Development and psychometric evaluation of a measure of perceived need for adolescents and young adults with cancer

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This thesis is dedicated to Greg

A more amazing husband there never was.

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List of Abbreviations

ALL	Acute Lymphoblastic Leukaemia
AQoL	Adolescent Quality of Life Instrument
AYA	Adolescent and Young Adult
CaSUN	Cancer Survivors' Unmet Needs measure
CI	Confidence Interval
CML	Chronic Myeloid Leukaemia
CNQ	Cancer Needs Questionnaire
CNQ–YP	Cancer Needs Questionnaire – Young People
CNS	Central Nervous System
CPNQ	Cancer Patient Need Questionnaire
CPNS	Cancer Patient Need Survey
CSF	Cerebrospinal Fluid
GP	General Practitioner
HPV	Human Papillomavirus
HRQOL	Health Related Quality of Life
ICC	Intraclass Correlation Coefficient
MMQL	Minneapolis-Manchester Quality of Life Instrument
NCI	National Cancer Institute
NCRI	National Cancer Research Institute
NEQ	Needs Evaluation Questionnaire
NHMRC	National Health and Medical Research Council
NIH	National Institutes of Health
NMSC	Non-Melanoma Skin Cancer
NSW	New South Wales
PCA	Principal Components Analysis
PCQL	Pediatric Cancer Quality of Life Inventory
PCQL–32	Pediatric Cancer Quality of Life Inventory - 32 Short Form
PedsQL	Pediatric Quality of Life Inventory
PFA	Principal Factor Analysis
PIE	Perceived Illness Experience Scale
PNAT	Patient Needs Assessment Tool
PNI	Psychosocial Needs Inventory
POQOLS	Pediatric Oncology Quality of Life Scale
QLD	Queensland
QOL	Quality of Life

QOL–CS	Quality of Life - Cancer Survivors
RCT	Randomised Controlled Trials
SA	South Australia
SCNS	Supportive Care Needs Survey
SEER	Surveillance, Epidemiology and End Results
SES	Standardised Effect Size
TCU	Teenage Cancer Unit
TYA	Teenage and Young Adult
UK	United Kingdom
US	United States
VIC	Victoria
WA	Western Australia
WHO	World Health Organization

Synopsis

An overview of the unique challenges that adolescent and young adult (AYA) cancer survivors may face due to their cancer diagnosis occurring at a critical phase of physical, psychological and social development is presented in Chapter 1. A review of the literature revealed that no psychometrically rigorous measures of unmet need for AYA cancer survivors currently exist, discussed in Chapter 2. The initial steps (face and content validity) in the development of a measure specifically designed to capture the needs of this population are described in Chapter 3. The measure was pilot tested with 32 AYAs recruited through a state-based cancer registry, discussed in Chapter 4. The construct validity and internal consistency of the measure were established with a sample of 139 AYAs recruited through seven treatment centres, presented in Chapter 5. Test-retest reliability was examined with a sub-sample of 34 AYAs. The final measure consists of 70 items and six factors. All factors achieved Cronbach's alpha values >0.80. Item-to-item test-retest reliability was also high, with most items reaching weighted kappa values >0.60. The prevalence of high levels of unmet need related to the availability of good food and leisure spaces at the treatment centre, body image, fertility, peer interaction, physical functioning, and tailored information were experienced by a large proportion of AYAs, described in Chapter 6. Participants who were female, diagnosed with haematological cancer, experienced a recurrence, received more than two types of treatment, or who were less than two years post-diagnosis had significantly greater odds of experiencing high levels of unmet need for a number of issues. Recommendations for further psychometric evaluation of the measure (including longitudinal studies to establish responsiveness and predictive validity) with a larger sample are discussed in Chapter 7.

CHAPTER 1

The psychosocial health of adolescent and young adult (AYA) cancer survivors

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Chapter 1

The psychosocial health of adolescent and young adult (AYA) cancer survivors

Introduction

Adolescents and young adults (AYAs) have gained increased attention in recent years as a population with unique health needs.[1] Adolescents and young adults are, for the most part, healthy and not affected by other causes of mortality that affect children or older adults such as pneumonia or heart disease.[1-3] However, chronic disease is prevalent and includes diabetes, asthma, mental illness and cancer.[2 4 5] Due to the ongoing nature of these diseases and the young age at which they occur, the lifelong burden of disease for AYAs is high.[6] Cancer is the second most frequent cause of death in AYAs after accidental injury and accounts for 11% of the total mortality for this age group in the United Kingdom (UK).[7]

Defining who is an adolescent and young adult (AYA)

Adolescents and young adults aged 15 to 29 years account for over one quarter of the world's total population.[8] However, there is some debate regarding the age at which adolescence begins and young adulthood ends. The World Health Organization (WHO) defines adolescents as being between 10-19 years of age, youth as between 15-24 years and young people as between 10-24 years.[2 9] Across the medical and health literature the age range used to define AYAs also varies from 10-40 years.[10-14] However, referring to individuals aged 15 to 30 years as AYAs is generally accepted.[5 15 16]

Reasons for variations in the definition of the AYA age group may be related to biological, cultural, societal or clinical reasons.[10] The term "AYAs" itself may also be culturally specific. For example, in the UK the term "TYAs" (teenagers and young

adults) is commonly used.[7 17] In this thesis, for the purpose of consistency and in line with Australian expression[16] the term “AYAs” will be used and will generally refer to individuals aged 15 to 30 years unless otherwise specified.

Developmental stages of AYAs

Adolescence and young adulthood have been defined as unique stages of physical, psychological, social and cognitive growth and maturation in the life of an individual.[10 18] The transition into adulthood requires that a number of developmental milestones be achieved that will enable AYAs to become financially independent, make their own decisions and accept personal responsibility in the future.[19]

Physical developmental milestones

The physical beginning of adolescence is often associated with the onset of puberty.[20] Puberty describes the biological changes which affect a young person’s anatomy, physiology and appearance and prepare the body for sexual reproduction.[21] These changes can include: growth spurts; increased bone density; development of muscle and fat; development of sex organs; enlargement of ovaries and testes; experiencing menarche, breast enlargement and widening of hips (females); experiencing spermarche and deepening of the voice (males); growth of pubic, body and facial hair; and an increase in sweat glands and body odour.[21-23] In western and industrialised countries puberty can begin as early as eight years of age in girls and nine years of age in boys, or as late as 13 years for both males and females.[19] Full maturation can take up to six years following the onset of puberty, however females will usually mature around two years ahead of males with 12.5 years being the average age of menarche for young women.[19]

Due to the multiple physical changes which occur with the onset of puberty, physical appearance is a major focus of AYAs.[24] Many AYAs become highly sensitive and self-conscious about how other young people will judge their appearance and the self-esteem of an individual is strongly associated with their own perceived attractiveness.[22] Female AYAs are more likely to develop a negative body image and be more critical of their appearance than males, and for some young people this can lead to adverse health behaviours such as anorexia or bulimia nervosa.[23 25 26]

Psychological and social developmental milestones

Erikson's theory of development outlines the psychological and social milestones AYAs need to achieve in order to successfully adapt to society.[27 28] According to Erikson, the two most important milestones during adolescence and young adulthood are 'Identity versus Role Confusion' (12-20 years) and 'Intimacy versus Isolation' (18-34 years).[24] In the formation of identity, Erikson proposes that three areas of the young person's life need to be consolidated: ideology (values and beliefs); occupation; and love (personal relationships).[27 28] Being able to experience and test out different sets of beliefs, jobs, and intimate relationships allows an individual to clarify who they are and how they wish to live their life.[19] Failure to successfully establish commitments in these areas can lead to Role Confusion, making it more difficult for AYAs to achieve other developmental milestones in adulthood.[24]

A young person also needs to be able to achieve intimacy, or the opportunity to share knowledge, thoughts and feelings with another person.[29] During adolescence, the child-parent dynamic begins to shift, and relationships between young people and their parents tend to become less warm and close, with an increase in conflict.[30 31] Subsequently, for most AYAs their friends rather than family become the focus of companionship and emotional intimacy.[32] Friends provide AYAs with an outlet to talk

about personal feelings and intimate issues (particularly romantic and sexual), that they may otherwise be uncomfortable talking about with parents or other family members.[29] Within these intimate peer relationships, young people are also able to share concerns, problem solve, and receive support and advice.[33] When AYAs are unable to successfully form these types of intimate relationships, feelings of isolation can occur.[19 24]

In addition to establishing identity and intimacy, psychological and social development which occurs during adolescence is focused around achieving independence and autonomy.[34] Three types of independence in adolescents have been identified: behavioural (e.g. breaking a curfew); emotional (e.g. indifference to parental anger); and value (e.g. setting goals without concern for parental values).[23] The degree to which a young person successfully achieves independence is influenced by the family unit and patterns for inclusion in decision making, resource allocation and discipline.[35] Opportunities to participate in these activities can influence the level of autonomy and responsibility a young person obtains.[35]

Cognitive developmental milestones

During adolescence and young adulthood a young person's cognitive abilities also reach a new level of sophistication. Specifically, the young person's ability to think and solve problems, and their capacity for attention and memory, significantly improve.[36] Compared to children, adolescents have a much greater ability to apply selective attention (focusing on relevant information and screening out irrelevant information), and to divide their attention (focusing on more than one task at a time).[37] An individual's capacity for short and long term memory also increases during adolescence.[38] Short term memory capacity and working memory is important for allowing storage while comprehending written words, spoken language, and space for

analysing, reasoning and making decisions.[38] Long term memory is necessary for storing accumulated knowledge, making it easier to acquire new information and form patterns, categories and associations.[19 22]

Piaget's stages of development also indicate that after about 12 years of age, a young person's cognitive abilities start to evolve from the purely concrete and logical thinking of childhood (Concrete Operations), into more abstract and complex thought processes observed in adolescence and adulthood (Formal Operations).[24 39] Abstract thinking refers to purely mental thoughts or processes which are not experienced directly through the five senses.[19 40] It involves the capacity to think about concepts which cannot be physically observed, such as time or morality. Complex thinking describes the young person's ability to see the greater complexity of a situation or to think about an idea from multiple perspectives.[19] It allows the detection of underlying meaning in everyday language such as the use of metaphors or sarcasm. Metacognition, or the ability to 'think about thinking' is also important for monitoring thoughts related to learning, problem solving and social relationships.[41]

More recently, theories regarding continued cognitive development in young adulthood (beginning at around 20 years of age) have emerged, and this developmental stage is referred to as Post-formal thinking.[42] The cognitive processes which are said to define Post-formal thinking are pragmatism and reflective judgement.[19] Pragmatism is described as the ability to realise that logical thinking has practical constraints when applied to complex and ambiguous real-life situations.[42] It recognises that sometimes non-logical, social factors must be considered when attempting to solve problems.[43] This ability is complemented by dialectical thought, which allows for the possibility that there may be more than one answer or solution to a problem, and that a number of perspectives may each have their own merit.[44] This type of thinking differs to the

thinking of adolescents who often engage in dualistic thought where situations are polarised into right or wrong, with no in-between.[19] Reflective judgement is also part of Post-formal thinking and refers to an individual's capacity to assess the degree to which presented evidence and arguments are logical and coherent.[19] This cognitive ability allows young adults to commit to the beliefs they regard as most valid, while still remaining open to re-evaluating their beliefs if new evidence is presented.[19]

The global burden of AYA cancer

Incidence and prevalence

Adolescents and young adults account for 0.5-2% of the total cancer population reported to be diagnosed with invasive cancers worldwide.[17 45 46 47] However, as most of the world's AYA population reside in developing countries, it is likely many more AYAs may experience cancer but are never diagnosed, let alone treated.[2] The world Incidence of cancer in AYAs has increased over the last 25 years, and is currently around 1102 per million for 15-29 year olds in the United States (US).[45] Approximately three times more AYAs are diagnosed with cancer over the age of 15, than children under the age of 15 years, and approximately half of all AYAs are diagnosed between the ages of 25 and 29 years.[45 46] In Australia, the incidence of cancer in AYAs aged 15 to 25 years in 2004 was 0.9% of all new cases diagnosed.[48]

Types of cancer

The types of cancer most frequently diagnosed in AYAs, and their distribution, is unique to this age group and differs to child or older adult cancer populations.[7 10 46] For example, carcinomas make up 80% of all cancers diagnosed, but are only attributable to 16% of AYA cancer types.[49] Furthermore, lung, breast, colorectal and bladder cancers which contribute to over half of all adult cancers, only account for around 2%

of AYA cancers.[45 50] The high rates of leukaemia observed in early childhood also steadily decline as the age of AYAs increases.[45]

Cancer distributions within the AYA population can also vary from early adolescence to early adulthood. For this reason Birch proposed that AYA cancer types fit into one of three groups: late paediatric cancers; true AYA cancers; and early onset adult cancers.[7 51] Late paediatric cancers refer to those cancer types that generally occur in younger children such as Wilms' tumour, rhabdomysarcoma, and neuroblastoma, whereas early adult onset cancers reflect those more commonly observed in older adults such as melanoma and thyroid cancer.[7] In contrast, true AYA cancers have their peak incidence between the ages of 13 and 24 years, and include Hodgkin's lymphoma, Ewing's sarcoma, osteosarcoma and testicular and ovarian cancers.[45 50]

In terms of overall prevalence by cancer type, the US National Cancer Institute (NCI) Surveillance, Epidemiology and End Results (SEER) AYA Monograph reported that the most common cancers for AYAs aged 15-29 years from 1975-2000 were lymphomas (20%), melanoma/invasive skin (15%), testicular/male genital (11%), thyroid/endocrine (11%), cervical/ovarian/female genital (9%), brain/central nervous system (CNS) (6%), and leukaemia (6%).[45] This prevalence is mirrored by epidemiological data in other countries[10 47 50-52] In Australia, the pattern of distribution is almost identical except for a slightly higher prevalence of melanoma.[16 53]

Risk factors

For young people under the age of 30 years, most cancers appear to occur spontaneously and are unrelated to behavioural or genetic risk factors.[45 46] A very small proportion of cancers diagnosed in AYAS have been linked to an environmental or biological risk factor. Some examples include cancers of the vagina and cervix (linked

to prenatal maternal use of diethylstilbestrol), melanoma (linked to ultraviolet light exposure), cervical cancer (linked to human papillomavirus (HPV)), Karposi sarcoma and Non-Hodgkin's Lymphoma (linked to human immunodeficiency virus (HIV)), and Hodgkin's lymphoma and Burkitt lymphoma (linked to Epstein-Barr virus).[45] However, the proportion of all diagnosed cases in AYAs that are directly attributable to these risk factors is small.[45 46]

There is some evidence to suggest specific cancer types may vary by socio-economic and geographic locations. Alston and colleagues analysed data on 35,291 young people aged 13-24 years and diagnosed with cancer between 1979 and 2001.[49] They found the incidence of chronic myeloid leukaemia (CML) and cervical cancer were significantly higher in areas of lower socio-economic status.[49] In contrast, the incidence of lymphoma, melanoma, gonadal germ cell tumours and cancer of the central nervous system (CNS) were much higher in more affluent areas, showing that socio-economic and environmental factors may play a role in the development of certain cancers in AYAs.[49] Gender may also play a part in the occurrence of cancer, with males up to 1.2 times more likely to be diagnosed with cancer during adolescence and young adulthood than females.[7 45 52]

Mortality and survival rates

Cancer in AYAs results in approximately 134,000 deaths worldwide, each year.[52 54 55] However, advances in treatment mean that between 73-82% of AYAs diagnosed with cancer will now survive at least five years post-diagnosis.[6 10 17 45 47 52] Gender seems to play a role. For example, the five year survival of all 13-24 year old AYA cancer survivors diagnosed between 1979 and 2001 in England was significantly better for females (73%) than males (69%), although the differences were small.[17] In Australia, the five year relative survival rate for AYAs aged 10-24 years of age was also higher for

females (85%) compared to males (81%).[56] Prognosis is somewhat dependent on cancer type, with thyroid cancer (99%), melanoma (97%), testicular cancer (94%) and Hodgkin's lymphoma (94%) having the highest five-year relative survival in Australia.[56] In contrast brain cancer (69%), bone cancer (61%), and leukaemia (54%) have the lowest survival at five years.[56]

Compared to children and adults, AYAs have had the smallest increase in survival rates over the past 30 years.[16 45] Some aspects of survival may be related to the demographic, behavioural, and psychosocial characteristics of AYAs. For example, a lack of private health insurance is thought to be associated with a longer time to diagnosis for AYAs.[57] However, delays in diagnosis may also be related to the invincible attitude displayed by young people during adolescence and young adulthood, or a general unawareness of symptoms.[58] Symptoms of cancer in AYAs often include: persistent or progressive fatigue, growth of masses, swelling of glands, abnormal discharges, pain or swelling of joints, increased inter-cranial pressure, neurological changes or changes in skin or moles.[46] Because some of these symptoms may overlap with other physical changes which occur during adolescence, such as increased hours of sleep and changes in skin, delays in the recognition and diagnosis of cancer in AYAs may occur.[46 59]

Lower inclusion or recruitment into clinical trials may have also contributed to a lack of improvement in survival rates for this group.[46 60] There is evidence to suggest that patients, including young people, who are treated on clinical trials may have better outcomes than those who are not.[61] However, for AYAs aged 15-24 years only around 20% are entered into clinical trials compared to 56% of children aged 5-14 years.[7]

Common treatments for AYA cancer

The most common treatment for the types of cancer diagnosed in AYAs include surgery, radiotherapy and chemotherapy.[15] AYAs diagnosed with cancer generally have fewer comorbid medical conditions compared with older adults (65 years and older), and therefore can tolerate higher doses and more aggressive treatment.[46] The acute side-effects of treatment can be defined as those which occur immediately following cancer treatment (days or weeks) and are usually reversible. In contrast, long-term physical side-effects are those which have a late onset (months or years) and are often irreversible.[62] Such side-effects can affect not only the young person's physical health, but also their psychological well-being.[62]

Surgery

Surgery is a common part of treatment for cancer in AYAs. However, as solid tumours differ in their biology, location and degree of metastases the purpose and process of surgery varies from patient to patient.[63] Potential reasons for surgery can include: tumour and regional lymph node biopsies; tumour resection; and the insertion of vascular catheters to allow for the delivery of chemotherapy.[64] Given the low rate of comorbid health conditions, surgery is often performed with less risk in AYAs than with adults and small children.[46] However, as with most surgical procedures post-operative side-effects such as pain, infections or haemorrhaging can occur.[63]

Radiotherapy

Radiotherapy involves the external or internal administration of radiation to destroy cancer cells and has been used successfully as a local, targeted treatment for cancer in AYAs.[65] Radiotherapy can be given to young people pre- or post-surgery to assist in tumour resection and to destroy any residual disease. It can also be used before, during and after chemotherapy, or as an alternative to other forms of treatment.[66] The

radiation dose and length of treatment is determined by the young person's age, tumour type, tumour location, stage of disease and degree of metastases.[65] Although most tumours will diminish with exposure to sufficiently high doses of radiation, doses must be moderated to prevent damage to normal tissue.[64] Radiation can permanently damage major organs such as the lungs, kidneys and liver.[65] Bone marrow is also highly radiosensitive, and AYAs may require bone marrow or stem cell transplantation if severe damage occurs.[66] Even with low doses of radiation, side-effects from radiotherapy are common. Acute side-effects can include skin rash, skin desquamation (weeping and peeling) and mucositis (painful inflammation and ulceration of the mucous membranes lining the mouth or gastrointestinal tract).[67] Long-term side-effects in AYAs have also been observed and include cognitive deficits, inhibited growth of bones, cardio-vascular disease and infertility.[65] Infertility is of particular concern when the site of radiation is related to the reproductive system (uterus, ovaries and testes).[46]

Chemotherapy

Chemotherapy is a systemic (whole body) treatment whereby anti-cancer agents are given either intraveneously or orally to a patient with cancer.[62] The anti-cancer agents interfere with the metabolic pathways of malignant cells to stop them from dividing. However, as the cytotoxic agents used are usually non-selective, pathways for healthy cells may also be affected.[67] Anti-cancer drugs can be either synthetic (e.g. alkylating agents and antimetabolites) or from natural sources such as plants and micro-organisms (e.g. topoisomerase inhibitors and tubulin-binding agents).[64] The dose of the agent is decided based on the cancer type, level of metastases and the patient's body weight or surface area.[64] Chemotherapy is usually most effective when a number of different agents are used in combination, and each agent is administered to the maximally tolerated dose.[62]

Like radiotherapy, chemotherapy can be used as a neoadjuvant (pre- surgery or radiotherapy), or adjuvant (post- surgery or radiotherapy) treatment.[66] Prior to the use of adjuvant chemotherapy, between 80-95% of young people with solid tumours treated with surgery or radiotherapy alone experienced a recurrence of the disease.[62] However, as most anti-cancer agents have difficulty making their way into the cerebrospinal fluid (CSF), treating brain tumours with chemotherapy can be very difficult.[62]

Some of the most common acute side-effects of chemotherapy include nausea, vomiting, mucositis, diarrhea, constipation, alopecia (loss of hair) and myelosuppression (suppression of bone marrow).[64] Peripheral neuropathy (nerve damage) can also occur. Motor nerve damage can result in muscle weakness, muscle cramping, or wrist and foot drop, while sensory nerve damage leads to numbness, tingling, burning, pain and loss of tendon reflexes.[62] In order to overcome some of the acute toxicity of chemotherapy and its related symptoms rescue agents, such as antimetics to relieve nausea and vomiting, have been developed.[64]

Chemotherapy can be detrimental in the long-term as well, damaging organs, prohibiting physical development, and affecting reproductive function. As with radiotherapy, bone marrow or stem cell transfusion following chemotherapy may be required if myelosuppression occurs.[66] AYAs treated with high dose alkylating agents during puberty are at greatest risk of later infertility.[62] The long term risk of developing a secondary cancer (predominantly leukaemia) following chemotherapy is also increased, particularly if the young person received radiotherapy as part of their treatment as well.[62]

Paediatric versus adult treatment centres and treatment regimes

Due to the age at which the cancer diagnosis occurs, AYAs may receive their treatment from either a children's or adult hospital, and sometimes both. The majority of patients aged over 15 years are treated in adult hospitals or oncology centres.[46 68 69] The choice of treatment centre is usually based on the recommendation or decision of the referring general practitioner (GP) or specialist (e.g. dermatologist or neurologist).[46] However, there is some evidence to suggest that AYAs may be more suited to either an adult or paediatric treatment centre, depending on their cancer type.

