The Self Reported Psychosocial Wellbeing of Adolescent Childhood Cancer Survivors in New Zealand

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ATTESTATION OF AUTHORSHIP

"I hereby declare that this submission is my own work and that, to the best of my knowledge and belief, it contains no material previously published or written by another person (except where explicitly defined in the acknowledgements), nor material to which to substantial extent has been submitted for the award any other degree or diploma of a university or other institution of higher learning"

Signed: Date:

Kathy Yallop

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ABSTRACT

There has been increased recognition of the psychosocial impact of a diagnosis of cancer as well as the intensive treatment necessary to effect a cure. However there is a lack of consensus across studies on the degree and type of psychosocial difficulties experienced by young survivors. The aim of this study was to describe the self-reported psychosocial wellbeing of adolescent childhood cancer survivors. In this case-control study, 170 childhood cancer survivors aged 12 to 18 years completed an internet based survey. This was a modified version of the Youth'07 Health and Wellbeing Survey of Secondary School Students in New Zealand. The comparison group were the 9,107 students who took part in the Youth'07 survey. Psychosocial wellbeing was assessed by four standardised measures of: a) wellbeing (WHO-5), b) anxiety (MASC-10), c) depression (RADS2-SF) and d) emotional and behavioural difficulties (SDQ). The majority of childhood cancer survivors scored within the normal range across all four measures; WHO-5 (89%), MASC-10 (93%), RADS2-SF (94%) and SDQ total difficulties (82%). Compared to a normative sample of their peers, they reported greater psychosocial wellbeing (very good or excellent 60.2% vs. 49.9%, $p < 0.01$), greater prosocial behaviour (86.5% vs. 78.2%, $p < .01$) and a trend towards less depression (6.1% vs. 10.6%, $p = .09$). There was however, a small but important minority who reported significant depression (9%), anxiety (7%), poor emotional wellbeing (11%) and increased emotional and behavioural difficulties (18%). Survivors of CNS disease, older age and older age at diagnosis were shown to have the greatest psychosocial difficulties. Following a diagnosis of childhood cancer, intensive therapy and the subsequent risk of adverse health outcomes, one might expect childhood cancer survivors as a group to not be doing as well as their peers in terms of psychosocial wellbeing. The findings of this study, however, show that childhood cancer survivors

are doing as well, and in some cases better, than their peers. This is the first study to report on the psychosocial wellbeing of adolescent childhood cancer survivors within New Zealand.

CHAPTER 1
INTRODUCTION AND OVERVIEW

> *It is as if we have invented sophisticated techniques to save people from drowning, but once they have been pulled from the water, we leave them to cough and splutter on their own in the belief we have done all we can*. Fitzhugh Mullan, MD & cancer survivor*, 1985* (as cited in Richardson, Nelson, & Meeske, 1999, p136).

1.1 Background

Care of cancer survivors has come a long way since that quote was written. The survival rates for children and young people who have had a childhood cancer have risen dramatically in the past 20 - 30 years with current estimates of an overall 5 year survival rate of greater than 80%. This is an impressive statistic when compared with adult cancer survival rates of 66% (Ries et al., 2007). The majority of children with cancer can now expect to survive for many years after diagnosis.

Increased survival rates have been brought about by a combination of advances in treatment, improved supportive therapies and collaborative multi-centred clinical trials (Hewitt, Weiner, & Simone, 2003). It is now estimated that in developed countries about 1 in every 1,000 adults reaching the age of 20 will be a long term survivor of cancer (Last, Grootenhuis, & Eiser, 2005). In New Zealand each year approx 160 children 15 years of age or younger are diagnosed with a childhood malignancy, therefore with an estimated 80% or greater survival, every decade will see an additional 1200 survivors within our population. However, cure has come at a cost, as cancer survivors are at risk for physical or psychosocial late effects from their disease, chemotherapy, radiation therapy and surgery (Hudson et al., 2003; Richardson et al., 1999). The U.S. Child Cancer Survivorship Study Group (CCSS) is one of the largest

multi-centred research groups and they have followed a cohort of 14,000 survivors diagnosed between 1970 and 1986. They concluded that two out of three childhood cancer survivors are likely to experience at least one chronic health problem and one in every four survivors is likely to experience a severe late effect as a consequence of their treatment or malignancy (Mody et al., 2008; Oeffinger & Hudson, 2004; Oeffinger et al., 2006; Richardson et al., 1999). The diagnosis and subsequent treatment of cancer in children usually occurs during the formative development years and impacts on normal growth and development, organ development including cardiovascular, endocrine, sensory (hearing & vision) and musculoskeletal, as well as neurological and neurocognitive development. As this is a childhood disease any late effect has the potential to significantly impact across the adult age-spectrum (Leisenring et al., 2009).

There is a sound body of knowledge around the medical late effects for childhood cancer survivors based on risk-related exposure to therapies. Evidence-based guidelines are established for follow-up surveillance. The most comprehensive being: a) *Long Term Follow-up Guidelines for Survivors of Childhood, Adolescent and Young Adult Cancers,* produced by the Children's Oncology Group (COG) Late Effects Committee and the Nursing Discipline in 2003, revised 2006 and 2008 (Children's Oncology Group, 2006), and b) "*Practice Statement*" issued by the United Kingdom Children's Cancer Study Group; Late Effects Group in 1995 and revised in 2005 (United Kingdom Children's Cancer Study Group, 2005). The risk of late effects for childhood cancer survivors is varied and dependant on the disease, type and intensity of treatment and individual personal characteristics, with those at greatest risk for significant cognitive and endocrine late effects being survivors of brain tumours and central nervous system directed therapies (Richardson et al., 1999; Shaw, 2009).

The psychosocial consequences of living with medical late effects and the journey through cancer are less well understood. For adolescents, medical and

psychosocial effects intersect with the already difficult transitions involved in normal young adult development (Richardson et al., 1999). While there is increasingly rich and varied data describing the effects on quality of life, risk taking behaviours, posttraumatic stress and social interactions of survivors, there is no clear consensus on psychosocial outcomes for child cancer survivors (Zebrack & Chesler, 2001; Zebrack & Chesler, 2002).

1.2 New Zealand Childhood Cancer Survivorship experience

The New Zealand Paediatric Haematology/Oncology service provides a national service for children diagnosed with cancer and non-malignant haematological diseases from birth to 18 years of age at diagnosis. This service is provided through two tertiary treatment centres (Auckland and Christchurch) and multiple regional shared care facilities throughout the country. The national service holds membership to several international study groups, the most significant being the large cooperative multi-centred Children's Oncology Group (COG) with the majority of patients diagnosed in this country now being enrolled in phase 3 and 4 clinical trials.

Long term follow-up care of survivors of childhood cancer became formalised in 2006 with the establishment of a national Paediatric Haematology/Oncology Late Effects Assessment Programme (LEAP) with provisional Ministry of Health funding for an initial period of three years. This was the result of a Paediatric Haematology/Oncology Steering Group (POSG) position paper and proposal in response the NZ Cancer Control Strategy Action Plan 2005-2010. The 2005-2010 plan identified the need to "ensure all survivors of childhood and adolescent cancer receive timely and ongoing support and rehabilitation, including the early identification of and intervention in, late effects" p.73. Survivors of childhood cancer transition into the LEAP programme at approximately two to five years from the end of treatment. They continue to be offered medical surveillance and psychosocial support until late

adolescence or early adulthood, this typically includes an annual clinic visit with the multidisciplinary team.

LEAP long term follow-up care has been modelled on overseas programmes, incorporating evidence based guidelines and recommendations into the programme. While this is "Best Practice" and provides us with the advantage of learning from the experiences and knowledge of much larger institutions, when we look at the psychosocial and quality of life issues these are less well translated across to New Zealand. Within New Zealand society the effects of culture, community, education, healthcare practice protocols are very different from those of European and America cultures.

For young people living in New Zealand who have survived a childhood cancer, our knowledge of the consequences of living with late effects as they try to deal with the normal tasks of adolescence and adulthood is limited. This limited knowledge is based on assumptions made from data drawn from other cultures. It is timely to gather our own information on the issues faced by young people who have survived cancer so we better understand them and develop supportive care interventions to improve outcomes.

1.3 Youth2000 and Youth'07 – National Health and Wellbeing Surveys of New Zealand Secondary School Students.

In 2001 the first national health and wellbeing survey of New Zealand youth was undertaken, this was a seminal study that provided the most comprehensive, accurate and up to date information on the health and wellbeing of young people growing up in New Zealand (Adolescent Health Research Group, 2003a). The aim of the Youth2000 survey was to determine the prevalence of selected health behaviours and protective factors in a representative population of New Zealand youth comprising approximately 10,000 secondary school students from 133 randomly selected colleges throughout New Zealand. The cross sectional self report survey incorporated 523 questions using an

innovative multimedia (pictures, music and animation) computer assisted self interview format (M-CASI). The domains of interest were ethnic diversity, health status, psychosocial wellbeing, health service utilization, social and environmental protective factors and health behaviours (both positive and negative). The findings of the survey supported the implementation of the Youth Health Development Strategy and the Youth Health Action Plan (Adolescent Health Research Group, 2003a)

A second national health and wellbeing study called Youth'07 was carried out in 2007 and 2008, the aim was to update and extend the original survey conducted in 2001 by tracking trends and investigating new issues for young people (Adolescent Health Research Group, 2008b). For this survey, approximately 10,000 students from 96 randomly selected secondary schools throughout New Zealand participated. The opportunity to use the Youth'07 questionnaire to survey survivors of childhood cancer and compare the results with a cohort of their peers who had not had cancer was made possible through the generous offer of access to the web-based questionnaire by the researchers, the Adolescent Health Research Group (AHRG) and funding from Rotary and CanTeen.

This survey of the psychosocial wellbeing of childhood cancer survivors in New Zealand, given the acronym ACSIS (*Adolescent Cancer Survivor Impact study)* was carried out in 2009 to 2010. My role in the study was that of principal investigator. A section of the total survey that comprised four standardised measures of adolescent wellbeing has been used for the purpose of this thesis.

1.4 Aims of the Study

The primary aim of this study is to describe the self-reported psychosocial wellbeing of childhood cancer survivors aged between 12 and 18 years. Specific research objectives include the following:

a) Describe the psychosocial wellbeing of adolescent childhood cancer survivors.
b) Test whether the type of childhood cancer diagnosis and treatment are associated with the psychosocial wellbeing of adolescent survivors.
c) Compare psychosocial wellbeing of adolescent childhood cancer survivors with a control group of their peers.

This will be a non interventional case control study with matched control design, where participants will complete an internet-based, computer administrated branching questionnaire using M-CASI (multimedia computer assisted self interview programme). The comparison group will be the 9,107 students throughout New Zealand who completed the same questionnaire for the Youth'07 health and wellbeing survey.

This study aims to identify differences, if any, in the psychological wellbeing of survivors compared with their peers. In particular, identify the risk factors and possible resiliencies that are significant to them as a unique group and identify common areas of difficulty. This information may be used to improve health outcomes and minimise the impact of late effects of their cancer experience. This study contributes to the rapidly increasing body of knowledge that exists internationally and provides for the first time a unique view of the emotional wellbeing of these young people in Aotearoa New Zealand. This will enable us to identify particular areas of need, gain an understanding of what may contribute to these and from this we can better plan assessments, interventions, services and supports in survivorship care.

The term "survivor" has become synonymous with young people who have been cured of a cancer and achieved a long term remission. It is difficult to find literature that doesn't use the term survivor in relation to childhood cancer. This is equally true of those cured or in remission from an adult cancer. The term was initially adopted by the National Coalition for Cancer Survivorship (NCCS) in 1986 at a time when cancer was a disease with poorer outcomes and was something to be fought (Ellen Stovall, as cited

in Twombly, 2004). The NCCS was established to move the perception from victim to survivor by Dr Fitzhugh Mullan, a survivor of cancer himself and whose quote is cited at the beginning of this chapter. He coined the phrase to describe the " seasons of survival" in an essay of his own cancer experience (Mullan, 1985). There has been a recent resurgence of debate about the term cancer survivor. In one study that examined the "survivor identity" of women with breast cancer, Kaiser (2008) noted that while some women identified with the survivor label which assisted them in adjusting to life after cancer, others rejected it and rarely referred to themselves by this term. In my experience this view is shared by young people who have had a childhood cancer. However as was reported by Twomby (2004) "no one has yet come up with a better term". For these reasons the term childhood cancer survivor is used throughout this thesis but it is acknowledged that this may not be a term young people who have had a cancer use to define themselves.

1.5 Structure of the Thesis

Chapter 1 has identified the purpose of this study: to describe psychosocial wellbeing of childhood cancer survivors. The long term consequences, both physical and psychosocial following a childhood cancer and/ or treatment have been introduced and rationale for the study design explained. The aim and significance of the study have been stated.

Chapter 2 presents a critical review of the existing literature and research on the aspects of the psychosocial wellbeing of survivors of childhood cancer. A summary of this chapter identifies the lack of consensus of the psychosocial impact for survivors and the absence of New Zealand specific research.

Chapter 3 describes the research objectives and the methodological approach for this study. The research instrument – the MCASI questionnaire is described as are the four standardised measures of psychosocial wellbeing. The procedures followed in

collecting and analysing the data is stated. Cultural and ethical considerations conclude the chapter.

Chapter 4 presents the key findings from an analysis of the research data using quantitative research methods.

Chapter 5 discusses the findings of the study with reference to each of the research questions and identifies the study strengths and limitations. The significance of the findings and the implications for practice and future research are discussed.

CHAPTER 2
LITERATURE REVIEW

2.1 Introduction

The aim of this study is to explore the self reported psychosocial well being of adolescent child cancer survivors in New Zealand. A review of current literature was carried out by reviewing those studies and reviews that looked specifically at the late effects of childhood cancer, in particular, psychosocial outcomes, posttraumatic stress symptoms, quality of life, risk taking behaviours and resiliencies of childhood cancer survivors (CSS). The primary focus was on published literature within the past ten years. Databases searched for this literature review were MEDLINE, CINAHL, PsychINFO and Cochrane Library using EBSCO and OVID, and Google Scholar. Keywords and phrases used were; childhood cancer, survivors, late effects, psychosocial wellbeing, quality of life, cancer treatment toxicities, post traumatic stress, anxiety, and health status of CCS. Additional literature was identified from reference lists and journals within the Haematology/Oncology Unit, Starship Children's Hospital.

2.2 Medical late effects

Childhood cancer therapy affects growing and developing tissues, so children and adolescent survivors are at increased risk of morbidity, mortality and diminished quality of life associated with their previous cancer therapy (Oeffinger & Hudson, 2004). Cancer therapies frequently include irradiation and certain chemotherapy agents that may significantly increase the risk of cognitive dysfunction, liver damage, endocrine, cardiac dysfunction, lung disease including fibrosis and precocious emphysema (Mody et al., 2008; Wallace et al., 2001). These physical late effects can have a significant impact on the psychosocial wellbeing and quality of life for survivors (Friedman, 1999; Speechley, Barrera, Shaw, Morrison, & Maunsell, 2006). It is also

known that childhood cancer survivors have an increased incidence of second malignancies with an estimated 30-year cumulative incidence of 9.3% (Meadows et al., 2009).

Many long term effects from childhood cancer therapy do not plateau but increase with age, often becoming apparent decades after therapy (Oeffinger & Hudson, 2004). Cancer survivors in the United States report higher use of special educational resources, lower graduation rates, lower employment and difficulty obtaining life insurance. Female survivors who received cranial irradiation and brain tumour survivors are at greatest risk of these outcomes (Mody et al., 2008; Richardson et al., 1999; Zeltzer et al., 1997)

2.3 Psychological health status in adolescent survivors of childhood cancer

Over the past 10 years there has been rich and varied data exploring the effects on quality of life, risk taking behaviours, posttraumatic stress and social interactions of survivors. However, there is no clear consensus on the impact of these psychosocial issues for child cancer survivors. Several studies have suggested that a large percentage of adolescent survivors may be at increased risk for adverse behavioural and social outcomes and been found to have consistently poorer health related quality of life compared to healthy matched peers (Hobbie et al., 2000; Mody et al., 2008; Speechley et al., 2006; Zebrack & Chesler, 2001; Zebrack & Chesler, 2002). The Childhood Cancer Survivor Study (CCSS) of 7147 adult survivors (Ness et al., 2008), found that limitations in physical performance, executive function and emotional health are all negatively associated with self reported health related quality of life (HRQL). Conversely, other studies report finding a generally positive quality of life, increased happiness and better adjustment psychosocially than their peers (Parry & Chesler, 2005; Zebrack & Chesler, 2002). Poorer HRQL outcomes of acute and chronic illness for females are reported in many studies, and female survivors of childhood cancer

demonstrate a higher likelihood of an adverse health status (Hudson et al., 2003; Nathan et al., 2007). Survivors of central nervous system (CNS) tumours and those who were treated with CNS directed therapy had the poorest outcomes (Schultz et al., 2007; Vannatta, Gerhardt, Wells, & Noll, 2007; Zeltzer et al., 2009). In a report from the Childhood Cancer Survivor Study on the psychological status of childhood cancer survivors, when compared with siblings, they were overall relatively healthy, however they were 80% more likely to report impaired mental health QOL and twice as likely to report emotional distress (Zeltzer et al., 2009). The authors also reported that up to forty percent of CCS demonstrate neurocognitive deficits in one or more domains and this can result in behavioural or emotional disorders.

