on their education, overall survivors were satisfied with their educational attainment. Survivors reported a variety of factors such as missing school

as well as issues with memory, concentration, fatigue and impacted ability to learn to have negatively affected their education. However, as found in the qualitative study, a few survivors felt that their experienced had impacted their education positively and again this may help to explain findings where some groups of survivors achieve better than norms as found in Lancashire *et al.* (2010).

A small number of survivors were reported to have problems with employment in terms of being unemployed, unable to work or perceiving issues in gaining employment and as reported by previous research (e.g. Dolgin *et al.* 1999; Dumas *et al.* 2015) several survivors reported they had been unable to pursue a desired career. Perceived reasons were due to their past cancer, physical limitations, learning difficulties as well as mental health issues. However, of those in full-time employment, almost three quarters were very satisfied with their current employment. This suggests that despite the positive outcomes in employment, there are a small number of survivors who may benefit from further support in this area.

In line with previous findings, overall, survivors reported good HRQoL and low levels of distress. In particular, survivors reported above expected norms for the physical component summary and most of its associated health domains on the SF36v2. The SF36 was developed for use in both healthy and ill populations, however, the young age of the sample may explain their tendency to score high across several of the subscales.

As in this study, substantial ceiling effects were reported by Reulen *et al.* (2006) who validated the SF36 (version 1) in childhood cancer survivors. However, modifications to the response options in the SF36v2 aimed to reduce the potential for these ceiling

effects (Jenkinson *et al.* 1999). Ceiling effects may be problematic in that the measure will lack precision and not distinguish between survivors who report very good health (Maruish, 2011). The SWEMWBS measure in comparison only had a ceiling effect of 4%. However, unlike the SF36v2, SWEMWBS does not provide health status across multiple domains, only mental wellbeing, which was highly correlated with both the MCS and the mental health subscale of the SF36v2.

It is argued that high HRQoL and low distress outcomes achieved via self-report measures may be subject to self-deception response bias in which survivors minimise or deny negative aspects of the cancer. Although it is unclear whether this would be considered adaptive or maladaptive coping (O'Leary *et al.* 2007). Alternatively, positive results may be the product of a response-shift, a change in the individuals self-evaluation of QoL which results from a combination of factors including antecedents (e.g. sociodemographics, personality, expectations, spiritual identify), mechanisms (e.g. coping, social comparison, social support, reframing expectations) in response to a catalyst such as a cancer diagnosis (Sprangers & Schwartz, 1999). Although these theories have been suggested in childhood cancer survivors, they have not been extensively researched and studies tend to focus on the potential for PTG in survivors as assessed by the PTGI (Tedeschi & Calhoun, 1996). It is also possible that measures such as the SF36v2 may also lack specificity and fail to capture a dimension of health which is critical to the patient group under study (Petitti, 2006). The limits of using a generic HRQoL measure were outlined in the literature review, however, this study also used the IOC-CS which has been designed to use alongside HRQoL measures. The IOC-CS covers several domains pertinent to young adult

survivors of childhood cancer such as issues with thinking, memory, challenging thoughts about cancer, body image and relationship with parents.

In comparing the IOC-CS subscales to the SF36v2 subscales, correlations were of moderate strength. Associations the SF36v2 mental health subscales and the MCS were stronger than those relating to physical function and the PCS, therefore, suggesting that the IOC-CS is able to reflect survivors' perceptions and the impact these potentially have on subjective well-being (Zebrack *et al.* 2010). In particular, the body/health and the socialising scale showed moderate to high relationships to SF36v2 scales of role-physical, general health, vitality, mental health and social function, suggesting they are tapping into similar construct or that the IOC-CS scales are associated to HRQoL. The small to medium correlations of the remaining subscales suggest a weaker relationship and that the IOC-CS may be measuring different domains of HRQoL not assessed by the SF36v2.

Supporting previous findings, socio-demographic factors significantly associated with poorer HRQoL as measured by the SF-36v2 were suggested to be being female, unemployed/unable to work, having lower personal income, lower education and older age at diagnosis (Cantrell *et al.* 2011; Klassen *et al.* 2011). Only lower income and being single were significantly associated with higher levels of distress on the BSI-18. While perceived impact of cancer as assessed with the IOC-CS was associated with education, employment and relationship status, with those with a university education, in employment and in relationships reporting a less negative and more positive impact of cancer. It also appeared that survivors who were older at diagnosis reported a more negative impact of cancer. These findings in part support those of

Zebrack & Landier (2011), however, in addition they found education and income were associated with the impact scores.

Overall, few significant results were found throughout the bivariate analyses of socio-demographic variables and measures of HRQoL, distress and impact of cancer. Factors associated with lower socioeconomic are often reported to be associated with HRQoL and distress, as is being female (Cantrell *et al.* 2011; Gianinazzi *et al.* 2013; Klassen *et al.* 2011; McDougall & Tsonis, 2009; Michel *et al.* 2010; Zebrack *et al.* 2011; Zeltzer *et al.* 2009). However, this reflects risk factors seen in general populations (Stuber *et al.* 2009).

The only significant medical factor found by this study to be associated with lower scores on physical aspects of HRQoL was radiotherapy. Several diagnostic and treatment variables have previously been linked with poorer psychological outcomes such as being a CNS survivor and receiving cranial radiation (Klassen *et al.* 2011; Zeltzer *et al.* 2009). CNS survivors have previously been found report a higher negative impact of cancer, although this just reached significance in a sample of 621 survivors including 79 CNS survivors (Zebrack & Landier, 2011). Survivors who reported late effects, somatic and psychological, were more likely to report poorer HRQoL in physical and mental health respectively, and late effects were also predictive of higher distress levels as previously reported (Gianinazzi *et al.* 2013; Recklitis *et al.* 2003). Survivors without psychological late effects reported a higher positive impact and a lower negative impact of cancer, while survivors with somatic late effects reported a higher negative impact of cancer. Zebrack and Landier (2011)

report a similar pattern for survivors reporting 'current health problems', although they do not specify whether this includes psychological as well as physical health.

Despite the lack of significance, trends were often observed in that the differences between the mean scores of the groups were in the expected direction. The lack of significant results may be due to a lack of power within the study. For the SF-36, Maruish (2011) states to detect a difference between T-scores of less than 5, a sample size of over 200 may be required (Maruish, 2011).

Scarring in this study was not associated with differences in HRQoL, but survivors reporting scarring to the face/head/neck reported higher levels of anxiety. This supported findings from chapter 3 which suggested that it was the visibility of scars which determined whether the scarring led to feelings of anxiety or not. Kinahan and colleagues (2012) have previously reported that after adjusting for cranial radiation and sociodemographic variables, head and neck disfigurement was associated with an increased risk of depression, while hair loss was associated with increased risk of anxiety. In the present study, low numbers of survivors reported persistent hair loss, therefore, analysis of this variable was not possible. Although small effects were found for scarring to the head, face or neck, it is possible that the wording of the item may not have been specific enough. Kinahan *et al.* (2012) used the phrase 'scarring or disfigurement' whereas this study used the phrase 'marks or scars', therefore, implying a less serious complaint.