Adolescents and young adults with more paediatric-related cancer types, such as non-Hodgkin's lymphoma and leukaemia, have been shown to have better survival outcomes when treated at specific paediatric cancer institutions compared to adult focused hospitals.[60] Adolescents and young adults with Ewing's sarcoma were also found to have better outcomes when treated at paediatric cancer centres.[70] This may be because adolescents treated at children's hospitals are more likely to be enrolled in clinical trials (35%) than those treated at adult hospitals (12%).[71] In contrast, AYAs with more adult-related cancers, such as colorectal, breast, and ovarian cancers, or melanoma, may have better outcomes when treated at adult hospitals by clinicians who specialise in adult cancer.[46]

A similar outcome has been found for the treatment regime or protocol used to treat cancer in young people. For example, AYAs with cancers such as acute lymphoblastic leukaemia (ALL), who are treated on paediatric clinical trials have considerably better outcomes than those treated on adult clinical trials.[72-74] Adolescents and young adults with lymphoma and rhabdomyosarcoma also showed better outcomes when paediatric therapy protocols were used, compared with adult treatment regimes.[75]

Despite these findings, a study conducted through the Utah cancer registry revealed that only half of all patients aged 15-19 years with leukaemia or soft tissue sarcomas had been seen by paediatric providers at a children's hospital.[68] This was also true for one third of patients with lymphoma or brain tumours.[68] These results were not significantly related to geographical distance to the children's hospital, and it is possible that factors such as age eligibility (many children's hospitals will not see patients over 16 years of age), available equipment size, provider preference and referral patterns contributed.[68] Patient preferences and the maturity of the young person may also influence choices. Some young people may perceive that paediatric treatment centres are for infants, and feel that at this stage in their life they identify more with adults.[68] Teenage Cancer Units (TCUs) have been recommended as a solution to this problem,[7] however the low incidence of AYA cancer in many countries would make these units unfeasible.

Treatment adherence in AYAs

Treatment adherence in AYA cancer patients, as with all cancer patients, is strongly aligned with prognosis.[4] However, as adolescence and young adulthood is often a time of autonomy and rebellion against parents and authority figures, gaining adherence in AYA patients can be challenging.[24] Studies of non-adherence of AYAs with cancer treatment in has been reported to be consistently higher than with children or adults, ranging from 2-59%.[76-79] Low levels of adherence appear to be especially prevalent in AYAs who have a family background of mental illness, low socioeconomic status, or cultural or linguistic differences.[76 77 80] The young person's accuracy of understanding regarding the course of the disease, and their communication with parents and physicians, has also been associated with adherence.[77 80] Whether non-adherence is intentional (rebellion) or non-intentional (confusion or lack of understanding), it can lead to poorer treatment outcomes for AYAs.

The impact of cancer on AYA development

Although AYAs diagnosed with cancer are likely to experience some of the same psychosocial concerns reported by all cancer survivors, they can also experience additional problems due to their unique stage of personal development.[24 81 82] As described earlier, adolescence and young adulthood is an important time in an individual's life when milestones such as: physical and sexual development; exploration of identity, intimacy, independence, and autonomy; and cognitive maturation, are supposed to be reached.[24 39] A life-threatening disease such as cancer can severely interfere with the achievement of these milestones.[24]

The acute psychosocial impact of cancer and its treatment

The acute psychosocial impact of cancer and its treatment can be substantial in young people. It has been reported that up to 60% of AYAs being treated for cancer experience pain,[83] with one study showing almost half of all adolescents report pain as the most common symptom experienced.[84] Other common side-effects experienced by AYAs include vomiting and nausea.[60 85] These physical symptoms can lead to high levels of distress in young people, affecting adherence with treatment and limiting their ability to engage in normal activities such as social gatherings with peers, or attending school or work.[86] Subsequently, adolescent rites of passage such as gaining a drivers licence, staying out late with friends, and dating are all affected.[24] The inability to participate in these activities can mean that experiences necessary for the formation of identity (trying out different ideologies, occupations and relationships) are inadequately achieved.[24 87 88]

Poor self-esteem and self-image can also occur due to significant changes in physical appearance during cancer treatment. Loss of hair, scars from surgery, changes in weight and visible central venous catheters are reported to be highly distressing to AYA

cancer patients.[24 60 89] Barrera and colleagues investigated health-related quality of life (HRQOL) and found that that parents perceived the self-esteem of adolescents (12-17 years) diagnosed with cancer to be lower than that of children (6-11years) with cancer.[90] Poor self-esteem related to physical appearance can impact on the achievement of intimacy with friends. Although some young people manage to socialise with their peers, many avoid social interaction and miss the opportunity to form close relationships, leading to feelings of isolation and lonliness.[24 87 89]

The achievement of independence requires that an adolescent has the ability to break free of their dependency on their parents.[24] However, being diagnosed with cancer and receiving treatment can mean that a young person actually becomes more dependent on their parents, often needing practical, financial and emotional assistance. Adolescents and young adults may be dependent on their parents for transport to hospital, covering the costs of medication, and providing assistance with dressing, eating and mobility.[24] Parents may also become over protective of the young person or act as the primary decision maker with regard to providing informed consent for cancer treatment.[24] As the young person is attempting to establish their own autonomy, this can lead to feelings of frustration and resentment, and difficulties in family relationships.[24]

The development of a sexual identity and knowledge of sexual health issues which usually occur during this stage of development can also be impacted by cancer and its treatment. Sexual health knowledge is often obtained from personal development classes at school and through interaction with peers, however many AYAs undergoing treatment are unable to attend school or have limited contact with people their own age.[24] Additionally, a combination of concerns related to body image and limited

opportunity to form close, intimate, and romantic relationships can further challenge the development of a sexual identity.[24 81]

The long-term psychosocial well-being of AYA cancer survivors

Cancer survivorship has been defined as beginning from the time of cancer diagnosis and includes individuals at various stages of the disease trajectory, including patients currently receiving treatment and survivors who are at any period of time post-treatment.[91 92] Survivorship is acknowledged as a growing area of concern due to the long term psychosocial morbidity experienced by many cancer survivors and the subsequent increased burden on the health care system.[93] Increasing survival rates for AYAs mean that a greater number of young people are living longer with the psychosocial sequelae of their cancer diagnosis and treatment.[13 17 56] However, although the five year survival rates for AYAs are promising, overall shorter life expectancy is still an issue.[94-96] While mortality due to the recurrence of the same cancer is decreasing, young cancer survivors are still at risk for other causes of early mortality attributable to their treatment, such as the development of new cancers and cardiac or pulmonary failure[6] Adolescents and young adults exposed to radiotherapy, or chemotherapy involving alkylating agents, epipodophyllotoxins or anthracyclines, appear to be most at risk.[6]

Ongoing physical, psychological and cognitive issues have also been reported in long-term survivors. A large proportion of young people (36%) still experience pain post-treatment,[83] and some long-term survivors are more likely to report suffering from fatigue compared with healthy controls.[97] Issues related to self-perception and body image can continue long-term, even when the physical changes related to treatment, such as hair loss, are no longer present.[24 98] Young cancer survivors also report

psychological problems such as anxiety, depression, mood disturbances, anger and feelings of hopelessness.[15 99]

Some AYAs experience cognitive impairment as a long-term side-effect of treatment which can impact on employment and educational attainment.[18 100-102] Barrera and colleagues undertook a population-based study of 800 child and adolescent cancer survivors aged 6-16 years and matched peers who had never had cancer.[103] Parental report showed that, compared with peers who had never had cancer, young people with cancer were significantly more likely to repeat a grade, attend learning disability or special education classes and experience educational or other school problems.[103] The highest levels of educational problems were reported for survivors of CNS tumours, leukaemia and neuroblastoma.[103] A study by Lansky also reported that almost half of all young people who had been diagnosed with cancer had to alter their academic plans, including making a reassessment of their career goals.[104]

Concerns related to fertility are also prevalent among AYA cancer survivors.[101 105 106] Infertility or reduced fertility may arise as a result of previous chemotherapy,[24] or radiotherapy of the ovaries or testes.[65] The effects of these cancer treatments are especially pertinent to female survivors because, although semen cryopreservation is a possibility for male cancer patients, equivalent forms of oocyte cryopreservation for females are generally less successful and not widely available.[24 107] Despite cryopreservation being possible for pubertal males prior to treatment, it may not be routinely offered due to their young age, or may not seem to be an important issue at the time. Therefore, treatment-related infertility is still of concern to some young men.[108 109] If AYA cancer survivors are able to conceive, they may still be concerned about the likelihood that the child may have health problems due to their past cancer experience and the treatment they received.[110]

Despite the potential long-term consequences associated with cancer treatment, many AYA cancer survivors (between 80 and 100% in some studies) report being in good or excellent physical health.[111-113] Further, a review of seven studies showed that a high proportion of long-term survivors also report reasonable psychological health.[111] Nevertheless, compared with other young people their age, long-term AYA cancer survivors are observed as having higher rates of obesity, anxiety, depression and suicide, and lower rates of marriage and parenthood.[111 114 115] Many survivors report barriers to obtaining work and have greater difficulty obtaining health and life insurance than their siblings who have never been diagnosed with cancer.[116] Therefore, it is not only the length of survival in AYAs which is important, but also the quality of that survival.[94]

The psychosocial impact of cancer on AYAs and their family

The diagnosis of cancer in a young person not only has an impact on the individual, but can also lead to changes in family dynamics and cause significant upheaval in the lives of family members including parents, siblings, and significant others.[24 82 87]

For many AYAs with cancer, it is their parents who are responsible for providing physical, emotional and financial support while the young person undergoes treatment.[87 117] For some parents it is as though they are caring for an infant again, and assisting the young person to wash, dress and eat can be physically demanding.[118] Losing their independence with such basic tasks can be extremely difficult for AYAs, and in their efforts to assist, parents can often be the target of the young person's anger or frustration.[117] Like the young person, parents may also experience a sense of isolation, as caring for the young person and taking them to treatment can be enormously time consuming.[119] To be able to take care of the young person, some

parents may also have to give up work which can add additional financial pressure and stress to an already disrupted family.[120]

Parents may need to provide emotional support not only to the young person with cancer, but also to siblings who can similarly suffer from psychosocial distress.[119 121] Siblings may experience a range of feelings including anxiety and resentment.[82] Fears about their own health and the likelihood of being diagnosed with cancer are high.[117] Anger about having to alter their life to fit around the young person and their treatment, or jealousy about the amount of attention the young person with cancer receives from their parents, is reasonably common.[82] These feelings, although understandable, might also increase the sibling's sense of guilt as they might feel they should be more concerned about the young person, and less concerned with their own needs.[82]

Given that many AYAs are in their late teens and early to mid twenties when diagnosed with cancer, a small proportion are married or have partners or significant others.[122] Many partners of AYA cancer survivors will have a similar experience to that reported by parents, such as taking on more responsibility around the home, caring for the partner with cancer and providing emotional support.[123] Problems surrounding the potential infertility of the young person following treatment, and the health of any future children may also be an area of concern for partners of AYAs.[123]

Palliative care and end of life issues for AYAs

Palliative care refers to care provided to patients who are no longer responsive to curative treatment and often focuses on pain and symptom control, as well as addressing the psychological, spiritual and social needs of the individual.[124] Due to their developmental stage and cognitive and emotional abilities, AYAs with advanced stage disease are generally able to recognise that their cancer is incurable.[125] The reality of

facing one's own mortality can be overwhelming for AYAs, and is often accompanied by feelings of fear and distress.[126] Families of patients with a life-limiting illness may also experience feelings of grief,[127] and the commencement of palliative care may be particularly distressing due to the sense of untimely death for the young person.[126] A general principle for the delivery of palliative care to AYA cancer patients is to support and involve members of the patient's family.[124] However, due to the developmental stage of AYAs, a balance may be required between the level of patient versus family involvement in decision-making regarding end-of-life care.[128] Adolescents and young adults are at a period of their life where they are attempting to establish autonomy. Therefore, if AYAs are not provided with the opportunity to contribute to decisions regarding their own care, conflict with parents, partners or other individuals may occur.[129]

Palliation can be provided to AYAs with advanced cancer in a number of settings including the home, day care centres (e.g. patient attends a palliative care centre during the day) or as an inpatient (e.g. care is provided full-time in an institution such as a hospital or hospice).[124] A large proportion of cancer patients, including AYAs, generally state a preference for wishing to receive palliative care in their own home.[126] [130] [131] For AYAs, the hospital setting may contribute to a lack privacy, isolate them from their peers or induce anxiety responses associated with receiving previous cancer treatment at the centre.[132] Palliative care delivered in an inpatient setting may also be more distressing to AYAs, as it is likely that the young person would be surrounded by people who are at a more advanced stage of life.[133] This situation may lead to increased feelings of isolation for AYAs and contribute to diminished quality of life.[125] Palliative care delivered in the home may provide a more age appropriate setting for AYAs, however this setting may also create other challenges for the young person and their family in terms of disruption to normal family routines and the burden placed on

caregivers.[124] For this reason additional practical and emotional support may need to be provided to AYAs and their families who are entering the phase of end of life care.[130]

Need to reduce the psychosocial burden of cancer for AYAs

Adolescents and young adults with cancer may face a myriad of physical, psychological and social challenges. In order to improve the psychosocial outcomes of this group it is important to understand and address the diverse needs and challenges they experience.[134] A first step in achieving this may be to assess the extent to which existing health services are currently meeting the needs of AYAs across a range of relevant items and domains.[135] Such an assessment should help to identify not only the areas of greatest need for AYAs, but also particular sub-groups within the population who may require special assistance.

Conclusion

Adolescent and young adult cancer survivors are a unique population who may experience both immediate and long-term psychosocial deficits as a result of their diagnosis and treatment. Short-term concerns of AYAs related to pain, vomiting, nausea, changes in physical appearance and feelings of frustration have all been reported. Adolescents and young adults are at risk of experiencing long-term consequences such as early mortality, the development of new cancers, cardiac or pulmonary failure, fatigue, anxiety, depression, anger, feelings of hopelessness, cognitive impairment, infertility, higher rates of obesity and suicide, and lower rates of marriage and parenthood. In addition, periods of missed schooling, difficulties gaining employment and problems obtaining health insurance are common to this group. The impact of cancer and its short- and long-term consequences can mean that the achievement of important milestones for AYAs such as the development of positive self-esteem and body-image, attainment of independence and autonomy, and the

opportunity to form close, intimate and romantic relationships, can all be affected. To improve outcomes for AYAs, gaps in the delivery of care and the ways that the health system respond to their needs, should be identified. An assessment of how well the health system meets the needs of this group may be an important first step in identifying which areas require attention and where efforts should be focussed to improve psychosocial health outcomes for this group.

CHAPTER 2

Measuring the psychosocial health of adolescent and young adult (AYA) cancer survivors: a critical review

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Data from this chapter have been published (Appendix 2.1):

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Chapter 2

Measuring the psychosocial health of adolescent and young adult (AYA) cancer survivors: a critical review

Introduction

Chapter 1 identified some of the physical, psychological and social challenges facing young people who are diagnosed with cancer. Measuring the prevalence and predictors of psychosocial health in this population is the first step towards developing interventions and targeting resources aimed at improving psychosocial outcomes.[1 2] Given their unique experiences, adolescents and young adults (AYAs) require psychosocial health measures specifically developed and validated for them in order to assess their well-being accurately. This chapter aims to review currently available measures of psychosocial health for AYA cancer patients and survivors.

Considerations when selecting health assessment measures

One of the most important factors to consider when selecting an appropriate health assessment measure is its performance characteristics, or psychometric properties.[3] Psychometrics can be described as the science of measuring qualitative or abstract phenomena such as personality or well-being, using a scale or instrument in an attempt to quantify them.[4] The psychometric properties of the scale therefore refer to how reliably and validly these concepts are able to be captured.[3 5] A measure of psychosocial health with poor psychometric properties may be unable to detect important changes in health outcomes following an intervention, lead to an over- or under-estimation of disease-related morbidity, or fail to predict future health outcomes or identify those individuals who are most at risk.[3] In addition to the psychometric quality of the measure, it is important to consider whether to use: 1) generic or disease-

specific scales; 2) uni-dimensional or multi-dimensional instruments; or 3) proxy or self-report measures.

Generic versus disease-specific measures

Generic health assessment measures enable the collection of health-related information from patients with any type of disease or condition, as well as from members of the general population.[6] For AYAs with cancer, generic health measures allow comparisons with young people who have other illnesses, such as asthma, or with population norms that have been established using reference data from healthy individuals.[3 7 8] Generic measures are also an effective method of collecting data from AYA cancer patients who have co-morbid diseases or conditions.[3 9] However, a criticism of generic health measures is that they may fail to reflect the scope of issues relevant to the AYA cancer experience (i.e. content validity may be a problem).[10] Furthermore, generic measures may not be able to detect small differences between important clinical sub-groups, such as patients receiving particular types of cancer treatment.[3]

In contrast, disease-specific measures are exclusively designed for use with patients with particular illnesses or health conditions.[3] The content of domains and items is often more specific, allowing the measure to be more sensitive to disease- and treatment-related changes.[3 6 11] Disease-specific measures can not only be specific to the wider cancer population, but can also be developed to assess the health outcomes of AYAs with particular tumour types or symptoms.[12 13] For young people with cancer this means that the domains and items in these measures are more relevant to the particular physical, psychological or social problems they face. As with generic health measures, disease-specific measures are known to have some disadvantages. For example, when a young person with cancer has an additional, non-cancer-related

illness or health problem, multiple disease-specific instruments may need to be administered, increasing the burden on respondents.[3] In these situations, the use of a core generic health instrument supplemented by a disease-specific module has been advocated.[10 12 14] However, due to the cost involved in measure development and the need for large samples for psychometric testing, it is generally not feasible to develop disease-specific measures for rare or low-incidence cancer types.[3]

Uni-dimensional versus multi-dimensional measures

Uni-dimensional measures of health are those which focus on one aspect or characteristic of a disease (e.g. physical symptoms) to the exclusion of all other disease-related deficits.[15] However, for populations such as AYAs with cancer, morbidity is often experienced across many facets of the individual's life, with many problems occurring concurrently and influencing each other in potentially complex ways.[15] For example, the physical limitation caused by an amputated limb due to cancer may be a measurable aspect of health on its own. However, a uni-dimensional measure may fail to capture the impact of this on a young person's body image, emotional status and self-esteem, and subsequently their level of participation in social activities.[16]

It has been proposed that understanding the impact of a disease on a young person's life requires the examination of all influencing factors simultaneously, so that an overall picture of psychosocial well-being can be ascertained.[15] The World Health Organization (WHO) has defined health as encompassing physical, mental and social dimensions of well-being, all of which are linked and contribute to the global health of the individual.[17] This definition necessitates the use of multi-dimensional rather than uni-dimensional measures, in order to develop a comprehensive assessment of the health of an individual. Multi-dimensional measures are able to assess numerous domains of

health, such as role, emotional, social, physical and spiritual functioning.[15] Due to the interaction of all these areas on both the long- and short-term outcomes of cancer patients and survivors, the use of multi-dimensional measures across the disease trajectory has been recommended.[2 15 18]

Proxy versus self-report measures

Another area of debate in the measurement of psychosocial outcomes in AYA cancer survivors is the use of measures completed by a proxy, compared with measures completed by the young person.[19] A proxy is an external rater who provides information regarding the health of a patient on the patient's behalf.[20] Proxy measurement can allow for the collection of data from patients who are too ill or do not have the necessary cognitive or literacy skills to participate alone.[19]

For AYAs with cancer, proxies are generally parents, partners, health care providers or teachers.[3 20] However, a number of disadvantages in using proxies to assess the psychosocial health of patients with chronic diseases have been identified. While parents of AYAs with cancer have been shown to have good agreement with the young person regarding observable behaviours such as physical functioning, poor agreement regarding social and emotional issues is often reported.[19] This may be especially true for parents of AYAs whose responses may be influenced by their own levels of stress or mental health, and may represent the parent's expectations or hopes for the young person, rather than a true reflection of the young person's psychosocial well-being.[9] When the proxy is a health care provider, there may also be large discrepancies between the proxy's and the patient's perceptions of needs.[21] While health care providers, including nurses and physicians, appear to have acceptable agreement with AYA patients regarding physical concerns, agreement for psychological and social aspects of health is poor, with staff frequently over-estimating impairment in these

areas.[22] For this reason, proxy ratings can only ever be viewed as an approximation of a patient's actual well-being and should always be interpreted with caution.[23]

Self-report instruments obtain the individual patient's perspective regarding health and well-being, rather than relying on the interpretation of an external rater or observer.[20] An advantage of self-report measures is that they do not require interviewers or health professionals to administer them, thus saving on costs and resources.[24] For this reason self-report measures are usually acceptable to health care providers. This may be important if the purpose is ongoing screening or data collection in a clinical setting.[23] Self-report measures are also user-friendly, and often allow the patient some flexibility in the time and location of completion.[24] Low response rates with self-report measures have been highlighted as an area of concern.[24] However, due to the low agreement between young people and proxies regarding perceptions of psychosocial health, self-report measurement is generally preferred for AYAs who are able to complete a measure without assistance.[6 13]

Types of health measurement scales

Measurement scales with all of the above characteristics have been developed for the assessment of psychological health or well-being in cancer patients. However, the purpose of measurement and function of the data collected will vary depending on the type of measure used. The four main types of health measurement scales include symptom, satisfaction, quality of life and perceived need scales.