A major limitation of CCS studies identified by Shultz et al. (2007) and Speechley et al. (2006) is that several used parent-report questionnaires that may not accurately reflect the HRQL self reports of adolescents. This is confirmed by a study by Levi and Drotar (1999) comparing self report and parent report of HRQL. The parent-reported HRQL was significantly lower than the adolescents' self report, suggesting that the two groups viewed the HRQL of the adolescents very differently. An additional limitation discussed by Last, Grootenhuis and Eiser (2005) in the quality of psychosocial research in child cancer survivors, is that most studies have focused on broad adjustment issues e.g. depression, anxiety, educational attainment and social functioning rather than quality of life. The use of quality of life (QOL) instruments has been limited, partly because no standardized instrument exists at present that can be applied with equal relevance in different European paediatric populations.

2.4 Posttraumatic Stress, depression and anxiety in adolescent survivors of childhood cancer.

Posttraumatic stress disorder (PTSD) is defined as an anxiety disorder that can develop after exposure to one or more traumatic events that threatened or caused great

physical harm (National Institute of Mental Health, 2011). It is well documented that survivors of adult cancer and parents of children with cancer are at increased risk for PTSD, though fewer studies have identified PTSD in childhood cancer survivors (Hardy et al., 2008; Kazak et al., 1997; Schwartz & Drotar, 2006). Kazak et al. (1997) suggested that while there were no significant differences between survivors and comparison children in levels of posttraumatic stress symptoms (PTSS), the posttraumatic distress found in a high proportion of parents of survivors had an effect on their children's understanding of illness and overall adjustment and warranted further study. This was supported by Stuber et al. (1997) who noted that a mother's perception of the effects of the disease and treatment contributed to the survivor's anxiety but did not independently contribute to post traumatic stress symptoms.

PTSD was also associated with older age in a study of 18 to 40 year old survivors by Hobbie et al. (2000). The authors found that one fifth met the criteria for PTSD, much higher than that seen in younger survivors and hypothesised that as young adult survivors became more independent and less protected by childhood they face increased uncertainties and limitations caused by late effects of the cancer.

Kazak et al. (1997) noted that the earlier age of diagnosis for many childhood cancers indicated the limited ability of pre-school and school-age children to cognitively process the life threat of a cancer diagnosis and associated trauma may be the reason for lower rates of PTSD seen in this group. No association was found between ratings of treatment intensity and medical late effects and many of the studies agreed that those with or without PTSD did not differ on duration and intensity of cancer treatment or number of relapses.

Survivors of brain tumours, leukaemia, neuroblastoma and bone tumours demonstrated elevated rates of psychosocial distress including depression, anxiety and somatization compared with siblings (Zeltzer et al., 2009). In a secondary analysis using

data from the Childhood Cancer Survivor Study, a comparison of survivors (n =2979) with siblings (n = 649) aged 12 to 17 years, showed survivors were 1.5 times more likely to have symptoms of depression and anxiety and that CNS disease or CNS - directed therapies were specific risk factors (Shultz et al., 2007).

In a review of the research into depression in paediatric cancer, Dejong and Fombonne (2006) noted that there have been few studies that have looked at depression in this population and of those most have excluded brain tumours. However, brain tumours make up twenty percent of all childhood cancer diagnoses and survivors have increased vulnerability to mood alterations. The authors concluded that there was only a modest prevalence of depression but concluded that minor depression may still have a significant negative effect on quality of life. In a study of QOL and depression symptoms in a group of seventy adult CCS, Sharp, Kinahan, Didwania and Stolley (2007) found that while most survivors were doing well, 21% had elevated symptoms of depression (n =15) and reported significantly poorer QOL scores across all domains.

To summarise, post traumatic stress appears evident in parents of childhood cancer survivors and is also evident but to a lesser degree in young adult survivors. One theme that seemed to be evident in the review of the literature on post traumatic stress was the frequent mention of anxiety either as a symptom of PTSD or a trait that could predispose people to experience the disorder. Depression was not identified as a significant psychosocial consequence for this population, however, the potentially vulnerable group of brain tumour survivors were excluded from most studies.

There were no studies I identified that looked at anxiety in child cancer survivors as a research question but anxiety was interwoven with, and used as a measure of both psychosocial distress and quality of life.

2.5 Limitations of studies

A large number of studies of HRQL and psychosocial wellbeing of survivors of childhood cancer have come from using data from two main longitudinal cohort study groups namely the Childhood Cancer Survivor Study (CCSS) and the Late Effects Study of the Canadian Childhood Cancer Surveillance and Control Programme (CCCSCP). The use of siblings as a control group has been common in many of these studies. There is a potential bias inherent in sibling controls associated with the effects that childhood cancer can have on those siblings (Speechley et al., 2006). A further potential limitation of the CCSS is that much of the data has been collected from a cohort of survivors treated between1970 and1986, differences in modern therapies may mean that findings may not correlate with those experienced by those survivors treated in later years. The CCSS are now expanding the cohort to include those survivors diagnosed between 1987 and 1999.

Several studies as previously discussed, have used parent questionnaires as proxy reports on the quality of life of survivors which has been shown not to always correlate with adolescent's view of their own well being.

In a report from the Childhood Cancer Survivorship Study, Zeltzer et al. (2009), wrote "a review of the literature on psychosocial outcomes of childhood cancer survivors demonstrates varied, and sometimes contradictory results". The authors go on to say that these inconsistencies are likely caused by "small sample sizes, varied outcome measures and population norms used for comparisons including demographic differences" (p. 2396).

2.6. Consideration of research methods

This study involves internet data collection. In this section, literature addressing research surveys is presented.

2.6.1 Internet surveys and studies

The internet is becoming an increasingly popular form of data collection, with many benefits as it is quick to administer, flexible and inexpensive compared to mail and phone surveys (Best, Krueger, Hubbard, & Smith, 2001). However, issues such as internet accessibility, skill in computer use and privacy of personal information are of concern. Studies have confirmed that younger respondents show a preference for internet surveys over mail surveys (Lusk, Delclos, Burau, Drawhorn, & Aday, 2007), (Kaplowitz, Hadlock, & Levine, 2004), but they can only be administered to those who have access to the internet and those who have the skills to use it. This questions the generalizability of internet surveys as they may not be representative of the entire population being tested (Best et al., 2001). This is supported in a study that found internet accessibility is strongly associated with younger age groups, demographic variables, and male gender, suggesting the use of an internet survey may not yield a representative sample (Couper, Kapteyn, & Schonlau, 2005).

Prior to the initial Youth2000 study, Watson et al. (2007) pilot tested (n =110) the multimedia computer assisted self-administered survey using both desk top and laptop computers to ascertain young peoples' perceptions of this form of questionnaire and concluded that it was an acceptable instrument for the administration of such a survey.

2.6.2 Increasing participation

Higher compliance rates are associated with greater statistical power, lower survey error, and are more representative of the target population thus producing results that are more generalizable and higher in external validity (Lusk et al., 2007). Incentives have shown to be effective in increasing response in internet surveys, a $10 incentive provided greater than 20% response rate in one mailed survey (Rosoff et al., 2005). Non-response rate to mail and internet surveys is often cited as a major disadvantage

and may importantly bias the conclusions of a study, the importance of sending follow-up reminders to improve response rates has been demonstrated (Kaplowitz et al., 2004).

In conclusion, factors such as follow up, the age of the respondent and the effective use of incentives are all associated with higher response rates. It appears there are many benefits to using internet surveys within certain age populations, specifically adolescents, as they are quick to administer, inexpensive and flexible compared to mail and telephone surveys. However, issues such as internet accessibility and privacy concerns need to be taken into account.

2.7 Summary

In the context of New Zealand childhood cancer survivorship, there has been no research into the psychosocial impact on these young people, so the implementation of a long term follow up programme is based on anticipated needs identified in overseas studies. The review of the literature confirms that late effects of childhood cancer therapy can include disruption to normal growth and development especially during adolescence. Adverse effects on quality of life, risk taking behaviours, post-traumatic stress and social interactions of survivors are all identified by various authors. However, there is no clear consensus on the impact of these psychosocial issues for child cancer survivors (Zebrack & Chesler, 2001; Zebrack & Chesler, 2002; Zeltzer et al., 2009).

It is clear from the review of existing literature that there are a significant number of validated, peer reviewed studies providing valuable insight into not only the medical consequences of treatment to effect a cure, but also the psychosocial impact on the rapidly growing numbers of survivors world wide. While much of the findings of these studies extrapolate across to the New Zealand childhood cancer survivor experience, the opportunity to repeat the Youth'07 survey and compare findings with a cohort of peers provides a unique opportunity to capture information on the psychosocial wellbeing of young people who have survived childhood cancer in

Aotearoa/New Zealand. The following chapter introduces the research questions and the study design and methods employed.

CHAPTER 3
METHODS

3.1 Introduction

This chapter describes the aims of the current research, study design, methods and ethical and cultural considerations. The primary aim of this study is to describe the self reported psychosocial wellbeing of young people aged 12 years to 18 years of age, living in New Zealand who have survived childhood cancer.

3.2 Research Objectives

a) Describe the psychosocial wellbeing of adolescent childhood cancer survivors.
b) Test whether the type of childhood cancer diagnosis and treatment are associated with the psychosocial wellbeing of adolescent survivors.
c) Compare psychosocial wellbeing of adolescent childhood cancer survivors with a control group of their peers.

The Adolescent Survivor Impact Study (ACSIS) was an adaption of the Youth'07 questionnaire. The ACSIS survey covered a number of areas including the broader domains of psychosocial wellbeing of childhood cancer survivors. Psychosocial wellbeing, for the purposes of this thesis is measured by the four standardized questionnaires for the screening of anxiety, depression, emotional wellbeing and emotional and behavioural disorders in survivors of childhood cancer included in the broader ACSIS questionnaire.

3.3 Study design

This was a case-control study in which participants completed an internet-based, computer administrated branching questionnaire using M-CASI (multimedia computer assisted self interview programme). The decision to describe this as a case-controlled study followed a discussion with the biostatistician. However, it is acknowledged that it

could also be reported as a nested case-control study with an historical comparison group.

Advantages of case-control studies are described as being able to study rare diseases with a relatively small sample size and are generally less expensive. The two most commonly cited disadvantages of case-controlled studies however are described as confounding and bias, that is, the difficulty in describing cause and effect (confounding variables) and bias, in particular recall bias with cases over reporting and cases under reporting an exposure (Bombardier, Kerr, Shannon, & Frank, 1994). The case-controlled design is appropriate for this study as childhood cancer is an uncommon disease and it would have been difficult to recruit a large sample. Data had already been collected from a normative control group that was current and provided a large representative sample of the same source population i.e. New Zealand youth aged between 12- 18 years. This large control group allowed for multivariate comparison with child cancer survivors with allowance for differences between the groups, in age, sex, ethnicity and socioeconomic status to control for those potentially confounding variables.

3.4 Cases: Childhood Cancer Survivors

There are two primary cancer treatment centres (Auckland and Christchurch) supported by a network of regional outreach services throughout New Zealand. These provide supportive and follow-up care to patients domiciled outside the main treatment centres. These links made it easier to identify and obtain current contact details for prospective participants.

Inclusion criteria were all survivors of a childhood disease that met the International Classification of Childhood Cancers version 3 (ICCD-3), aged between 12 years and 18 years inclusive at the commencement of the study and who were at least two years from completion of therapy and disease free. Participants had to have English

language skills equivalent to year 6 (10 years of age) the same criteria as that set for the Youth2000 and Youth'07 surveys. In addition, participants needed to be able to physically use a computer as well as understand instructions to competently interface with the computer and questionnaire. Those who met the criteria but had not been the recipient of medical surveillance for their cancer within the past 4 years were excluded, because it couldn't be determined if the young person was aware of having had a cancer diagnosis and it would be unethical to approach them. An example of this would be a child diagnosed at a young age with a pilocytic astrocytoma that is classified as a cancer but is frequently successfully treated with surgery only and is often not referred to as cancer. In those cases where eligibility criterion was unclear due to cognitive ability and/ or physical impairment, recommendation for inclusion was sought from the primary oncologist and/or clinical psychologist attached to the prospective participant's oncology service.

Participants were recruited from the New Zealand Child Cancer Registry (NZCCR). The NZCCR was established in 2000 to provide a complete database of all children diagnosed with cancer. Funding for the setting up and maintenance of the registry is provided through the National Paediatric Oncology Steering Group (POSG).

In using the NZCCR to identify participants it was recognised that while retrospective data had been included on cancer diagnoses prior to 2000 it was incomplete. Additional sources of recruitment were used, namely the Paediatric Haematology/Oncology service databases at both tertiary centres (Auckland and Christchurch). Advertisements were also taken out in the family publications of the two charitable organisations that support young people with cancer, namely the Child Cancer Foundation (CCF) and CanTeen NZ. A data file was established by identifying all childhood cancer survivors who met the selection criteria (Table 1).

Table 1. *Childhood cancer survivor characteristics -data sourced from the NZCCR and hospital demographic database.*

Variable	**Comments**
Name	Deleted after recruitment
Ethnicity	Primary ethnicity only
Gender	
Date of Birth	
NHI	Deleted after recruitment
Age at time of survey	Calculated from DOB for participation criteria
Residential address	Deleted after recruitment
Domicile Code	Measure of socioeconomic status –mapped to New Zealand Deprivation (NZ Dep) score1-10
Diagnosis	12 diagnostic groups/ sub groups drawn from the ICCD-3
Date of Diagnosis	Calculated time from completion of treatment
Date treatment complete	
Treatment modality	Surgery Chemotherapy Radiotherapy (site) Haemopoetic Stem Cell Transplant (check all that apply)

The data file was reviewed and updated using the inclusion criteria detailed previously and checked against the National Hospital Index (NHI) to confirm live status. Once finalized, a unique login code was generated for each entry on the database. To ensure anonymity, identifiers were removed from the database once the survey was completed and prior to data retrieval and analysis, these identifiers were: name, address and NHI. Three hundred and ninety nine eligible childhood cancer survivors who met all the criteria were identified and all were invited to participate. Given the relatively small number of adolescent survivors in New Zealand and the individual variables of disease type, treatment therapies, and late effects, a whole population approach was utilized.

For the majority of participants the setting for completing the survey was their home, if privacy was difficult earphones were offered by phoning the 0800 number. For those who didn't have access to a computer or internet or sufficient privacy to complete the survey, the local library, school health centre or library or internet café was used. Both CanTeen and Child Cancer Foundation, the charitable arms of child cancer services were engaged in assisting with access where needed. This differed significantly from the Youth'07 research setting of classrooms with a research team at hand.

3.5 Controls: New Zealand College Students aged 12years to 18years inclusive

The control population was the 9,107 students from 96 colleges throughout New Zealand who completed the questionnaire for the Youth'07 project in 2008. Inclusion criteria was that students had to be 18 years of age or younger with English language skills equivalent to year 6 (Adolescent Health Research Group, 2003b). Eligible year 9 to year 13 students were randomly selected and invited to participate. The final 9,107 students who took part represented 74% of those selected and 3.4% of the total New Zealand secondary school roll (Adolescent Health Research Group, 2008b).