A factor which requires further investigation is the cut-off point which indicates caseness on the BSI-18. Although Derogatis (2001) recommends using the original cut-off, it is stated that caseness should ideally be established on a large sample of

the target group. An alternative case-rule of ≥ 50 has been suggested for adult survivors of childhood cancer (Merport & Recklitis, 2012; Recklitis & Rodriguez, 2007), and a case-rule of ≥ 57 has been recommended for adult cancer patients (Zabora *et al.* 2001). However, studies often continue to use the original rule (Michel *et al.* 2010; Zebrack *et al.* 2004; Zeltzer *et al.* 2009). Using the ≥ 50 case-rule, 42% ($n=39$) of survivors would have been distress cases, while 25% ($n=23$) would have been using the ≥ 57 rule. Use of the original case-rule identified 14% of survivors as cases, which is similar to previous findings. Prevalence of cases using this case-rule is variable from 8% (Gianinazzi *et al.* 2013) to 25% in adult cancer survivors (Michel *et al.* 2010), when 10% is expected in the normal population (Derogatis, 2001). Michel *et al.* (2010) suggests that while survivors as an overall group report low distress, there may be a significant minority who have very high distress.

Both the positive and the negative impact scales and the subscales of the IOC-CS had moderate to strong correlations with the subscales of the SF-36v2 and the BSI-18. However, this was not the case for the personal growth and financial problem scales. Zebrack and Landier (2010) comment that the lack of substantial correlations for the personal growth subscale, suggests that there is a new aspect of survivorship which is not measured by other instruments. Future investigations of the IOC-CS may benefit from administering it alongside the PTGI (Tedeschi & Calhoun, 1996). The low alpha score for the financial problems scale may be due to the measure being developed in the context of the U.S. health system in which survivors and families are more likely to incur expenses compared to U.K National Health Service which provides free treatment. Administering the IOC-CS to a large enough sample to

enable a further psychometric analysis and a factor analysis to confirm the eight subscales which Zebrack *et al.* (2010) propose, would be beneficial.

4.6.2 Strengths and limitations

Recruitment to the study was a lengthy process and required continued communication with NHS contacts. The final response rate for the questionnaire was 44% in a population which is documented to be hard to reach (Aubin, 2011). However, the response rate was favourable in comparison to Zebrack and colleagues (2010) previous administration of a survey containing the IOC-CS (30%). However, it is acknowledged that in administering a comprehensive questionnaire, the questionnaire sent to survivors was of considerable length and took approximately 40 minutes to complete. Therefore, a greater response rate may have been reached with a shorter and less burdensome questionnaire. However, in turn the scope of the questionnaire would be reduced. Results from this current study can be used to identify any redundant items or measures, such as the SWEMWBS which do not contribute additional and important data and, therefore, do not need to be included in future administrations.

Due to ethical constraints, it was not possible to know whether responders differed on key variables to non-responders. It also meant that it was difficult for the researcher to know the characteristics of those invited to the study which made it impossible to ensure that all diagnoses were represented appropriately. This highlights the need to discuss further with the registry and consultants and devise strategies to address this in future work.

Overall, the questionnaire appeared to be acceptable to survivors, the response rate was reasonable, the feedback was positive, and missing data was minimal throughout. However, approximately 40% of survivors had some level of agreement (either slightly, moderately or strongly) that the questionnaire was too long. The measures and items appeared to fulfil the goal of the questionnaire. However, it is acknowledged that the questionnaire will need redrafting prior to administration in future studies. The wording of particular items needs to be more effective. It is also possible that in large scale study, analyses of free-text responses will be problematic. Therefore, the answers gained in this study could be used to develop response options for future use.

Although diagnoses, date of diagnosis and date of birth were confirmed in the registry, self-reported data for treatment and the existence of health problems was not confirmed by medical records. Participants also self-selected to be in the study, therefore, the results may be biased on a sample of high functioning survivors. As noted, the CNS survivors who would be expected to have significant impairment were under-represented. However, the nature of the questions may also be too demanding for some survivors.

The IOC-CS shows promise as a tool to assess survivors' perceptions quantitatively, but as stated above would benefit from further testing in a larger sample. Additionally, correlations only suggest a relationship between variables, and do not imply causation. Therefore, multivariate regressions would be of benefit to explore whether perceptions as assessed by the IOC-CS predict survivors scores on HRQoL and distress measures, whilst also controlling for potentially confounding variables.

However, the sample size in the current study was not sufficient as sample size should be a minimum of 50, plus the number of independent variables multiplied by 8 (Tabachnick & Fidell, 2007). In addition, although perceptions of the impact of cancer can influence HRQoL outcomes, it is acknowledge that the reverse is also possible. Therefore, by treating symptoms of distress, an individual's perceptions of the impact of cancer may be improved (Zebrack & Landier, 2011).

4.6.3 Conclusion

Overall, the questionnaire shows promise in comprehensively investigating psychosocial outcomes and the factors associated with poorer outcomes. Piloting has provided information on effects and associations which may be useful to investigate further in a larger sample. The process has identified potential issues in the recruitment phase and the questionnaire which need to be addressed in future studies.

Chapter 5 Final discussion

5.1 Integration of findings

Cornish and Gillespie (2009) suggest that in health psychology, knowledge should be judged on whether it successfully solves the problems in relation to a specific goal or interest, and not whether it accurately mirrors reality. Therefore, the research methods should be chosen on the basis of which best achieves the aim of the study, and not based on claims about ontology and epistemology. The aim of this thesis was to use an exploratory sequential study design to comprehensively investigate the psychosocial outcomes of survivors, to explore the perceptions of survivors and how they may be connected to psychosocial outcomes. Therefore, mixed methods enabled a broader exploration than would be achieved by using only quantitative or qualitative methods alone, thus resulting in a more complete picture (Johnson & Onwuegbuzie, 2004).

The qualitative study allowed the views of survivors to be gained prior to embarking on the development and administration of the quantitative questionnaire. This was seen as vital due to the lack of qualitative literature (at the time), emerging research evidence about the importance of survivors' views in influencing psychosocial outcomes, and the lack of studies adopting an explanatory approach to understand why and in what way cancer affects a young person in later life. The questionnaire also enabled assessment of survivors' perceptions to be considered as potential risk factors for psychosocial outcomes. However, the benefit of the qualitative study was that it allowed survivors to give greater depth to their experiences and perceptions. Although free-text items in the questionnaire allowed survivors to expand on

answers or provide explanations, this resulted in relatively shallow data as survivors' responses were brief, often just two or three words in length. However, it is possible that in a shorter and less burdensome questionnaire, the inclusion of free-text items may result in more in-depth data.

However, an advantage of the questionnaire is that it enabled an investigation into what factors may influence psychosocial outcomes by using validated measures of distress and HRQoL. However, as these measures are generic it is possible they may not detect issues specific or important to cancer survivors, particularly young adult survivors. Although the IOC-CS is a cancer specific tool, and showed promise in this study, it is essential that further investigations of the IOC-CS are carried out. However, due to the number of items included in the IOC-CS, doing so will require a large sample of survivors (approximately 600) to enable a full factor analysis. Therefore, such efforts will require much time and resources, particularly from a relatively small treatment centre such as the RVI.