Symptom scales

A symptom can be defined as a change in the normal appearance or function of the body and is an important aspect in understanding the morbidity caused by a disease or its treatment.[13] Symptom scales are useful for measuring the impact a symptom has on

a person in terms of its frequency and intensity, and the level of distress or suffering caused.[13 25] For AYA cancer patients and survivors, measures for symptoms including pain, nausea, vomiting, fatigue, loss of appetite and constipation have been developed,[25-30] with a number of these scales being uni-dimensional.[30] While uni-dimensional measures may be able to explore the characteristics of a particular symptom in greater detail, they can fail to discern how a symptom interacts with other important aspects of the cancer experience. Many quality of life scales for cancer populations include items related to symptoms as part of the broader psychosocial assessment.[25] However, the reported presence of a symptom may not necessarily indicate that assistance for the symptom is needed. Therefore, additional enquiry or follow-up by health care professionals may be required.[30]

Satisfaction scales

Satisfaction can be defined as the degree to which an individual's experiences match their levels of expectation.[31] It is important that patients are satisfied with the care received, as satisfied patients are more likely to continue to utilise medical services and adhere to treatment regimes.[32-35] Satisfaction measures have been promoted as useful for providing health systems and health care providers with feedback regarding areas of improvement and resource allocation.[31] However, a limitation of satisfaction measures is that they generally only enquire about satisfaction with existing services, and do not have the scope to capture what additional resources may be needed.[36] Furthermore, as with symptom scales, measures of satisfaction ask patients to indicate if they are experiencing a problem (i.e. dissatisfaction), but do not address whether or not the patient wants or requires assistance.[37] This means that if low satisfaction is reported, patient responses may fail to indicate in concrete and specific terms what action, if any, needs to be taken to remedy the problem.

Satisfaction scales are also subject to a number of social and psychological biases.[38] Across many samples and settings, high levels of satisfaction have been reported,[39 40] and positive response bias due to social desirability in measures of satisfaction has been observed.[38] This may occur because patients are unwilling to complain or criticise the health care system or providers for fear of consequences such as unfavourable treatment or withdrawal of care.[36 41] Alternatively, answers may be influenced by gratitude, or a patient trying not to appear uncooperative or demanding.[38] The demographic characteristics of some patient groups also appear to be important. Patients who are older[42-45] or who have lower levels of education[35 45] tend to report higher levels of satisfaction. Therefore, reported levels of satisfaction may be a product of a patient's expectations, values or past experiences, rather than the actual quality of care received.[34]

Quality of life measures

Quality of Life (QOL) measures assess an individual's view of current abilities and lifestyle compared with expectations.[9] Perceptions of QOL will vary among individuals depending on their hopes, goals, ambitions, culture, values and past experiences.[9 46] Health-related quality of life (HRQOL) specifically measures the degree to which factors such as disease, treatment and health policies impact on a person's QOL. However, the terms, QOL and HRQOL, are often used interchangeably in the health literature.[7]

As QOL generally covers the broad domains of physical, psychological and social well-being, multi-dimensional measures are needed to capture these constructs.[9] Quality of life measures can have a number of uses in the clinical setting: 1) they assist health care providers to identify previously hidden health problems, especially those which may be non-observable, such as psychological or social concerns;[47 48] 2) they help to

chart a patient's progress over time;[49] 3) they act as a prompt to initiate and facilitate communication between health care providers and patients;[47 50] and 4) they provide patients with opportunities to disclose information about their overall well-being in a non-threatening way.[49] However, like many other types of instruments, QOL measures are unable to determine which health problems cancer patients would most like help with.[51] Instead, a judgement is made by an expert, usually a health care provider or researcher, that the absence or presence of an issue indicates areas where help must be required.[31] While often used to assess the QOL of populations or sub-groups of patients (e.g. patients receiving a particular treatment), QOL measures have also been used to assess individual well-being. Some randomised controlled trials (RCTs) have indicated that routine QOL assessment can result in improvements in the QOL and satisfaction of cancer patients compared to usual care,[50 52] while other studies found no significant differences between the QOL of patients who received routine QOL assessment and those who did not.[53]

Needs measures

A measure of unmet or perceived need allows patients to indicate whether or not they have a need in a particular area, to prioritise the needs which are most important and to indicate the level of assistance required.[31] Unmet needs measures have the potential to provide more useful information for the purpose of informing service delivery improvements than either QOL or satisfaction measures. If needs are met it implies that existing services are satisfactory; if needs are unmet it can indicate that some additional action or resource may be required.[54] The urgency or priority of the need can also be determined depending on the magnitude or level of need reported.[37 55 56] Need assessments have the potential to provide the same benefits as QOL measures (outlined above).[49] However, they have the advantage of removing the assumptions made by health care providers or researchers regarding the type of help a

patient wishes to receive.[57] They can also assist with the identification of those individuals with the highest levels of need, so that interventions can be targeted to those at greatest risk of psychosocial morbidity.[55]

Previous reviews of measures of psychosocial health for AYA cancer survivors

A small number of previous reviews have been conducted to assess the availability and performance characteristics of measures developed for AYAs with chronic diseases including cancer.

Linder[13] conducted a review of physical symptom measures developed between 1988 and 2003 for children and adolescent cancer patients up to 18 years of age. Nineteen symptom scales were identified. Of these, five scales measured a single symptom, with four measuring nausea and vomiting and one measuring fatigue.[13] Fourteen scales measured multiple symptoms, with five assessing symptoms as part of a larger quality of life assessment.[13] Psychometric properties for the identified scales were also reported, with the majority deemed to be reliable and valid.[13]

In 1996, Spieth and Harris[7] assessed HRQOL measures designed for children and adolescents. Of the six measures identified, four were generic measures of HRQOL and two were disease-specific.[7] One measure, the Pediatric Oncology Quality of Life Scale (POQOLS),[58] was developed for young people with cancer. However, data collection was by parent proxy rather than self-report.[7] A subsequent review of QOL measures for chronically ill children and adolescents up to 20 years of age was undertaken by Eiser and Morse.[9] They identified 43 measures developed between 1980 and 1999.[9] Twenty-four measures were disease-specific, and five of these were developed for young people with cancer. However, only two of these measures were self-report.[9]

A more recent review of generic and disease-specific HRQOL measures for children and adolescents up to 19 years of age was conducted by Solans and colleagues.[6] The review identified 94 measures that were developed between 1980 and 2006 (30 generic QOL measures and 64 disease-specific measures).[6] The 64 disease-specific measures represented 27 different illnesses, with eight of these measures designed specifically for children and adolescents with cancer. While six of these measures were self-report, only three were available in English.[6] Two of the measures were for young people with any type of cancer,[59] [60] whereas one measure was specific to bone marrow transplant patients.[61] All three measures reported acceptable psychometric properties for at least one form of reliability or validity.[6]

Findings from these previous reviews reveal a strong focus on measures for very young children and adolescents up to 20 years of age. However, no reviews focussing on measures developed for AYAs were found. Furthermore, the reviews only focused on symptom scales and measures of QOL. There is a need for a review of instruments developed for AYAs up to 30 years of age with cancer which assesses all multi-dimensional measures of psychosocial health, not just QOL.

Aims

The aim of this review is to critically examine the psychometric properties of multi-dimensional, self-report measures developed to assess the psychosocial health of AYA cancer survivors.

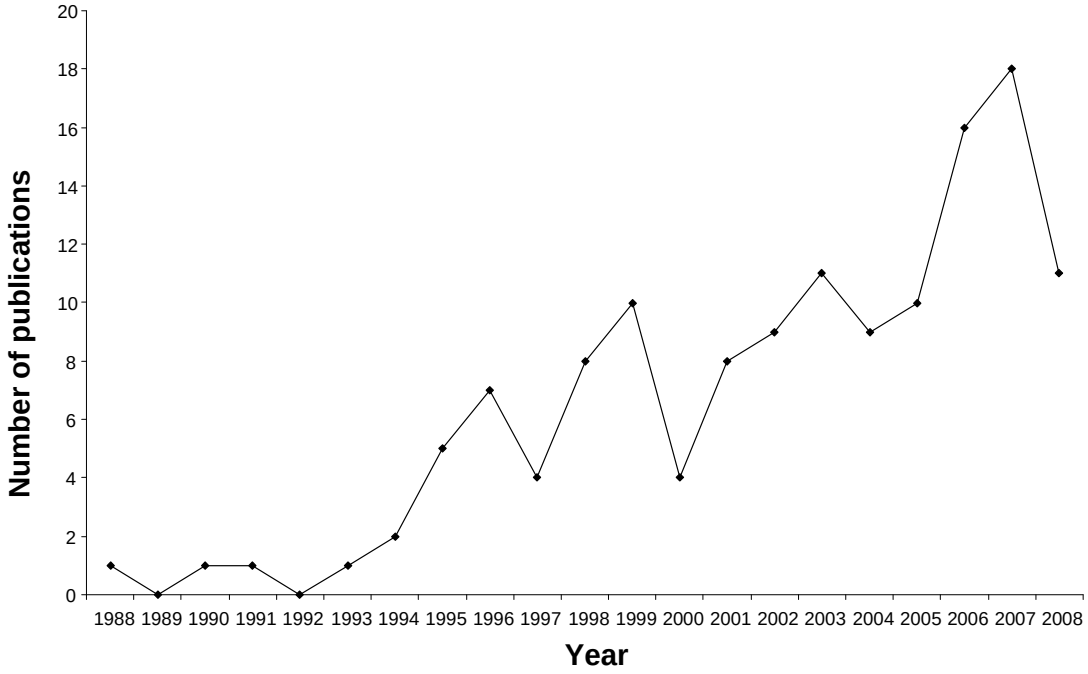
Methods

Database search to identify relevant publications

Medline, PsycINFO, EMBASE and CINAHL databases were searched to identify publications which described the development of measures for assessing psychosocial outcomes in AYA cancer survivors. These databases were chosen as they all provide extensive coverage of journals in the field of cancer research.

The database search was performed using the following combinations of keywords: [neoplasm or cancer or oncol*] and [adoles* or teenager or young adult or youth] and [perceived need* or unmet need* or quality of life or psychosocial or distress] and [develop* or questionnaire or survey or measure or scale] and [psychometric or reliability or validity or acceptability]. Results of the search were limited to the English language and covered the ten years from 1998 to 2008. This time-frame was selected as a preliminary search of Medline for all AYA-related psychosocial research without a year limitation revealed that there had been minimal (<17%) research output in the field prior to 1998 (Figure 2.1), with only one publication before 1988 identified (i.e. one publication in 1976). Appraisal of papers published prior to 1998 also revealed that no measures met the inclusion criteria (outlined below).

Figure 2.1: Number of publications related to the assessment of psychosocial well-being in AYA cancer survivors by year (1988-2008)



Duplicate publications and publications which did not specifically describe the development, psychometric properties or acceptability of a measure were excluded. Full-text articles of the remaining publications were obtained and reviewed to identify relevant measures.

Inclusion and exclusion of measures

While AYAs are commonly defined as aged 15-30 years, definitions in the literature vary.[62-64] Therefore, an inclusive approach was employed whereby scales developed for use with young people less than 15 years but with an upper age limit between 15 and 30 years were included (e.g. 12-20 years). Similarly, scales developed for use with populations older than 15 years but less than 30 years were included (e.g. 16-28 years).

Measures were included in the study for coding if they were: 1) quantitative; 2) developed or validated in English; 3) multi-dimensional and measured at least the following three psychosocial domains – physical psychological and social; 4) cancer-specific; 5) assessed the well-being of patients or survivors; 6) developed specifically for AYAs (i.e. they included participants aged between15 and 30 years in the sample); and 7) completed by self-report.

After identifying measures which met all of the inclusion criteria, a second search of all databases by “measure name” was performed to ensure that all publications related to each identified measure were obtained.

Measure coding

Sample characteristics

In order to assess the psychometric properties of a measure accurately, the sample used to develop the measure should be described.[65] Measure development papers were examined to determine whether the following sample characteristics were reported: inclusion and exclusion criteria; setting; response rate; sample size; age of participants; proportion of male and female participants; cancer type; and cancer treatment stage.

Table 2.1: Summary of criteria used to review measures

Psychometric Property	Criteria
Reliability	
Internal consistency degree to which responses to all items on a scale are consistent [66]	Calculated correlations for total scale and domains [67] – Cronbach's alpha (α) >0.70 [65 67] – Kuder–Richardson 20 (KR–20) >0.70 [65 67]
Test-retest reproducibility of scores on a scale over repeated administrations [67]	Second administration within 2-14 days [68] Calculated correlations for total scale, domains and items [69] – Cohen's kappa coefficient (κ) >0.60 [67] – Pearson correlation coefficient (r) >0.70 [65 67] – Intra-class correlation coefficient (ICC) >0.70 [65 67]
Validity	
Face subjective assessment of whether a scale "appears" to measure what it is designed to measure [66]	Considered reasonable by those who administer/complete it [66]
Content degree to which the content of a scale is representative of the issue being measured [66]	Reported item selection process [65 67] Content assessed by experts [65 67] Reported which aspects of the measure were revised [65 67]
Construct way in which the internal structure of a scale relates to other conceptual constructs [67]	Stated hypothesis about correlations between measures [67] – Convergent (r) >0.40 or Divergent (r) <0.30 [70] Calculated correlations between known groups [65] Performed factor analysis [67] – Eigenvalues >1 [71]
Criterion how well a scale agrees with existing "gold standard" measurement of the same issue [67]	Provided rationale for "gold standard" measure [67] Stated type of criterion validity (concurrent or predictive) [66] Reported proportions [67 72] – Sensitivity: % with issue correctly classified [67 72] – Specificity: % without issue correctly classified [67 72]
Responsiveness sensitivity of a scale to detect clinically important change in an outcome/behaviour over time [65 72]	Reported floor/ceiling effects [73] – <5% of respondents have highest or lowest score [73] Reported magnitude of change [65] – Effect size >0.5 [65 67 72]
Acceptability level of burden placed on those who complete the measure [65]	Reported response rate, missing items, reading level, time to complete [65]
Feasibility level of burden placed on those who administer the measure [65]	Reported perceived time to administer, score, interpret [65]
Cross-cultural adaptation conceptually, linguistically equivalent and displays similar psychometric properties to the original form [65]	Confirmed reliability and validity reflects the original version [65]

Psychometric properties

Measures were coded using pre-defined criteria considered important for scale development and health outcome measurement.[65-74] The rigour of each measure was assessed against the following criteria: reliability; validity; responsiveness; acceptability; feasibility; and cross-cultural adaptation. These criteria are summarised in Table 2.1.

Reliability

Internal consistency

Internal consistency is the degree to which responses to all items on a scale are consistent.[66] Cronbach's alpha (α) >0.70 is generally considered to indicate acceptable internal consistency in both continuous and dichotomous scales.[65 67] For dichotomous scales, the Kuder–Richardson formula 20 (KR–20) can also be used.[67] Measures were reviewed to determine whether internal consistency had been examined using either of these two methods, and whether it had been calculated at both the total scale and domain levels.

Test-retest

Test-retest reliability refers to the reproducibility of scores on a scale over repeated administrations.[67] The same individual completes the same measure on two separate occasions, and the correlation between the two sets of scores is calculated. The length of the interval needs to be not so short that the individual can recall previous responses, and not so long that large permanent changes (rather than small day-to-day fluctuations) may have occurred.[66] The generally agreed interval for the second administration of a measure when attempting to determine test-retest reliability is 2-14 days.[68]

Correlations can be calculated at the overall scale, domain or individual item levels. Item-level test-retest correlations are the best reflection of measure stability.[69] Cohen's kappa coefficient (κ) is used for ordinal and nominal scales, with a kappa of 0.60 commonly accepted as the minimum value.[67] The Pearson product-moment correlation coefficient and the intra-class correlation coefficient (ICC) can be calculated for interval scales.[65] Pearson correlations or ICC scores >0.70 are generally considered acceptable.[65 67] Measures were coded to indicate whether test-retest reliability had been examined, the length of the interval between each administration, and the correlations at the total scale, domain and item levels.

Validity

Face

Face validity is a subjective assessment of whether an instrument "appears" to measure what it is designed to measure, to those who administer it and to those who complete it.[66] Measures were coded as demonstrating face validity if both potential administrators (health care professionals or researchers) and subjects (young adult cancer survivors) agreed that the measure was plausible.[67]

Content

Content validity involves determining the degree to which the content of a scale is representative of the issue being measured.[66] Systematic analysis of scale items should be performed to ensure that all aspects of the issue are covered in the correct proportion.[66] Measures were reviewed to determine if they reported the following criteria for content validity: 1) how measure items were selected (e.g. review of the literature, existing measures, interviews, focus groups); 2) by whom the measure content was assessed (e.g. number of experts, qualifications); and 3) what aspects of

the measure were revised after expert assessment (e.g. comprehensiveness, redundancy).[65 67]

Construct

Construct validity refers the way the internal structure of a measure relates to other conceptual constructs.[67] Measures were reviewed to determine whether construct validity was examined using any of the following commonly used methods: 1) the relationship between the newly developed measure and other existing measures (i.e. convergent/divergent validity); 2) the ability of the measure to distinguish between groups with known differences; and 3) factor analysis.[67] Pearson's correlation coefficients of (r) >0.40 and (r) <0.30 are generally used to indicate convergent and divergent validity respectively.[70] For "known-groups" discriminative validity, the comparison of scores on the measure between groups with known differences should be reported.[65] In factor analysis, inter-correlations between responses on a scale are grouped into factors which appear to measure common themes.[67] Each factor is distinct from another so that groups are homogenous and unrelated.[67] The principal component extraction method with Eigenvalues set at >1 is commonly used.[71]

Criterion

Criterion validity is an assessment of how well a scale agrees with existing "gold standard" measurement of the same issue.[67] It measures the ability of a scale to predict an individual's performance or behaviour, either in the present (concurrent validity) or future (predictive validity).[66] The proportion of people who actually have the characteristic of interest who are correctly classified as such by the measure (sensitivity), and the proportion of individuals who are negative for the characteristic who are correctly classified as such by the measure (specificity) should be reported.[67 72]

Responsiveness

Measures were also coded to indicate whether responsiveness was examined. Responsiveness refers to the sensitivity of a measure to detect clinically important change in an outcome or behaviour over time.[65 72] It is often expressed in terms of effect size, which estimates the magnitude of change.[65] The statistical methods used to calculate the effect size should be reported, with a standardised effect size (SES) of 0.5 generally considered a reasonable threshold of change.[65 67 72] When there are ceiling or floor effects in pre- or post-test scores on a measure, however, difference scores may not be meaningful.[73] To ensure that a scale is able to detect clinically important change, less than 5% of respondents should achieve the lowest (floor) or highest (ceiling) possible scores.[73]

Acceptability

Acceptability was assessed in terms of the burden placed on those who complete the measure.[65] Measures were coded to indicate whether they reported acceptability in terms of response rate, missing items, completion time and reading level.[65]

Feasibility

Feasibility refers to the level of burden placed on those who administer the measure.[65] Measures were assessed to determine whether the times needed to administer, score or interpret the measure were reported.[65]

Cross-cultural adaptation

Cross-cultural adaptations of measures were assessed to determine whether the adapted versions were conceptually and linguistically equivalent and displayed similar psychometric properties, compared with their original forms.[65]

Quality assurance of coding

In the present study, one coder used the inclusion and exclusion criteria to identify measures for inclusion in the review. A second coder cross-checked 15% of the measures to confirm their inclusion and exclusion status. The psychometric criteria of all included measures were reviewed by the first coder and checked by the second.

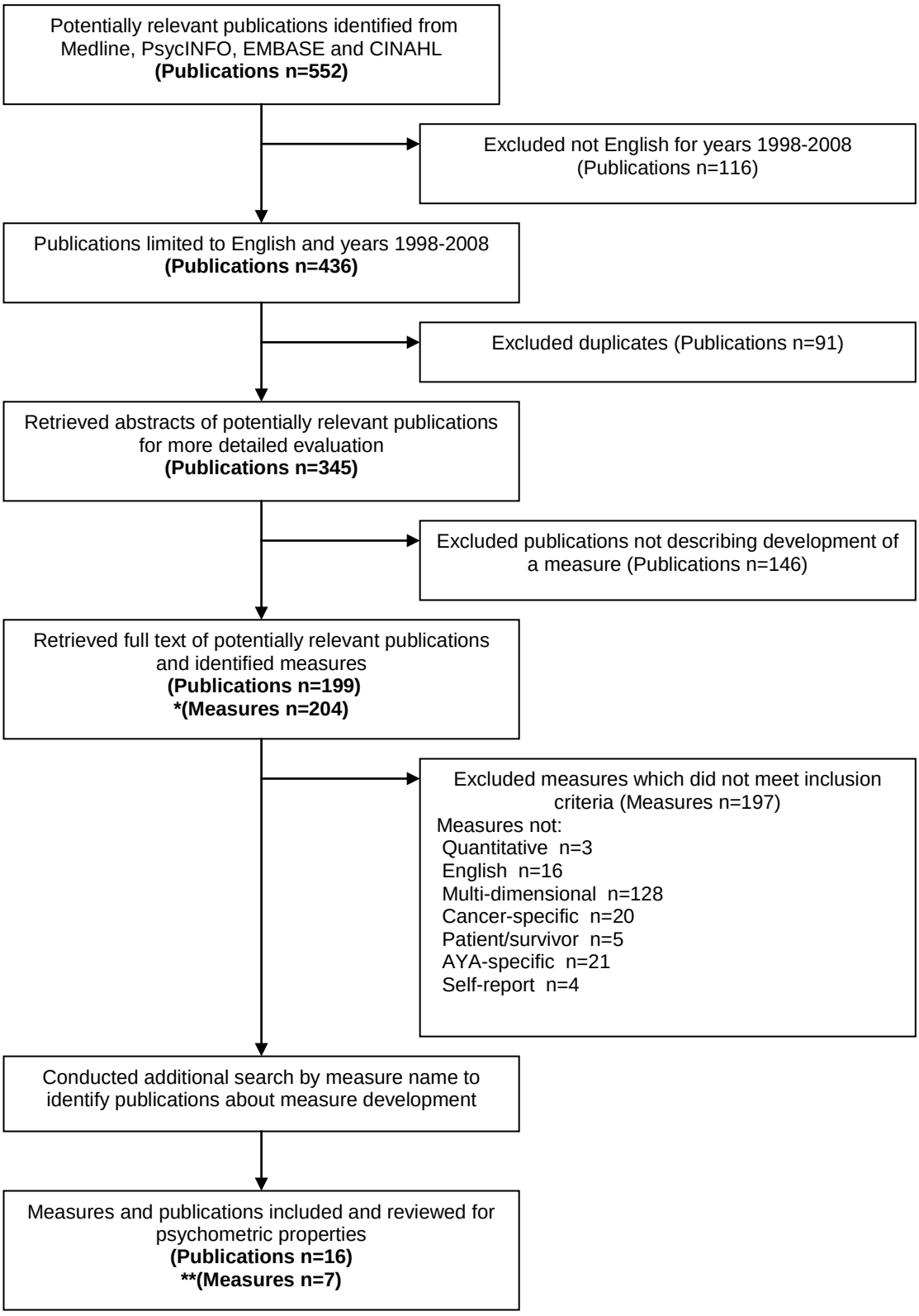
Results

Database search to identify relevant publications

The initial search of the Medline, PsycINFO, EMBASE and CINAHL databases identified a total of 552 publications related to assessing psychosocial outcomes in AYA cancer survivors, with 436 papers published in the previous ten years (1998-2008). Of these 436 publications, 91 were duplicates and 146 did not describe the development of a measure. The remaining 199 publications described the development of 204 measures.