The Youth'07 study was chosen as the control as it provided a unique opportunity to use recent, comparative data from a large normative cohort of 12 to 18 year old New Zealand students providing current and accurate information that is representative of young adolescents in New Zealand. Specific measures included had proven validity and reliability and built on the information gained from the Youth2000 survey providing two time points for comparison (Adolescent Health Research Group, 2008a). The survey was carried out in school venues including gymnasiums, classrooms or school Marae with enough space to ensure privacy using hand held computers. Headsets were provided for privacy as well as assisting those participants with reading difficulties. The questionnaire used the M-CASI audio-visual format with questions

answered by touch screen action with a stylus and response options also read out with headsets ensuring privacy.

3.6 Survey Method

The survey tool initially developed for the Youth 2000 and Youth'07 surveys used a multimedia computer assisted self-interviewing (M-CASI) survey and was designed as a youth-friendly tool. Hand-held computers with headsets were provided and questions displayed on the screen with a mouse click and point action required instead of keyboard use. The questions were in an audio-visual format. Computer assisted self-interview (CASI) methods have become more widely used, particularly with surveys involving adolescents and young adults. Watson et al. (2001) of the Youth2000 study piloted the survey tool in 1999 and found a high level of student enjoyment and acceptability with reported ease of use of the M-CASI tool and concluded that "M-CASI was an acceptable instrument for the administration of a youth health survey" (p. 520). In another study (Webb, Zimet, Fortenberry, & Blythe, 1999) comparing CASI data collection and a self-administered written questionnaire (SAQ) with adolescents no significant differences were found in reporting of information.

3.7 The Youth '07 questionnaire

The Youth'07 survey contained questions on important health and wellbeing outcomes as well as risk taking and protective behaviours. The main domains were: culture and ethnicity; home, families and school; general health; emotional wellbeing; substance use; sexual health; health-risk behaviours and community (Adolescent Health Research Group, 2008a). The Youth'07 questionnaire included a possible 622 questions, however because the branching design removed irrelevant questions, most students were reported as only answering half that number, with the average completion time reported as approximately 73mins (Adolescent Health Research Group, 2008b).

The questionnaire was also translated into Māori with the student able to move between Māori and English.

3.8 Modification of the questionnaire

In using this survey tool to describe the psychosocial wellbeing of childhood cancer survivors, the Youth'07 questionnaire was reviewed and those questions that were not pertinent to the research question were excluded. The decisions on the exclusions were informed by the findings from the literature review and the clinical expertise of the research team. In addition to answering the research question the aim was also to reduce the questionnaire to a manageable time to complete, as it was acknowledged that the participants would be completing it in their own home and their own time, not in a controlled school environment as was the setting for the Youth'07 survey. An additional consideration to the inclusion/ exclusion of questions were those questions of a sensitive nature that could pose an unacceptable risk to participants in an uncontrolled setting, this is addressed in further detail in the section on ethical considerations.

A focus group consisting of young adult child cancer survivors who were just outside the study age range (e.g.19 years of age or older) was formed to look at the design and delivery of the questionnaire including duration, recruitment methods, incentives and privacy concerns (Mayne, 2008). A completion time of, on average no more than half an hour was decided on based on feedback from the focus group, any longer was thought be a deterrent to completion rates. While it was important that the final questionnaire was kept to a reasonable length, several additional questions were included. These focused on the participants' knowledge of their cancer diagnosis, treatment modalities and time since diagnosis. This was to enable comparisons between the different disease groups, length of time from treatment, and where possible the intensity of treatment.

The final survey questionnaire was composed of sets of standardized closed-ended questions with response categories using a mix of forced choice, multi-choice and checklist responses and included a possible 250 questions if all branching questions were answered. The section of the survey questionnaire that contained the four standardized instruments that inform this thesis is included as a text document with summary headings for the reader's reference that weren't included in the online version (please see Appendix C on page 90). Once the questionnaire was completed and the audio files for the new questions added, the survey was tested with several young child cancer survivors who were not eligible to take part because they were outside the age range or still on treatment. They were asked to comment on clarity of the information sheet and instructions, problems encountered (i.e. access, freezing or crashing of the site, ease of use, length of time to complete) and how they felt after completing it. Feedback was positive and no changes were suggested by the group and no modifications were made. The framework for the study is detailed in figure 1.

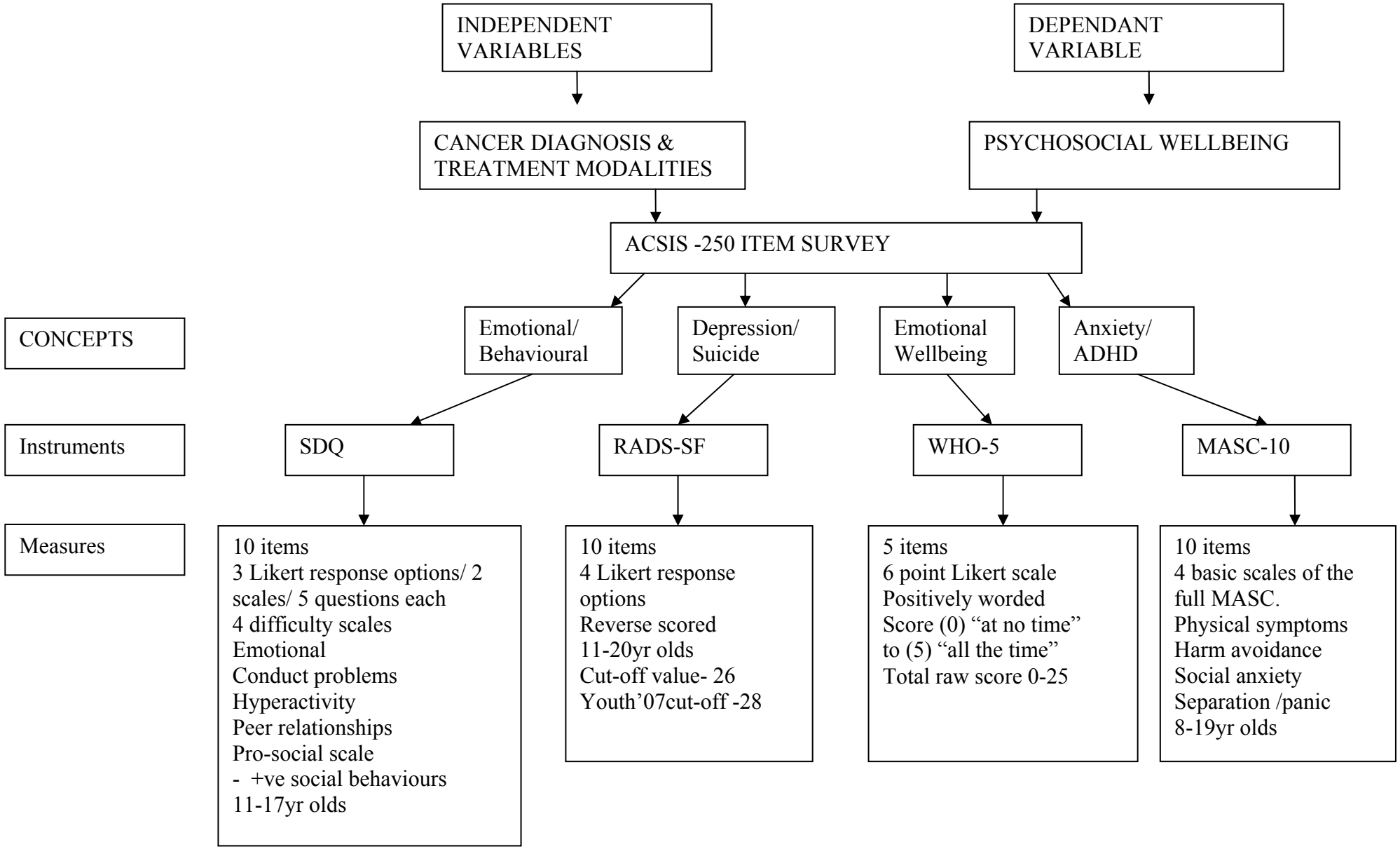


Figure 1. Study framework

3.9 Standardized measures of psychosocial wellbeing

Four standard outcome measures of psychosocial well-being and quality of life were included in the questionnaire, three from the Youth'07 and one from the Youth2000. These included the following:

- The World Health Organisation (Five) Wellbeing Index (WHO-5)
- The Reynolds Adolescent Depression Scale - short form (RADS2-SF)
- The Strengths and Difficulties Questionnaire (SDQ)
- The Multidimensional Anxiety Scale for Children - short form (MASC-10)

3.9.1 The WHO-Five Well-being Index

Who-5 was derived from a larger rating scale (WHO-10) developed for a World Health Organisation project in 1990 and is a measure of psychological wellbeing. It is a 5 item positively worded scale designed to assess the emotional wellbeing within the previous 2 weeks, covering positive mood, vitality and general interest and scored on a 6-point Likert scale from 0 (not present) to 5 (constantly present) (Bech, 1999; Bech et al., 2003). The raw score is calculated by totalling the figures of the five answers and ranges from 0 to 25, 0 representing worst possible and 25 representing best possible. The WHO-5 has been found to have a good internal consistency (Cronbach's $\alpha = 0.91$, (Lowe et al., 2004) A study to evaluate the psychometric properties of the WHO-5 in adolescents with Type 1 diabetes (de Wit, Pouwer, Gemke, Delemare-van de Waal, & Snoek, 2007) found good reliability and validity and concluded it was a suitable tool to use with adolescents in measuring the emotional wellbeing, allowing for comparison with healthy peers.

3.9.2 The Reynolds Adolescent Depression Scale-Short Form (RADS2-SF)

The Reynolds Adolescent Depression Scale is a self-report tool with proven reliability and validity that measures symptoms of depression in adolescence (Reynolds, 2002). It has a five factor structure that assesses generalized demoralization, despondency and worry, externalized somatocism, anhedonia, and self worth.

The RADS2-SF is a shorter version developed to provide a brief measure for the assessment of depression. For the Youth'07 survey the RADS2 –SF was examined and found to have psychometric properties similar to the full RADS version with acceptable reliability and validity for New Zealand adolescents across the major different ethnic groups (Milfont et al., 2008). The authors found the RADS2-SF had Cronbach's alpha of 0.88 and was strongly correlated (0.95) to the RADS. Therefore the RADS2-SF was an appropriate tool for replacing the full length version in the Youth'07 survey. The RADS2-SF cutoff of ≥ 26 for symptoms of significant depression was replaced by the Y'07 survey with a cutoff of ≥28. This was based on the analysis of Youth2000 data which showed a level of ≥28 best matched the cut-off level of the RADS and more closely matched the percentage identified with significant levels of depressive symptoms (Milfont et al.,(2008). The authors acknowledged the cutoff of 28 proposed could lead to a drop in sensitivity but was more appropriate in looking at time trends in a comparison between Youth2000 and Y'07. To provide an accurate comparison with the control group a cutoff of ≥28 was chosen for this study.

The RADS2-SF is the copyright of Psychological Assessment resources, Inc., Florida 2002 and permission was obtained for use in this survey.

3.9.3 The Strengths and Difficulties Questionnaire (SDQ)

The SDQ is a standardized tool that has been widely used and validated (Cronbach's a = 0.73). It is a measure of psychosocial functioning in 4 to 18 year olds and was developed as a parent- and self screen tool. SDQ has four difficulties scales:

emotional, conduct problems, hyperactivity-inattention and peer problems. The SDQ also included a positive strength scale, prosocial domain. The 25 items comprise 5 scales of 5 items with each of the scales 0-10, with the total difficulties score being the summary of all four difficulties scores excluding the prosocial scale (Goodman, 1997, 2001; Klasen et al., 2000). For this survey as with the Youth'07, the self-report SDQ with the 11-17 year age supplement was used.

3.9.4 The Multidimensional Anxiety Scale for Children –short form (MASC-10)

The MASC-10 is a one-dimensional, ten item measure and is the modified form of the 39-item, 4-point Likert self-report MASC scale. It measures four basic anxiety dimensions: physical symptoms, harm avoidance, social anxiety and separation anxiety in children aged 8 to 18 years (March et al., 1999). Comparison is made with a normative sample of the same gender and age range. Scores of >19 for females and >17 for males indicate significant anxiety.

In a study of youth with attention deficit hyperactivity disorder (ADHD), March and colleagues (1999) concluded that the MASC was a valid measure of anxiety in diverse populations. In a study evaluating the psychometric properties of the short form MASC-10, good scale reliability and validity was reported, though this was in a population of psychiatric inpatient youth (Osman et al., 2009).The MASC-10 was included in the Youth2000 survey but not in Youth'07 however in our experience in working with young survivors over the past four years, anxiety is a common problem and it was therefore considered important to include in this study.

Permission was obtained for the use of the MASC-10 in this study.

3.10 Data Collection Procedures

All eligible child cancer survivors were sent a pack inviting their participation that included a cover letter explaining the purpose of the study and intended outcomes

and an information sheet outlining the process for completing the survey with their anonymous login code. An entry form for a competition to win one of 5 iPods and a reply paid envelope was also included. For those young people under the age of 16 years, the letter was addressed to the parent/ caregiver with a request to pass the information to their child to complete if they agreed to their participation. As well as the contact details for the research team an 0800 number was set up that was diverted to my mobile phone for 24hour support if required. A follow-up reminder was sent to all participants at three to four weeks and three months after the initial contact. This had been decided in the planning stage as a review of the literature had identified that reminders as well as incentives improved response rates (Kaplowitz et al., 2004; Rosoff et al., 2005).

The data collection period was initially set for three months, however unanticipated problems occurred for some participants in accessing the survey. The access issues were more common with laptops. The cause was identified as being the high security settings of some computer software which blocked the site. This hadn't been evident during the testing of the survey, predominantly on hospital computers and laptops which allowed access. The web-based survey had been developed for the two Youth Health surveys where the hand-held internet tablets were part of an integrated system i.e. connected by a router to a laptop used as a server for the survey software programme. The same website development company set up and hosted the site for this survey and the difficulties with access were not foreseen.

In discussion with the hosting company it was felt that the best option was to advise those young people experiencing problems how to turn off the " pop-up blockers" and with the second mail out a slip was enclosed explaining the simple steps to bypass the security settings. This was successful for some but did not allow access for all. The web development company was ultimately able to refine the software

programme without destabilizing existing data, however this only occurred 3 months after the problems had been identified. The access issues were resolved entirely but we had lost the window of opportunity in engaging a percentage of participants some of whom did not retry entering the survey site. The final mail out was at three months after the initial contact and had been held back until the access issue was resolved. No further issues were reported following the final mail out. The final data collection period was from August 2009 to January 2010.

3.11 Data Analysis

Data analysis was conducted using JMP V5.1 (SAS Institute Inc software). All measures were scored using the standard criteria detailed for each measure. Comparisons between cancer survivor survey responders and non-responders were undertaken using the Chi-square test for categorical variables and the Mann Whitney U test for continuous variables (non normally distributed). Comparisons between the Youth'07 control group and the childhood cancer survivor cohort were undertaken by multivariate logistic regression using PROC SURVEYREQ in SAS. For these analyses, the psychosocial wellbeing measure of interest was incorporated as the outcome of a multivariate model which included as predictor co-variates: the study group (childhood cancer survivor or Youth'07), age, gender, ethnicity and NZDep06. The cancer survivor data were weighted (by ethnicity and NZDep06) to match the total childhood cancer survivor population, and the Youth'07 data were weighted and allowance made for the clustered sampling design as instructed by the providers of the Youth'07 data. Hence, the predictor effect (and associated p-value) of the study group co-variate (childhood cancer survivor vs. Youth'07) provides a comparison of the two groups, while controlling for differences in age, gender, ethnicity and NZDep06. This method allowed for the complex study design of the Youth'07 data, and enabled use of the entire data from both study groups. Because this study involved the testing of the psychosocial

wellbeing and numerous demographic and cancer variables, a suitable corrected threshold of statistical significance would be less than the conventional $p = .05$. Therefore all p-values are presented as actual values and a p value of $< .05$ was used only as an arbitrary indicator of significance difference. Internal consistency of the scales were tested by Cronbach's alpha coefficient. A level above .7 is recommended by Nunally (1978).

Due to the small number of responders (170) and the broad spread of cancer diagnosis, in answering the research questions, the groups were combined into three categories, leukaemia/ lymphoma, CNS tumours and all others. In comparing the characteristics of the childhood cancer survivors by treatment type, these were grouped into chemotherapy, radiation, Haemopoetic Stem Cell Transplant (HSCT) and surgery. It is important to note that cancer treatment often includes more than one of the treatment modalities and surgery particularly, is often used as an adjuvant rather than a stand alone therapy.