Both studies confirmed that survivors experience and perceive both negative and positive consequences of cancer. In both studies, the majority of survivors appeared to be in good psychosocial health, with a minority reporting concerns and poorer outcomes. The studies have been able to give an insight into the psychosocial outcomes of young adult survivors treated at the RVI. Although it has been acknowledged that the samples may not have been truly representative of the survivors at the RVI as a whole. Greater efforts in future studies will need to be made to ensure survivors with various diagnoses and abilities are invited to take part.

Although both studies provided evidence of positive outcomes in survivors, it is acknowledged that it is beyond the scope of this thesis to state which theory of cognitive adaption may be more appropriate to explain these results. In terms of reports of high HRQoL and low distress, O'Leary *et al.* (2007) suggests this may be due to the survivor systematically denying psychological and emotional difficulties as assessed by the measures. Although this may be true for some survivors, it is unlikely to be the case for the majority. Survivors may also experience a shift in their values and internal standards (Sprangers & Schwartz, 1999), which may indeed be the case in this thesis due to survivors reporting that cancer had changed their views of what was important in their lives. Resilience, in which survivors demonstrate positive outcomes despite encountering adverse or traumatic circumstances is also of relevance (Phipps *et al.* 2014).

In terms of survivors reporting positive changes due to the cancer, research in childhood cancer survivors has focused on PTG which suggests that positive changes occur due to a struggle with a traumatic event. Qualitative evidence of growth was seen in chapter 3 and by using the IOC-CS in chapter 4. However, interestingly Phipps *et al.* (2014) report that in a sample of 255 childhood cancer survivors, approximately 50% who were less than 5 years from diagnosis stated that cancer was their most traumatic event in life, while only 24% of survivors more than 5 years from diagnosis stated the same. Phipps *et al.* (2014), therefore, question whether the traumatic impact of childhood cancer has been overstated.

The use of PTG in survivors has been questioned due to the lack of evidence for posttraumatic stress symptoms in survivors, which are stated to be a necessary

precursor for developing PTG (Phipps *et al.* 2014; Tillery *et al.* 2015). Tillery *et al.* (2015) states a small subgroup of survivors may indeed be experiencing true PTG but suggests that in the context of cancer, many survivors appear to experience growth without posttraumatic stress. Therefore, they suggest the term 'challenge-related growth' may be more appropriate. However, research into the different explanations for survivors cognitive adaptations are complex and will require investigation of several inter- and intra-personal factors. As with psychosocial outcomes in general, investigation of psychological growth within survivors would benefit from longitudinal study designs to study their development over time and enable investigations of causality (Bitsko, *et al.* 2016; Tillery *et al.* 2015).

5.2 Implications for research and practice

In terms of research implications, recruitment to both studies was a challenge and this process has highlighted the need to improve procedures to identify and approach the survivors at the RVI. With most forms of data collection, certain groups of survivors will be excluded from participating. They may not feel comfortable meeting with an interviewer or feel unable or uninterested in completing a questionnaire. In addition, although recruitment for focus groups failed for this study, and it has been stated that recruiting young adult cancer survivors for focus groups is particularly challenging (Ford, 2011), this type of data collection could enable survivors to participate in a group discussion and engage with each other using their own everyday vocabulary which may lead to generating new ideas, reaching consensus and rich data (Kitzinger, 1995). Ford (2011) also states that participation in focus groups can be a meaningful personal experience for survivors, and this appeared to

be the case in the one group that took place for this study. Therefore, new and innovative ways of accessing survivors' views via group discussion such as online discussions/forums may address many of the practical challenges of bringing survivors together and may be valuable. The process of undertaking this thesis also emphasised the need to engage more with survivors and the potential for establishing a Patient and Public Involvement group for childhood and adolescent cancer survivors treated at the RVI will be explored. This will enable survivors to give their views on proposed research and may assist with the challenges of recruitment for future studies.

Both studies indicated that to varying extents, survivors have concerns of both a medical (e.g. health and fertility) and a non-medical nature (e.g. relationships, employment). Therefore, underlining the importance of survivors' being able to access reliable information and support from healthcare professionals. For survivors in LTFU care, age appropriate and developmentally relevant information is crucial and it may be necessary for information given at an earlier stage of survivorship to be revisited at a later point.

For survivors no longer in LTFU, there needs to be a way in which they can access accurate information about late adverse effects of treatment and fertility issues. This will most likely be via their GP, although there is evidence that GPs may feel unexperienced and uncomfortable in caring for childhood and adolescent cancer survivors and unfamiliar with clinical practice guidelines for the LTFU care of survivors (Nathan *et al.* 2013). Equally, childhood cancer survivors may have concerns about being followed up in primary care due to a perceived lack of GPs knowledge about

their medical history (Blaauwbroek *et al.* 2008). Therefore, survivors could be referred to LTFU specialists at their treating hospital who can help to address gaps in knowledge and address concerns. Alternatively, providing GPs with treatment summaries, evidence-based clinical practice guidelines and a LTFU specialist as a contact point may improve care of survivors. However, it is also important to develop interventions which ensure that survivors have accurate knowledge of their cancer history and skills to enable them to advocate for appropriate care (Nathan *et al.* 2013).

The outcomes from this research indicate that health professionals should monitor the psychosocial health of even long-term survivors of childhood and adolescent cancer. The potential importance of survivors' perceptions of their health and the impact of cancer suggest that healthcare professionals should initiate assessment of survivors' views, beliefs and comprehension. Identifying negative beliefs or concerns and intervening may improve patient outcomes such as distress or HRQoL. Recent LTFU guidelines state that yearly ongoing psychosocial assessments should be made with specific attention to depression, anxiety, post-traumatic stress and suicidal ideation (Bitsko *et al.* 2016). In the U.K, holistic psychosocial assessments are stated to be an important aspect of care for every cancer patient (National Cancer Action Team, 2011). However, there is no concordance in what measures should be used to carry out psychosocial assessments at LTFU with survivors of childhood and adolescent cancer.

For a consultancy project, the student previously developed a brief holistic assessment tool for the LTFU clinic at the RVI. The tool aimed to facilitate a

conversation between the survivor and the LTFU nurse to help identify any concerns the survivor may have across several areas of their life. This tool was reported to provide a focus for the consultation and identify needs of the survivor which may have otherwise been undetected. However, this tool did not include reliable and valid screening tools for outcomes such as distress or HRQoL. Therefore, it may be useful to re-develop this tool using information gained from this thesis. However, the student has also been invited to join a multidisciplinary group to develop pan-European LTFU guidelines for psychosocial issues in survivors of childhood and adolescent cancer. This group aims to produce evidence based recommendations for the screening of psychosocial issues of survivors in LTFU care.