One hundred and ninety-seven measures did not meet the inclusion criteria (Figure 2.2), leaving seven measures to be included in the review. These included the following measures: Adolescent Quality of Life Instrument (AQoL);[75 76] Minneapolis–Manchester Quality of Life Instrument (MMQL) – Adolescent Form;[59 77 78] Pediatric Quality of Life Inventory (PedsQL) 3.0 Cancer Module Child and Adolescent (C&A) Forms;[14 79-81] Quality of Life – Cancer Survivors (QOL–CS) validation in childhood cancer survivors;[82] Pediatric Cancer Quality of Life Inventory – 32 Short Form (PCQL–32);[60 83 84] Pediatric Cancer Quality of Life Inventory (PCQL) Modular Approach;[12] and Perceived Illness Experience Scale (PIE).[85 86]

Figure 2.2: Flowchart of the publication and measure inclusion and exclusion process



**Some publications described the development of more than one measure*
***Development of some measures were reported across more than one publication*

Six measures were developed in the United States (US) and one in the United Kingdom (UK).[85 86] A description of each measure's domains and number of items is presented in Table 2.2.

Table 2.2: Items and domains of measures included in the review

Measure	Items	Domains	Description	Reference
AQoL Adolescent Quality of Life Instrument	16	5	normal activities, social/family interactions, health status, mood, meaning of being ill	75 76
MMQL Adolescent Form Minneapolis–Manchester Quality of Life Instrument	46	7	physical, psychological, social and cognitive functioning, body image, outlook on life, intimate relations	59 77 78
PedsQL 3.0 Cancer Module (C&A) Pediatric Quality of Life Inventory Child and Adolescent Forms	27	8	pain and hurt, nausea, procedural anxiety, treatment anxiety, worry, cognitive problems, perceived physical appearance, communication	14 79-81
QOL–CS Quality of Life – Cancer Survivors	41	4	physical, psychological (distress and fear), social and spiritual well-being	82
PCQL–32 Pediatric Cancer Quality of Life Inventory – 32 Short Form	32	5	Disease- and treatment-related symptoms, physical, psychological, social and cognitive functioning	60 83 84
PCQL Modular Approach Pediatric Cancer Quality of Life Inventory Modular Approach	23	5	(core) physical, psychological, social (modules) pain, nausea	12
PIE Perceived Illness Experience Scale	34	9	physical appearance, interference with activity, peer rejection, integration in school, manipulation, parental behaviour, disclosure, preoccupation with illness, impact of treatment	85 86

Sample characteristics

Overall, reporting of the sample accrual method and the sociodemographic and clinical characteristics of participants for each measure was comprehensive (Table 2.3). Of the seven measures, three did not report a response rate, one did not describe the inclusion and exclusion criteria, and one measure did not report the proportion of male and female participants or cancer type.

All measures were developed using samples recruited through hospitals or medical centres. Sample sizes ranged from 41 to 291 participants, and age of participants ranged from 5 to 28 years (mean range of 10.9-21.8 years). The proportion of males and females was reasonably equal. For the majority of studies, the greatest proportion of young people had been diagnosed with leukaemia. Cancer treatment stage ranged from newly on treatment to 3-27 years post-diagnosis.

Psychometric properties

An overall summary of the psychometric properties reported for each measure can be seen in Table 2.4.

Reliability

Internal consistency

Table 2.5 shows that five measures had at least one domain with poor internal consistency (Cronbach's alphas <0.70), although their total scale internal consistency was adequate. Two measures (AQoL and PCQL Modular Approach) did not report internal consistency at the domain level. However, the pain and nausea modules of the PCQL Modular Approach had Cronbach's alpha values >0.70.

Table 2.3: Reported sample characteristics for each measure

Measure	Sample characteristics							
	Exclusion	**Setting**	**Response rate (%)**	**Sample size (n)**	**Age (yrs)**	**Gender (%)**	**Cancer type (%)**	**Cancer treatment stage (%)**
AQoL[75]	Reported	Haematology/ oncology clinic	95	75	9-20 mean 12.4	M (55) F (45)	Leukaemia (50) Bone/joint (17) Lymphoma (9) Neurological (9) Hodgkin's (5) Other (9)	In treatment (55) Pre/post-treatment(45)
MMQL Adolescent Form[59]	-	Nine hospitals	-	268	13-20.9 median 16.6	M (56) F (44)	Leukaemia ALL (37) Leukaemia AML (8) Hodgkin's (11) Non-Hodgkin's (11) Brain (6) Other (27)	On therapy (41) Off therapy >1 year (59)
PedsQL 3.0 Cancer Module (C&A)[14]	Reported	Haematology/ oncology centre and Centre for Cancer and Blood Diseases	-	220	5-18 mean 10.9	M (56) F (44)	Leukaemia (50) Brain (7) Non-Hodgkin's (6) Hodgkin's (3) Wilm's (6) Other (28)	On treatment (54) Off treatment <1 year (18) Off treatment >1 year (28)
QOL–CS[82]	Reported	University medical centre	53	176	16-28 mean 21.8	M (43) F (57)	Leukaemia (30) Brain/CNS (11) Lymphoma (21) Wilm's (10) Sarcomas (16) Other (11)	3-27 years post-diagnosis (100) (average 13.3 years)
PCQL–32[83]	Reported	Three paediatric cancer centres	89.5	291	8-18 mean 11.78	M (61) F (39)	Leukaemia ALL (44) Leukaemia AML (6) Hodgkin's (6) Non-Hodgkin's (9) Other (35)	Newly on treatment (37) Relapsed; on treatment (8) Remission; off treatment (11) Long-term off treatment (44)
PCQL Modular Approach[12]	Reported	Three paediatric cancer centres	89.5	291	8-18 mean 11.78	-	-	On treatment (45) Off treatment (55)
PIE[85]	Reported	Children's cancer unit	-	41	8-24 mean 14.6	M (49) F (51)	Leukaemia ALL (68) Wilm's (15) Sarcomas (12) Non-Hodgkin's (5)	Maintenance treatment (41) Follow-up only (59)

Table 2.4: Summary of psychometric properties reported for each measure

Measure	Internal consistency	Test-retest reliability		Face/content validity	Construct validity			Responsiveness	Acceptability	Cross-cultural
		Time	ICC		Convergent/ divergent	Known-groups	Factor analysis			
AQoL	√	√	-	√	-	√	√	-	√	-
MMQL Adolescent Form	√	√	√	√	√	√	-	-	-	√
PedsQL 3.0 Cancer Module (C&A)	√	-	-	√	√	√	-	-	√	√
QOL–CS	√	-	-	-	√	√	√	-	√	-
PCQL–32	√	-	-	√	√	√	-	√	√	-
PCQL Modular Approach	√	-	-	√	-	√	-	√	√	-
PIE	√	-	-	√	√	√	-	-	√	-

Table 2.5: Coding of reliability criteria for each measure

Measure*	Internal consistency		Test-retest reliability		
	n	Cronbach's alpha α > 0.70	n	Administration Period	Intra-class correlation ICC >0.70
AQoL[75]	75	Total scale = 0.77 No domains reported	17	Pre-weekend to post-weekend Post-weekend to one month[76]	-
MMQL Adolescent Form[59]	397	Total scale = 0.78 6/7 domains >0.70 Physical = 0.88 Psychological = 0.83 Social = 0.81 Cognitive = 0.89 Body image = 0.80 Outlook = 0.85	87	Two-week interval	Total scale = 0.71 5/7 domains >0.70 Physical = 0.90 Cognitive = 0.88 Body image = 0.73 Outlook = 0.76 Relations = 0.81
PedsQL 3.0 Cancer Module (C&A)[14]	220	Total scale = 0.72 6/8 domains >0.70 Pain and hurt = 0.70 Nausea = 0.79 Procedural anxiety = 0.82 Treatment anxiety = 0.79 Worry = 0.74 Cognitive = 0.76	-	-	-
QOL–CS[82]	176	Total Scale = 0.87 5/6 domains >0.70 Physical = 0.81 Psychological = 0.82 Fears = 0.88 Social = 0.76 Spiritual = 0.78	-	-	-
PCQL–32[60]	291	Total scale = 0.91 4/5 domains >0.70 Disease/treatment = 0.83 Physical = 0.78 Psychological = 0.76 Cognitive = 0.81	-	-	-
PCQL Modular Approach[12]	281	Total scale = 0.83 No domains reported All modules >0.70 Pain = 0.82 Nausea = 0.71	-	-	-
PIE[85]	41	Total scale =0.84 2/9 domains >0.70 Manipulation = 0.70 Parental behaviour = 0.73 Total scale = 0.91 4/9 domains >0.70 Peer rejection = 0.79 Parental behaviour = 0.71 Preoccupation illness = 0.73 Food = 0.70 [86]	41	-	-

**Data taken from the publication referenced in the Measure column unless otherwise referenced within the table.*

Test-retest

Two measures examined test-retest reliability. For both studies, the second administration of the measure was within the recommended time-frame of 2-14 days. Only the MMQL Adolescent Form reported the intra-class correlations for the two administrations, with five of the seven domains having intra-class correlations >0.70.

Validity

Face/content

Table 2.6 shows that six of the seven measures explored face and content validity. Most studies involved both AYA cancer survivors and health care providers in the development of the measures.

Construct/criterion

Five measures examined convergent or divergent validity against other existing measures. Hypotheses were supported by correlations >0.40 or <0.30. All of the measures were able to discriminate between known groups. Factor analysis was performed for two measures. No measures were examined for criterion (concurrent or predictive) validity.

Responsiveness

Only two measures reported floor and ceiling effects (Table 2.7). None of the studies reported a measure's ability to detect clinically important change over time.

Table 2.6: Coding of validity criteria for each measure

Measure*	Face/content validity	Construct validity		
		Convergent *r* > 0.40 Divergent *r* < 0.30	**Known groups (discriminative)**	**Factor Analysis Eigenvalues > 1**
AQoL[75]	Assessed by survivors Review of literature Item wording, redundancy Pilot test (n=7)	-	Receiving treatment (n=41) Not receiving treatment (n=34) P = 0.000	6 factors represented 66.5% of variance
MMQL Adolescent Form[59]	Assessed by survivors Focus group (n=20) Interviews (n=20) Pilot 1st (n=10) Pilot 2nd (n=10)	Child Health Questionnaire – Child Form Hypotheses supported 42 correlations >0.40	Healthy adolescents (n=129) On therapy (n=110) Off therapy (n=158) P < 0.05 for 4 domains	-
PedsQL 3.0 Cancer Module (C&A)[14]	Adapted from Pediatric Cancer Quality of Life Inventory (PCQL), PedsQL 1.0 Cancer Module, and PedsQL	PedsQL 4.0 Generic Core Scale PedsQL Multidimensional Fatigue Scale Hypotheses supported 34 correlations >0.40	On treatment (n=106) Off treatment <1 year (n=41) Off treatment >1 year (n=73) P < 0.05 for 3 domains	-
QOL–CS[82]	-	Cancer Specific Worry Scale Psychosocial Worry Scale General Health Worry Scale Hypotheses supported 9 correlations >0.40	Other condition (Y=28, N=148) After-effects (Y=86, N=90) Income (<$25K=36, >$25K=127) Gender (F=101, M=75) Marital status P < 0.05 for 5 factors	6 factors represented 56.2% of variance
PCQL–32[60]	Assessed by survivors Review of literature Interviews/pilot test Item wording, relevance, redundancy, reduction [83]	Children's Depression Inventory State-Trait Anxiety Inventory – 32 (Child) Social Support Scale (Child/Adolescent) Self-Perception Profile (Child/Adolescent) Child Behaviour Checklist Hypotheses supported 10 correlations >0.40 15 correlations <0.30	On treatment (n=125) Off treatment (n=156) P < 0.05 for total scale and 3 domains	-
PCQL Modular Approach[12]	Adapted from the PCQL long form and PCQL–32	-	On treatment (n=125) Off treatment (n=156) P < 0.05 for the core and symptom modules	-
PIE[85]	Assessed by survivors Interviews (n=15) Item reduction	Rotterdam Symptom Checklist Functional Disability Inventory Restrictions Scale Psychological Symptoms Hypotheses supported 9 correlations >0.40 20 correlations <0.30 SF-36 Functional Evaluation Scale Hypotheses supported 38 correlations >0.40 44 correlations <0.30 [86]	Younger children Older children Maintenance treatment Completed treatment P < 0.05 for 2 domains	-

**Data taken from the publication referenced in the Measure column unless otherwise referenced within the table.*

Table 2.7: Responsiveness, acceptability and feasibility of each measure

Measure*	Responsiveness	Acceptability	Cross-cultural
AQoL[75]	-	Response rate 95% Reading level Flesch–Kincaid grade 6.2 [76]	-
MMQL Adolescent Form[59]	-	-	Anglicised for UK and shortened to the MMQL–29 [77] Internal consistency in an online format [78] Reliability and validity demonstrated
PedsQL 3.0 Cancer Module (C&A)[14]	-	Missing items 0.5%	Initial development in English and Spanish [14] Adapted to Brazilian, German and Australian cultures [79-81] Reliability and validity demonstrated
QOL– CS[82]	-	Response rate 53%	-
PCQL–32[84]	On treatment Floor 1.6-20.0% Ceiling 0% Off treatment Floor 1.9-32.7% Ceiling 0%	Response rate 89.5% Missing items 0.01%	-
PCQL Modular Approach[12]	On treatment Floor 0-3.1% Ceiling 3.1-22.9% Off treatment Floor 0-1.9% Ceiling 10.6-35.6%	Response rate 95% Missing items 0.01% Reading level Flesch–Kincaid grade 1.8	-
PIE[85]	-	Reading level Flesch–Kincaid grade 7	-

**Data taken from the publication referenced in the Measure column unless otherwise referenced within the table.*

Acceptability and feasibility

Table 2.7 also shows that the acceptability of the measures was poorly described, with only four measures reporting missing items and only three measures reporting reading levels. The reading levels that were reported, however, were appropriate for the

population group. Feasibility, i.e. the time needed to administer, complete and score the measure, was not reported for any of the measures.

Cross-cultural adaptation

Two measures, the MMQL Adolescent Form and PedsQL 3.0 Cancer Module (C&A), have been adapted for cultures other than the United States. For the culturally adapted measures, similar reliability and validity to the original measure was reported. The reliability of MMQL Adolescent Form in an online format has also been verified.

Discussion

All of the psychosocial measures developed for AYA cancer survivors included in this review showed high total-scale internal consistency. Only one measure reported test-retest reliability coefficients. Although intra-class correlations were reported for total scales and domains, item-level test-retest correlations were not examined. This can present a problem because while the same overall domain score may be achieved from the first to the second administration, it is possible that the individual item scores that make up the domain score differ between administrations. This may compromise the stability of the measure over time.

Face, content and construct validity for all of the measures were also psychometrically adequate. However, no measures reported predictive or concurrent criterion validity. This may reflect difficulties in identifying an appropriate “gold standard” against which to compare AYA perceptions of their health, or difficulties related to longitudinal study designs, such as cost and participant attrition. The implication of this is that the ability of these measures to predict the risk of future health outcomes in AYA cancer survivors remains unknown.

Reporting of measure responsiveness, acceptability and feasibility was poor. No measures reported ability to detect clinically important change over time, raising questions about the sensitivity of these instruments. Reading level was only reported for three measures. This is of concern because, due to their illnesses, AYA cancer survivors may have missed significant proportions of their schooling.[87 88] Poor readability and comprehension of items may lead to misinterpretation or missing items, thereby reducing the accuracy of results obtained.

Given the absence of findings regarding test-retest reliability, responsiveness or acceptability for all of the identified measures, it is difficult to recommend any of them as outcome measures for use in intervention studies. For some, the unknown ability of the measure to remain stable over time would make it difficult to assess whether changes on the measure were due to the intervention alone. For others, the undetermined responsiveness of the instrument would mean that if no change were observed, this could be either due to lack of sensitivity in the measure or lack of an intervention effect.

Both the MMQL Adolescent Form and the PCQL–32 show promise as measures of quality of life for AYAs. The MMQL Adolescent Form demonstrated good internal consistency (6/7 domains $\alpha > 0.70$) and test-retest reliability at the domain level (5/7 domains ICC > 0.70). The PCQL–32 also reported good internal consistency, validity and acceptability. Further psychometric testing to establish item-level test-retest reliability and responsiveness for the MMQL and test-retest reliability for the PCQL–32 is needed.

A search of the literature did not reveal any other reviews of psychosocial measures for AYA cancer survivors. However, the results of the current review appear to be

commensurate with the findings of similar reviews of measures developed for use with other cancer populations. A review of quality of life instruments for use with adult cancer survivors[89] found that, of the nine measures identified, readability, acceptability, feasibility and predictive validity were rarely or (as in the case of predictive validity) never examined. Of the four measures that examined test-retest reliability, only one reported acceptable test-retest coefficients.[89] A comparable review of needs assessment instruments for cancer patients and their families also found that reading levels and sensitivity to change were poorly examined.[90] Similar trends were reported in a systematic review of instruments for the assessment of fatigue in cancer patients.[91] Of 14 instruments identified, only six were examined for test-retest reliability, and only seven analysed responsiveness.[91] In a review of cancer symptom assessment instruments, only one out of 21 identified instruments reported predictive validity.[92]

It is interesting to note that all of the multi-dimensional measures included in this review assessed quality of life in AYA cancer survivors. However, no measures of perceived need were identified. Using only measures of quality of life may lead to assumptions being made about the type of help AYA cancer survivors would like, rather than allowing individuals to indicate specific areas in which they would like to receive help.[55] [93] In addition, all of the samples used in the development of these measures were recruited through hospitals or medical centres. The extent to which these samples were representative of the broader AYA population, including under-served AYA populations such as those living in rural or remote areas, is unknown.

Limitations

The literature search for this review was conducted using four online publication databases. As the grey literature was not included, it is possible that some relevant measures were missed. However, it is likely that measures identified in this review are

of the best quality as they have been published in peer-reviewed, indexed journals. The step of conducting a second search by measure name would have also minimised the chance that publications related to relevant measures were overlooked.

The definition of AYA cancer survivors used in this review was young people between the ages of 15 and 30 years. However, as a group, the AYA population is not defined well in literature, and ranges from 12 to 40 years.[62-64] To overcome this discrepancy, any measures developed for an age cohort which overlapped the age bracket of 15 to 30 years were included. This may mean that some of the results reported in this review reflect measure performance with individuals outside the AYA definition used for this review.

Conclusion

The psychometric properties of existing quality of life measures to assess the psychosocial well-being of AYA cancer survivors require improvement. The MMQL Adolescent Form and the PCQL–32 have provided the most evidence for their psychometric properties to date. However, without sufficiently robust measures, the prevalence of any reported concerns or issues and the effectiveness of interventions which aim to ameliorate them, remain uncertain. Studies which focus on the test-retest reliability, responsiveness, acceptability, feasibility and predictive validity of the measure are essential. There is also a need to develop a measure of perceived need for AYA cancer patients and survivors. Such a measure is necessary for assessing the prevalence of unmet needs in this population as a whole, as well as for identifying groups of AYAs who are at risk of experiencing high levels of unmet need.

CHAPTER 3

Development of the Cancer Needs Questionnaire – Young People (CNQ–YP): identifying domains and items, and establishing face and content validity

Table of Contents

Chapter 3

Development of the Cancer Needs Questionnaire – Young People (CNQ–YP): identifying domains and items, and establishing face and content validity

Introduction

A critical review of the literature in Chapter 2 revealed that only seven existing measures of psychosocial health were specifically developed for adolescents and young adults (AYAs) who had been diagnosed with cancer. Of these seven measures, all assessed quality of life, and no measures of unmet need were identified. This represents a key gap in the types of psychosocial measures available for this population; although quality of life measures can assess the cancer-related issues a young person is experiencing, they do not indicate whether help for the issue is needed.[1 2] Measures of unmet need can determine both issues for which help is desired, and the level of help required.[1 2] This chapter describes the initial process of development of an unmet needs measure specifically for AYA cancer patients and survivors, and the establishment of face and content validity of the measure.