3.12 Ethical and Cultural Considerations

Health and Disability Multi-regional Ethical approval was granted by the Upper South B Regional Ethics Committee for the Health and Disability Ethics Committees in July 2009 for a period until December 2011 (Ethics Reference Number: URB/09/05/017). Prior to submission, through the locality assessment process, ethical approval was granted by the Auckland District Health Board Research Review Committee (ADHB-RRC) (Approval number: A+4391) and the Canterbury District Health Board Research Review Committee (CDHB-RRC). Support had been sought and granted by the Māori Research Review Committees (MRRC) for the District Health Boards.

In their granting of support the ADHB MRRC queried "whether it was ethical to only involve English speaking participants in a country that has three official languages

approved in Crown Health Agencies", they are English, Māori and sign. The omission of not offering the survey in Māori as had been done in the Youth2000 and Youth'07 surveys is acknowledged, but given the small number of potential participants and the costs involved, translation into Māori was not deemed achievable in this instance. Simon Denny, principle investigator for Youth'07, when asked the percentage of those completing the Youth2000 and Youth'07 surveys in Māori said "We weren't able to collect this information. Our sense from talking with the survey teams was that it was very small outside of the wharekura settings" (personal communication, February 22, 2010).

In response to an initial poor participation in the survey, the recruitment process was amended to include a phone call to check an invitation to participate had been received and the young person had the means to do so, i.e. had access to the internet, a computer, and privacy. An amendment was submitted to the Upper South B Regional Ethics Committee and approved in September 2009 to permit eligible participants to be contacted by a third party, specifically CanTeen and Child Cancer Foundation support staff.

3.13 Voluntary participation and informed consent

The information sheet clearly stated that participation was voluntary and all information would remain confidential. The process for consent of those under 16years of age was to send the information to the parent/ caregiver with a cover letter explaining the survey and requesting that it be given to their child if they agreed to their participation. Consent for those 16years of age and over was implicit in logging on to the questionnaire using a unique identifier code. The front screen was the consent page which required the participant to accept by ticking a box before being able to enter the site.

3.14 Confidentiality and right to privacy

A unique identifier code was generated from the web based programme and randomly allocated to ensure anonymity and confidentiality. All data entered by participants was immediately transferred to the online spreadsheets and no individual files or hardcopies were generated. From the commencement of the study, data files were kept in a secure, password protected database accessible only by the research team. Once the data collection was completed all identifiers, for example, name, contact details and NHI number on the database were removed and deleted. At completion of the study all data will be stored for 14 years as anonymous data in a password protected database.

3.15 Minimisation of harm

There was an acknowledged risk of harm in the nature of the research questions on sexuality, social connectedness, risk taking behaviours, and mood alteration. In addition, completion of the questionnaire may not have been conducted in a secure or protective environment with appropriate counselling support at hand. As the Youth'07 survey was conducted in schools during class time, counsellors were on hand if required. Minimisation of harm was carefully considered and questions in the Youth'07 questionnaire that could cause distress and were not absolutely necessary to our research were removed. At the start of sensitive sections of the questionnaire, an information box was displayed with the words "*if these questions have been upsetting for you and you wish to talk with someone you can phone the 0800 free call number on your letter, Youthline 0800 376633, free txt 234 or 0800 WHATSUP (0800 942 87 87)".* All questions that had potential harm were branching questions with an opt-out box. An 0800 number was set up with 24 access and as well as being clearly identifiable on the information sheet, was also displayed on screen at the start and finish of the questionnaire, with a recommendation to make contact if there is anything within the

questionnaire that was upsetting in any manner. By completion of data collection, no calls had been made to the free call number due to concern or distress at any of the questions.

3.16 Māori Participation in Research

The Treaty of Waitangi is the founding document of New Zealand and acknowledgement of the principles of the Treaty i.e. partnership, participation and protection is fundamental to any research involving Māori. The Health Research Council of New Zealand states that there is a need to increase Māori participation in health research to reduce health disparities. Consultation is important to ensure the research is acceptable, appropriate and outcomes contribute to improving Māori health and wellbeing as much as possible (Health Research Council of New Zealand, 2008).

As per ADHB research guidelines, ethical approval was granted by the Māori Research Review Committees (MRRC) prior to submitting the proposal to the Health and Disability Ethics Committee. Participation and protection were upheld through Māori childhood cancer survivors being offered equal opportunity to participate and protection through the anonymous data collection process and secure storage of data. In acknowledgement of the importance of consultation and partnership with Māori in this survey, I met with the Māori advisory team within the Auckland District Health and Canterbury District Health Boards. The research proposal and aims were presented and discussed and the questionnaire was made available for review prior to the final decision on question inclusion/ exclusion. Support was granted following both meetings. The principal of partnership was integral to the Youth2000 and Youth'07 survey development which had a Māori advisory group established for both surveys and Māori health researchers as co-investigators. The changes made to the Youth'07 questionnaire were minimal, which provided a level of confidence in the appropriateness and acceptability to Māori for the advisory groups consulted, the ethics

committees and the research team. In addition, I met with representatives of Asian and Pacific Peoples working within ADHB to explain the aims and projected outcomes of the survey, at both these meetings there were no specific concerns or requests made and support was offered.

In disseminating the findings of this research all groups consulted were offered access to the results in a format requested by them, this may take the form of Hui, written report or a presentation.

This chapter has described the methodology and methods of this case-controlled study of the psychosocial wellbeing of childhood cancer survivors compared with a group of their peers. Ethical and cultural considerations have been discussed. The following chapter presents the results of this study.

CHAPTER 4
RESULTS

4.1 Introduction

This chapter presents the findings that answer the following research questions:

a) Describe the self-reported psychosocial wellbeing of childhood cancer survivors as determined by standardized measures, WHO-5, RADS.2-SF, SDQ and MASC-10.

b) Test whether demographic or cancer characteristics are associated with the psychosocial wellbeing of adolescent survivors.

c) Compare the psychosocial wellbeing of childhood cancer survivors to a normative control group of New Zealand youth aged between 12- 18 years.

The following section details characteristics of the study sample and compares the socio-demographic and cancer characteristics of those who completed the survey (respondents) and those who did not (non-respondents). The subsequent sections address the research questions listed above.

4.1.1 Characteristics of the study sample

A total of 396 childhood cancer survivors aged between 12 and 18 years and at least 2 years from the end of treatment were invited to complete the survey, 170 (43%) responded. Survey respondents were similar to non-respondents in gender and age at time of the survey (Table 2). While not statistically significant, a lower proportion of respondents were Māori compared to non responders (12% vs.20%), and fewer living in low socio-economic areas (high deprivation) responded (28% v. 40 %).

In comparing respondents with non-respondents by cancer diagnosis, the largest group for both had been diagnosed with leukaemia (38% vs. 42%) followed by CNS

disease (13% vs. 19%) and lymphoma (11% vs. 12%). Survivors with a diagnosis of a renal tumour were the one diagnostic group with a higher response than non-response rate (11% vs. 7%) but the total number was small. In general there were no differences between the groups based on treatment type (Table 2). Respondents were slightly younger at age of cancer diagnosis (5.6 years) compared to non-respondents (6.5 years, $p = .02$).

In summary, respondents tended to be European, approximately 15 years of age, from middle to higher socio-economic backgrounds with a diagnosis of leukaemia that was treated primarily with chemotherapy. There were potentially important differences between respondents and non-respondents in ethnicity and socioeconomic status. Therefore, all analyses in the following sections have been weighted for ethnicity and socioeconomic status (New Zealand Deprivation Index 06).

Table 2. *Socio-demographic and cancer characteristics of the childhood cancer survivor cohort and sample*

	NZCCS Cohort (n=396)		Non-respondents (n=226)		Respondents (n=170)		*P*
Socio-demographic characteristics	**n**	**%**	**n**	**%**	**n**	**%**	
Gender							
Female	163	41	90	40	73	43	0.53
Male	233	59	136	60	97	57	
Ethnicity							
European	269	68	146	65	123	72	0.07
Māori	66	17	45	20	21	12	
Pacific	38	10	25	11	13	8	
Asian	23	6	10	4	13	8	
Age							
Median	15.4		15.3		15.4		0.8
NZ Deprivation Index 06							
Low	126	32	66	29	60	35	0.05
Medium	131	33	69	31	62	36	
High	138	35	90	40	48	28	
Cancer Characteristics							
Diagnosis- ICCD-3							
l Leukaemia	159	40	92	42	65	38	0.02
ll Lymphoma	47	12	28	12	19	11	
lll CNS	64	16	42	19	22	13	
lV Neuroblastoma	14	4	8	4	6	4	
V Retinoblastoma	8	2	1	0	7	4	
Vl Renal Tumours	32	9	16	7	18	11	
Vll Hepatic Tumours	9	2	2	1	7	4	
VIII Bone Tumours	22	6	11	5	11	6	
lX Soft Tissue sarcoma	20	5	15	7	5	3	
X Germ Cell/ Gonadal Tumours	8	2	2	1	6	4	
Xl Epithelial /Melanoma NOS	10	3	7	3	3	2	
Xll NOS malignant neoplasm	1	0	0	0	1	1	
Cancer diagnosis grouped							
Leukaemia/Lymphoma	206	52	122	54	84	49	0.06
CNS	64	16	42	19	22	13	
Other	126	32	62	27	64	38	
Age at diagnosis							
Median	5.3		5.8		4.8		0.02

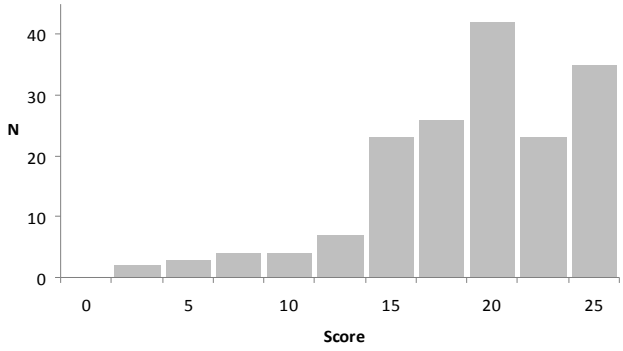
	NZCCS Cohort (n=396)		Non-respondents (n=226)		Respondents (n=170)		*P*
Age range			0 - 16		0 - 15		
Time since diagnosis							
Median	9.4		9.0		10.0		0.02
Surgery							
No	188	47	100	44	88	52	0.14
Yes	208	53	126	54	82	48	
Chemotherapy							
No	45	11	24	11	21	12	0.59
Yes	351	89	202	89	149	88	
Radiation							
No	295	74	165	73	130	76	0.43
Yes	101	26	61	27	40	24	
HSCT							
No	357	90	206	91	151	89	0.44
Yes	39	10	20	9	19	11	

p values test for differences between respondents and non-respondents. Chi-square was used for categorical variables and Mann Whitney U for continuous variables (non normally distributed).

4.2 Research Question 1. Describe the Psychosocial Wellbeing of Childhood Cancer Survivors

4.2.1 Wellbeing of Childhood Cancer Survivors (WHO-5)

One hundred and sixty nine childhood cancer survivors (CCS) answered the five items that comprise the WHO-5 wellbeing scale (Cronbach's $\alpha = 0.87$). The WHO-5 score ranged from a minimum of 2 to a maximum of 25 (possible range 0 to 25). As figure 2 shows, the distribution was not normally distributed, with a negative skew (Skew = -1.0, Kurtosis = 1.17, Kolmogorov-Smirnov = 0.10, p = .05). The mean score was 18.25 and the median was 19. The majority (89%) of respondents reported good to excellent wellbeing. Eighteen respondents (11%) scored below the threshold of 13, indicating poor wellbeing.



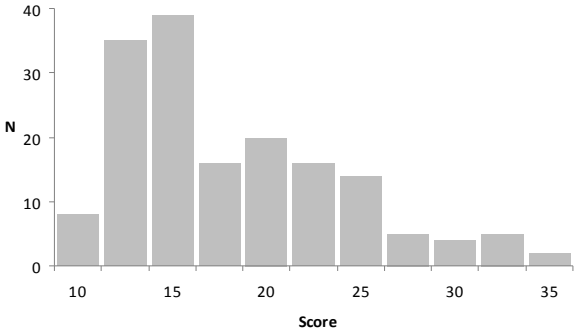
Poor = 0-12, Good = 13-17, Very Good = 18-21, Excellent = 22-25

Figure 2. Histogram of the WHO-5 scores

(Weighted for ethnicity and SES)

4.2.2 Depression in Childhood Cancer Survivors (RADS2-SF)

One hundred and sixty four CCS answered the RADS.2 SF (Cronbach's α = 0.89). The RADS2-SF raw score ranged from a minimum of 10 to a maximum of 34 (possible range 10 to 40). The distribution was positively skewed (Figure 3) and not normally distributed (Skew = +0.90, Kurtosis = 0.13, Kolmogorov-Smirnov = 16, p = .05). The mean score was 17.3 and the median was 15.5. The majority of respondents (91%) were within the normal range. Ten respondents (9%) scored greater or equal to 28, consistent with depression (Milfont et al., 2008).



Normal <28, Mod-severe depression ≥28

Figure 3. Histogram of RADS2-SF Scores

(Weighted for ethnicity and SES)

4.2.3 Strengths and Difficulties in Childhood Cancer Survivors (SDQ)

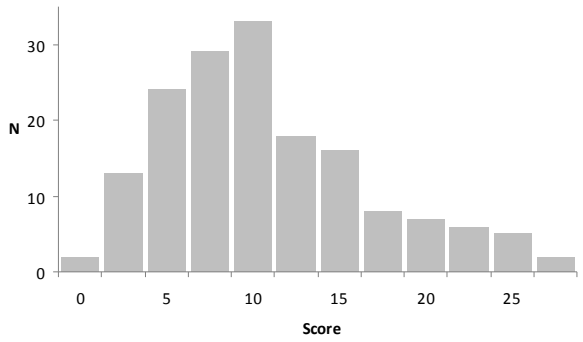
One hundred and sixty three CCS completed the 5-scale SDQ. Descriptive statistics for the scales are provided in Table 3. Three of the scales (emotional symptoms, hyperactivity and pro-social scale) had Cronbach's alpha greater than 0.7, two scales (conduct problems and peer problems) had Cronbach's alpha of 0.64-0.65. The proportion of CCS scoring in the borderline /abnormal category ranged from 13.5% (emotional difficulties) to 20.8% (hyperactivity).

Table 3.*Comparisons across the five SDQ scales.*

Measure	**α**	**Min (0)**	**Max (10)**	**Median**	**Skew**	**Kurtosis**	**K-S**	**Borderline/ abnormal** n (%)
Difficulties Scales								
Emotional Symptoms	0.71	0	9	2.0	0.73	-0.15	0.17 <.05	22 (13.5)
Conduct Problems	0.64	0	6	1.0	1.06	0.28	0.23 <.05	24 (14.8)
Hyperactivity	0.77	0	10	3.0	0.50	-0.26	0.12 <.05	34 (20.8)
Peer Problems	0.65	0	9	2.0	1.10	1.16	0.16 <.05	25 (16)
Strengths Scale								
Prosocial (reverse score)	0.73	1	10	8.0	-1.04	0.76	0.20 <.05	22 (13.5)

Note: All scales have 5 items; α=Cronbach's Alpha; K-S=Kolmogorov-Smirnov p-value

Figure 4 presents the total difficulties score (Cronbach's α = 0.84) which is the summary of all four difficulty scales. One hundred and thirty four (82.2%) CCS scored in the normal range with 29 (17.8%) scoring in the abnormal/ borderline range.



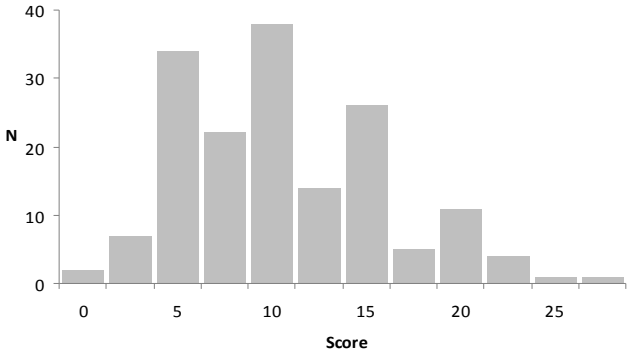
Normal range 0-15, borderline 16-19, abnormal range 20-40.