Overall, the studies within this thesis have enabled an investigation into the psychosocial outcomes of young adult survivors of childhood and adolescent cancer treated at the RVI, Newcastle upon Tyne. The results of this thesis will inform future research directions, particularly around administration of the questionnaire in a larger sample of survivors, but also research into the concerns and needs of the survivors at the RVI and how these can be addressed. As stated above, the results have highlighted the need for long-term survivors to receive monitoring for their psychosocial health either from a health psychologist or other health professionals. As yet, there is no concordance on how best to monitor the psychosocial health of childhood and adolescent cancer survivors who attend follow-up care clinics. Therefore, efforts are required to implement and evaluate different approaches to identify how this can be done effectively in order to improve the quality of care these survivors receive.

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Appendix A REC approval for qualitative study (Chapter 3)



Health Research Authority NRES Committee North East - Newcastle & North Tyneside 2

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26 February 2013

Ms Morven Brown
Research Assistant/Trainee Health Psychologist
Institute of Health & Society
Newcastle University
Newcastle upon Tyne
NE1 4LP

Dear Ms Brown

Study title: Quality of life, socio-economic status, relationships and risk perceptions in survivors of childhood or young adult cancer: a pilot study
REC reference: 11/NE/0187
Amendment number: Amendment 1
Amendment date: 15 February 2013
IRAS project ID: 69178

Thank you for submitting the above amendment, which was received on 22 February 2013. I can confirm that this is a valid notice of a substantial amendment and will be reviewed by the Sub-Committee of the REC at its next meeting.

Documents received

The documents to be reviewed are as follows:

Document	Version	Date
Covering Letter	Morven Brown	19 February 2013
Notice of Substantial Amendment (non-CTIMPs)	Amendment 2 (16.1.13)	16 February 2013
Interview Schedules/Topic Guides	3 (Patients)	08 February 2013
Consultant Ltr to discharged patients	2	06 February 2012
Consultant Ltr to LTFU patients	2	06 February 2013
Participant Consent Form: Interviews	4	08 February 2013
Participant Consent Form: Verbal	5	08 February 2013
Participant Information Sheet	v 4	06 February 2013
Protocol	v 6	06 February 2013

Notification of the Committee's decision

The Committee will issue an ethical opinion on the amendment within a maximum of 35 days from the date of receipt.



Health Research Authority

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14 March 2013

Ms Morven Brown
Research Assistant/Trainee Health Psychologist
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Dear Ms Brown

Study title:	Quality of life, socio-economic status, relationships and risk perceptions in survivors of childhood or young adult cancer: a pilot study
REC reference:	11/NE/0187
Amendment number:	Amendment 1
Amendment date:	15 February 2013
IRAS project ID:	69178

The above amendment was reviewed by the Sub-Committee in correspondence.

Ethical opinion

Members noted that whilst they had no real concerns with the researchers offering telephone interviews in addition to the focus groups, further information was required as follows -

- Clarify how the researchers will deal with participants who may become distressed during the interview.
- Clarify what action would be taken if the researchers have concerns with regard to the well-being of participants arising from the interviews.
- Clarify how support referrals will be managed in these scenarios.

You provided the following information in response to the issues raised, in your role as researcher -

Prior to the interview during informed consent, the participants will be told that they can stop at any time and do not need to give a reason why. Participants will also be told that they do not have to answer all questions that are put to them should they not wish to discuss certain aspects of their experience. If it is noticed that the participant is becoming emotional/upset/distressed during the telephone interview, they would be asked if they want to stop the interview. If they wish to continue, they will be asked if they would like a break. Before discontinuing the call, similar to the focus groups, interviewees will be told that they can be put in touch with a member of the paediatric oncology team should the interview have

raised any issues/questions for them. With their agreement you would pass on the request to the participant's consultant who will arrange this.

Additionally, if you notice that the participant is upset/distressed it will be suggested to them that they may find it helpful to access the psychological support which is available from the oncology service at the RVI. This will be fed back immediately to the consultant who will then inform psychological services to contact the patient. If a case arises where there is particular concern for the participant's wellbeing, and in the case that they say they do not want to speak to psychological services, their consultant will still be informed. You informed that as a trainee health psychologist, you have conducted previous interviews concerning sensitive subjects (treatment and diagnosis in childhood/adolescent cancer and future cardiovascular risk in women who have experienced pre-eclampsia).

The sub-committee was satisfied with this further explanation and clarification.

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Appendix B Consultant's letter and patient information sheet for qualitative study



DEPARTMENT OF PAEDIATRIC AND ADOLESCENT HAEMATOLOGY AND ONCOLOGY

Consultant and Honorary Clinical Senior Lecturer in Paediatric and Adolescent Oncology

Dear <LTFU patient's name>,

Re: Quality of life, socio-economic status, relationships and risk perceptions in survivors of childhood or young adult cancer.

I hope that you are well and that you do not mind me contacting you.

The reason for writing is to ask whether you might be interested in helping in a research project that is currently being performed in Newcastle. The aim of the research is to look at the views of young people such as yourself, about how their experience of cancer or leukaemia and its treatment has affected their lives, for example by influencing how they view their future health, their education or their future career aims and life goals.

The research will involve participating in a **“focus group”**. This is a group of between 4 and 8 young people such as yourself, and will be facilitated by a researcher who will help raise relevant questions and listen to the discussions that follow. There will be no hospital staff present at this discussion to enable you to talk freely, however, should you wish to talk with a member of the oncology team after taking part, we will of course be glad to see you. Alternatively, we are offering **one-to-one telephone interviews** with our researcher as we realise it may not be convenient for some people to attend focus groups, or that some may simply feel more comfortable speaking to one person, as opposed to talking in a group.

An information leaflet about the study is enclosed. I hope that you may be interested in helping, but recognise that you may feel unable to if you are too busy, or it may simply be that you feel you would rather not be involved for another reason. The fact that I am writing to you in no way assumes that I think that you will be willing to participate, but I thought you would be interested to be given the opportunity.

If this would be of interest to you and you would like more information, please contact either myself by telephone or email, or the researcher at Newcastle University (Morven Brown) whose details are on the information leaflet enclosed.

Yours sincerely,

[Redacted signature block]

What will happen to the results of the study?

We will write a report which will be fed back into the oncology services at the R.V.I. The results of the study may also be published in a medical journal. We may use direct quotes in publications that result from the research but you will not be identifiable from these.

What are the risks and benefits of taking part?

It is possible that you may find it upsetting to discuss your experiences or the issues which affect you. You should consider this carefully before agreeing to take part. Your views may help to inform future services here at the RVI. However, you will remain anonymous.

What if there is a problem?

If you have a concern about any aspect of this study, you should speak to the researchers who will do their best to answer your questions. If you remain unhappy, and wish to complain formally, you can do this through the NHS Complaints Procedure. Details can be obtained from the hospital. In the event that something does go wrong and you are harmed during the research and this is due to

someone's negligence, then you may have grounds for legal action for compensation against Newcastle upon Tyne Hospitals NHS Foundation Trust and Newcastle University but you may have to pay your own legal costs.



Further Information

This research study is being organised by Newcastle University and is sponsored by NHS Foundation Trust. All research in the NHS is independently examined by a Research Ethics Committee to protect your safety, rights, wellbeing and dignity. This study has been reviewed and approved by a local Research Ethics Committee.