Ensuring a needs measure for AYA cancer survivors assesses a broad range of experiences relevant to this group

Quality of life measures developed for AYA cancer survivors have included psychosocial items and domains related to the following issues: physical functioning and ability;[3-6] disease- and treatment-related symptoms (e.g. pain and nausea);[4 5 7 8] interference with normal daily activities;[7 9] psychological distress (e.g. mood, anxiety, worry and fear);[3-6 8 9] meaning of illness and outlook on life;[3 7 9] communication;[8] cognitive functioning;[3 5 8] school integration;[7] body image and perceived physical appearance;[3 7 8] social interaction and support;[3-6 9] relationships with parents and other family members;[7 9] intimate relationships;[3] peer rejection;[7] and spiritual well-being.[6] It is

likely that an unmet needs measure developed for this population would need to reflect a similar scope of concerns.

Establishing face and content validity of the unmet needs measure

Following the identification of items and domains to be included in a measure, an important next step is the establishment of face and content validity. As described in Chapter 2, face validity refers to whether an instrument "appears" to measure what it is designed to measure, assessed by those who administer and complete it.[10] Content validity involves determining how comprehensively the content of a scale represents the issue being measured.[10] Steps for establishing face and content validity include: 1) examining previous measures and literature in the area to ensure all relevant topics are covered; and 2) obtaining input from consumers (i.e. patients and their families) and experts (i.e. health care and other professionals) as to whether the measure is comprehensive, relevant and easy to understand.[11 12] It is important that the process of determining face and content validity involves both experts and consumers to ensure the measure does not miss important aspects of the consumer experience.[13] Input from experts and consumers can also help determine how feasible the measure is for those who will administer it, and how acceptable the measure is for those who will complete it.[11]

Aims

The aims of this preliminary research were: 1) to identify domains and items which would form the basis of a measure to assess the unmet needs of young people with cancer; and 2) to establish the face and content validity of the measure.

Methods

Identifying domains and item generation

Database search

A review of the literature was performed in order to derive potential items and domains for the draft measure. Medline, PsycINFO, Embase and CINAHL databases were searched to identify existing measures which had been developed to assess the unmet needs of cancer patients or survivors. The following keyword combinations: [neoplasm or cancer or oncol*] and [perceived need* or unmet need*] and [questionnaire or survey or measure or scale] were used in the database search. As the previous critical review (Chapter 2) revealed that there were no existing measures of unmet need for AYA cancer survivors, perceived needs measures for adult cancer patients and survivors were identified.

Inclusion/exclusion criteria for the literature review

Results of the database search were limited to the English language, but were not limited to any year. Publications reporting the development of any unmet need measures were retained if they were developed specifically for cancer populations and were quantitative, multi-dimensional, not developed for a specific cancer type, assessed the needs of patients or survivors, and could be completed by self-report. Review articles which reported the development or psychometric properties of multiple unmet needs measures for adult cancer patients and survivors[14-16] were also included.

Derivation of items and domains

Existing measures of unmet need which were identified in the published literature and retained included the Supportive Care Needs Survey (SCNS),[1] Cancer Survivors' Unmet Needs measure (CaSUN),[17] Cancer Patient Need Questionnaire (CPNQ),[18 19] Cancer Patient Need Survey (CPNS),[20 21] Needs Evaluation Questionnaire (NEQ),[22]

Patient Needs Assessment Tool (PNAT)[23] and Psychosocial Needs Inventory (PNI).[24] Measures were examined for items and domains considered relevant to AYA cancer patients and survivors. These items and domains were then modified to suit the life stage, language and reading age of young people.

Draft list of items and domains

One-hundred and eight items (Table 3.1) which were conceptually relevant to young people's experience of cancer were listed within the following seven domains:

1) *Daily Life:* focused on practical needs, such as needing assistance to move around, manage jobs and chores at home and maintain a normal life.
2) *Education and Work:* covered needs such as being able to catch up on school work, being able to continue studying, or having the option to work part-time.
3) *Relationships:* covered needs related to friends (e.g. help dealing with being left out by friends), family (e.g. help dealing with all the attention from my parents) and other young people (e.g. being able to talk to someone my own age who has been through a similar experience).
4) *Feelings*: explored emotions (e.g. loneliness, anxiety and frustration), coping mechanisms (e.g. help finding my inner strength) and self-esteem.
5) *Cancer Treatment Centre:* included items related to the location and environment of the hospital or clinic, such as having privacy, leisure space and nearby accommodation.
6) *Cancer Treatment Staff:* focused on staff-patient interactions and processes of care delivery, including the communication skills of staff, involvement in decision-making about treatment, and continuity of care.
7) *Information:* covered both the content of the information (e.g. effects of treatment on fertility) as well as the delivery of the information (e.g. having information presented in different ways).

Format of the draft questionnaire

The list was formatted into a draft questionnaire (Appendix 3.1). The 108 items from all seven domains were spread randomly across the measure. It was anticipated that this would help prompt participants to identify whether they felt any important items were missing. The stem used was "In the last 12 months, I needed…". This 12-month time-frame was initially chosen to allow the small number of participating young people to reflect on a wide range of needs experienced over time, which may be relevant to the larger AYA population. The response scale for the measure was adapted from the Cancer Needs Questionnaire (CNQ) and Supportive Care Needs Survey (SCNS), as this response format has been rated as easy to follow and use by adult cancer populations.[1 25 26] The four response levels were as follows: "No Need" – This has not been a problem for me as a result of having cancer; "Low Need" – This item caused me concern or discomfort. I had some need for help; "Moderate Need" – This item caused me concern or discomfort. I had a moderate need for help; "High Need" – This item caused me concern or discomfort. I had a strong need for help. There was also a "Yes/No" column at the far end of the response scale so that participants could indicate whether or not the need had been met.

Items were worded so that the overall reading age of the measure was suitable for young people with a primary school education (reading age of 10-11 years confirmed by the Flesch–Kincaid Grade Level in Microsoft Word). This reading level was chosen because young people undergoing treatment for cancer may have missed substantial proportions of their schooling. Reading levels below a 12-year-old level are also recommended for questionnaires to ensure answers are reliable and to avoid missing values.[27 28]

Table 3.1: List of 7 domains and 108 items identified from the literature

Domains	Proposed Items
Daily life	
Physical	assistance coping with pain
	assistance with moving around
Travel	assistance getting to social events
	assistance getting to and from the hospital/clinic
Home environment	assistance with taking medication
	assistance with jobs and chores at home
Eating	assistance with eating
	advice about what to eat
Other	assistance participating in social activities
	help maintaining a normal life
Education/Work	
School	assistance catching up with school work
	assistance continuing my education
	assistance in getting back into school
	assistance in not missing school work
	assistance in travelling to and from school
	guidance about further study or future career paths
	to know how much school I would miss
	to know that people at school understood my situation
	to know that there were support services for people at my school
Work	access part-time work
	assistance in getting back to work
	assistance in travelling to and from work
	to know that people at work understood my situation
	to know that there were support services for people at my workplace
Relationships	
Friends	to be able to spend time with and talk with people my own age
	help when I lost my friend(s)
	assistance with asking for support from my friends
	help dealing with being left out by friends
Family	my parent(s)/carer(s) physically near me while in hospital
	support dealing with changes in relationships with my parent(s)/carer(s)
	help dealing with all the attention from my parent(s)/carer(s)
	assistance with asking for support from my parent(s)/carer(s)
Friends and family	to know how to tell family, friends and other people about how I felt
	to know how to tell family, friends and other people about my cancer
Other	to talk to someone my own age who has been through a similar experience
	help dealing with missing the people I care about
Feelings	
Loneliness	help dealing with loneliness
Anxiety − treatment	help with anxiety about going to the hospital/clinic or seeing a doctor
	help with anxiety about treatment side-effects
	help with anxiety about painful medical procedures and treatment
	help with anxiety when waiting for test results
	help dealing with concerns about whether the treatment has worked
Anxiety − disease	help dealing with the possibility of the disease returning
Anxiety − other	help dealing with uncertainty about the future
Depression	help dealing with sadness
	help dealing with feelings of helplessness
Coping	to find enjoyment in my life
	to keep hope for the future
	help finding my inner strength
	help finding meaning in my experience
	help dealing with changes to who I am
Frustration/anger	help dealing with feelings of frustration
Protective	help dealing with my parent(s)/carer(s) being over-protective
Emotions	help thinking about my future
	help dealing with feelings of distress

Domains	Proposed Items
Emotions (cont.)	help dealing with embarrassment
	help with feeling vulnerable
	help to feel secure
	help dealing with boredom
	help dealing with being scared
	help to be independent
Relationships/roles	help dealing with other people's reactions
	help dealing with the worries of those close to me
Other	help focusing on tasks and/or remembering things
	help coping with being unable to do the same things as others my age
Cancer Treatment Centre	
Location of care	easy access to local treatment centres
Access to providers	quick and easy access to someone skilled in cancer care
	quick and easy access to professional counselling
Setting/ resources	privacy while in hospital
	a place to stay with my parent(s)/carer(s) while getting treatment
	leisure space and activities in hospital (e.g. internet access)
	pleasant clinic surroundings
	access to better food while in hospital
Cancer Treatment Staff	
Care management	the same health care provider to take care of me throughout treatment
Communication	friendly health care providers who I could have a laugh with
	health care providers who talked to me, not just my parent(s)/carer(s)
	my parent/carer with me when I talked to health care providers
	to be treated as an individual by health care providers
	to be spoken to by health care providers in a way that I could understand
	to know that my health care providers are part of a team
	health care providers who listened
	health providers who were skilled
	health care providers who were approachable
	to be able to talk to health providers in private, without my parent(s)/carer(s)
	to talk to health care providers informally
Decision-making	to know that I had the right to refuse treatment
	the opportunity to make decisions about when I had appointments
	the opportunity to make decisions about my treatment
	more time to make decisions about my care
Other	assistance with filling out forms
Information	
General cancer	complete information about cancer
	complete and honest information about the long-term impact of cancer
Individual AYA	complete and honest information about my diagnosis
	complete and honest information about how I am going
	complete and honest information about being able to have children
	complete and honest information about the chances of recovery
Treatment/side-effects	information about the effects of treatment on contraception
	complete and honest information about treatment options
	complete and honest information about the side-effects of treatment
	information about what I should do if I notice particular side-effects
	information about how the treatment would be given
	information about what to expect when going to the hospital
	information about what happens after treatment
Services/resources	information about support services and available help
Information format	information presented to me in different ways
Other	to be told by health care providers that the way I felt was normal
	information about relaxation
	information about feelings caused by the experience

Establishing face and content validity of the measure

The draft measure was assessed by a number of consumer and professional groups to establish face and content validity. Groups included AYA cancer patients and survivors and their parents, health care providers, researchers and the general population (e.g. professionals, AYAs and parents who were not affected by cancer or working in the health field).

Sample

AYA cancer survivors and their carers

Young people were eligible to participate if they had been diagnosed with cancer, were currently aged 14 to 19 years of age, and were members of either the Western Australia (WA) or New South Wales (NSW) branches of CanTeen Australia. CanTeen is the peak support organisation in Australia for young people aged 12 to 24 years affected by cancer. A stratified random sample of young people and their carers who met the eligibility criteria was identified from the CanTeen Australia database. The sample was stratified on the basis of the young person's cancer type, age and sex, and attempts were made to balance focus groups by gender, age of parents, cancer type and time since diagnosis.

Health care providers

Twelve health care providers from a regional hospital in NSW were nominated and invited by a paediatric oncologist to join a panel. Members of the panel included one paediatric oncologist, two paediatric haematologists, three oncology nurses, one cancer care coordinator, one social worker, one psychologist, one oncology pharmacist, one physiotherapist and one occupational therapist. All health care providers had experience working directly with the AYA cancer population.

Researchers in the field

Eight researchers in the field of cancer from Australia and Canada were selected on the basis of their relevant expertise with development of measures to assess the psychosocial needs of cancer populations.

General population

A convenience sample of twelve individuals who were known to the research team and were not from a medical or research background was selected. These included professionals, such as school teachers and engineers, as well as parents and young people who had not experienced cancer.

Procedure

Focus groups with AYA cancer survivors and their carers

Approval for the study was granted by the Human Research Ethics Committee of the University of Newcastle (Appendix 3.2). CanTeen sent a letter of invitation, a project information sheet, a "top five needs" sheet (Appendices 3.3–3.5) and a consent form to AYAs and their carers identified from the database. The invitation letter informed participants about the date and location of the focus group, and what they would be asked to do if they attended. Participants were offered incentives (i.e. pizza and movie tickets) in return for their attendance at the focus group. The "top five needs" sheet asked young people to think about the needs which were most important or difficult for them, and then list the top five in the space provided. Prior to attending the focus group, participants were asked to telephone their local CanTeen offices to confirm their attendance. Participants were also asked to complete both the consent form and the "top five needs" sheet beforehand and bring them along to the meeting.

Focus groups were facilitated by either a clinical psychologist or a social worker from CanTeen, and a member of the research team. The meetings lasted for approximately two hours and were split into two parts. In the first hour, each participant was asked to read out the list of top five needs. This process continued until a master list of needs was formed. The facilitators helped the group identify areas of overlap and redundancy. In the second hour, participants were asked to look at the draft questionnaire and provide feedback about the content, wording of questions and ease of completion.

Once data from the focus groups were collected, follow-up packages were posted to participants. These contained a thank you letter (Appendix 3.6), a summary of the feedback from the discussion groups, a re-drafted version of the measure, and a revised response scale and feedback sheet. Participants were asked to provide written comments on the new response scale and return them to the researchers. This feedback was then incorporated into a new version of the measure.

Feedback from the panel of health care providers

Panel members were emailed the new version of the unmet needs measure and the summary of outcomes from the focus groups with AYA cancer survivors and their carers. Providers were asked to review these materials and provide feedback to the research team prior to the panel meeting. Once the suggestions of the provider panel had been incorporated into the measure, a meeting between all members of the panel and the research team was organised so that providers could give feedback on the revised measure. Two members of the research team facilitated the panel meeting. Questions for discussion included whether or not any important topics were missing, which items or domains were most important, and which were least important. To prompt discussion, providers were given a revised list of domains and items for the unmet needs measure, and then asked to consider their content, identify any potential

redundancies and offer suggestions for refinements or additions. Based on this feedback the unmet need measure was further refined. Items and domains which were redundant, unclear or overlapping were revised. After incorporating these changes, the measure was re-checked to ensure that the reading age had not changed substantially.

Feedback from researchers and the general population

Researchers in the field of cancer were sent a copy of the draft questionnaire and asked to provide critical feedback on the domains, items and response scale *via* email. Members of the general population were also asked to provide feedback on the questionnaire in terms of its language and clarity.

Results

Focus groups with AYA cancer survivors and their carers

Six young people participated in the focus groups, three per state. In WA, ten new items were identified by the focus group and were subsequently added to the questionnaire (Appendix 3.7).

In NSW, concerns were raised that the response scale used in the questionnaire was too complex and that the time-frame used, “In the last 12 months”, was not specific enough (Appendix 3.8). It was suggested that focusing on time-frames, “When you were in hospital”, “When you were receiving treatment” or “When you had finished treatment” might be easier to understand. Comments also reflected the need for two questionnaires: one questionnaire for during treatment; and one questionnaire for post-treatment. However, further discussion revealed that determining whether a young person was in treatment or had finished treatment was difficult, and participants’ definitions of treatment differed from those of health professionals and researchers.

Having two different measures would also make it difficult to compare changes in the needs of AYAs with cancer over time. For these reasons it was agreed that one version of the questionnaire that captured both stages of the cancer journey would be the better option.

Following the second round of feedback from the focus groups, the stem of the response scale was changed from "In the last 12 months, I needed…" to "During the past month, my needs were not met for the following issues…". A one-month time-frame was determined to be less susceptible to recall bias, and it was hoped that having a more discrete time period would more accurately reflect the needs of those young people who were currently receiving treatment, compared with those whose treatment was completed.[29] Feedback from participants also led to further revision of the response scale (Appendix 3.9). All "need" response options were rephrased as "unmet need" responses. This meant that participants only had to respond whether the need was met ("No Unmet Need") or unmet and, if unmet, indicate the level of the unmet need ("Low Unmet Need", "Moderate Unmet Need", "High Unmet Need" or "Very High Unmet Need") (Appendix 3.10).

Feedback from the panel of health care providers

Items that were considered to be most important by the panel of health care providers were compared with those considered most important by AYA cancer survivors who participated in the focus groups (Table 3.2). Each tick indicates the number of times the issue was raised by a member of the panel or the focus group. The panel of health care providers also suggested a number of additional needs, some of which were included in the final measure (Table 3.3).

Table 3.2: Feedback on items from the panel of health care providers

Domains	Proposed Items	Perceived important by AYAs	Perceived important by clinicians
Daily life			
Physical	assistance dealing with physical pain	✓	✓✓
	assistance with moving around	✓	✓✓
Travel	assistance getting to social events	✓	✓
	assistance with travelling to and from hospital	✓✓	✓✓
Home environment	assistance with taking medication	✓✓✓	✓✓
	assistance with doing jobs and chores at home	✓	✓✓
Eating	advice about diet	✓	✓✓
Other	assistance participating in social activities	✓	✓✓
	help maintaining a normal "young person" life	✓✓	✓✓
Education/Work			
School	assistance catching up with school work	✓✓	✓
	assistance to ensure I continue my education	✓	✓
	assistance in getting back into school	✓✓	✓
	assistance to ensure I don't miss out on learning	✓	✓
	assistance in travelling to and from school	✓	✓
	guidance about further study or future careers	✓	✓
	to know how much school I would miss	✓	✓
	to know that people at school understand cancer and its impact	✓✓	✓✓
	to know that there was ongoing support for people at my school	✓	✓✓
Work	access to age-appropriate part-time work	✓	✓✓
	assistance in getting back into work	✓✓	✓
	assistance in travelling to and from work	✓	✓
	to know that people at work understand cancer and its impact	✓✓	✓✓
	to know that there was ongoing support for people at my workplace	✓	✓✓
Relationships			
Friends	to be able to spend time with and talk with people my own age	✓✓	✓
	help dealing with loss of friendships	✓✓	✓
	assistance with asking my friends for support	✓✓	✓✓✓
	help dealing with being left out by friends	-	-
Family	my parent(s)/carer(s) physically near me while in hospital	✓✓	✓✓
	support dealing with changes in relationships with my parent(s)/carer(s)	-	-
	help dealing with all the attention from my parent(s)/carer(s)	-	-
	assistance with asking for support from my parent(s)/carer(s)	✓	✓✓✓
Friends and family	to know how to tell family, friends and others about how I feel	✓✓	✓
	to know how to tell family, friends and other people about my cancer	-	-
Other	to talk to someone my own age who has been through a similar experience	✓✓	✓✓
	help dealing with missing the people I care about	-	-
Feelings			
Loneliness	help dealing with feelings of loneliness	✓✓	✓✓✓
Anxiety − treatment	help with anxiety about going to the hospital	✓	✓✓✓
	help dealing with my worry about treatment side-effects	✓✓	✓✓
	help dealing with my worry about painful medical procedures and treatment	✓✓	✓✓
	help with anxiety when waiting for test results	-	-
	help dealing with concerns about whether the treatment will work or has worked	-	-
Anxiety − disease	help dealing with the possibility of the disease returning	✓✓	✓✓
Anxiety − other	help dealing with uncertainty about the future	✓✓	✓✓
Depression	help dealing with feelings of sadness	✓	✓✓
	help dealing with feelings of helplessness	✓	✓✓✓
Coping	to find enjoyment in my life	✓	✓✓
	help in finding hope for the future	✓✓	✓✓
	help in finding my inner strength	✓	✓✓
	help finding meaning in my experience	-	-
	help dealing with changes to who I am	✓	✓✓
Frustration/anger	help dealing with feelings of frustration or anger	✓✓	✓✓✓✓✓
Protective	help dealing with my parent(s)/carer(s) being over-protective	✓✓	✓
Emotions	help thinking about my future and life priorities	✓✓	✓
	help dealing with feelings of distress	✓✓	✓✓✓
	help dealing with feelings of embarrassment	✓	✓✓
	help with feeling vulnerable	-	-
	help to feel secure	✓✓	✓
	help dealing with being bored	✓	✓✓✓✓
	help dealing with being scared	-	-
	help to be independent	✓✓	✓✓
Relationships/roles	help dealing with people's reactions towards me	✓✓	✓
	help dealing with the worries of those close to me	-	-
Other	help focusing on tasks and/or remembering things	-	-
	help dealing with not being able to do the same things as other young people	✓	✓✓

Domains	Proposed Items	Perceived important by AYAs	Perceived important by clinicians
Cancer Treatment Centre			
Location of care	access to local treatment centres	✓	✓✓
Access to providers	quick and easy access to providers skilled in cancer care	✓	✓✓✓
	quick and easy access to professional counselling	✓	✓✓
Setting/ resources	privacy while in hospital	✓✓	✓✓
	a place to stay with my parent(s)/carer(s) while in hospital to have treatment	✓	✓✓
	access to leisure space and activities in hospital that are appropriate for my age (e.g. TV, mobile phone, computer)	✓	✓✓✓
	pleasant and inviting clinic surroundings	✓✓	✓
	access to better and wider food choices while in hospital	✓✓	✓
Cancer Treatment Staff			
Care management	the same health care provider to take care of me throughout treatment	✓✓	✓
Communication	friendly health care providers who I could have a laugh with	-	-
	health care providers who talked to me, not just my parent(s)/carer(s)	✓	✓✓
	health care providers who talked to me and my parent(s)/carer(s) together	-	-
	to be treated as an individual by health care providers	✓✓	✓✓
	to be spoken to by health care providers in a way that I could understand	✓	✓✓
	to know that my health care providers are part of a team	✓	✓
	health care providers who listened	✓✓	✓
	health providers who were skilled	✓	✓✓
	health care providers who were approachable and let me ask questions	✓✓	✓
	to be able to talk to health providers in private without my parent(s)/carer(s) to talk to health care providers informally	✓✓✓	✓
Decision-making	to know that I had the right to refuse treatment	✓	✓
	the opportunity to make decisions about my appointment times	-	-
	the opportunity to make decisions about my treatment	✓✓✓	✓✓
	more time to make decisions about my care	✓	✓
Other	assistance with filling out forms	-	-
Information			
General cancer	complete and honest information about cancer	✓✓✓	✓✓
	complete and honest information about the long-term impact of cancer	✓✓	✓✓
Individual AYA	complete and honest information about my diagnosis	-	-
	complete and honest information about how I am going	-	-
	complete and honest information about being able to have children	✓	✓✓
	complete and honest information about the chances of recovery or being cured	✓	✓✓✓
Treatment/side-effects	complete and honest information about treatment options	-	-
	complete and honest information about treatment and its side-effects	✓✓✓✓✓	✓✓
	information about what I should do if I notice a particular side-effect	✓	✓✓✓
	information about how treatment would be given	-	-
	information about what to expect when going to the hospital and during treatment	✓✓	✓✓
	information about what to expect after treatment	✓✓	✓✓✓✓
Services/resources	information about support services and help that are available	✓	✓✓
Information format	to be able to access different formats of information (e.g. internet, verbal, written)	✓✓	✓
Other	to be told by health care providers that the way I felt was normal	-	-
	information about relaxation techniques	✓✓	✓
	information about feelings caused by the experience	-	-

Table 3.3: Additional needs suggested by panel of health professionals

Suggested needs added to the final measure	Suggested needs not added to the final measure
being able to talk openly with health care providers about my feelings or worries	ensuring I still felt part of the classroom/school while in hospital
being able to participate in new research or cancer treatment	knowing people at school/work were prepared for when I returned
help dealing with my worries about my future career prospects	help participating in activities without my parents being there
help dealing with going back to the real world post-treatment	assistance with personal care tasks such as dressing and showering
help dealing with changes in the way I look	access to space that is available just for young people while in hospital
help dealing with not being able to do the same things as I used to	being able to have efficient and coordinated appointments
getting information about how to stay healthy	having health providers who could inspire hope
help dealing with my worry about treatment or side-effects	information about the effects of treatment on contraception
help dealing with changes in relationships with siblings	getting support from people of my faith and/or culture

Feedback from researchers and the general population

The number of domains in the measure was increased from seven to eight following comments from researchers that items in the “Education and Work” domain should be separated into two domains.