Figure 4. Histogram of SDQ Total Difficulties Scores

(Weighted for ethnicity and SES)

4.2.4 Anxiety in Childhood Cancer Survivors (MASC-10)

One hundred and sixty five childhood cancer survivors completed the 10 item MASC-10 anxiety scale (Cronbach's α = 0.76) with scores ranging between 7 and 20 (possible range 0 to 25). The distribution was positively skewed (Figure 6; skew = +.51, Kurtosis – 0.16, Kolmogorov-Smirnov = 0.09, p = .05). The mean score was 11.95 and the median was 11.50. One hundred and fifty three (92.7%) scored within the normal range with 12 (7.3%) reporting moderate to severe anxiety.



Score ≥ 17- moderate to severe anxiety

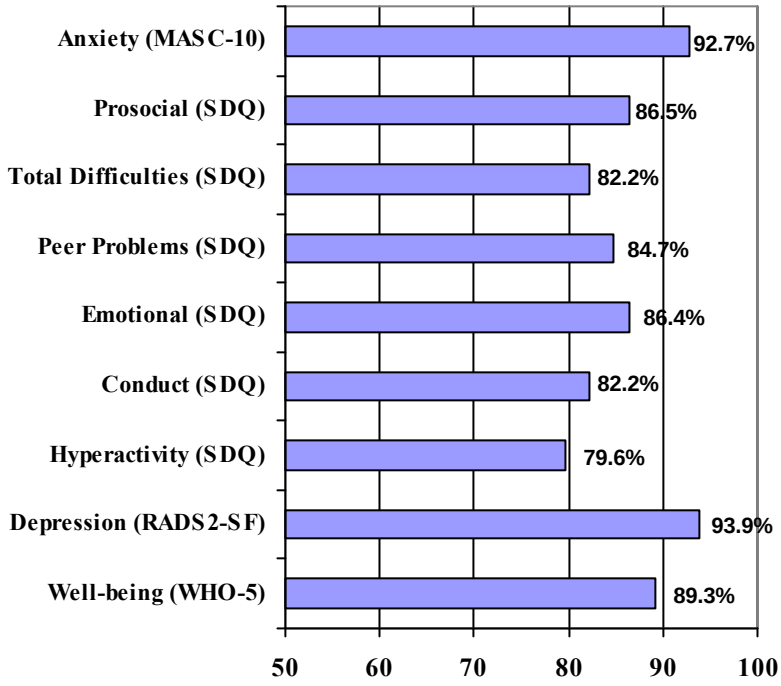
Figure 5. Histogram of MASC-10 scores

(Weighted for ethnicity and SES)

In summary, the majority of childhood cancer survivors scored within the normal range across all four instruments (Figure 6). The least reported problems were with anxiety (7.3%), depression (9%) and poor wellbeing (11%). The highest reported

problems were with SDQ total difficulties (17.8%) which included conduct, emotional, hyperactivity and peer problems. Total difficulties were reported by one in six CCS whereas anxiety was reported by only one in fourteen.

Figure 6. Percentage of Childhood Cancer Survivors scoring within the normal range across all measures.



4.3 Research Question 2. Test whether demographic or cancer characteristics are associated with the psychosocial wellbeing of adolescent childhood cancer survivors.

The reader is cautioned in interpreting the following data due to the small samples sizes for many of the reporting categories. The data are reported to inform future research and hypothesis generation.

4.3.1 Wellbeing of Childhood Cancer Survivors by Demographic and Cancer Characteristics (WHO-5)

Older age was associated with poorer wellbeing ($p = .003$) as was older age at time of cancer diagnosis ($p = .007$). There were no other demographic or cancer characteristics associated with wellbeing outcomes (Table 4).

Table 4. *WHO-5 score by demographic and cancer characteristics*

		Poor		Good		Very Good		Excellent		
Socio-demographic characteristics		n	%	n	%	n	%	n	%	p*
Gender	F	10	13.5	18	24.3	24	32.4	22	29.7	0.47
	M	8	8.2	32	33.0	29	29.9	28	28.9	
Ethnicity	Asian	3	33.3	2	22.2	2	22.2	2	22.2	0.29
	Euro	12	10.4	36	31.3	38	33.0	29	25.2	
	Maori	2	7.1	9	32.1	5	17.9	12	42.9	
	Pacific	1	6.3	3	18.8	6	37.5	6	37.5	
Age	Mean	16.5		16.0		15.1		14.9		0.003
	Median	16.6		16.1		15.0		14.3		
	Range	13.6 to 19.0		12.2 to 19.1		12.0 to 19.0		12.0 to 18.9		
NZDep06	Mean	5.2		5.5		5.8		5.7		0.90
	Median	5		5		5		5.5		
	Range	1 to 10		1 to 10		1 to 10		1 to 10		
Cancer Characteristics										
Diagnosis	CNS (intracranial & intraspinal)	3	15.0	4	20.0	8	40.0	5	25.0	0.86
	Leukaemia & Lymphoma	8	9.0	29	32.6	26	29.2	26	29.2	
	All other	7	11.9	16	27.1	18	30.5	18	30.5	
Age at diagnosis	Mean	7.4		6.6		4.5		5.3		0.007
	Median	6.5		6.2		3.9		4.9		
	Range	1.0 to 14.3		0.4 to 14.9		0.2 to 10.8		0.0 to 11.8		
Time since diagnosis	Mean	9.1		9.4		10.6		9.7		0.21
	Median	10.1		10		11.2		9.5		
	Range	3.8 to 16.4		2.5 to 14.7		3.6 to 16.0		3.3 to 17.3		
Surgery	N	11	12.8	27	31.4	23	26.7	25	29.1	0.62
	Y	7	8.4	23	27.7	29	34.9	24	28.9	
Chemotherapy	N	1	5.0	7	35.0	8	40.0	4	20.0	0.49
	Y	17	11.4	43	28.9	44	29.5	45	30.2	
RT	N	14	11.1	39	31.0	38	30.2	35	27.8	0.81
	Y	4	9.5	10	23.8	14	33.3	14	33.3	
HSCT	N	15	10.1	46	30.9	48	32.2	40	26.8	0.18
	Y	3	14.3	3	14.3	5	23.8	10	47.6	

* Pearson's Chi-Square was used for categorical variables and t-tests for continuous variables.

4.3.2 Depression in Childhood Cancer Survivors by demographic and cancer characteristics (RADS2-SF)

There were no differences in reported RADS2-SF scores by demographic or cancer characteristics (Table 5).

Table 5. *RADS2-SF by demographic and cancer characteristics*

		Mod-severe Depression		Normal range		
Socio-demographic characteristics		n	%	n	%	p*
Gender	F	4	5.7	66	94.3	0.82
	M	6	6.4	88	93.6	
Ethnicity	Asian	1	10.0	9	90.0	0.67
	Euro	8	7.1	104	92.9	
	Maori	1	3.8	25	96.2	
	Pacific	0	0.0	16	100.0	
Age	Mean	16.0		15.4		0.39
	Median	16.3		15.6		
	Range	13.1 to 19.0		12.0 to 19.1		
NZDep06	Mean	6.0		5.6		0.72
	Median	6		5		
	Range	4 to 8		1 to 10		
Cancer Characteristics						
Diagnosis	CNS (intracranial & intraspinal)	1	5.0	19	95.0	0.78
	Leukaemia & Lymphoma	5	5.7	82	94.3	
	All other	4	7.1	52	92.9	
Age at diagnosis	Mean	6.7		5.6		0.39
	Median	7.1		4.8		
	Range	1.0 to 14.3		0.0 to 14.9		
Time since diagnosis	Mean	9.3		9.8		0.64
	Median	10.6		10.1		
	Range	3.8 to 14.0		2.5 to 17.3		
Surgery	N	6	7.1	78	92.9	0.77
	Y	4	5.0	76	95.0	
Chemotherapy	N	1	5.0	19	95.0	0.76
	Y	9	6.2	136	93.8	

		Mod-severe Depression		Normal range		
RT	N	9	7.3	114	92.7	0.22
	Y	1	2.4	41	97.6	
HSCT	N	9	6.3	134	93.7	0.85
	Y	1	4.8	20	95.2	

* Pearson's Chi-Square was used for categorical variables and t-test for continuous variables.

4.3.3 Strengths and difficulties of Childhood Cancer Survivors by demographic and cancer characteristics (SDQ)

Difficulty with conduct problems was associated with lower socioeconomic status as recorded by the NZDep06 (p = .04). There were no other demographic or cancer characteristics associated with conduct problems (Table 6).

Table 6. *Conduct Problems by demographic and cancer characteristics*

		Abnormal/ Borderline		Normal		
Socio-demographic characteristics		n	%	n	%	p*
Gender	F	9	12.7	62	87.3	0.46
	M	15	16.5	76	83.5	
Ethnicity	Asian	0	0.0	9	100.0	0.20
	Euro	16	14.0	98	86.0	
	Maori	6	25.0	18	75.0	
	Pacific	1	6.7	14	93.3	
Age	Mean	15.3		15.5		0.53
	Median	14.8		15.7		
	Range	12.1 to 19.0		12.0 to 19.1		
NZDep06	Mean	6.7		5.4		0.04
	Median	7		5		
	Range	1 to 10		1 to 10		
Cancer Characteristics						
Diagnosis	CNS (intracranial & intraspinal)	4	19.0	17	81.0	0.88
	Leukaemia & Lymphoma	11	13.3	72	86.7	
	All other	9	15.5	49	84.5	
Age at diagnosis	Mean	5.6		5.6		0.99
	Median	5.6		4.8		
	Range	0.6 to 14.3		0.0 to 14.9		

		Abnormal/ Borderline		Normal		
Time since diagnosis	Mean	9.6		9.9		0.71
	Median	10.1		10.3		
	Range	3.8 to 14.7		2.5 to 17.3		
Surgery	N	12	15.0	68	85.0	0.84
	Y	12	14.5	71	85.5	
Chemotherapy	N	3	15.0	17	85.0	0.98
	Y	21	14.7	122	85.3	
RT	N	18	14.6	105	85.4	0.91
	Y	6	15.0	34	85.0	
HSCT	N	20	14.0	123	86.0	0.53
	Y	4	20.0	16	80.0	

*Pearson's Chi-Square used for categorical variables and t-tests for continuous variables

Difficulty with peer problems was associated with cancer diagnosis (Table 7). Among those with a CNS diagnosis, 36.4% scored in the abnormal/ borderline range compared to 12% and 12.1% for the leukaemia/ lymphoma and the other diagnosis groups respectively (p = .02).

Table 7. *Peer Problems by demographic and cancer characteristics*

		Abnormal/ Borderline		Normal		
Socio-demographic characteristics		n	%	n	%	p*
Gender	F	12	16.9	59	83.1	0.55
	M	13	14.1	79	85.9	
Ethnicity	Asian	1	11.1	8	88.9	
	Euro	19	16.7	95	83.3	0.26
	Maori	5	20.8	19	79.2	
	Pacific	0	0.0	15	100.0	
Age	Mean	15.6		15.5		0.70
	Median	16		15.5		
	Range	12.1 to 19.0		12.0 to 19.1		
NZDep06	Mean	5.8		5.5		0.67
	Median	5		5		
	Range	1 to 9		1 to 10		

Cancer characteristics						
Diagnosis	CNS (intracranial & intraspinal)	8	36.4	14	63.6	0.02
	Leukaemia & Lymphoma	10	12.0	73	88.0	
	All other	7	12.1	51	87.9	
Age at diagnosis	Mean	5.4		5.7		0.75
	Median	4.7		4.8		
	Range	0.2 to 12.8		0.0 to 14.9		
Time since diagnosis	Mean	10.2		9.8		0.56
	Median	11.2		10.1		
	Range	3.6 to 15.6		2.5 to 17.3		
Surgery	N	11	13.8	69	86.3	0.58
	Y	14	16.9	69	83.1	
Chemotherapy	N	4	20.0	16	80.0	0.70
	Y	21	14.7	122	85.3	
RT	N	16	12.9	108	87.1	0.11
	Y	9	23.1	30	76.9	
HSCT	N	21	14.7	122	85.3	0.58
	Y	4	20.0	16	80.0	

* Pearson's Chi-Square used for categorical variables and t-tests for continuous variables

Difficulty with emotional symptoms was not associated with any demographic or cancer characteristic (Table 8). Of note, CCS with a CNS diagnosis reported more difficulty with emotional symptoms (28.6%) compared to those with leukaemia/ lymphoma (12%) or other diagnosis (10%), though the differences were not statistically significant.

Table 8. *Emotional symptoms by demographic and cancer characteristics*

		Abnormal/ Borderline		Normal		
Socio-demographic characteristics		n	%	n	%	p*
Gender	F	12	16.9	59	83.1	0.31
	M	10	11.0	81	89.0	
Ethnicity	Asian	2	20.0	8	80.0	0.88
	Euro	17	14.9	97	85.1	
	Maori	3	12.0	22	88.0	
	Pacific	1	6.7	14	93.3	
Age	Mean	15.4		15.5		0.86
	Median	15.5		15.7		
	Range	12.2 to 19.0		12.0 to 19.1		
NZDep06	Mean	5.5		5.6		0.92
	Median	5		5		
	Range	1 to 10		1 to 10		
Cancer Characteristics						
Diagnosis	CNS (intracranial & intraspinal)	6	28.6	15	71.4	0.07
	Leukaemia & Lymphoma	10	12.0	73	88.0	
	All other	6	10.3	52	89.7	
Age at diagnosis	Mean	6.2		5.5		0.47
	Median	6.9		4.8		
	Range	1.0 to 14.9		0.0 to 14.7		
Time since diagnosis	Mean	9.2		9.9		0.36
	Median	10.1		10.1		
	Range	3.6 to 16.4		2.5 to 17.3		
Surgery	N	13	16.3	67	83.8	0.37
	Y	9	11.0	73	89.0	
Chemotherapy	N	1	5.0	19	95.0	0.21
	Y	21	14.8	121	85.2	
RT	N	18	14.6	105	85.4	0.64
	Y	4	10.3	35	89.7	
HSCT	N	18	12.6	125	87.4	0.36
	Y	4	21.1	15	78.9	

* Pearson's Chi-Square used for categorical variables and t-tests for continuous variables

Difficulty with hyperactivity was associated with time since diagnosis (Table 9). Children with more recent diagnoses (8.8 years ago vs. 10.1 years ago) were more likely to have scores indicative of hyperactivity. Hyperactivity difficulties were similar for males (22%) and females (19%).

Table 9. *Hyperactivity by demographic and cancer characteristics*

		Abnormal/ Borderline		Normal		
Socio-demographic characteristics		n	%	n	%	p*
Gender	F	14	19.4	58	80.6	0.71
	M	20	22.0	71	78.0	
Ethnicity	Asian	0	0.0	9	100.0	0.23
	Euro	26	22.8	88	77.2	
	Maori	6	24.0	19	76.0	
	Pacific	1	6.7	14	93.3	
Age	Mean	15.5		15.5		0.96
	Median	15.6		15.7		
	Range	12.1 to 19.0		12.0 to 19.1		
NZDep06	Mean	5.5		5.6		0.82
	Median	5.5		5		
	Range	1 to 10		1 to 10		
Cancer characteristics						
Diagnosis	CNS (intracranial & intraspinal)	5	23.8	16	76.2	0.87
	Leukaemia & Lymphoma	18	21.4	66	78.6	
	All other	11	19.0	47	81.0	
Age at diagnosis	Mean	6.6		5.4		0.09
	Median	5.1		4.8		
	Range	0.6 to 14.9		0.0 to 14.3		
Time since diagnosis	Mean	8.8		10.1		0.05
	Median	9.9		10.4		
	Range	2.5 to 13.9		2.5 to 17.3		
Surgery	N	16	20.0	64	80.0	0.82
	Y	18	21.7	65	78.3	
Chemotherapy	N	4	20.0	16	80.0	0.96
	Y	29	20.4	113	79.6	
RT	N	29	23.6	94	76.4	0.06
	Y	4	10.3	35	89.7	
HSCT	N	32	22.4	111	77.6	0.08
	Y	1	5.3	18	94.7	

* Pearson's Chi-Square used for categorical variables and t-test for continuous variables

There were no differences in reported total difficulties by any demographic or cancer characteristics (Table 10).