If you would like any further information please contact:

Morven Brown (Research Assistant):
Tel: 0191 282 1351
Email: morven.brown@newcastle.ac.uk



Quality of life, socio-economic status, relationships and risk perceptions in survivors of childhood or young adult cancer.

Information about the study

You are being invited to take part in a research study. It is important for you to understand why the research is being done and what it will involve before you decide whether to take part. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear or if you would like more information. Please take time to decide whether or not you wish to take part.



Why we are doing this study?

Recent research has highlighted that some young cancer survivors may face problems in education, employment and relationships. Other studies have also indicated that the experience of cancer may impact on a young person's life in other ways such as changing their views of life, their career aspirations or influencing how they view their health.

We would like to hear your views on these issues, and any other issues which you feel are of importance, regarding how an experience of cancer has impacted on your life. We would like to gain a better understanding of the long-term effects that a diagnosis of cancer brings for a young person. We are also hoping to explore any views survivors may have on preferences for information and support after a diagnosis of & treatment for cancer.



Why have I been invited to take part?

We have asked you to take part because you experienced a diagnosis of cancer when you were younger. It is up to you to decide whether or not to take part. If you do decide to take part you will be asked to sign a consent form, a copy of which you can keep. You are free to change your mind and withdraw at any time without giving a reason. A decision to withdraw or to not take part will not affect the care you receive. If you decide to withdraw from the study, any data collected from you will not be used.

What will I be asked to do?

You are invited to take part in a **focus group** with about 3-7 other survivors, who have had a similar diagnosis to yourself. This may last up to 60-90 minutes and it will take place at the Sir James Spence Institute at the RVI. We will provide snacks and refreshments and will also reimburse your travel/parking expenses. We hope that those who attend will feel comfortable enough to take part in an informal discussion on these topics and no staff from the hospital will be present. However, if you would prefer, we are happy to carry out **one-to-one telephone interviews**. We are interested in hearing about your

experience of life after cancer and how having cancer has impacted on you. Morven Brown, the researcher for this study, will facilitate focus groups and carry out the interviews. She can be contacted on 0191 282 1351 or emailed at morven.brown@newcastle.ac.uk

What will happen to the information I give you?

Information that is collected about you during the course of the research will be **strictly confidential**. Your personal details and any identifiable data will not be kept by us once the study has ended. As soon as the interviews are transcribed the recordings will be deleted and any information that you give will have your name, address and other personal details deleted so you cannot be identified from it. This resulting **anonymous** data will be stored securely for 10 years. All information will be stored securely in locked files and on password-protected computers. The information may be looked at by authorised members of the research team and by authorised people to confirm that the study is being carried out correctly.



Appendix C Consent forms for qualitative study



CONSENT FORM FOR PATIENTS

Quality of Life, socio-economic status, relationships and risk perceptions in survivors of childhood or young adult cancer.

Participant ID: _____

Please initial box:

1. I confirm that I have read and understood the information sheet (02/08/11, version 3) for the above study. I have had the opportunity to consider the information, ask questions, and had these questions answered satisfactorily.

2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving a reason, and that this will not affect my care. Any information I provide up to that point will also be withdrawn from the study.

3. I understand that data collected during the study may be looked at by regulatory authorities from the NHS Trust, where relevant to my taking part in this research. I give permission for these individuals to do so.

4. I understand that the focus group I am taking part in will be audio-recorded and typed out, and that all my identifying personal details will be removed.

5. I understand that direct quotations may be taken from what I say and used in publications, but that I will not be identifiable from these quotations.

6. I agree that relevant details regarding my health may be obtained from my medical records if necessary.

7. I agree to take part in the above study.

Name of participant

Date

Signature

Name of researcher

Date

Signature

CONSENT SCRIPT/FORM FOR PATIENTS (telephone interviews)

Quality of Life, socio-economic status, relationships and risk perceptions in survivors of childhood or young adult cancer.

Participant ID: _____

Interviewer:

“Before we start I need to check that you are happy to continue. I sent you a copy of a consent form recently, have you managed to read it?”

No.....*ask them if they have it at hand. If not, remind them to ask questions if not sure of something.*

Yes.....*continue*

“For ethical reasons, before we start the interview, I need to ask you the questions listed on the sheet and check whether you agree, disagree or have any questions you want to ask about the study. I also need to tape record this so there is a verbal record that you have been asked if you are happy to take part. After this we can go on with the interview. Is this OK with you?”

No.....*thank them for their time and finish call*

Yes.....*turn on tape recorder (“I’m just turning on the recorder”)*

“As you know, I’m Morven Brown, a researcher from Newcastle University and I am talking to you as you have shown an interest in taking part in our project. This project is exploring the impact that an experience of cancer can have on young people and how it may influence different areas of their lives.

Ok, so going through the points on the sheet...”

1. Do you confirm that you have read and understood the information sheet for the study and have had the opportunity to consider the information, ask questions, and have had these questions answered satisfactorily.

No.....*answer any questions and if don’t want to proceed thank them for their time and finish call*

Yes.....*proceed*

2. Do you understand that your participation is voluntary and that you are free to withdraw at any time, without giving a reason, and that this will not affect your care. Any information you provide up to that point will also be withdrawn from the study.

No..... *answer any questions and if don’t want to proceed thank them for their time and finish call*

Yes.....*proceed*

3. Do you understand that data collected during the study may be looked at by regulatory authorities from the NHS Trust, where relevant to your taking part in this research and that you give permission for these individuals to do so.

No..... *answer any questions and if don't want to proceed thank them for their time and finish call*
Yes.....*proceed*

4. Do you understand that the interview you are taking part in will be audio-recorded and typed out, and that all your identifying personal details will be removed.

No..... *answer any questions and if don't want to proceed thank them for their time and finish call*
Yes.....*proceed*

5. Do you understand that direct quotations may be taken from what you say and used in publications, but that you will not be identifiable from these quotations.

No..... *answer any questions and if don't want to proceed thank them for their time and finish call*
Yes.....*proceed*

6. Do you agree that relevant details regarding your health may be obtained from your medical records if necessary.

No..... *answer any questions and if don't want to proceed thank them for their time and finish call*
Yes.....*proceed*

7. And finally, do you agree to take part in the above study.

No..... *answer any questions and if don't want to proceed thank them for their time and finish call*
Yes.....*proceed and conduct interview using interview schedule*

This consent serves as documentation that the required elements of informed consent have been presented orally to the participant.

Verbal consent to participate in this telephone interview has been obtained by the participant's willingness to continue with the telephone interview by providing answers to the above questions

Researcher' Name (Printed)

Researcher's Signature

Date

Appendix D Topic guides for qualitative study



Topic guide:

Quality of Life, socio-economic status, relationships and risk perceptions in survivors of childhood or young adult cancer.

- ✓ Introduction of facilitators
- ✓ Thank participants for taking the time to attend.
- ✓ Brief summary of study, asking if there are any questions.
- ✓ The session will be recorded – is this OK with everyone?
- ✓ It is voluntary and if at any point they wish to leave, they can do so.
- ✓ They do not have to answer all questions that are posed to them.
- ✓ What is discussed in the focus group is confidential and should not be repeated out-with the group. Other's privacy needs to be respected.
- ✓ There are no right and wrong answers and it's important to be able hear everyone's ideas and opinions, the positive and the negative.
- ✓ Try not to talk over each other – just one person at a time if possible.
- ✓ Run through consent form and asking participants to sign this if they agree.
- ✓ Turn on recorder.
- ✓ Introductions of group.