Discussion

Final revision of the unmet needs measure

The face and content validity of the CNQ–YP was established. Feedback and advice received from participants led to a revised response scale and stem for the measure, and one extra domain. The initial number of items was also modified. Four items were removed, and the total number of items increased from 108 to 144 (36 items). It was expected that this number would be further reduced once factor analysis was performed on the measure (Chapter 5).

When formatting the final version of the measure, items were grouped into domains rather than being spread randomly across the scale. This decision was based on evidence suggesting that participants can become confused when items skip between different topics, and this can lead to errors in data collection.[30 31] In line with guidelines for measure development,[32] the most pertinent and non-threatening domains were presented at the beginning of the draft measure. These were followed by more sensitive and personal domains towards the end. For example, domains related to daily life, education and work were presented first, while domains related to relationships and emotional health were presented towards the end of the measure.

Comparison of methodologies for establishing face and content validity

As part of the critical review, Chapter 2 outlined the methods used to establish face and content validity for seven measures of AYA psychosocial well-being. Although the processes of confirming face and content validity for all but one of the measures were reported, the protocols used in each individual study varied. For example, two of the developed measures adapted their items from existing measures, but did not report gaining input from AYAs *via* a focus group or pilot test.[4 8] Furthermore, only four of the measures were assessed by AYA cancer survivors.[3 5 7 9]

As face and content validity are primarily subjective assessments consisting of qualitative feedback, one over-arching methodology is unlikely to be consistently used. The current study attempted to use a comprehensive approach to evaluate the face and content validity of the measure which incorporated a variety of methods including: a literature search; derivation of items from existing measures; input from AYA focus groups; input from a panel of health professionals; feedback from researchers; and assessment by other professionals. Consulting a range of stakeholders (consumer and professional groups) with potentially different perspectives has increased the likelihood

that all relevant issues and areas of need have been included. This process has strengthened the face and content validity of the CNQ–YP.

Although establishing the face and content validity of a measure is often regarded as the first step in establishing psychometric rigour, further evaluation of the measure's properties are required.

Limitations

Limitations in the study methodology need to be considered. First, the sample size of the two AYA focus groups was small (three young people per group) and views of these participants may have been biased as all of these young people were recruited through CanTeen. It could be argued that members of CanTeen may have greater levels of social support than the general AYA cancer population or, alternatively, that membership of CanTeen suggests that these young people may have higher unmet needs than the wider population and have therefore sought additional support. Therefore, the sample used may have implications for generalisability of the measure to the broader population of AYA cancer survivors. Pilot testing of the measure is needed to assess the relevance and acceptability of the CNQ–YP to a wider group of young people.

Conclusion

This study confirmed the face and content validity of a measure to assess the unmet needs of AYAs diagnosed with cancer. Further development of the CNQ–YP will require pilot testing to determine acceptability, and the recruitment of a large sample of AYAs to enable psychometric analysis.

CHAPTER 4

Development of the Cancer Needs Questionnaire – Young People (CNQ–YP): pilot test to determine acceptability

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Chapter 4

Development of the Cancer Needs Questionnaire – Young People (CNQ–YP): pilot test to determine acceptability

Introduction

Chapter 3 described the initial steps in the development of the Cancer Needs Questionnaire – Young People (CNQ–YP). The draft measure was developed based on identified domains and items, and the face and content validity of the measure was established. This chapter describes the process of pilot testing the newly developed measure to determine its acceptability.

Importance of pilot testing measures

A pilot test or study is a common way of assessing the feasibility and acceptability of the proposed design, materials and procedures for a larger research study or trial.[1 2] The specific aims of pilot studies will vary, but generally involve testing the following issues: suitability of participant inclusion and exclusion criteria; cost, time and resources needed for recruitment and data collection; commitment of health care providers to recruitment and data collection; effectiveness of reminders and prompts (e.g. follow-up letters or telephone calls); recruitment rates; consent rates; acceptability of the materials used in the study; and analysis of preliminary data.[1-4]

In the context of measure development, pilot testing allows participants to comment on the suitability of the measure in terms of its format, content, clarity, comprehensibility, time to complete and overall acceptability.[1 3] This process ensures that any items that are difficult to understand or distressing can be removed or modified prior to the main study or trial, and maximises the likelihood of accurate and complete data collection.[1 5] To ensure that a correct assessment of the research methodology and materials is

made, the sample selected for the pilot study should be as close as possible to the target population.[1]

Use of cancer registries for accessing representative groups of survivors

The degree to which cancer research findings are generalisable to the wider cancer population is, in part, dependent upon the representativeness of the study sample recruited.[6] One of the challenges facing population health research in cancer is the recruitment of large and representative samples of participants in a timely and cost-efficient manner.

Cancer registries are a potential mechanism for recruiting population-based samples of cancer survivors for research.[7] In many developed countries cancer registries are supported by Public Health Acts or other legislation.[8-10] As a consequence, notification of any cancer diagnoses by hospitals, general practitioners or pathology units may be legally required. Having a centralised source for recruiting a sample of cancer survivors has advantages, especially when conducting research with low-incidence cancer populations such as adolescents and young adults (AYAs).[7]

A sample of AYA cancer survivors recruited *via* a population-based cancer registry is likely to represent the relative distribution of all cases of AYA cancer diagnosed, with the exception of cancers with high mortality for this age group, such as brain tumours (mortality rate of 21%).[11] Therefore, for low-incidence cancer populations, registries offer potential access to all cases through a single access point and remove the need for researchers to recruit survivors from multiple sites.[7]

Process for recruitment of research participants through cancer registries

There can be up to three stages of consent required when recruiting cancer survivors

through registries for research studies.[12] At each stage the number of potential participants may be reduced, thereby potentially reducing the size and representativeness of the final sample.

In the first stage of recruitment, the registry may contact the responsible clinician and request a professional judgement as to whether the survivor is well enough for the registry to approach. Clinician consent can be active or passive, depending on the protocols in place within the registry.[13 14] Active consent requires clinicians to provide written or verbal confirmation regarding the suitability of all identified cancer survivors prior to the registry contacting the survivor.[13 14] Passive consent requires the clinician to respond to the registry only if an identified survivor should not be contacted. If the clinician does not respond within a specified time period, clinician consent is inferred and the registry can proceed with contacting the cancer survivor.[13 14]

The second stage of the recruitment process may require the cancer survivor to grant consent for the registry to provide their contact details to the researchers. Survivor consent can also be active or passive.[12] Active consent requires survivors to provide written or verbal consent if they wish to be contacted.[12 14] Passive consent allows all survivors to be contacted unless they opt out by providing written or verbal notification that they do not wish contact to occur.[12] In the third stage of recruitment, the researchers contact survivors in accordance with their approved research protocols and request participant consent to take part in the research.

Aims

The aims of this study were: 1) to conduct a pilot study to determine the acceptability of the CNQ–YP; and 2) to determine the representativeness of the pilot sample recruited *via* the cancer registry.

Methods

Setting

A cross-sectional study design was used to recruit AYAs diagnosed with cancer through the New South Wales (NSW) cancer registry in Australia. Under the Public Health Act for the state, notification of malignant neoplasms is a statutory requirement for public and private hospitals, departments of radiation oncology, nursing homes, pathology laboratories, outpatient departments and day procedure centres. When any of these institutions diagnose or treat someone with cancer, they are required by law to notify the state cancer registry. Notifications of cancer in NSW residents who are diagnosed and treated outside the state are also received from other state and territory cancer registries. Demographic information and clinical details about the cancer and treating clinician are collected from notifiers.[14]

Participants

Adolescents and young adults listed on the registry were eligible to participate in the study if they were: 1) diagnosed with an invasive cancer between the ages of 14 and 19 years inclusive; 2) diagnosed in the previous five years; 3) residents in the same state as the registry; and confirmed by their primary treating clinician as: 4) having a life expectancy of at least 12 months; 5) physically and mentally able to complete the survey; and 6) sufficiently literate in English.

As discussed in Chapter 1, the age range used to define AYAs varies across the literature and ranges from 10 to 40 years.[15-17] In this study the upper age limit of 19 years was selected as the World Health Organization (WHO) defines adolescents as being 10 to 19 years of age.[16] The lower limit of 14 years was chosen because, in Australia, AYAs aged 14 years and older have the legal right to make their own decisions about the types of health care they receive.[18] Limiting registrations to those

diagnosed in the previous five years represented a balance between: 1) accessing a large enough population to achieve required sample sizes; 2) eliciting views of those who had been affected by cancer for different periods of time; and 3) maximising the number of respondents still in treatment. Ethical approval to contact AYAs was only granted for NSW residents therefore AYAs who were listed on the NSW cancer registry but who resided in other states could not participate. Adolescents and young adults diagnosed with non-invasive cancers, such as non-melanoma skin cancer (NMSC), were excluded, for two reasons: 1) Notification of NMSC is not compulsory in most Australian states, with the exception of Tasmania (TAS), therefore records for NMSC listed on the NSW cancer registry are not representative of the population; and 2) it was expected that the number of survivors with NMSC would be very small as most cases are diagnosed in those aged 20 years and older.[11]

Procedure

Ethical approval for the study was granted by the Human Research Ethics Committees of the University of Newcastle (Chapter 3, Appendix 3.2) and the Cancer Institute NSW (Appendix 4.1). A description of the recruitment protocol used in the registry is presented in Appendix 4.2.[14] Data for all potentially eligible participants who were listed on the registry were cross-checked with the death register for NSW prior to contacting clinicians.

Stage 1 – Clinician consent to contact survivors

A letter was sent to each identified AYA cancer survivor's primary treating clinician (as recorded on the registry) to inform the clinician about the study, confirm eligibility and request permission to contact the eligible survivor (Appendix 4.3). Treating clinicians listed on the registry may have been the survivor's general practitioner, surgeon or oncologist, depending on who was the first provider to notify the registry of the

survivor's cancer diagnosis. Clinicians who did not respond to the letter within two weeks were telephoned by the registry at two-weekly intervals, up to five times, to determine the eligibility of the identified AYAs.

Stage 2 – Survivor consent to pass on contact details

Adolescent and young adult survivors whose clinicians consented for them to be contacted were sent a project information letter and a "consent to be contacted form" by the registry, seeking written permission to forward the survivor's contact details to the research team (Appendix 4.4). Adolescents and young adults were encouraged to discuss their possible involvement in the study with their parents and/or primary treating clinicians. Survivors who had not responded within two weeks of receiving their initial letters were sent reminder letters by the registry. Survivors who had not responded within two weeks of receiving the reminder letters received follow-up telephone calls at the four-week interval. Up to two attempts to contact survivors by telephone were made. The registry then provided researchers with the contact details of AYAs who agreed to be contacted.

Stage 3 – Survivor consent to participate

The researchers sent AYA cancer survivors who agreed to be contacted a letter of invitation, a study information sheet, (Appendix 4.5) and a copy of the Cancer Needs Questionnaire – Young People (CNQ–YP) (Appendix 4.6). Survivors who had not returned questionnaires within two weeks of receiving them were sent reminder letters. Survivors who had not returned questionnaires within two weeks of receiving the reminder letters received follow-up telephone calls at the four-week interval. Up to two attempts to contact survivors by telephone were made. Return of the questionnaire was taken as implied consent to participate in the study.

Measure

Cancer Needs Questionnaire – Young People (CNQ–YP)

The draft Cancer Needs Questionnaire – Young People (CNQ–YP) is a self-administered, paper and pencil questionnaire designed to capture the multi-dimensional unmet needs experienced by AYAs diagnosed with cancer (Appendix 4.6). The measure has 8 domains and 144 items, examples of which are presented in Table 4.1. The stem for each item is, “During the past month, my needs were not met for the following issues”. For each item the level of unmet need is rated using a five-point response scale: “No unmet need” – All my needs were met for this issue or this did not apply to me; “Low unmet need” – My needs were not met for this issue. My level of need was low; “Moderate unmet need” – My needs were not met for this issue. My level of need was moderate; “High unmet need” – My needs were not met for this issue. My level of need was high; “Very high unmet need” – My needs were not met for this issue. My level of need was very high. The reading age for the CNQ–YP is 10-11 years, as assessed by the Flesch-Kincaid Grade Level in Microsoft Word. The questionnaire is estimated to take approximately 30 minutes to complete.

Demographic questions

Participants were asked questions regarding the type of cancer treatment they had received (e.g. surgery or chemotherapy), the type of hospital where they had received most of their treatment, whether or not they had had a recurrence of their cancers, and where they were in their cancer journeys. Details about living arrangements, language spoken, education and employment were also collected. Demographic characteristics such as age, gender and cancer type were not asked in the questionnaire as these details had already been provided, with the survivor’s consent, from the cancer registry.

Table 4.1: Description of the eight domains in the CNQ–YP and examples of items

Domain	Items	Example items During the past month, my needs were not met for the following issues:
Daily Life	10	managing the physical side-effects of treatment doing chores or housework taking part in social activities
Education	12	knowing how much school, TAFE or university I would miss receiving guidance about further study options or future career paths knowing that my friends at school, TAFE or university understood my situation
Work	12	going back to work knowing how to ask for support from my friends or colleagues at work being able to work part-time
Relationships	19	coping with all the attention from my parent(s) or carer(s) being able to talk to people my own age who have been through a similar experience coping with changes in my relationships with friends
Feelings	35	feeling anxious or nervous making plans or thinking about the future coping with changes in my physical ability
Cancer Treatment Centre	11	being treated at a hospital, clinic or treatment centre in my local area having leisure space and activities while in the hospital, clinic or treatment centre being able to take part in new research at the hospital, clinic or treatment centre
Cancer Treatment Staff	36	having the same health care provider take care of me throughout my treatment being able to talk to health care providers in private, without my parent(s) or carer(s) having health care providers who I could talk openly with about my feelings
Information	9	having information that was specific to me knowing which information I should trust getting information about the feelings or emotions caused by the experience

Acceptability questions

Adolescents and young adults were asked to rate the acceptability of the CNQ–YP on a four-point Likert scale with responses ranging from “Strongly disagree” to “Strongly agree”. Participants responded to the following three statements: “I found the questionnaire clear”; “I found the questionnaire easy to understand”; and “I found the questionnaire distressing”. Participants were also asked approximately how long it took them to complete the measure.

Qualitative feedback and suggestions for additional items

Participants were invited to provide qualitative feedback and comments at the end of the questionnaire about improvements that could be made and items that could be added to the CNQ–YP in the future.

Statistical analysis

Proportions were calculated to estimate survivor consent rates overall and at each stage of recruitment. Chi-square tests (Fisher's exact test) were performed using Stata Version 11 statistical software[19] to identify whether there were any significant differences between non-participants (AYAs who were unable to be contacted or did not provide consent) and the final sample.

Data from the registry indicated that there were 468 potentially eligible survivors. Assuming a 75% consent rate at each stage, 351 survivors would receive clinician consent at Stage 1, 263 survivors would consent to be contacted at Stage 2 and 197 survivors would consent by returning a questionnaire at Stage 3. The anticipated sample size of approximately 200 would allow the estimation of consent rates within $\pm$7% and a detection of differences in consent rates between the groups of 25%, with 80% power at the 5% significance level.

Results

Recruitment and response rates

Excluding 54 AYAs who were deceased, the registry identified a total of 995 cases of AYAs aged 14 to 19 years when diagnosed with cancer. Four hundred and fifty-six of these were NMSC cases and were excluded. Of the 539 AYAs diagnosed with invasive cancer, 63 did not reside in the same state as the registry. Prior to contacting clinicians,

survivors' data were checked against the death register again; a further eight were deceased, leaving 468 potentially eligible survivors.

Recruitment rates, including rates within each stage of recruitment and rates for the overall sample, can be seen in Figure 4.1. Of the 468 potentially eligible survivors identified, clinicians reported that 57 did not meet the eligibility criteria. Of the remaining 411 potentially eligible survivors, almost a third of their primary treating clinicians were unable to be contacted (n=134, 33%) and 45 (11%) refused consent for other reasons. In total, clinician consent was provided for 232 (56%) of all potentially eligible survivors. Of the 232 AYAs who received their clinicians' consent to be contacted, the registry was unable to contact 150 (65%). Of the remaining 82 survivors, 18 AYAs (7.8%) did not provide consent for their contact details to be passed on to the research team. Survivor consent to contact was provided by 64 AYAs (28% of those with clinician consent, and 16% of all potentially eligible survivors). Of the remaining 64 survivors who agreed to be contacted by the research team and were sent a copy of the unmet needs measure, 12 (19%) could not be contacted, 32 (50%) returned the questionnaire and 20 (31%) did not return a questionnaire. The overall consent rate for all potentially eligible AYAs was 7.8%.

Reasons for clinicians not providing consent to contact survivors

Reasons reported by clinicians for not providing consent for the registry to contact survivors can be seen in Table 4.2. Clinicians reported that 56% (n=57) of AYAs did not meet the eligibility criteria for the study, and that 35% (n=36) were no longer their patients.

Figure 4.1 Flowchart of proportion of cancer survivors filtered at each stage, and proportion of overall sample

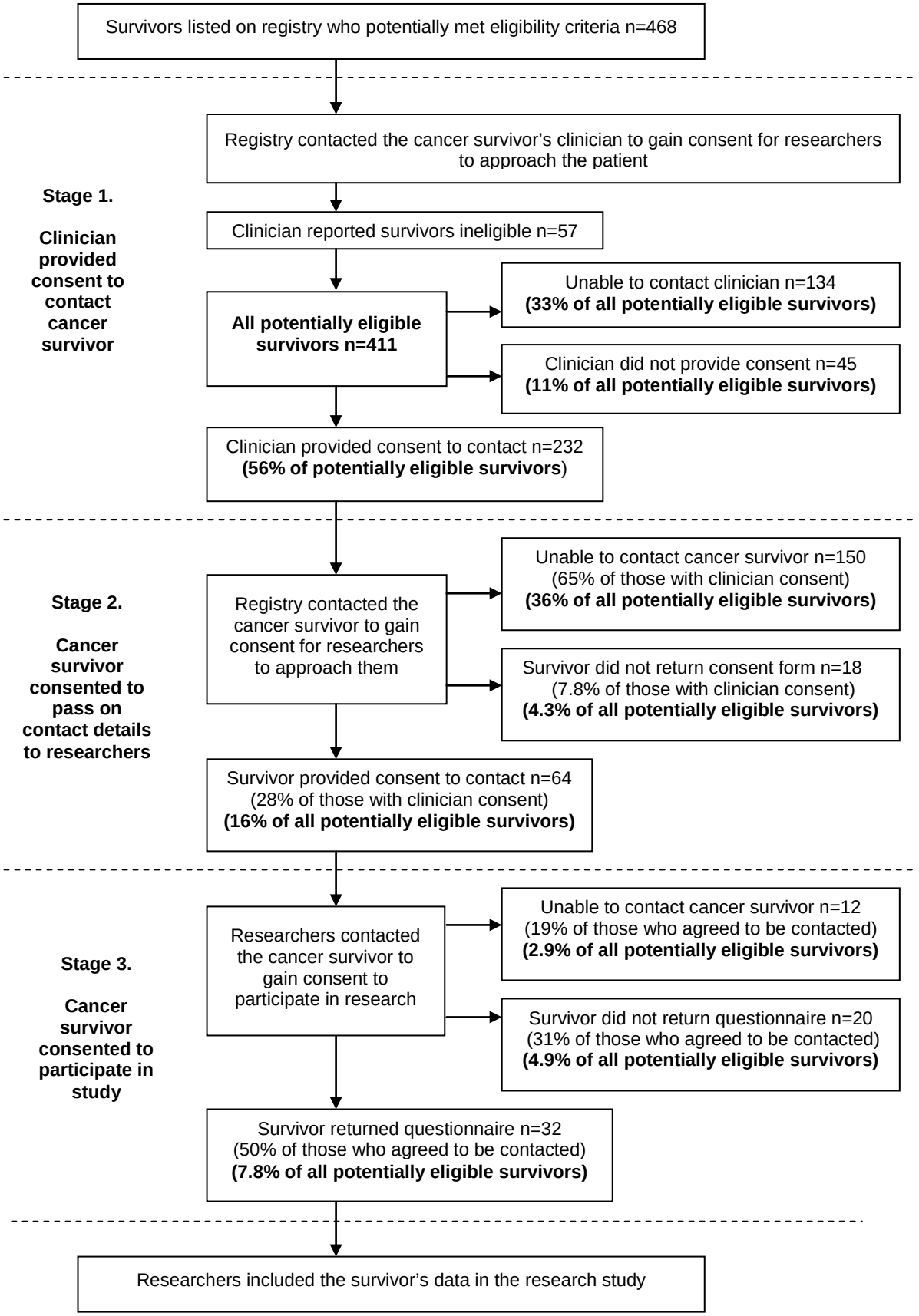

Table 4.2 Reasons provided by treating clinicians for not providing consent to the registry to contact survivors

	Reason for refusal	n=102	%
Survivors did not meet eligibility criteria n=57 (56%)	Not physically/mentally capable	12	12
	Diagnosis not appropriate	11	11
	<1 year life expectancy	9	8.8
	Survivor did not wish to participate	7	6.8
	Not in same state as registry	6	5.8
	Situation not appropriate	6	5.8
	Too ill	2	1.9
	Doesn't speak English	2	1.9
	Survivor unaware of cancer diagnosis	2	1.9
Other reason n=45 (44%)	Not current patient	36	35
	No reason given	7	6.8
	Clinician did not wish to participate	2	1.9

Participant demographic characteristics

Participants ranged from 14 to 19 years of age inclusive at diagnosis, with a mean age of 16.3. There was a slightly higher proportion of females (53%) than males (47%). Cancer diagnoses included leukaemia (25%), lymphoma (19%), bone cancer (19%) and thyroid cancer (9.3%). A large percent of participants had a combination of surgery and either chemotherapy or radiation therapy (47%), had finished treatment (90%), had not experienced a recurrence of their cancer (69%) and had been treated in an adult rather than a children's hospital (66%) (Table 4.3).