Table 10. *Total difficulties by demographic and cancer characteristics*

		Abnormal/Borderline		Normal		
Socio-demographic characteristics		n	%	n	%	p*
Gender	F	12	16.9	59	83.1	0.87
	M	17	18.5	75	81.5	
Ethnicity	Asian	0	0.0	9	100.0	0.08
	Euro	19	16.8	94	83.2	
	Maori	8	33.3	16	66.7	
	Pacific	1	6.7	14	93.3	
Age	Mean	15.3		15.5		0.58
	Median	15.4		15.7		
	Range	12.1 to 19.0		12.0 to 19.1		
NZDep06	Mean	6.1		5.4		0.29
	Median	5.5		5		
	Range	1 to 10		1 to 10		
Cancer Characteristics						
Diagnosis	CNS (intracranial & intraspinal)	7	31.8	15	68.2	0.12
	Leukaemia & Lymphoma	16	19.0	68	81.0	
	All other	7	12.1	51	87.9	
Age at diagnosis	Mean	6,0		5.6		0.61
	Median	5.1		4.8		
	Range	0.6 to 14.9		0.0 to 14.7		
Time since diagnosis	Mean	9.3		10.0		0.37
	Median	10.1		10.2		
	Range	3.6 to 14.7		2.5 to 17.3		
Surgery	N	17	21.3	63	78.8	0.20
	Y	12	14.5	71	85.5	
Chemotherapy	N	3	15.0	17	85.0	0.69
	Y	26	18.2	117	81.8	
RT	N	23	18.5	101	81.5	0.74
	Y	6	15.4	33	84.6	
HSCT	N	25	17.5	118	82.5	0.82
	Y	4	20.0	16	80.0	

* Pearson's Chi-Square used for categorical variables and t-tests for continuous variables

For the prosocial scale, younger age at survey was linked to poorer social behaviour (mean age =14.5years vs. 15.6years, p = .0007). Males reported higher abnormal/ borderline social behaviour than females (16.5% vs. 9.7%) (Table 11).

Table 11. *Prosocial Scale by demographic and cancer characteristics*

		Abnormal/ Borderline		Normal		
Socio-demographic characteristics		n	%	n	%	p*
Gender	F	7	9.7	65	90.3	0.18
	M	15	16.5	76	83.5	
Ethnicity	Asian	0	0.0	9	100.0	0.19
	Euro	17	14.9	97	85.1	
	Maori	5	20.0	20	80.0	
	Pacific	0	0.0	15	100.0	
Age	Mean	14.5		15.6		0.007
	Median	14		15.9		
	Range	12.2 to 16.9		12.0 to 19.1		
NZDep06	Mean	5.7		5.5		0.80
	Median	6		5		
	Range	1 to 10		1 to 10		
Cancer Characteristics						
Diagnosis	CNS (intracranial & intraspinal)	2	9.5	19	90.5	0.42
	Leukaemia & Lymphoma	14	16.9	69	83.1	
	All other	6	10.3	52	89.7	
Age at diagnosis	Mean	4.3		5.8		0.08
	Median	4.5		4.8		
	Range	0.2 to 9.3		0.0 to 14.9		
Time since diagnosis	Mean	10.1		9.8		0.67
	Median	10		10.1		
	Range	4.7 to 15.6		2.5 to 17.3		
Surgery	N	12	15.0	68	85.0	0.55
	Y	10	12.0	73	88.0	
Chemotherapy	N	4	20.0	16	80.0	0.35
	Y	18	12.6	125	87.4	
RT	N	20	16.3	103	83.7	0.08
	Y	2	5.1	37	94.9	
HSCT	N	21	14.7	122	85.3	0.28
	Y	1	5.3	18	94.7	

* Pearson's Chi-Square used for categorical variables and t-test for continuous variables

4.3.4 Anxiety in Childhood Cancer Survivors by demographic and cancer characteristics (MASC-10)

As illustrated by Table 12, there were no statistically significant differences in MASC-10 scores by demographic or cancer characteristics.

Table 12. *MASC-10 by demographic and cancer characteristics*

		Significant Anxiety		Not Sign Anxiety		
Socio-demographic characteristics		n	%	n	%	p*
Gender	F	5	7.0	66	93.0	0.81
	M	7	7.4	87	92.6	
Ethnicity	Asian	0	0.0	10	100.0	0.47
	Euro	8	7.1	105	92.9	
	Maori	2	7.7	24	92.3	
	Pacific	3	17.6	14	82.4	
Age	Mean	15.2		15.5		0.62
	Median	14.7		15.8		
	Range	13.1 to 18.9		12.0 to 19.1		
NZDep06	Mean	6.2		5.6		0.47
	Median	5.5		5		
	Range	2 to 10		1 to 10		
Cancer Characteristics						
Diagnosis	CNS (intracranial & intraspinal)	3	14.3	18	85.7	0.45
	Leukaemia & Lymphoma	5	5.7	82	94.3	
	All other	5	8.6	53	91.4	
Age at diagnosis	Mean	6.2		5.6		0.62
	Median	5.8		4.8		
	Range	1.0 to 12.1		0.0 to 14.9		
Time since diagnosis	Mean	9		9.9		0.40
	Median	9.9		10.1		
	Range	5.0 to 13.8		2.5 to 17.3		
Surgery	N	6	7.2	77	92.8	0.87
	Y	6	7.3	76	92.7	
Chemotherapy	N	2	10.0	18	90.0	0.76
	Y	10	6.9	135	93.1	
RT	N	9	7.3	115	92.7	0.84
	Y	3	7.3	38	92.7	
HSCT	N	12	8.3	132	91.7	0.17
	Y	0	0.0	21	100.0	

*Pearson's Chi-Square used for categorical variable and t-tests for continuous variables

In summary, older age was associated with poorer reported wellbeing. Lower socioeconomic status was associated with greater conduct problems but not any other scale. Of note, Māori reported the most difficulties with four of the SDQ subscales and while this did not reach statistical significance and the numbers were low it may be clinically important. Of the cancer characteristics which included diagnosis groups and treatment types, older age at cancer diagnosis was associated with poorer wellbeing. The CNS diagnosis group reported the greatest number of difficulties with the emotional and peer problem scales of the SDQ but not on any other measure. While not statistically significant males reported slightly more conduct problems than females (16.5% vs. 12.7%) and higher abnormal/ borderline social difficulties (16.5% vs. 9.7%), with females reporting increased difficulties on the emotional scale (16.9% vs. 11%). There were no gender differences in reported anxiety or depression. There were no other demographic or cancer characteristics that were significantly associated with adverse wellbeing.

4.4 Research Question 3. Compare the psychosocial wellbeing of childhood cancer survivors to a normative control group of NZ youth aged 12 to 18 years.

The gender distribution within the childhood cancer survivors and the normative control group of NZ youth was similar. Males accounted for 57% of CCS and 54% of Youth'07. Mean age at time of survey was 15 years for both groups. Europeans were more highly represented in the CCS group (68.2% vs. 52.6%) with fewer Asians than the Y'07 group (5.9% vs. 18.4%, $p = .001$).

Table 13. *Demographics of Childhood Cancer Survivors compared with Youth'07 samples*

		CSS		**Y'07**		p*
		n	%	n	%	
All		170		9107		
Gender	F	73	42.9	4178	45.9	0.58
	M	97	57.1	4920	54.1	
Ethnicity	Asian/Other	10	5.9	1671	18.4	0.001
	European	116	68.2	4780	52.6	
	Maori	28	16.5	1699	18.7	
	Pacific	16	9.4	930	10.2	
Age	Mean	14.9		14.9		0.76
	Median	15		15		
	Range	12 to 19		11 to 20		
NZDep06	Mean	5.6		5.1		0.13
	Median	5		5		
	Range	1 to 10		1 to 10		

* Adjusted for Y07 clusters (PROC SurveyMeans in SAS)

4.4.1. Comparison of Wellbeing in Childhood Cancer Survivor and Youth'07 samples (WHO-5)

The childhood cancer survivor group reported greater wellbeing than the Y'07 group ($p < .01$). The CCS scored higher in the excellent category (29.2% vs. 18.6%) with more Y'07 reporting poorer wellbeing (21.5% vs. 10.7%).

Table 14. *Comparison of WHO-5 score of CCS and Y'07 samples*

	Poor		Good		Very Good		Excellent		
	n	% (95%CI)	n	% (95%CI)	n	% (95%CI)	n	% (95%CI)	p*
CCS (n=168)	18	10.7 6.2, 15.3	49	29.2 22.1, 36.3	52	31.0 23.8, 38.1	49	29.2 22.0, 36.3	0.0003
Y'07 (n=8677)	1864	21.5 20.4, 22.6	2487	28.7 27.3, 30.0	2714	31.3 30.1, 32.5	1612	18.6 17.1, 20.0	

* Adjusted for age, gender, NZDep06, ethnicity & Y07 clusters (PROC SurveyFreq in SAS)

4.4.2 Comparison of depression in Childhood Cancer Survivor and Youth'07 samples (RADS2-SF)

There was no statistically significant difference in the proportion of CCS and Y'07 groups reporting depression. Of note, while not statistically significant there was a trend for lower rates of depression in the CCS group (6.1% vs. 10.6%).

Table 15. *Comparison of RADS2-SF score of CCS and Y'07 samples*

	Mod/ Severe Depression		Normal		
	n	% (95%CI)	n	% (95%CI)	p*
CCS	10	6.1 2.5, 9.7	154	93.9 90.3, 97.5	0.09
Y'07	904	10.6 9.7, 11.4	7664	89.4 88.6, 90.3	

* Adjusted for age, gender, NZDep06, ethnicity & Y07 clusters (PROC SurveyFreq in SAS)

4.4.3. Comparison of strengths and difficulties in Childhood Cancer Survivor and Y'07 samples (SDQ)

The proportion of adolescents meeting the criteria for difficulties was similar for the CCS and Y'07 groups. Fewer CCS were in the abnormal/borderline range on the conduct scale compared with the Y'07 (14.8% vs. 21.6%) but this did not reach a level of significance. There were no significant differences on the total difficulties score (summation of the four difficulty scales). In the Prosocial scale, fewer CCS scored social difficulties than the Y'07 group (13.5% vs. 21.8%) (Table 17).

Table 16. *Comparison of SDQ scores of CCS survivors and Y'07 samples*

	Abnormal/Borderline		Normal		
	n	% (95%CI)	n	% (95%CI)	p*
Emotional					
CCS	22	13.6 8.4 to 19.1	140	86.4 80.9 to 91.6	0.86
Y07	1174	13.4 12.3 to 14.5	7581	86.6 85.5 to 87.7	
Conduct					
CCS	24	14.8 9.1 to 20.5	138	85.2 79.5 to 90.9	0.06
Y07	1895	21.6 20.2 to 23.1	6859	78.4 76.9 to 79.8	
Hyperactivity					
CCS	33	20.4 14.3 to 26.9	129	79.6 73.1 to 85.7	0.43
Y07	2001	22.9 21.7 to 24.1	6734	77.1 75.9 to 78.3	
Peer Problems					
CCS	25	15.3 9.6 to 21.0	138	84.7 79.0 to 90.4	0.95
Y07	1341	15.3 14.4 to 16.2	7406	84.7 83.8 to 85.6	
Total Difficulties					
CCS	29	17.8 11.6 to 23.9	134	82.2 76.1 to 88.4	0.48
Y'07	1782	20.4 19.3 to 21.6	6941	79.6 78.4 to 80.7	

* Adjusted for age, gender, NZDep06, ethnicity & Y07 clusters (PROC SurveyFreq in SAS)

Table 17. *Comparison of SDQ prosocial score of CCS and Y'07 samples*

	Abnormal/Borderline		Normal		
	n	% (95%CI)	n	% (95%CI)	p*
CSS	22	13.5 8.1 , 18.8	141	86.5 81.2, 91.9	0.009
Y07	1911	21.8 20.3, 23.3	6852	78.2 76.7, 79.7	

* Adjusted for age, gender, NZDep06, ethnicity & Y07 clusters (PROC SurveyFreq in SAS)

4.4.3 Comparison of anxiety in Childhood Cancer Survivor and Youth2000 samples (MASC-10)

Comparison of reported anxiety in CCS with the previous Youth2000 group was not done as intended. The MASC-10 data was collected in the Youth2000 study but was not used in the published reports or collected in the Youth'07 survey. The Youth2000 survey reported an abnormal anxiety estimate of 4.8% (95% CI 4.1 to 5.5) but no further data was available. It is unclear why the team abandoned the reporting and future collection of anxiety data.

Overall, the evidence suggests that the childhood cancer survivors have higher rates of psychosocial wellbeing than a normative sample of their peers. CCS reported greater wellbeing as assessed by the WHO-5, less depression and greater prosocial behaviour.

This chapter has presented the findings of the research questions. Statistical analyses of association between reported wellbeing and demographic or cancer characteristics have been presented together with comparisons between the wellbeing of the childhood cancer survivor group and the Youth'07 control group. In the following chapter these findings are discussed and compared to current literature.

CHAPTER 5
DISCUSSION

This chapter summarizes and discusses the findings of this research study on the psychosocial wellbeing of childhood cancer survivors. How the findings relate to current research literature in the area of childhood cancer survivors and emotional wellbeing is also examined. The strengths and limitations of the study are discussed and the chapter concludes with the implications for delivery of care for childhood cancer survivors in New Zealand and suggestions for future research.

5.1 The Psychosocial Wellbeing of Childhood Cancer Survivors

Following diagnosis of a childhood cancer and the intensive treatment necessary to effect a cure, one might expect childhood cancer survivors (CCS) as a group to not be doing as well as their peers in terms of psychosocial wellbeing. The findings of this study show that childhood cancer survivors are doing as well, and in some cases better, than their peers.

Age at the time the young people were surveyed was a factor, with younger age associated with poorer prosocial behaviour as measured by the SDQ, but conversely also with greater emotional wellbeing i.e. happier as defined by the WHO-5 Wellbeing Index. Age at time of diagnosis was also a factor, with those diagnosed at a younger age reporting feeling happier with a trend to diagnosis at an older age being associated with poorer wellbeing on the WHO-5. There were no associations with age or age at diagnosis to wellbeing on any other measure.

Lower socioeconomic status (SES) was associated with greater conduct problems on the SDQ scale, however there was no such association with depression, anxiety or any other measure of psychosocial difficulty. This was in contrast to the findings of the national survey, Te Rau Hinengaro: The New Zealand Mental Health Survey (Oakley Browne,

Wells, & Scott., 2006), that reported the prevalence over a 12 month period of mental health disorders was higher for younger people, those with less education and income and those living in areas of higher deprivation (Wells, 2006). There was a lower response rate from CCS residing in communities with a lower SES as defined by the NZDep06. The reasons for lower responses from this group of young people are not clear. Low SES has been associated with lower participation in surveys; disengagement, unfamiliarity with technology, poor literacy and poor contact information have been cited as the reasons (Lorant, Demarest, Miermans, & Oyen, 2007; Goree & Marszalek, 1995). These possible barriers were identified at the beginning of this study, hence the offer of assistance to access computer and internet.

In this study, survivors of central nervous system (CNS) tumours were included which is at variance with many other studies. In these other studies, brain tumour survivors have been deliberately excluded because the psychopathology is considered different (DeJong & Fombonne, 2006). However, it should be noted that CCS with CNS disease were less likely to be included in this survey because the physical or cognitive difficulties experienced by some, meant they did not meet the inclusion criteria. In considering the type of diagnosis as a variable in the psychosocial wellbeing of CCS, CNS disease was the only diagnosis group that was shown to be associated with poorer wellbeing. Our findings showed there was a greater prevalence of peer problems among the survivors with a diagnosis of a CNS disease and though not statistically significant there was a trend to more emotional problems and total difficulties than any other diagnosis group.

Treatment modalities, specifically CNS directed chemotherapy and CNS radiation were identified in the literature as being significant contributing factors to poorer psychosocial outcomes for CCS. Radiation to all sites including total body irradiation used with Haemopoetic Stem Cell Transplant (HSCT) is also associated in

some studies with psychological distress and somatization (Schultz et al., 2007; Vannatta et al., 2007). In this study, CNS directed therapy including radiation and intrathecal chemotherapy was not linked to poorer reports of psychosocial functioning, which was unexpected. Based on the existing literature and our knowledge of the medical and neurological late effects that a number of these young people have, we expected the difficulties to be apparent, reaching a level of significance. One explanation that may account in part for this was suggested by Vannatta et al. (2007), who reported that while children who received CNS therapy with neurotoxic late effects had more peer problems and were more socially isolated, they did not report problems with social functioning to the same degree as parents or teachers and suggested that limited self-awareness of social difficulties was a factor. Other possible contributing factors are discussed later in this chapter.