Topic guide for patient focus groups

Impact on life:

Generally, how do you feel cancer has impacted on your life in the long-term, if at all?

Do you feel cancer has impacted on your life? (education, relationships, career, employment).

If yes, in what way/how did it impact on these areas? (education, relationships, career, employment).

Do you feel the experience of cancer has impacted on the course of your life in any of these areas?

Perceptions of life:

Has the experience of cancer influenced how you view life? (education, relationships, career, employment).

Do you have goals in life? If, so what is the most important goal in life to you?

Has this always been the case or do you think it has changed as a result of experiencing cancer?

Do you think there has been any positive outcomes as a result of having cancer? If yes, what are they?

Quality of life:

What does the term 'quality of life' mean to you?

How would you describe it? What are the important aspects of quality of life for you?

How would you describe your current quality of life?

If you had been asked what quality of life meant before your diagnosis, do you think you would have viewed it in the same way as you do now?

What would have changed the way you view it (if relevant)

Future health:

What do you think your experience of ill health in the past, means for your health in the future?

Are you concerned about your health in the future?

Are you aware of any late effects of treatment? Who discussed this with you?

What things do you think you can do to look after your health for the future? (diet, exercise, health behaviours)

Are there any things we haven't touched upon today which you would like to discuss?

- ✓ Ask if there is anything they want to ask about the study.
- ✓ Tell them they can contact a member of the research team, their consultant or key worker if they would like to discuss any issues raised in the focus group
- ✓ Thank them for their participation and ask if they would like a summary report.

Topic guide: interviews

Quality of Life, socio-economic status, relationships and risk perceptions in survivors of childhood or young adult cancer.

- ✓ Talk through study with interviewee
- ✓ Record verbal consent (as per YA Patient consent form version 4_verbal). If consent given, proceed with interview.
- ✓ Let them know they can take a break during the interview if they like, and to just say should they wish to.
- ✓ Let them know, that you understand that it may seem odd talking on the telephone and not face-to-face. However, the idea is that they can talk freely and it should be like a one-sided conversation.

Topic guide for patient focus groups

Impact on life:

Generally, how do you feel cancer has impacted on your life in the long-term, if at all?

Do you feel cancer has impacted on your life? (education, relationships, career, employment).

If yes, in what way/how did it impact on these areas? (education, relationships, career, employment).

Do you feel the experience of cancer has impacted on the course of your life in any of these areas?

Perceptions of life:

Has the experience of cancer influenced how you view life? (education, relationships, career, employment).

Do you have goals in life? If, so what is the most important goal in life to you?

Has this always been the case or do you think it has changed as a result of experiencing cancer?

Do you think there has been any positive outcomes as a result of having cancer? If yes, what are they?

Quality of life:

What does the term 'quality of life' mean to you?

How would you describe it? What are the important aspects of quality of life for you?

How would you describe your current quality of life?

If you had been asked what quality of life meant before your diagnosis, do you think you would have viewed it in the same way as you do now?

What would have changed the way you view it (if relevant)

Future health:

What do you think your experience of ill health in the past, means for your health in the future?

Are you concerned about your health in the future?

Are you aware of any late effects of treatment? Who discussed this with you?

What things do you think you can do to look after your health for the future? (diet, exercise, health behaviours)

Are there any things we haven't touched upon today which you would like to discuss?

- ✓ Ask if there is anything they want to ask about the study.
- ✓ Tell them they can contact a member of the research team, their consultant or key worker if they would like to discuss any issues raised in the focus group
- ✓ Thank them for their participation and ask if they would like a summary report.

Appendix E Development of the questionnaire (Chapter 4)

The content of the self-report questionnaire was informed by established questionnaire and survey design methodology (McColl *et al.* 2005; Oppenheim, 2005; Streiner & Norman, 2008) and followed the processes illustrated in Figure 6. The literature review identified important issues and existing measures which were relevant to the study. It is generally advised that, where validated and published measures exist, researchers utilise these (Boynton & Greenhalgh, 2004; Oppenheim, 2005). Doing so avoids attempts to ‘re-invent the wheel’, ensures scientific rigour and enables comparison of results across studies (Stone, 1993). The three main measures identified in the literature were the SF36v2 (a measure of health-related quality of life), the Brief Symptom Inventory-18 (BSI-18) (a measure of psychological distress) and the Impact of Cancer Scale for Childhood Cancer Survivors (IOC-CS) (a measure of survivors perceptions of the impact of cancer on their lives). These measures are described in detail in chapter 4 (section 4.4.3).

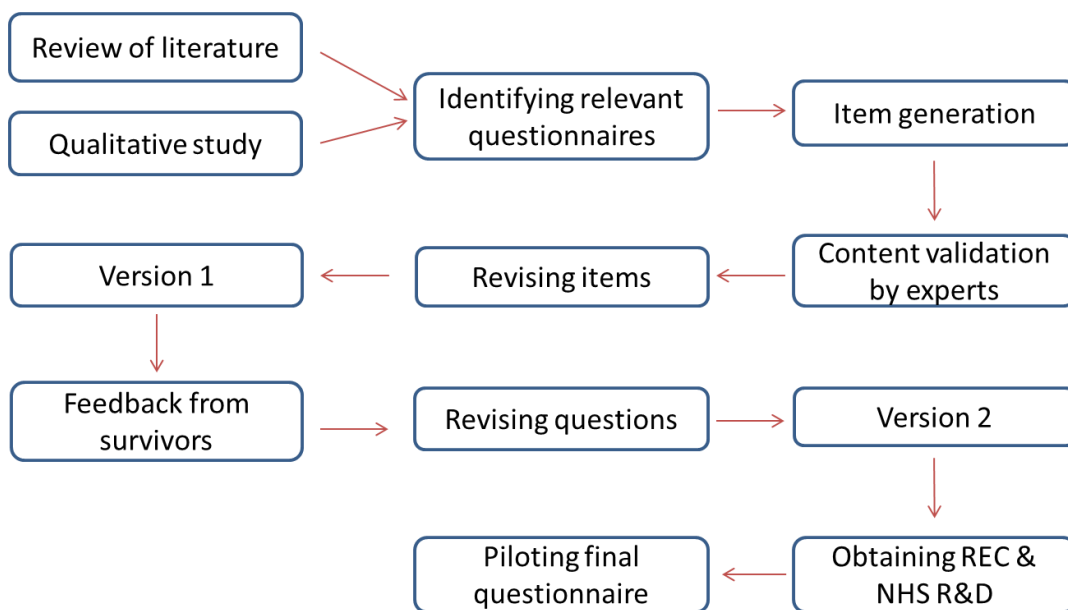


Figure 7 Process used to develop the questionnaire

In the absence of appropriate and standardised measures, it is acceptable to take individual items from existing questionnaires which fit the requirements of the study (Streiner & Norman, 2008) or develop a short bespoke questionnaire which can be administered alongside the standardised measures (Boynton & Greenhalgh, 2004). Important sources which can be used to inform the development of new items include the research literature, theory, expert opinion and importantly, patient views (Robson, 2002; Streiner & Norman, 2008). Therefore, the literature review and the results of the qualitative study aided identification of important areas to be included in the questionnaire.