Eighty-one percent of the AYAs who completed the survey lived at home with their parents, 90% spoke English at home, and most had completed high school education above year 10. Nineteen percent of participants were not employed.

Table 4.3: Demographic characteristics of participants (n=32)

Demographic Characteristics	n	%
Age at diagnosis		
14-15	11	34
16-17	16	50
18-19	5	16
Gender		
Female	17	53
Male	15	47
Cancer type		
Leukaemia	8	25
Lymphoma	6	19
Bone	6	19
Thyroid	3	9.3
Melanoma	2	6.2
Brain	2	6.2
Ovarian	2	6.2
Other	3	9.3
Treatment type		
Surgery or chemotherapy	7	22
Surgery + chemotherapy or radiation therapy	15	47
Chemotherapy + radiation therapy	5	16
Other	5	16
Cancer care stage*		
On treatment	3	9.6
Off treatment	28	90
Cancer recurrence		
Yes	6	19
No	22	69
Unsure	4	13
Hospital type		
Children's	11	34
Adult	21	66
Language		
English	30	94
Other	2	6.2
Living arrangements*		
Parent/s	25	81
Partner	3	9.6
Flatmates	3	9.6
Education completed		
University	1	3.1
TAFE/trade	7	22
Year 11 or 12	21	66
Year 10 or less	3	9.3
Employment		
Full-time	15	47
Part-time	4	13
Casual	7	22
Not employed	6	19

**n=31*

Acceptability

Sixty-six percent of participants agreed or strongly agreed that the questionnaire was clear, while 68% agreed or strongly agreed that it was easy to understand. Ninety percent of participants disagreed or strongly disagreed that the questionnaire was distressing (Table 4.4). Reported time to complete the measure ranged from 5 to 45 minutes, with a mean of 22.17 and standard deviation of 10.48.

Table 4.4: Acceptability of the CNQ–YP

I found the questionnaire:	Strongly disagree (%)	Disagree (%)	Agree (%)	Strongly agree (%)
Clear (n=32)	6.2	28	44	22
Easy to understand (n=31)	6.4	26	42	26
Distressing (n=31)	58	32	9.6	0

Qualitative feedback and suggestions for additional items

Additional items for the measure were suggested by four participants. These included: discussing ways to have children if infertile; coping with feelings at the time of treatment related to body image, depression, self-esteem, confusion and trying to interact normally; meeting other people the same age going through treatment; and coping with psychological aspects after treatment was completed, wanting reassurance that psychological help is available, and being offered psychological help during and after treatment.

Eight participants also commented that they found the response time-frame, “in the past month”, did not allow them to express all the concerns about unmet needs they had experienced throughout their cancer journeys. These AYAs reported that most of the problems they faced were at the time of treatment, and as the majority were now finished treatment not much had happened in the previous month. Two participants

noted that they had filled out the questionnaire from the perspective of how they felt while undergoing treatment, rather than how they felt in the past month, as the CNQ–YP instructions stated. They explained that the reason behind this was that they wanted to express how they felt going through treatment so that more of the questions could be answered and so that the researchers could receive more information. One young person also commented that memory of treatment was still strong, even two to five years later, and that their needs did not generally happen within a month, but rather were spread over years.

Comparison of the final sample compared with the overall sample

The demographic characteristics of AYAs who were included and excluded at each stage of recruitment are presented in Table 4.5. The final sample of participants was reasonably representative, with no significant differences between the proportions of males and females ($\chi^2(1)=1.10$, $p=0.36$) or different cancer types ($\chi^2(5)=8.04$, $p=0.14$) for those who participated and those who did not, although there was only adequate power to detect large differences. There was, however, a significant difference between the ages of the two groups, with those who participated being significantly younger at diagnosis than those who were unable to be contacted or did not consent ($\chi^2(2)=9.17$, $p<0.01$).

Table 4.5: Comparison of final participant group with eligible non-participants (did not consent or unable to contact)

	Demographics of survivors							
	Stage 1 (n=468)		Stage 2 (n=232)		Stage 3 (n=64)		**Total (n=468)**	
	Clinician did not consent (n=102)	Unable to contact clinician (n=134)	Survivor did not consent (n=18)	Unable to contact survivor (n=150)	Survivor did not consent (n=20)	Unable to contact survivor (n=12)	**Did not consent and unable to contact (n=436)**	**Final participant group (n=32)**
	n (%)	n (%)	n (%)	n (%)	n (%)	n (%)	**n (%)**	**n (%)**
Age at diagnosis								
14-15	27 (26)	30 (22)	9 (50)	40 (27)	2 (10)	2 (17)	**110 (25)**	**11 (34)**
16-17	27 (26)	51 (38)	2 (11)	46 (31)	8 (40)	6 (50)	**140 (32)**	**16 (50)**
18-19	48 (47)	53 (40)	7 (39)	64 (43)	10 (50)	4 (33)	**186 (43)**	**5 (16)**
Gender								
Female	44 (43)	59 (44)	7 (39)	68 (45)	4 (20)	8 (67)	**190 (44)**	**17 (53)**
Male	58 (57)	75 (56)	11 (61)	82 (55)	16(80)	4 (33)	**246 (56)**	**15 (47)**
Cancer type								
Lymphoma	16 (16)	36 (27)	5 (28)	36 (24)	4 (20)	2 (17)	**99 (23)**	**6 (19)**
Skin	25 (25)	21 (16)	3 (17)	31 (21)	2 (10)	2 (17)	**84 (19)**	**2 (6.2)**
Testicular	9 (8.8)	13 (9.7)	0 (0)	13 (8.6)	4 (20)	0 (0)	**39 (8.9)**	**1 (3.1)**
Thyroid	11 (11)	7 (5.2)	0 (0)	11 (7.3)	2 (10)	1 (8.3)	**32 (7.3)**	**3 (9.3)**
Leukaemia	13 (13)	17 (13)	3 (17)	21 (14)	2 (10)	2 (17)	**58 (13)**	**8 (25)**
Other*	28 (27)	40 (30)	7 (39)	38 (25)	6 (30)	5 (42)	**124 (28)**	**12 (38)**

** "Other" includes cancer of brain, bone, connective tissue, colorectum, small intestine, kidney, liver, ovary, bladder, breast, lung and heart.*

Discussion

Acceptability of the CNQ–YP

The CNQ–YP appears to be acceptable to AYAs who have experienced cancer. Ninety percent of survivors reported that the measure did not cause distress, suggesting that no items needed to be removed on this basis. The length and reading age of the CNQ–YP also appears to be acceptable, with 66-68% of AYAs reporting that the measure was clear and easy to understand. The reading age of 10-11 years seems appropriate for the sample of AYAs who completed the measure who had a mean age of 16.3 years at diagnosis. The average time to complete the questionnaire was below the expected 30 minutes (mean time 22.17 minutes).

Improvements for the next version of the CNQ–YP

Based on the qualitative feedback of participants, there are a number of improvements that could be incorporated into the next version of the CNQ–YP. It may help AYAs with cancer to better express their needs at the most difficult periods of their cancer journeys (i.e. during treatment) if the time-frame of the stem questions were expanded to include the treatment phase. Although the accuracy of memory recall of survivors may be a concern, participants reported that the memories of the treatment period were strong, and that they wanted to express their needs during this period. Two participants reported pretending they were still in treatment and answering the survey this way. A further eight participants felt that broadening the stem time-frame to the time of treatment would have helped to give a more accurate picture of the needs they faced early in their cancer experiences. This may be an early indication that the period of highest unmet need for young people is while they are undergoing treatment and that this phase in the cancer journey may warrant further investigation.

Three participants reported that they were confused by the wording in the measure. Therefore, revision of the stem, response scale and wording of items may need to be considered.

Representativeness of the sample recruited

Despite the potential of the registry to access a large, population-based sample of AYAs diagnosed with cancer, only a small percentage (7.8%) of the potentially eligible population was recruited into the study. Compared with AYAs who were unable to be contacted or did not provide consent, the resulting sample of participants did not differ significantly by gender or cancer type. However, compared with all eligible AYAs listed on the registry within the time period of interest, a greater proportion of those recruited into the study sample were younger at diagnosis. Given the potential promise of

registry recruitment to deliver population-based, representative samples, it is important to understand what factors may have contributed to these low recruitment rates and potential sample bias.

Possible reasons for low recruitment rates and non-representative samples

Clinician-related reasons

In the current study over one-fifth of potential participants were unable to be approached by the registry because their clinicians refused to provide consent. Surprisingly, survivor ineligibility was only cited as a reason for 56% of cases in which clinicians refused consent for the survivor to be approached. Furthermore, of the reasons for ineligibility provided, at least two appear to be less about the actual study eligibility criteria, and more about the clinician's own perception of the survivor's situation (e.g. diagnosis or situation not appropriate). There is evidence to suggest that clinicians often overestimate the psychological distress experienced by their patients.[20] [21] This may mean that at least some potentially eligible participants are unable to be contacted due to the assumptions made about their well-being by their clinician.

Clinicians no longer had contact with survivors

For the majority of the remaining survivors (35%) where clinician consent was not obtained, it was because they were no longer current patients of the clinicians. This situation may arise due to a number of reasons. Registries often receive cancer notifications from various sources including pathology laboratories, the survivors' general practitioners or the hospitals where the survivors are or were receiving treatment.[22] [23] Depending on the source, the clinician who is listed as the treating clinician on the registry file may not be the person who is overseeing the young person's cancer care. Factors such as increasing population mobility[24] [25] and increasing emphasis on consumer satisfaction in health care[26] may mean that AYAs

are likely to change health care providers as their needs change. This may contribute to a lack of continuity in the doctor-patient relationship.

Clinicians did not respond

Almost a third (n=134, 33%) of all identified potential participants could not be contacted because their clinicians did not respond to correspondence from the registry, including contact by mail and telephone. Reasons as to why clinicians did not respond can only be hypothesised. However, one possible explanation may be that, as outlined previously, the identified young person was no longer a current patient of the treating clinician. Further, clinicians are very time-poor.[27 28] This factor may be of particular importance when active clinician consent is required by the registry. Others may feel that responding to or participating in research studies is not a priority.[29 30] There is evidence to suggest that a large proportion of clinicians do not regard assisting patients to participate in research studies to be an important part of their professional role.[29]

It is also possible that some clinicians do not respond as they do not wish their patients to participate in the type of research study being offered, especially if they perceive that the research question being investigated may reflect badly on their profession.[28] A study of unmet needs, such as the one described in this chapter, has the potential to link patient outcomes to the knowledge, attitudes and skills of health care providers.[31] It is conceivable that attitudes toward the research topic may influence clinician behaviour with respect to responding to registry requests.

Non-response from clinicians can be a particular problem when studies involve survivors with all cancer types, as there is a resultant need to contact a diverse range of clinicians who cannot be easily targeted for study recruitment. Members of particular professional groups or clinical sub-specialities may have different attitudes towards

research participation, and this may in turn influence the types of patients who are permitted to be contacted and who subsequently participate in the study.[27 32] When studies are instead focussed on recruiting survivors with a particular type of cancer, specific professional groups can be targeted and educated about the purpose of the study. For example, a study on leukaemia could be promoted to haematologists through relevant professional groups. In the case of the current study, however, the focus was AYAs diagnosed with any type of cancer, making it impractical to target particular speciality groups to promote the study.

Survivor-related reasons

Participants were unable to be contacted

Almost two-thirds (65%) of the 232 AYAs who received their clinicians' consent to be approached by the registry were unable to be contacted due to inaccurate contact details (mail was returned to sender or telephone number was disconnected). Although this may present a problem for all retrospective studies recruiting through cancer registries, it may be a particular problem when recruiting AYAs, as individuals in this age group are highly mobile.[25 33] In Australia, half of all AYAs aged 15-24 years moved residence during 1997 to 2001, with a large proportion moving interstate.[25] This problem may not be as prevalent with older survivor groups. For example, in a previous study conducted in Germany by Pritzkuleit and colleagues, recruitment of an older adult sample (18-85 years) through a cancer registry showed that only 3.6% of patients were unable to be contacted due to the lack of a valid address.[34] Australian registries can cross-check adult survivors' names and dates of birth with the electoral roll and check for changes of address. However, the same procedure cannot be used to update contact information for AYAs under the age of 18 years who are not eligible to vote.

Participants did not want to be contacted by the researchers

It might be expected that the low consent rate achieved was the result of AYAs not wishing to be contacted by the researchers. However, this does not appear to be the case as only a small percentage (7.8%, n=18) of survivors did not return a "consent to be contacted form". The proportion of refusal may have been even lower, as we are assuming that all 18 AYAs received the information pack from the registry. It is possible that some of these AYAs did not receive the information. The low non-consent rate suggests that being contacted for participation in research studies is acceptable to this survivor group.

Implication of low recruitment rates and non-representative samples

There are a number of ethical, research, and cost-related implications of low recruitment rates and non-representative samples achieved through registries using active clinician consent.

Ethical implications

The process of seeking active clinician consent is underpinned by the ethical principle of beneficence. It is implemented to minimise avoidable psychological harm that survivors may experience by being contacted by the registry.[12 35] The main purpose for contacting clinicians is to confirm that survivors are aware of their cancer diagnoses and are not too ill to be approached by the registry.[12-14] However, in the current study the main reasons for potential participants being excluded at stage 1 of recruitment were related to clinicians no longer having professional relationships with patients, or clinicians simply not responding. Forty-three percent of all identified AYAs could not be approached due to clinician refusal or non-response. This suggests that a large proportion of survivors are being excluded for reasons not related to study eligibility or

emotional health, indicating that the balance between the ethical principles of beneficence and patient autonomy may need to be considered.[12 36 37]

In weighing up these two ethical principles, the potential level of harm associated with the research and its probability of occurrence need to be explored. It is generally accepted that most research studies will involve some potential for harm.[35 37] Harm may range from simple inconvenience to psychological distress or, in the case of drug trials, unforseen side-effects. Prior to commencing a research study researchers need to provide a justification to ethics committees as to why the potential benefits of their research outweigh any potential risks.[35 37] Therefore, risk of harm needs to be considered on a study-by-study basis. In many studies, risk of harm may be small compared with the potential benefits of the research.[38-40]

Research implications

Participation in research may have benefits for both the individual and the community. Patients involved in a clinical trial may have better outcomes than those who are not enrolled.[41 42] Patients have also reported that they enjoy participating in research as they feel as though they are helping others who may be facing a similar situation.[43] As well as benefits for the individual, research also provides results which may lead to improved outcomes for the wider cancer population.[44] Love and colleagues recruited patients with axillary node negative breast cancer through a cancer registry to take part in a randomised controlled trial (RCT) which highlighted the effectiveness of tamoxifen for reducing future risk of cancer.[45 46] Similarly, Irwin and colleagues recruited breast cancer survivors through a cancer registry to take part in an RCT which resulted in increased exercise in the intervention group, again reducing future cancer risk.[47] In instances such as these, it is necessary to weigh up the small risk of harm to individuals from being contacted by a registry if they are unaware of their diagnoses or

too ill to participate, with the potential research benefit to the individual and to the wider cancer population.[39 40 44]

If neither a high consent rate nor a representative sample is achieved, the results of a study may not generalisable to the wider cancer population.[48] In this study, the needs reported by the 32 AYAs who completed the questionnaire may not represent a true reflection of the overall needs of the AYA population. The small sample size also means that it is difficult to do anything meaningful with the data as there is limited statistical power to test hypotheses or validate instruments, and parameter estimates will not be very precise. For example, predictors of risk for AYAs with high levels of unmet need would not be able to be examined due to the small sample size.

Recruiting through more than one registry may be one way to increase the sample size. However, if low consent rates are achieved in all registries, the potential for bias in the sample remains. For example, if other registries also use an active clinician consent protocol and the consent rates at each stage of recruitment are the same as in this study, recruiting an adequate sample would appear unlikely. The current study was conducted with the largest state cancer registry in Australia; even if four additional registries were used, the number of AYAs who consent and participate in the research would still be under 200 participants due to the smaller populations in these states. Other issues, including the burden of gaining approval from numerous cancer registry ethics committees and the cost of research and registry personnel, would also need to be taken into account.[49]

Cost-related implications

There are high public and individual costs involved in conducting research. The vast majority of research is publicly funded through either large government organisations

such as the National Cancer Institute (NCI) in the United States and the National Health and Medical Research Council (NHMRC) in Australia, or through charitable organisations which rely on donor contributions.[50 51] For this reason it is of paramount importance that the public gets a good return on investment. However, the process of recruitment through registries, especially when active clinician consent is required, can be expensive in terms of time, energy and resources[49], and for some population groups may not be cost-efficient. Many registries charge researchers for costs associated with staff time for identifying eligible cases, mail-outs and telephone calls to clinicians and potential participants. The research team will also incur staff costs associated with development of the study protocol, ethics applications, study management and liaison with the registry. In the present case where only 32 AYAs (7.8%) were recruited from 411 potentially eligible AYAs, the enormous staff and material costs of the research appear to be out of proportion to the number of participants recruited and the usefulness of the data collected.

In addition to costs to the public, registry and the researchers, there are also costs to the individual clinicians and survivors who participate. The active consent protocol requires clinicians to provide consent for all their eligible survivors to be contacted by the registry. This adds an additional burden to already time-poor clinicians, especially if a large number of their patients have been identified.[49] The utility of this approach is doubtful when subsequent contact with AYA survivors is only successful in a small proportion of cases, due to registries not having current contact details for survivors. This would appear to be an inefficient use of time for both the registry and the clinician, particularly if clinicians no longer have contact with these patients.[12]

The active consent protocol also places a burden on survivors. In the current study there is a cost to the 32 survivors who participated in the study and completed the

survey. They have expended time and effort reporting their unmet needs while, given the low overall response rate, it is difficult to do anything meaningful with their data. In light of the cost and limited usefulness of the data obtained, it is important to explore possible alternatives that could be used to overcome these low consent rates.

Resource-efficient alternatives for achieving representative research samples

There are a number of alternative protocols which could be considered as feasible options when attempting to recruit a population-based sample through a cancer registry.

Passive clinician consent

The state where the current study was conducted uses an active clinician consent protocol, while in other countries and Australian states only passive clinician consent is required.[12 13] This means that clinicians are not required to respond to the registry unless they wish to indicate specifically that a patient should not be approached by the registry. Passive clinician consent may help to reduce the burden placed on clinicians, and has been used successfully by a number of international registries and studies.[12 13] However, given the fact that many clinicians may no longer have contact with AYA survivors, this method may still not be viable.

Other approaches may involve the registry giving clinician and survivor details directly to the researchers. The researchers then make the initial contact with the clinician, rather than the registry. Over 60% of cancer registries in the United States use this approach.[12] However, in Australia, because registries are notified usually under a Public Health Act, information cannot be provided to a third party without survivor consent. The majority of registries using this researcher-initiated protocol required passive clinician consent (70% of registries) and a patient opt-out approach (86% of

registries).[12] The process of researchers notifying a survivor's clinician prior to contacting the survivor about a research study, however, may still be viewed as paternalistic. Beskow and colleagues reported that over two-thirds of patients (68%) said that they preferred that researchers contact them directly about opportunities for research participation, rather than checking with their physicians first.[52] Therefore, alternatives to a passive clinician consent model should be explored.

Direct survivor consent

A novel method for assisting registries to provide cancer survivors with information about the range of research studies for which they are eligible may be through direct survivor contact.[52 53] This could involve a one-off contact with a survivor by the registry which would allow the survivor to indicate whether or not they would like to be contacted about research studies in the future.[7] At this point the survivor could determine which, if any, types of research they would like to be contacted about, and how often these contacts could be made, for example every six months or once every two years. This consumer-driven approach would allow survivors to choose the focus of research studies they are interested in participating in and negate the need for clinician consent to contact the survivors.