Higher rates of depression following a childhood cancer diagnosis and some treatment modalities specifically CNS irradiation and HSCT, have been identified as being a significant issue for this group of young people in a number of studies (Schultz et al., 2007; Vannatta et al., 2007; Zeltzer et al., 2009). This was not found in this survey. Depression as defined by the RADS2-SF was seen in approximately 6% of those who completed the survey and there was no correlation to gender, ethnicity, disease or treatment modality. While the numbers reporting significant depression were low in this survey, it is important to acknowledge that for those that did so, it is clinically important. In addition, as suggested by DeJong and Fombonne (2006), any level of depression is pertinent as subclinical levels have been shown to have a negative effect on quality of life, social relationships and compliance with heath interventions.

5.2 Comparison of Childhood Cancer Survivors and Youth'07 Sample

Youth'07 (Y'07) was a national health survey of a representative sample of college students in New Zealand. The rates of participation, as a percentage, were very

similar between both the study and control groups for age, socioeconomic status and gender. Ethnicity was also comparable except for Asian childhood cancer survivors with a response rate of 5.9% compared with 18.4% in the Youth'07 study. This difference could be an effect of the rise in immigration and student visa holders from Asian countries being more highly represented in the Youth'07 survey and, as the highest incidence of childhood cancer diagnoses occur in the two to seven year age group, Asian adolescents were less represented in the study sample.

CCS overall were happier, with 29.2% reporting excellent emotional wellbeing compared with 18.0% from the Y'07 group on the WHO-5. Reports of good to excellent wellbeing were more prevalent for males in the CCS (92% v. 72%) and females were similar for both groups (86.4% v. 85%). The Y'07 group was twice as likely to report poorer wellbeing.

CCS reported significantly less depression than their peers, with only 6.1% scoring in the depression range compared with 10.6% in the Y'07 cohort. Of interest, both the Y'07 and CCS groups overall had lower rates of depression than the 12-16% expected in normative adolescent samples as described by Reynolds (2002). The percentage of depression in males for both groups was similar (6.4% vs.7%) but surprisingly only 5.7% of female CCS scored in the depression range compared with 15%, a threefold increase in the Y'07 survey.

There is considerable literature that describes the higher rate of depression in adolescent females as compared to males. The Dunedin and Christchurch longitudinal studies reported increased rates of depression for New Zealand females aged 15 to 18 (Hankin et al., 1998), and this was also observed in the Youth2000 Survey where 14% of students (males 9.0% vs.18.3% females) reported significant levels of depression. There was a decrease in the later Y'07 findings however this was still a higher rate of depression than that found in the CCS cohort.

Comparison of the two groups on the SDQ subscales of emotional problems, peer problems, hyperactivity and total difficulties showed both groups were similar. CCS reported slightly better conduct and social behaviour than the Youth'07 cohort.

Overall, this study found that the majority of CCS reported more positive emotional wellbeing, less depression and less anxiety than may have been expected from much of the published literature and by comparison with a group of normal New Zealand students, faring equally as well, and in some measures, better.

There are themes emerging from the more recent studies that a large majority of CSS do not show elevated levels of anxiety, or depression, or lower self esteem than their peers and may have a greater sense of wellbeing (Kazak et al., 2010; Parry & Chesler, 2005; Phipps, Jurbergs, & Long, 2009; Williams, Davis, Hancock, & Phipps, 2010). Newer therapies and advancing knowledge of the mechanisms of these diseases mean that the intensity of many treatment protocols have been modified, reducing the potential for the late effects of treatment that has been reported in earlier studies. It is also very possible that with the comparatively small number of childhood cancer diagnoses and subsequent survivors in New Zealand, there is a health protective effect as most are still involved in a long term follow-up programme with multi-disciplinary health professionals and not many are lost to follow-up in this age bracket. Parry and Chesler (2005) note that many thrive, and report that the cancer experience made them stronger, self reliant, able to deal with problems better than their peers, and indicated that they felt they were "more mature" than others their age. This is congruent with the sense the LEAP team get from working with these young people in a clinical setting.

By necessity, many of these young survivors form a close bond with their family during the period of diagnosis, treatment and follow-up care which often spans a number of years. This is often at a time when most of their peers are becoming increasingly independent. In addition, the age for participation in this survey was 12 to

18years, when the protective factors of family, targeted health care, school and friends are still predominant and may have a positive impact on their psychosocial wellbeing. As suggested by Zebrack and Chesler (2002), at this age they have not yet had to deal with significant changes to home, employment, financial status or sexuality as a result of having had cancer.

One of the goals of this study was to survey adolescent childhood cancer survivors in the context of a New Zealand culture and whether that would make a difference. New Zealand has a national framework for child and adolescent cancer services that offers a comprehensive service for the diagnosis and treatment of childhood cancer providing a free, equitable service regardless of ethnicity or socioeconomic status. The multi-disciplinary approach includes psychologists, social workers, play therapists and bedside teachers, with supported transition back into school and communities. On transition into a LEAP long term follow-up programme, the core multidisciplinary healthcare teams comprise a paediatric oncologist, nurse specialist and clinical psychologist/ neuropsychologist. Special childhood cancer charitable agencies provide educational and social support for many years from the time of diagnosis to long after treatment has been completed. It could be argued that these are all protective factors that have a positive effect on the cancer experience for many survivors and may have contributed to the positive findings of this survey.

5.3 Study Strengths and Limitations

This study has several strengths; it is the first survey of the effects of a childhood cancer diagnosis and subsequent treatment on the coping and wellbeing of adolescent childhood cancer survivors in New Zealand. The use of a nationally representative sample of New Zealand secondary school students as the control group provides a strong and valid comparison. The four instruments used to measure the mental health and emotional wellbeing had been used previously with the two large

population youth health surveys (Youth2000 and Youth'07) and have been shown to have acceptable reliability and validity in a New Zealand adolescent population.

Time between the Youth'07 and this survey was less than two years and provided relatively up to date comparison with a same-generation population.

The criticism made of the varied and often contradictory reports on the psychosocial outcomes for childhood cancer survivors is the lack of consistency in use of outcome measures, sample sizes and control groups used (Zeltzer et al., 2009), and though we used a normative group of peers as a control group this study was not able to address the issues of small sample size or instruments used. The choice of the standardized instruments was dictated by the decision to use the Youth07 survey as opposed to building on findings from previous studies using similar measures.

There are a number of limitations to this study, acknowledged limitations of a case- control survey are that of selection bias and recall bias. While self-report surveys can be questioned in regard to recall bias, both groups used the same survey tool and were the same age. Given the dynamic nature of mood, life events at that time and the recall ability of the participants, the findings are limited to the time of completing the survey. However as Schwartz wrote in an editorial (JAMA 2003), "critical information for understanding the psychological and behavioural responses to survival is revealed by self-report" (p 1641).

Due to the relatively small number of childhood cancer diagnoses in New Zealand each year it was not possible to randomly select a sample, in addition there was a possible self-selection bias as a greater number of those young people who are engaged in follow-up care or feeling more positive were more likely to participate. Of the Maori and Pacific Island young people only a third who were invited completed the survey. This may have been due to limited access to, or familiarity with, the internet or computer or lack of confidence in accepting the offer of assistance. Some more remote

parts of New Zealand have either limited or no internet connection e.g. Northland with a proportionately higher Māori population (Statistics New Zealand. New Zealand census of population and dwellings, 2006). The use of an internet-based survey tool may have been a barrier to those with limited access to a computer or internet service even though steps were taken to overcome this, unfamiliarity and lack of confidence with this medium may have stopped some young people from taking part. The issue of some computers being blocked by security settings also had an impact on the participation numbers.

On first impressions, while some of the results may appear significant, with the small sample size, the precision of the estimates is wide and may negate the importance of some of the findings, e.g. in the SDQ conduct problems, 13.5% of CCS reported in the abnormal range (95% CI =8.1-18.8) and 21.8% of Y"07 (95% CI =20.3-23.3), (p<.009).

It is acknowledged that this study does not adequately address the issues of older adolescent and young adult cancer patients. The overall survival rates of this group of young people with cancer have not kept pace with those diagnosed at a younger age, this has been due to factors such as differences in disease sub types, lack of specialised care guidelines and access to clinical trials and research (Andrea & Archie, 2007; Bleyer et al., 1997). Their exclusion from this study is solely because they fall outside the criteria for comparison with the pre-existing control group data.

Those with significant late effects affecting cognitive ability or vision impairment for example, were excluded but may have reported significant psychosocial distress that could have altered the findings.

5.4 Implications for Practice

The dedicated long term follow-up programme for survivors of childhood cancer in New Zealand that was established in 2006 has become an integral component of the continuum of childhood cancer care in New Zealand. It could be argued that the multidisciplinary support provided through the Late Effects Assessment Programme (LEAP) is a factor in the majority of these young people doing so well to date. Though difficult to assess, a CCS and parent clinic evaluation of the LEAP programme demonstrated a high level of satisfaction from both survivors and parents and confirms the importance of ongoing knowledgeable multidisciplinary support (Bartle, 2008).

While it is reassuring that the findings of this study demonstrate that for a majority they appear socially and emotionally well adjusted, there is a small but significant subgroup of young survivors who remain at risk for difficulties with psychosocial functioning. As a group, survivors of CNS disease have been shown to have the greatest difficulties in this survey. As described by a number of studies, survivors of CNS disease and CNS directed therapy are one group who are particularly vulnerable and at risk of significant poorer health outcomes as adults (Zeltzer et al., Oeffinger et al., 2006; Speechley et al., 2006; 2009).

Adolescence is a time of transition and adjustment, as all these young survivors enter adulthood, health problems that are minimal when young may become worse with age. Many will certainly have to deal with significant long term morbidity and mortality that increases long after treatment is completed. There is clear evidence that they are more like to get a second cancer, be infertile and be at greater risk of developing a chronic health condition than the general population (Armstrong et al., 2009; Meadows et al., 2009; Oeffinger et al., 2006). In translating the findings of this study into the clinical setting it is important for health care practitioners to remember that these young survivors are individuals and assumptions cannot be made that they will all thrive.

Key implications for practice include;

1. The need for routine emotional health screening, neurocognitive assessment and psychosocial support. Ministry of Health funding currently enables this, however continued access to this resource needs to be guaranteed in the future.
2. Continue to develop and strengthen the multidisciplinary model of care for survivors of childhood cancer incorporating medical surveillance, psychosocial support and health education based on individual risk-related health outcomes.
3. Develop strategies to ensure that the follow-up care continues to engage young survivors by maintaining relevance to their changing needs and based on individual needs rather than age cut-offs. This will ensure they remain supported until an appropriate time for transition to confident self-care and skill in engaging with adult healthcare services.

5.5 Implications for Future Research

Based on the results of this study, there are several recommendations for future research. First some of the limitations outlined in this study may be minimized by increasing the sample size to improve the statistical power. As childhood cancer only makes up 1% of all cancers in New Zealand, this will always be a small population to survey. However by making the survey tool more accessible, response rates would be higher and provide greater statistical power. Second the survey could be repeated in several years time, much in the same method as the Youth2000 and Youth'07 studies to capture changing trends and more accurately reflect the experiences of survivors exposed to different treatment modalities and health outcomes. Third, it would be interesting to repeat this study with older adolescent and young adult survivors aged greater than 18years, when the issues of negotiating work, family, relationships and the potential for increased health concerns may produce very different findings. Fourth, the

majority of research into childhood cancer survivorship to date has not been theoretically driven, with care primarily delivered and studied through medical models focused on specific late effects. As stated by O'Hair et al. (2003), there is a need for more theoretically driven research in the growing field of cancer and survivorship. There is a need for frameworks addressing the individual and environmental influences on survivors (Karian, Jankowski, & Beal, 1998; Skalski, DiGerolamo, & Gigliotti, 2006). Neuman's systems theory has been applied as a nursing framework for survivor care (Karian et al., 1998).

Finally, the findings of this study leave questions that provide an opportunity for qualitative research to help heath professionals working with these young people gain a greater understanding of what factors may contribute to the overall positive findings. This thesis has reported on only one aspect of the wider ACSIS study in which data was collected on the broader health and wellbeing domains that include social connectedness, spirituality, risk taking behaviours and health protective behaviours. Once the findings of this study are analysed there will be a much richer profile of this population of young survivors that may lead to further research opportunities that are not indicated from the findings of this thesis.

5.6 Conclusion

This research began with questions about the impact on an adolescent population in New Zealand of surviving a childhood cancer. Did they experienced more psychosocial difficulties after their experience or conversely had a greater sense of self protective behaviours than their peers? The young childhood cancer survivors who took part in this survey demonstrated that for many of them they see themselves as mostly happy, well-adjusted young people. There are however, an important minority who reported significant depression, poorer emotional wellbeing and increased difficulties. As Oeffinger and Hudson (2004) observed, many late effects from childhood cancer

therapy do not plateau but increase with age, often becoming apparent decades after therapy. While the majority of these young people do not report significant problems there are still protective factors as discussed earlier in this chapter that support and cushion them and the challenges of independence and adulthood haven't yet needed to be met, it would be a mistake to assume " all is well" for the majority. The numbers of young people surviving cancer will continue to increase, with approximately 160 childhood cancer diagnoses a year in New Zealand and survival rates of greater than 80% there will be at least 1200 additional survivors of a childhood cancer each decade becoming adults who may be burdened by the late effects of their childhood disease.

This study does provide for the first time valuable information on the self-perceived emotional wellbeing of adolescent childhood cancer survivors in New Zealand. Health professionals engaged with these young people from diagnosis through to completion of treatment and long term follow-up need to continue to work towards a better understanding of the unique challenges and difficulties they face, developing appropriate, holistic care for not only those with identified late effects but to ensure the general wellbeing of all childhood cancer survivors.

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APPENDICES

Appendix A: Ethics

Research Office
Level 14, Support Bldg
Auckland City Hospital
PB 92024, Grafton, Auckland
Phone: 64 9 307 4949 Extn. 23854
Email: SamanthaJ@adhb.govt.nz
Website: www.adhb.govt.nz/ResearchO

14 August 2009

Ms Kathy Yallop
LEAP
Paediatric Haematology/Oncology
Starship Children's Health
Auckland City Hospital

This is the ADHB Management Approval. Please keep in your Trial Master File.

Dear Ms Yallop

RE: Research project A+ 4391 (URB/09/05/017) - Paediatric Haematology/Oncology Late Effects Assessment Programme (LEAP) study of the quality of life and wellbeing of survivors of childhood cancer in New Zealand

The Auckland DHB Research Review Committee (ADHB-RRC) would like to thank you for the opportunity to review your study and has given approval for your research project.

Your Ethical and Institutional approval is dependant on the Research Office having up-to-date information and documentation relating to your research and being kept informed of any changes to your study. While the Research Office endeavours to send reminders for annual approvals and missing documents, it is your responsibility to ensure you have kept Ethics and the Research Office up to date and have the appropriate approvals. Please refer to the overleaf of this letter for guide to maintaining your research approval.

If you have any questions please do not hesitate to contact the Research Office.

Yours sincerely

On behalf of the Research Review Committee
Dr Samantha Jones
Manager, Research Office
Auckland DHB

c.c. Lochie Teague, Elizabeth Woods, Kay Hyman

..../continued next page

Health and Disability Ethics Committees

Upper South B Regional Ethics Committee
Ministry of Health
4th Floor, 250 Oxford Tce
PO Box 3877
Christchurch
Phone (03) 372 3018
Fax (03) 372 1015
Email: uppersouth_ethicscommittee@moh.govt.nz

10 August 2009

Ms Kathy Yallop
Auckland District Health Board
Paediatric Haemotology/Oncology Service
Starship Hospital
Private Bag 92 024
Auckland

Dear Ms Yallop

Ethics Reference Number: URB/09/05/017
Paediatric Haematology / Oncology Late Effects Assessment Programme (LEAP) study of the quality of life and wellbeing of survivors of childhood cancer in New Zealand
Investigators: Ms Kathy Yallop, Dr Heather McDowell, Dr Simon Denny, Belynda Wynn, Ms Rosemary Simpson
Localities: Starship Children's Health, Christchurch Hospital, Wellington Hospital

The above study has been given ethical approval by the **Upper South B Regional Ethics Committee**.