Items to assess social outcomes of education, employment and income were adapted from existing surveys including the Health Survey for England 2012 (Health and Social Care Information Centre, 2012), the British Childhood Cancer Survivor Study (Hawkins, 1999), and the Childhood Cancer Survivor Study (Robison *et al.* 2002).

For new items, care was taken to keep them short and the language simple (Robson, 2002). The nature of each question informed the response options presented to respondents. The majority of questions were closed questions with limited response options (Oppenheim, 2005). Basic and factual questions requested a categorical judgement by indicating 'yes', 'no' or 'I don't know', or ticking the appropriate box(es) from a list of options. Items which ask the respondent to make a quantitative estimate of the size of an attribute, such as their level of agreement, are called continuous judgements (Streiner & Norman, 2008). Therefore, items which asked respondents for their level of agreement, desire or satisfaction used a 5-point adjectival scale which, in most cases, ranged from *not at all (1)* – *very much (5)*. These scale options were chosen as they reflected the response categories used in the IOC-CS and the BSI-18, therefore, providing consistency throughout the questionnaire.

Additional items were developed which aimed to capture detailed information on the survivors' experiences and views in education, employment and relationships, for example: their level of satisfaction with educational attainment; experiences relating to education (e.g. time missed at school due to illness, educational support received); satisfaction with current job; problems encountered in gaining or keeping employment; perceived reasons for not being married or being a parent and level of desire to marry and to have children.

To assess survivors beliefs regarding their future health and the perceived likelihood of developing ill health, survivors were asked to indicate on a 5-point Likert scale the extent to which they felt it was *very unlikely (1)* or *very likely (5)* that they would develop each of 21 health issues in the future. These 21 listed health issues are well known late adverse effects of treatment in survivors (e.g. heart problems, diabetes, poor hearing, anxiety, depression). If respondents felt they had already developed this health issue, they were asked to tick '*I already have this problem*'. To detect the presence of scarring, single items asked if survivors had scarring on three different areas of the body (face or neck; chest or stomach; legs or arms) and required a *yes/no* response.

To collect additional information, open questions were included in which the participants' were invited to expand on their answer to a previous question (e.g. if you have never been married, why do you think this is?). Open questions allow respondents freedom to answer as they wish (Oppenheim, 2005).

The main focus of the questionnaire booklet, and this thesis, was to investigate the psychosocial impact of cancer which, as stated in the introduction, are considered to be the psychological, emotional and social aspects of a disease and its treatment (National Cancer Institute, 2015). However, to gain a more complete picture of survivor's health and potential needs, questions were also included about the wider issues of health behaviour

and views of health. These items are not addressed in chapter 4 which focuses only on the psychosocial aspects of survivorship for the purpose of this thesis. Therefore, the items not included in chapter 4 are highlighted in grey in Table 18.

At the end of the questionnaire booklet, to assess acceptability of the questionnaire in survivors, supplementary questions invited feedback regarding their views on the questionnaire, their experience of completing it and suggestions to improve it.

Reviewing the draft questionnaire

Clinical and research staff who work with survivors of childhood and adolescent cancer (a consultant paediatric oncologist, two clinical psychologists, a long-term follow-up nurse specialist and a senior epidemiologist) were all invited to give their views on the draft questionnaire.

A valid questionnaire is one which measures what it intends to and there are many approaches to assessing validity (Streiner & Norman, 2008). Face validity is a process by which experts subjectively judge the questionnaire on whether it appears to measure what it aims to measure (Streiner & Norman, 2008). In the context of the present study, face validation was used in order for experts to judge whether the questionnaire booklet as a whole reflected the overall goals of the study. A related concept, content validity, entails a subjective judgement of whether the questionnaire covers all relevant domains (Streiner & Norman, 2008), therefore, experts were asked to comment if important aspects were missing. Experts were also asked for their view on the wording and appropriateness of the questionnaire, as well as the design.

There was a general concern that the questionnaire was lengthy. However, it was acknowledged that the questionnaire was aiming to be comprehensive. The nurse was concerned about sensitive questions surrounding sexual satisfaction/function and it was

agreed to remove these items. The rewording of some of the newly developed items was suggested, discussed and agreed as a group.

It is also important that the questionnaire assesses the outcomes in a way which is relevant and acceptable to the target participants (Rattray & Jones, 2005). Therefore, following feedback from experts, a revised version was given to two survivors. They were asked to give feedback on their experiences of completing the questionnaire, identify any questions which were unclear, comment on the design and layout, and provide suggestions for any missing content. Both survivors gave positive feedback stating that the design was appealing. There were no suggestions for further content, although the re-wording of a couple of items was suggested. The reported length of time to read the study information and complete the questionnaire was approximately 40 minutes. An overview of the final questionnaire is presented below in Table 18.

Table 18 Overview of questionnaire content

Page	Section	Measure/source of questions	Outcome
1-6	Your health-related quality of life	SF-36	Health-related quality of life
7-15	Your views on the impact of cancer	The Impact of Cancer Scale (IOC-CS)	Survivors perceived impact of cancer
16-17	Your views on health	Multidimensional Health Locus of Control (MHLC) Scales	General health locus of control beliefs: a measure of people's beliefs that their health is or is not determined by their own behaviour
18	Your views on health	Cancer Awareness Measure	Views of factors that increase the chances of getting cancer
19	Your physical health	Adapted from British Childhood Cancer Study (BCCSS)	Diagnosis Treatment received
20	Your physical health	New items	Presence/perceived likelihood of late effects of treatment
21	Your physical health	New items	Other health problems Presence of hair loss/scarring Timing of scarring

			Found scarring difficult to deal with
22	Your physical health	Items adapted from Newcastle Thousand Families cohort study	Height Weight
23	Your physical health	From Adult Oral Health Survey Adapted from NHS North of Tyne Cancer Awareness Research	Attendance at dentist Attendance at screening for breast cancer & cervical cancer (Females only)
24	Your feelings	Brief Symptom Inventory – 18 (BSI-18)	Presence of psychological distress symptoms
25	Your feelings	Shortened Warwick-Edinburgh Mental Wellbeing Scale (SWEMWBS)	Mental wellbeing
26-28	Your education	Item from BCCSS New items	Educational level Satisfaction with education achieved Illness affected education Missed time at school Repeat a year at school Educational support Delay in sitting exams What school year were in when left Experience of bullying/teasing
29-30	Your employment	Items from (BCCSS) New item New item New item	Employment status Job title/description Satisfaction with employment Problems with employment Cancer prevented from pursuing career
31	Your income	Adapted from Health Survey for England	Level of income
32	Insurance	Items adapted from EURO 2K cohort study for childhood cancer survivors	Life insurance and problems obtaining this Travel insurance and problems obtaining this
33	Your family	Items adapted from Newcastle Thousand Families cohort study	How many children in family Mother's employment Father's employment Mothers qualifications Father's qualifications
34-35	Relationships	Item adapted from BCCSS New items	Relationship status Desire to be married in future Reasons for not being married Cancer influenced views of marriage Age of first girl/boyfriend Current living circumstances