Approved Documents
Information Sheet (Starship Children's Health) version 2 dated 7.7.09
Information Sheet Canterbury DHB) version 2 dated 7.7.09 (front page only)
Information Sheet Capital & Coast DHB) version 2 dated 7.7.09 (front page only)
Questionnaire
Consent Form version 2 dated 7.7.09*

**For administrative clarity please include a footer on the Consent Form with the full study title, version number 2 dated 7.7.09 and send a copy to the Committee Administrator.*

Certification
The Committee is satisfied that this study is not being conducted principally for the benefit of the manufacturer or distributor of the medicine or item in respect of which the trial is being carried out.

Accreditation
The Committee involved in the approval of this study is accredited by the Health Research Council and is constituted and operates in accordance with the Operational Standard for Ethics Committees, April 2006.

Progress Reports
The study is approved until **31 December 2011**. The Committee will review the approved application annually and notify the Principal Investigator if it withdraws approval. It is the Principal Investigator's responsibility to forward a progress report covering all sites prior to ethical review of the project in **August 2010**. The report form is available on http://www.ethicscommittees.health.govt.nz. Please note that failure to provide a progress report may result in the withdrawal of ethical approval. A final report is also required at the conclusion of the study.

Requirements for SAE Reporting
The Principal Investigator will inform the Committee as soon as possible of the following:

- Any related study in another country that has stopped due to serious or unexpected adverse events
- withdrawal from the market for any reason
- all serious adverse events occurring during the study in New Zealand which result in the investigator breaking the blinding code at the time of the SAE or which result in hospitalisation or death.
- all serious adverse events occurring during the study worldwide which are considered related to the study medicine. Where there is a data safety monitoring board in place, serious adverse events occurring outside New Zealand may be reported quarterly.

All SAE reports must be signed by the Principal Investigator and include a comment on whether he/she considers there are any ethical issues relating to this study continuing due to this adverse event. It is assumed by signing the report, the Principal Investigator has undertaken to ensure that all New Zealand investigators are made aware of the event.

Amendments
All amendments to the study must be advised to the Committee prior to their implementation, except in the case where immediate implementation is required for reasons of safety. In such cases the Committee must be notified as soon as possible of the change.

Please quote the above ethics committee reference number in all correspondence.

The Principal Investigator is responsible for advising any other study sites of approvals and all other correspondence with the Ethics Committee.

It should be noted that Ethics Committee approval does not imply any resource commitment or administrative facilitation by any healthcare provider within whose facility the research is to be carried out. Where applicable, authority for this must be obtained separately from the appropriate manager within the organisation.

The committee would like to take this opportunity to wish you all the best with your study.

Yours sincerely

Diana J. Whipp

Mrs Diana Whipp
Upper South B Regional Ethics Committee Administrator
Email: diana_whipp@moh.govt.nz

Appendix B: Survey Participant Information and covering letters

Service: Paediatric Haematology / Oncology
Long Term Follow Up Clinic
Tel No: 0800 022 747

27 August 2009

Parent of …

Dear Parent …

A study of the quality of life and wellbeing of survivors of childhood cancer in New Zealand

You have been sent this letter and attached information sheet because your child is younger than 16yrs of age and we would like to invite them to take part in a research study. This study looks at what life after cancer is like for young people aged 12 to 18 years of age who have survived a childhood cancer or an illness that was not called a cancer but needed the same treatment.

Please read the attached information sheet and if you are happy for your child to take part please pass this onto them.

To accurately reflect your child's own view it is important that they are the one to complete the questionnaire and are able to complete it on their own and in private.

If you have any questions at all please contact me or any of the research team - our contact details are on the information sheet.

Yours sincerely,

Kathy Yallop
Principal Investigator

Service: Paediatric Haematology / Oncology
Long Term Follow Up Clinic
Tel No: 0800 022 747
Email: LEAP@adhb.govt.nz

27 August 2009

Dear …….

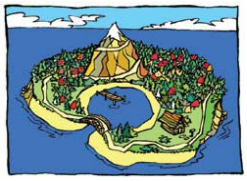
A study of the quality of life and wellbeing of survivors of childhood cancer in New Zealand

You have been sent this letter and information sheet (attached) because we want to invite you to take part in a research study that looks at what life is like for young people aged 12 to 18 years of age who have survived a childhood cancer. You may have been sent this invitation because you had an illness that was not called a cancer, but needed the same treatment.

We hope the information sheet will answer most of the questions you may have, but there is a lot of writing so if you have any questions or concerns, please contact me or any of the research team – our contact details are on the information sheet. We are happy to answer your questions.

Yours sincerely,

Kathy Yallop
Principal Investigator



Information Sheet

A study of the quality of life and wellbeing of survivors of childhood cancer in New Zealand.

Kathy Yallop
Principal Investigator
Nurse Specialist -*LEAP*
Email: kyallop@ADHB.govt.nz
Ph: 09 3074949 ext 23119
Mob: 021475451

Dr Heather McDowell
Investigator
Clinical Psychologist – *LEAP*
E-mail: HeatherM@adhb.govt.nz
Mob: 021938014

Paediatric Haematology/Oncology Service
Starship Children's Health
Private Bag 92024
Auckland 1142

You are invited to take part in an online questionnaire to find out what life is like for young people who are survivors of childhood cancer. This is the first time this has ever been done in New Zealand.

If you take part in this questionnaire, your answers together with those of the others who complete it will give us a greater understanding of the good stuff and hard stuff that happens after treatment finishes. This will be used to develop services to better support survivors of childhood cancer.

How do I do it?

- Before starting make sure you can do it in private and on your own.
- The questionnaire is answered online and should take about ½ hour to complete.
- The questions are spoken (audio) as well as in writing and to respond you just click on the right answer for you. The answer choices are also spoken if you click on the words.
- We will provide headphones for privacy if you need these.
- The questionnaire is called a branching questionnaire so you only get the questions that are relevant for you.
- There is also an opt-out box for any questions you don't want to answer.
- We will organise access to a computer and the internet if you need this.

You can call the following free-phone number to organise headphones, internet access or help at any time (24hrs/7days) this will be answered by Kathy Yallop or Heather McDowell.

0800 0 ACSIS (0800 0 22747)

The questionnaire has some questions on sensitive areas such as use of alcohol and drugs, sexual activity, and fertility, however, it is important to remember two things: 1) you can choose not to answer questions you don't want to answer and 2) you will only be asked questions about these areas if you are engaging in these behaviours (e.g. if you answer "No" to a question about drinking alcohol, then the questionnaire branches so you won't get asked any other questions about alcohol). If you have any questions about this, please just call us on the free-phone 0800 number.

Why should I take part in this study?

By taking part in this research you have the opportunity to say what life is like for you now and hopefully this will help many young people like you in the future. The more people who answer this questionnaire the more useful the study will be.

What are the risks for me?

We don't expect there to be any risks for you, however if answering the questionnaire brings up issues or questions that you would like to talk with someone about you can phone 0800 0 ACSIS (0800 0 22747) and we will organise someone for you to talk with.

How confidential is this?

To send this letter out we have your name and address and the type of cancer you had (database A). This information is kept confidential like your medical information.
Once we sent out this letter to you, we replaced your name and contact information with a randomly generated code number (database B).
From here on only this code number will be used, so people looking at the questionnaire answers will not know who has provided them.
The anonymous answers will be stored in a secure central database which is password protected to prevent unauthorised access. This data will be securely stored for fourteen years and will then be safely destroyed.
No material which could personally identify you will be used in any reports on this study.

To protect your own confidentiality and allow you to give your own answers we recommend you do this in a room on your own where you will not be interrupted or overheard.

If you decide to take part this is the code number for you to use:

XXXX

Do I have to participate?

Your participation is entirely voluntary (your choice). You do not have to take part in this study, if you choose not to take part this will not affect any future care or treatment. Remember also that your participation is anonymous.

How will I find out about the results of this study?

It is estimated that it will take up to 12months to review the data and for the results of the study to be finalised. A report will be available through CanTeen and Child Cancer Foundation at that time and will be advertised in their magazine.
If you would like an electronic or hard copy of the results when the study is finished please contact one of the researchers whose details are above.

How do I answer the questionnaire?

- Before starting make sure you can do it in private and on your own
- Connect to the internet on your computer (remember if you do not have access to a computer or internet at home, free-phone 0800 number on the front page to ask for an alternative to be arranged for you.)
- Go to **www.leapin.ac.nz**
- Enter your confidential code number, as above and press **SUBMIT**
- Make sure the volume is turned up enough for you to hear and if you are using ear phones for privacy plug them into the computer.
- While we recommend you complete the questionnaire in one session, if you need to stop before you have finished, log in again and it will take you back to where you left off.-

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If you have any questions or concerns about your rights as a participant in a research study you can contact an independent health and disability advocate. This is a free service provided under the Health and Disability Commissioner Act.
Ph (NZ wide): 0800 555 050
Free Fax (NZ wide): 0800 2787 7678 (0800 2 SUPPORT)
Email (NZ wide): advocacy@hdc.org.nz

For Māori health support, or to discuss any concerns or issues regarding this study, please contact;
Mata Forbes RGON, Maori Health Services Co-ordinator / Advisor
Auckland City Hospital.
Ph: (09)307 4949 extn 23939
Mobile 021 348 432

If you have any questions please contact us, Kathy or Heather- contact info above or phone

0800 0 ACSIS (0800 0 22747)

Thank you for reading this and considering taking part.

This project has been approved by the Upper South B Regional Ethics Committee. Reference number URB/09/05/017

WOULD YOU LIKE TO WIN AN IPOD?

We want to thank you for taking part in the ACSIS study and to help us say thanks we have 5 iPods to give away.
To be in the draw, just fill your details below & send them to us in the envelope provided, how easy is that?

Yes, I answered the ACSIS on-line questionnaire, please put me in the draw to win 1 of 5 iPods

NAME _____

ADDRESS _____

_____

_____

CONTACT PHONE NUMBER _____

E-MAIL _____

THANKS FOR YOUR HELP AND BEST OF LUCK FOR THE DRAW!

ACSIS
ADOLESCENT
CANCER
SURVIVOR
IMPACT STUDY

Appendix C: M-CASI questionnaire- Introduction and consent page

Consent Form

ACSIS: A Paediatric Haematology/Oncology study of the quality of life and wellbeing of childhood cancer survivors on New Zealand.

- I have read and understood the participant information sheet that was sent to me.
- I know I can ring the contact people listed on the participant information sheet at any time I want more information.
- I understand that taking part in the survey is entirely my choice.
- I understand that the questionnaire is confidential and anonymous.
- I understand that I will not be able to be identified from any reports arising from this research.

I Accept	I Don't Accept

This study has been approved by the Upper South B Regional Ethics Committee.
Reference number: URB/09/05/017

Text version of the M-CASI questionnaire- Section: Emotional health

Info: **Emotional health**
We would now like to ask some questions about how you have been feeling.

a) Who-5 Wellbeing Index

Over the last two weeks...

	all of the time	most of the time	more than half of the time	less than half of the time	some of the time	at no time
I have felt cheerful and in good spirits	☐	☐	☐	☐	☐	☐
I have felt calm and relaxed	☐	☐	☐	☐	☐	☐
I have felt active and vigorous	☐	☐	☐	☐	☐	☐
I woke up feeling fresh and rested	☐	☐	☐	☐	☐	☐
my daily life has been filled with things that interest me	☐	☐	☐	☐	☐	☐

b) Strengths and Difficulties

For each of the following statements please mark the box for Not True, Somewhat True or Certainly True. It would help us if you answered all the questions as best you can even if you are not absolutely certain. Please give your answers on the basis of how things have been for you over the last six months

Over the last six months...

	not true	somewhat true	certainly true
I try to be nice to people, I care about their feelings	☐	☐	☐
I am restless, I cannot stay still for long	☐	☐	☐
I get a lot of headaches, stomach-aches or sickness	☐	☐	☐
I usually share with others, for example CDs, games, food	☐	☐	☐
I get very angry and often lose my temper	☐	☐	☐

Over the last six months...

	not true	somewhat true	certainly true
I would rather be alone than with people of my age	☐	☐	☐
I usually do as I am told	☐	☐	☐
I worry a lot	☐	☐	☐
I am helpful if someone is hurt, upset or feeling ill	☐	☐	☐
I am constantly fidgeting or squirming	☐	☐	☐

Over the last six months...

	not true	somewhat true	certainly true
I have one good friend or more	☐	☐	☐
I fight a lot. I can make other people do what I want	☐	☐	☐
I am often unhappy, depressed or tearful	☐	☐	☐
Other people my age generally like me	☐	☐	☐
I am easily distracted, I find it difficult to concentrate	☐	☐	☐

Over the last six months...

	not true	somewhat true	certainly true
I am nervous in new situations. I easily lose confidence	☐	☐	☐
I am kind to younger children	☐	☐	☐
I am often accused of lying or cheating	☐	☐	☐
Other children or young people pick on me or bully me	☐	☐	☐
I often volunteer to help others	☐	☐	☐

Over the last six months...

	not true	somewhat true	certainly true
I think before I do things	☐	☐	☐
I take things that are not mine from home, school or elsewhere	☐	☐	☐
I get along better with adults than people my own age	☐	☐	☐
I have many fears, I am easily scared	☐	☐	☐
I finish the work I am doing, my attention span is good	☐	☐	☐

Overall do you think you have difficulties in any of the following areas: emotions, concentration, behaviour or being able to get along with other people?
yes - minor difficulties
yes - definite difficulties
yes - severe difficulties
no-

How long have these difficulties been present?
less than a month
1-5 months
6-12 months
over a year

Do the difficulties upset or distress you?
not at all
a little
a medium amount
a great deal

Do the difficulties interfere with your everyday life in the following areas?

	not at all	a little	a medium amount	a great deal
home life	☐	☐	☐	☐
friendships	☐	☐	☐	☐
classroom learning	☐	☐	☐	☐
leisure activities	☐	☐	☐	☐

Do the difficulties make it harder for those around you (family, friends, teachers, etc)?
not at all
a little
a medium amount
a great deal

Info: **Thank you**
You are almost finished this section. The last few questions are about how you have been feeling.

c) RADS-2 SF

We will now ask some questions about how you feel. After each one decide if you feel this way almost never, hardly ever, sometimes, or most of the time. Remember there are no right or wrong answers. Just choose the one answer that tells how you usually feel.

How do you usually feel?

	almost never	hardly ever	sometimes	most of the time
I feel lonely	☐	☐	☐	☐
I feel happy	☐	☐	☐	☐
I feel like hiding from people	☐	☐	☐	☐
I feel sad	☐	☐	☐	☐
I feel like hurting myself	☐	☐	☐	☐

How do you usually feel?

	almost never	hardly ever	sometimes	most of the time
I feel I am no good	☐	☐	☐	☐
I feel I am bad	☐	☐	☐	☐
I feel mad about things	☐	☐	☐	☐
I feel bored	☐	☐	☐	☐
I feel like nothing I do helps anymore	☐	☐	☐	☐

d) MASC-10

We would like to ask you some questions about how you have been thinking, feeling or acting recently

	never true about me	rarely true about me	sometimes true about me	often true about me
The idea of going away to camp scares me	☐	☐	☐	☐
I'm afraid that others will make fun of me.	☐	☐	☐	☐
I try to stay near my mum or dad.	☐	☐	☐	☐
I get dizzy or faint feelings	☐	☐	☐	☐
I feel restless and on edge.	☐	☐	☐	☐

Here are some more questions about how you have been thinking, feeling or acting recently

	never true about me	rarely true about me	sometimes true about me	often true about me
I feel sick to my stomach.	☐	☐	☐	☐
I get nervous if I have to perform in public	☐	☐	☐	☐
Bad weather, the dark, heights, animals or bugs scare me.	☐	☐	☐	☐
I check to make sure things are safe	☐	☐	☐	☐
I feel shy.	☐	☐	☐	☐

Info: **Well done!**
Thank you for answering these questions. If these questions have been upsetting for you and you wish to talk with someone, you can phone 0800 xxxx the free call number on your letter, Youthline 0800 376633, free txt 234 or 0800 WHATSUP (0800 942 87 87)