36	Pregnancies & children	Items adapted from BCCSS New item New item Question adapted from study by Hohmann <i>et al.</i> 2011	Have children Been pregnant/partner pregnant Told unlikely to have children Desire to have children Reasons for not having children
37-38	Physical activity	Physical activity questions taken from Health Survey for England/BCCSS	Limitations in sports/activities Use of mobility/walking aids Amount of physical activity
39	Diet	Adapted from the Newcastle Thousand Families cohort Study	Consumption of meat Consumption of fruit/veg
40	Alcohol	Questions taken from Health Survey for England/BCCSS.	Alcohol consumption
41-43	Smoking	Questions taken from Health Survey for England/BCCSS Exposure to tobacco smoke questions taken from Newcastle Thousand Families study	Smoking status Reasons for giving up smoking Reasons for starting smoking Exposure to tobacco smoke If mother/father smoked as child
44	Sun sensitivity	Items adapted from CCSS questionnaire	Use of tanning devices How often sunburnt Protection from sun

Highlighted items are not reported in this thesis

Appendix F REC approval for piloting of questionnaire



Health Research Authority

NRES Committee North East - Newcastle & North Tyneside 1

Jarrow Business Centre

Room 001

Viking Industrial Park

Rolling Mill Road

Jarrow

NE32 3DT

Telephone: 0191 428 3384

11 February 2015

Miss Morven Brown
Sir James Spence Institute
Newcastle University
Royal Victoria Infirmary
Queen Victoria Road
Newcastle upon Tyne
NE1 4LP

Dear Miss Brown

Study title:	Developing and piloting a questionnaire to help explain psychological and social outcomes in young adult survivors of childhood and adolescent cancer
REC reference:	15/NE/0003
IRAS project ID:	99312

Thank you for your submission of 9th February 2015. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 19 January 2015



Health Research Authority

North East - Newcastle & North Tyneside 1 Research Ethics Committee

Jarrow Business Centre
Room 001
Viking Industrial Park
Rolling Mill Road
Jarrow
NE32 3DT

Tel: 0191 428 3384

30 October 2015

Miss Morven Brown
Sir James Spence Institute
Newcastle University
Royal Victoria Infirmary
Queen Victoria Road
Newcastle upon Tyne
NE1 4LP

Dear Miss Brown

Study title:	Developing and piloting a questionnaire to help explain psychological and social outcomes in young adult survivors of childhood and adolescent cancer
REC reference:	15/NE/0003
Amendment number:	Substantial Amendment 1. 01/10/2015
Amendment date:	12 October 2015
IRAS project ID:	99312

The above amendment was reviewed at the meeting of the Sub-Committee held by correspondence.

This amendment was submitted to inform the committee of going over the target substantially (i.e. more than about 70 responses).

Ethical opinion

The members of the Committee taking part in the review gave a **favourable** ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

What is the study about?

Previous research has suggested that a young person's psychological and social development may be affected by the experience of cancer. While some survivors report a negative effect on areas such as their education, employment or relationships, others do not. In fact, some survivors may feel that they have benefitted in some way from their experience of ill-health.

We have developed a questionnaire which we hope will help us to better understand why and in what way cancer can affect a young person's life several years after they have completed treatment. By filling this questionnaire in, you will help us to collect important and useful information for our research. We would also like to explore how different medical factors (e.g. diagnosis, age at diagnosis, treatment received) may influence outcomes for survivors.

This study builds on previous work carried out by Newcastle University and the Great North Children's Hospital which involved interviews with survivors about their views of how cancer had impacted on their lives. This study is being led by Dr Rod Skinner at the Great North Children's Hospital and Dr Mark Pearce at Newcastle University.

As this is a new questionnaire, your views would be of great value to us. Also as you are one of the first survivors to be sent this questionnaire, it will help us to know if there are any problems with it and if there are ways in which we can improve it for future use.

What are the possible advantages and disadvantages of me taking part?

This work will help us to identify ways in which survivors can be better supported.

Personally, you may find it rewarding to contribute to research.

Findings from this research (which will be anonymous) may be published in a medical journal. You will be sent a summary of the results once the study is complete.

This application has been reviewed by the Newcastle & North Tyneside 1 Research Ethics Committee. We do not expect there to be any harm from you taking part. However, it may be possible that you find thinking about your experiences upsetting. If this is the case and you would like to talk to someone, you are asked to contact the Long-term Follow-up Nurse Specialist (Sandra Barlow, Tel: [REDACTED] [REDACTED] who will be happy to speak with you.

If you are concerned about any aspect of the study, the researcher (Morven Brown) will do their best to answer your questions. If you remain unhappy you can contact the Patient Advice and Liaison Service (PALS) for Newcastle upon

Tyne Hospitals NHS Foundation Trust (Freephone: 0800 0320202) who can provide guidance. If you wish to complain formally, you can do so through the NHS Complaints Procedure. Details can be obtained from the hospital.

If you are interested by what you have read so far, please read the information at the beginning of the questionnaire.

The Newcastle University researcher for the study is: Morven Brown
(Research Assistant & Trainee Health Psychologist). Tel: **0191 282 1351**
Email: morven.brown@ncl.ac.uk

Appendix H Consent form for piloting of questionnaire



Study ID number: _____

CONSENT FORM

Title of Project: **Developing and piloting a questionnaire to help explain psychological and social outcomes in young adult survivors of childhood and adolescent cancer**

Please initial all boxes

1. I confirm that I have read and understand the information enclosed in this booklet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.

3. I understand that the relevant sections of my medical notes and the data collected during the study may be looked at by the researchers from Newcastle University, or individuals from regulatory authorities or the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my records.

4. I agree to take part in the above study.

Please sign below

Name of Participant Signature Date

If you have any questions about this form, please contact Morven. Tel: 0191 282 1351, Email: morven.brown@ncl.ac.uk

Appendix I Survivor refusal form for piloting of questionnaire

Study ID _____

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If you have read the information and decided that you do not want to fill-in the questionnaire, would you mind letting us know why?

If survivors do not feel this questionnaire is something they would like to fill-in or feel uncomfortable doing so, your feedback could help us improve the questionnaire for future use.

I did not want to fill-in this questionnaire because (please tick all the reasons):

The questionnaire was too long	<input type="checkbox"/>
I felt the questionnaire was boring	<input type="checkbox"/>
I do not have the time to fill-in the questionnaire	<input type="checkbox"/>
I didn't think the questionnaire applied to me	<input type="checkbox"/>
I felt the questions were too personal	<input type="checkbox"/>
I was concerned about confidentiality of my answers	<input type="checkbox"/>
I did not understand what I was expected to do	<input type="checkbox"/>
I did not understand what the point of the questionnaire was	<input type="checkbox"/>
Other reason?	

Thank you for taking the time to read about the study.