In a national household survey involving a random sample of the British public, Barrett and colleagues found that over 80% of participants did not feel having their names and addresses listed on a cancer registry, or being sent letters inviting them to take part in a research study, was an invasion of privacy.[53] In addition, although 82% of participants did not know the registry existed, 95% thought that the information collected was useful.[53] In anticipation of low survivor awareness of the registry, the cancer registry could send an information leaflet as part of their initial contact with

survivors to help survivors better understand the role of the registry and why they are included on it.

The perception of the cancer registry by the public and professionals would also need to be considered. There is a risk that the role of the registry in fostering and encouraging research may come under criticism or impact on the completeness of case ascertainment, particularly if notification to the registry is raised as an issue of concern by individuals who are approached for study participation. Protocols for removing the names of survivors from registry research participation lists would also need to be carefully planned in terms of registry workload.

Recruitment of AYAs via treatment centres and clinics

Direct consumer contact may provide greater survivor autonomy and overcome problems such as low clinician response rates. However, it may still not be feasible with highly mobile groups such as AYAs, as the issue of difficulty in obtaining up-to-date contact details remains. An alternative mechanism for achieving representative samples of AYAs diagnosed with cancer may be recruitment from treatment centres and clinics. The low prevalence of AYAs diagnosed with cancer would usually mean that a sufficiently large sample is unlikely to be achieved through just one clinic. Recruitment from a number of different clinics or treatment centres may help to increase the size and representativeness of the sample. This would also have the advantage of providing more up-to-date contact details for individuals, especially if AYAs have attended subsequent follow-up appointments once their active cancer treatments have ceased. In the case of retrospective studies, electronic medical records could be searched for eligible participants, and clinicians at the treatment centres would be able to confirm their eligibility. A number of AYA research studies have successfully recruited samples which are generally representative of the larger

AYA population group in this way.[54 55] One complication may be the need to target both adult and children's hospitals to obtain a sample which adequately covers the age range of the AYA population.

Limitations

Limitations of the study should be considered when interpreting the findings of the current study. First, recruitment was only conducted through one state registry in Australia. Therefore, it is possible that the low recruitment rates obtained may have been specific to this registry. Studies which have compared recruitment rates between registries using active or passive clinician consent, however, have generally found that recruitment rates are lower in registries requiring active consent.[13]

Second, the age group that was targeted in this study was quite small, with only AYAs aged 14 to 19 years at diagnosis being eligible. However, the number of AYAs listed on the registry as meeting this criterion was adequate to meet the necessary sample size if clinician and survivor consent rates had been about 75%. Other studies with AYA cancer patients have achieved response rates above 75%, indicating that this expectation would not seem unreasonable. Increasing the upper age limit of AYAs to 25 or 30 years[41] might have helped to increase the sample size.

Conclusion

Modifications to the stem, response scale and wording of items may help to increase the acceptability of the CNQ–YP and will be incorporated into the next version of the measure. Further psychometric testing of the measure, including the establishment of internal consistency, test-retest reliability, construct validity and responsiveness will be performed. The length of the CNQ–YP will also be reduced once the psychometric properties of the questionnaire have been established.

Despite the potential for cancer registries to provide researchers with access to large and representative samples, current registry protocols, such as active clinician consent, may inhibit this process. Alternative methods such as passive clinician consent or direct survivor consent may help to overcome some of these barriers. In the case of AYAs diagnosed with cancer who are highly mobile, recruitment through registries may not be feasible, and other alternatives such as recruitment through treatment centres and clinics may need to be considered.

CHAPTER 5

Psychometric properties of the Cancer Needs Questionnaire – Young People (CNQ–YP): reliability and validity

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Chapter 5

Psychometric properties of the Cancer Needs Questionnaire – Young People (CNQ–YP): reliability and validity

Introduction

Chapters 3 and 4 of this thesis have outlined the initial steps in the development of the Cancer Needs Questionnaire – Young People (CNQ–YP): item generation; development of a response scale; establishment of face and content validity; and pilot testing with a population-based sample to eliminate any ambiguity in item wording and establish acceptability. The following chapter describes the next steps in the process of measure development. These include performing factor analysis, assessing internal consistency and test-retest reliability and determining the discriminative validity and potential responsiveness of the scale.[1 2]

Methods of item reduction and selection

Reducing the number of items in a scale is essential for ensuring that all items capture the most important elements of the issue being measured, without being redundant or unrelated.[3] Shorter measures have benefits for patients, providers, researchers and the health system by reducing the burden placed on individuals and the time necessary to administer and complete questionnaires.[3 4]

In addition to establishing the reliability and construct validity of a measure, the three methods which can assist in item reduction include factor analysis, examining internal consistency and establishing test-retest reliability.[2]

Factor analysis

Factor analysis is a statistical method that can be used to assess the internal structure of, and reduce the number of items in a multi-dimensional scale.[1 5] By determining the patterns of inter-correlations between responses to items, factor analysis attempts to group items which appear to measure a common underlying theme or factor.[1 5 6] Each factor should be unique, measuring a different concept or construct from any other factors identified.[1]

Factor analysis can either be exploratory or confirmatory. Exploratory factor analysis is used when no pre-existing assumptions regarding the structure of the measure have been made.[2 5-7] This method begins with the items and determines how they best group together to represent underlying factors.[1] In contrast, confirmatory factor analysis seeks to test how well individual items fit within an existing conceptual structure or model.[1 2 7] A factor structure is hypothesised and analysis is performed to test whether data fit the structure.[5 6]

Exploratory factor analysis can be conducted using one of two methods: principal components analysis (PCA); or principal factor analysis (PFA). The principal components method aims to explain all variance, both common and unique, in the data.[5 6] It is the most appropriate method to use when the purpose of the analysis is item reduction.[5 6] The principal factor method only explains the common variance in the items.[5 6] This method is most useful when the aim of analysis is to identify correlations and covariance among items.[5 6]

Once the initial factor analysis has been performed, factors are usually rotated to simplify and clarify the factor structure.[8] Rotation can be either orthogonal or oblique. Orthogonal rotation methods (i.e. varimax, quartimax and equamax) produce factors

that are uncorrelated.[8] Oblique rotation methods (i.e. oblimin, quartimin and promax) allow correlation between factors.[8]

When performing factor analysis, a number of assumptions or principles should be observed: 1) items should be measured using an interval level response scale; 2) responses should be normally distributed; and 3) the number of participants in the sample should be at least five times the number of items in the measure.[1] Factor analysis should also be performed with caution, as there is some evidence to suggest that relying on factor loadings alone for item reduction can lead to the elimination of items of high importance to participants.[9] For this reason, the proportion of participants endorsing an item, as well as the correlation of an item with other items, should be observed when making decisions regarding the exclusion of an item from a measure.[9]

Internal consistency

It is important for scale development that all items within a sub-scale or domain are homogenous, that is, all items should be related to each other, as well as to the domain they are designed to measure.[7] To determine how homogenous the items are, a test of internal consistency is performed. Items within a domain or factor should be: 1) moderately correlated with each other; and 2) correlated with the total score of the measure.[7] Item-to-item correlations should be moderate rather than high, as high correlations suggest that the items are measuring the same issue and therefore at least one of the items is redundant.[7]

When performing tests of internal consistency two methods are usually applied: item-total correlation; and Cronbach's alpha.[1 7]

Item-total correlation

Item-total correlations provide an indication of the homogeneity of items in a scale by describing how much each individual item contributes to the overall construct being measured.[1] The score for an item within a domain or scale is correlated with the total score of all remaining items.[1 7] Either the point-biserial correlation (for items with dichotomous response options) or the Pearson product-moment correlation (for items with more than two response options) can be used.[7] It is usually agreed that items with low total score correlations (<0.20) should be removed from the scale.[7 10]

Cronbach's alpha

Cronbach's alpha[11] is defined as the square of the correlation between a domain or scale and the underlying factor the scale is supposed to measure.[1 12] The value of alpha can range from 0 (no correlation among items) to 1 (perfect correlation among items).[1] There are limitations to Cronbach's alpha, however, as the value of alpha is often dependent on the number of items within a domain or scale.[1] A scale or domain with a large number of items (e.g. greater than 11 items) will usually have a very high Cronbach's alpha, even if items are only moderately correlated.[7 13] A domain or scale with a Cronbach's alpha between 0.70 and 0.95 is generally recognised as having good internal consistency.[13] Cronbach's alpha is used when an item has more than two response choices.[1] For items with dichotomous response options, the Kuder–Richardson formula 20 (KR–20) can be used as an equivalent to alpha.[1 14]

Test-retest reliability

Test-retest reliability is concerned with the stability or reproducibility of scores on a measure over subsequent administrations when changes in the underlying state are not expected.[15] The recommended time-frame between baseline and retest measure completion is usually two to fourteen days.[16] Depending on the type of response scale,

one of two statistics are appropriate for assessing agreement between scores at test and retest: the intra-class correlation coefficient; or Cohen's kappa coefficient.[15]

Intra-class correlation coefficient

The intra-class correlation coefficient (ICC) is the most appropriate statistic for assessing the test-retest reliability of continuous measures as it takes into account not only agreement from baseline to retest, but also the average similarity of participant scores at both administrations.[1 13 17] Regular correlations (such as the Pearson product moment correlation) are able to determine correlations between scores at time one and time two, but fail to detect systematic differences in the mean values of scores between the two time points.[1 17] For example, if all individuals systematically score better on the second administration, the scores will be highly correlated, but will have low agreement.[1 17] The ICC allows for this by taking into account consistent variance among participants over time.[7] Coefficients range from -1 to +1, with an ICC value >0.70 indicating good reliability.[1 15 18]

Cohen's kappa coefficient

Agreement between baseline and retest scores for categorical measures (ordinal or nominal) is calculated using Cohen's kappa coefficient.[1 13 19] Kappa accounts for the possibility that agreement between the two scores is by chance alone. It estimates the amount of agreement that would be expected by chance and removes it from the estimation.[1] A weighted kappa can be used to distinguish between minor and major discrepancies in scores at baseline and retest.[1] The most commonly used weighting scheme involves quadratic weights, where disagreement weights are based on the square of the amount of discrepancy between scores.[7] When a quadratic weighting scheme is used, the kappa coefficient is identical to the ICC.[7 13 20] Items that have a

kappa or weighted kappa: ≤0.40 are considered to have poor/slight agreement, 0.41-0.60 moderate/fair agreement, and >0.61 substantial/excellent agreement.[7 21 22]

Methods of determining discriminative validity

Discriminative validity, a form of construct validity, is based on the theory that different groups of participants will achieve different scores on a measure.[2 15] The ability of a measure to distinguish between different participant characteristics is usually examined using a known-groups comparison.[2]

Known-groups comparison

In a known-groups comparison, an hypothesis is generated that one group of participants (for example, individuals with a particular health state or disease) are expected to produce significantly better or worse mean/median scores on the measure, compared with another group which does not possess the characteristic.[7] The hypotheses should be directional and made *a priori*, and may be based on previous literature, research or recognised clinical differences.[7]

Methods of determining responsiveness and sensitivity to change

In the context of health research, responsiveness and sensitivity to change refer to a measure's ability to detect changes in an individual's health state over time.[1] While sensitivity to change can refer to any change in a health state, responsiveness is usually concerned with clinically important change.[7 23] Many statistics to assess the responsiveness of health measurement scales have been proposed,[24] and are generally concerned with measuring the effect size or the magnitude of the change. To be able to detect change, a measure must first be able to demonstrate an absence of floor and ceiling effects.[24]

Floor and ceiling effects

Calculating floor and ceiling effects is necessary to determine how well a measure reflects the various levels of health reported by different groups of patients. If a large proportion of respondents achieve the lowest (floor) or highest (ceiling) possible scores on a measure, existing items may be unable to distinguish between different levels of well-being.[25] Responsiveness will also be limited in the sense that any improvement or worsening of health in these patients will not be detected, as higher or lower possible scores do not exist.[13]

Aims

The purpose of this research was to assess the psychometric properties of the Cancer Needs Questionnaire – Young People (CNQ–YP). Specifically, the aims were to evaluate the reliability and validity of the measure and to reduce the number of items by: 1) performing exploratory factor analysis to identify underlying factors; 2) establishing the internal consistency of items and identified factors using item-total correlation and Cronbach's alpha; and 3) assessing the test-retest reliability of the measure using kappa. Further aims were to: 4) determine the discriminative validity of the measure using a known-groups comparison; 5) identify floor and ceiling effects to determine the potential responsiveness of the scale; and 6) reassess the acceptability of the CNQ–YP following a revision of the question stem, response scale and wording of items (as recommended in Chapter 4).

Methods

Setting

Adolescents and young adults (AYAs) diagnosed with cancer were recruited from seven major adult cancer treatment centres across Australia. These included two

centres from New South Wales (NSW), two from Victoria (VIC), one from Queensland (QLD), one from South Australia (SA) and one from Western Australia (WA). Recruitment from treatment centres rather than cancer registries was undertaken due to the low response rates experienced in the registry-based pilot study (Chapter 4). It was anticipated that recruiting young people from seven treatment centres across five Australian states would optimise the representativeness of the sample and the generalisability of the results. Given that adult hospitals treat the majority of adolescent and young adult patients diagnosed with cancer,[26-28] children's hospitals were not approached.

Participants

Adolescents and young adults were eligible to participate in the study if they had not participated in the pilot study, had received treatment for cancer at one of the seven identified treatment centres, and were: 1) diagnosed with an invasive cancer in the previous five years; 2) aged 14-25 years at the time of diagnosis; 3) residents of NSW, VIC, QLD, SA or WA; and confirmed by their treating clinician as: 4) having a life expectancy of at least 12 months; 5) physically and mentally able to complete a survey; and 6) sufficiently literate in English to complete the survey.

As described in Chapter 4, the lower cut-off age of 14 years conforms to Australian laws specifying that young people over 14 years of age have the legal right to make decisions regarding the types of health care they receive.[29] The upper cut-off of 30 years (e.g. diagnosed at 25 years of age and up to five years post-diagnosis) conforms to existing definitions of young adulthood in the oncology literature[30-33] and reflects the unique types of cancer which affect this age group.[28 34 35] Identifying young people diagnosed with cancer in the previous five years also allowed young people who were at different stages of their cancer journeys to be recruited.

Procedure

Ethical approval for the study was granted by the Human Research Ethics Committees of each hospital (Appendices 5.1-5.6), as well as by the University of Newcastle (Appendix 5.7). Data about eligible young people were extracted from patient databases at each treatment centre. Due to the high volume of patients identified at two of the treatment centres, data for each potential participant were cross-checked against the death register in the relevant states to confirm the young person's survival status. For the remaining five hospitals, the survival status of the young person was confirmed by the primary treating clinician.

Clinicians' contact with patients to obtain consent

In NSW, QLD, SA and WA, the principal clinician at each hospital sent a letter of invitation, project information sheet, consent form and reminder letter to eligible AYAs (Appendices 5.8-5.12). In VIC, the principal clinician sent eligible young people a letter of invitation, project information sheet and a "do not contact" form (Appendices 5.13-5.14). The letter of invitation sought the young person's consent to forward contact details to the research team. Methods of obtaining consent varied slightly in accordance with the requirements of each state. Adolescents and young adults in VIC only received one invitation letter and were asked to return a "do not contact" form within four weeks if they did not wish to participate in the study. Adolescents and young adults who did not return a "do not contact" form were considered to have provided passive consent to receive correspondence from the research team. Those in the remaining states were sent initial letters of invitation, and received reminder letters after two weeks and reminder telephone calls after four weeks if they had not returned the consent forms (i.e. active AYA consent was required).

The principal clinician provided the research team with the contact details of participants who agreed to be contacted. For AYAs who did not consent to be contacted, the principal clinician provided de-identified demographic details (i.e. gender, age at diagnosis, date of diagnosis and cancer type) to the research team so that any differences in the characteristics of consenters and non-consenters could be determined.

Data collection by the research team

The research team sent participants who had returned a consent form, or had not returned a "do not contact" form, an information package which comprised of a study invitation letter (Appendix 5.15), information sheet (Appendix 5.16), copy of the Cancer Needs Questionnaire – Young People (CNQ–YP) (Appendix 5.17) and a reply-paid envelope. The original study plan approved by the ethics committees included a randomised controlled trial to compare the acceptability of the paper and pencil version of the measure with an online version. However, due to the limited number of eligible young people identified at the participating hospitals, randomisation was not possible. It was decided that only one version of the measure should be administered to all participants and, given that the pencil and paper version had been pilot tested and demonstrated acceptability (Chapter 4), this was the format chosen.

As the pilot study with AYA cancer survivors recruited through the cancer registry had achieved a poor response rate (Chapter 4), five strategies recommended by the Cochrane Systematic Review on methods to increase response rates to posted questionnaires were used.[36] These included: 1) *pre-notification of the study* – a study information sheet was included in the initial contact with patients by clinicians; 2) *follow-up contact* – participants who had not returned questionnaires were sent reminder letters after two weeks (Appendix 5.18) and received reminder telephone calls after

four weeks; 3) *providing a second copy of the questionnaire at follow-up* – all reminder letters were accompanied by a second copy of the measure; 4) *an assurance of confidentiality* – both the letter of invitation and study information sheet clearly stated that all responses would be kept strictly confidential; and 5) *university sponsorship* – the University of Newcastle logo appeared on all envelopes, letterheads and copies of the measure.[36] All correspondence clearly indicated that the study was being conducted by researchers at the university, formed part of a PhD thesis and had the approval of the University of Newcastle Human Research Ethics Committee.
Return of the completed measures was taken as participants' consent for their data to be included in the study. In order to evaluate the test-retest reliability of the measure, all participants who agreed to be contacted again were sent copies of the CNQ–YP one week later and asked to complete the measure a second time (Appendix 5.19). The one-week time-frame was chosen to minimise the chance that patients had substantial changes in their unmet needs or could recall their previous responses.[13]

Measure

Cancer Needs Questionnaire – Young People (CNQ – YP)

The CNQ–YP was developed and piloted with AYA cancer survivors from across Australia, and has face and content validity (Chapter 3). The 144 items in the measure were presented in eight domains derived from the literature and focus groups: 1) Cancer Treatment Staff (36 items); 2) Cancer Treatment Centre (11 items); 3) Education (12 items); 4) Work (12 items); 5) Information (9 items); 6) Feelings (35 items); 7) Relationships (19 items); and 8) Daily Life (10 items). For all domains, items were rated using a five-point response scale from "No Need" to "Very High Need". This was a revision of the previous response scale which rated items from "No Unmet Need" to "Very High Unmet Need".

Following feedback from participants in the pilot study (Chapter 4), the time-frame of the response scale was also modified to “any time since your cancer diagnosis” for the first five domains (Cancer Treatment Staff, Cancer Treatment Centre, Education, Work and Information). For the remaining three domains (Feelings, Relationships and Daily Life), the response time-frame remained “in the last month”. Screening questions were added to the Education, Work and Relationships domains so that AYAs were able to skip these questions if they were not relevant to their current situations. Four screening items determined whether the young person was currently employed, studying or both: 1) “Since my cancer diagnosis, I have had problems enrolling at (place of study)”; 2) “Since my cancer diagnosis, I have attended (place of study)”; 3) “Since my cancer diagnosis, I have had problems finding work (type of work)”; and 4) “Since my cancer diagnosis, I have been employed (type of work)”. Questions in the Relationships domain related to partners and siblings were only answered if the AYA reported having these relationships, again determined by a screening item: “Do you have: a spouse/partner or boyfriend/girlfriend; sibling/s or step-brothers/sisters; none of the above”.

Demographic characteristics and measure completion

Participants were asked to provide demographic information including their age, gender, cancer type, time since diagnosis and types of treatment received. Participants were also asked where they had completed the measure (e.g. home, work or cancer treatment centre). Information concerning the average time between measure administration and completion was obtained.

Measure acceptability

Four questions regarding the acceptability of the measure were asked by using the following questions: 1) “I found the instructions easy to follow”; 2) “I found the questions

clear"; 3) "I found the answer choices easy to understand"; and 4) "I found the questions distressing".

Additional questions

Additional questions included the following: marital status; language preferences; living arrangements; education; preferences regarding the survey format and locations for survey completion; and feedback of results to health professionals, treatment centres, researchers and other organisations. The results of analyses relating to these items are reported in Chapter 6.

Statistical analysis

Statistical analysis was performed using Stata Version 11 statistical software.[12]

Demographic characteristics, consent bias and measure completion

The demographic characteristics of participants and details regarding measure completion were reported using descriptive statistics (i.e. frequencies, proportions, means, standard deviations, medians and quartiles). The demographic characteristics of AYAs who consented and AYAs who did not consent to participate in the baseline and test-retest phases of the study were compared using either t-tests (for continuous variables) or Chi square tests (for categorical variables).

Item reduction

Proportions and missing values

The proportions of responses at each level of need, for each item, were calculated and histograms produced to assess the distribution of each item. Items which had greater than 90% of respondents indicating the same level of need were eligible for exclusion.

The proportion of missing values for each item was also calculated. Items which had not been answered by >10% of respondents were also eligible for exclusion.

Aim 1: Perform exploratory factor analysis to identify underlying factors

The remaining items were eligible for inclusion in the exploratory factor analysis. Exploratory, rather than confirmatory, factor analysis was used, as the intention of the analysis was to determine previously unknown relationships between items in the measure (factor structure).[1 2 5-7] Observations which had missing values for any item were excluded from the analysis using list-wise deletion.[37] List-wise deletion refers to the removal of all observations which have a missing value for any item. Although list-wise deletion may result in a smaller number of cases being included in the analysis, and therefore less statistical power, it provides more accurate estimates of correlations than other methods of dealing with missing values, such as pair-wise deletion or mean substitution.[37] Pair-wise deletion (deletion of an observation only for the missing item) can lead to inconsistent correlations due to variable sample sizes.[37] Mean substitution (replacing missing items with the average of all other scores for that item) can result in a decrease in estimates of variance.[37] For factor analysis, list-wise deletion is particularly recommended over mean substitution, as replacing missing items with average scores can lead to an artificial increase in the clarity of factor structures.[37]

The principal components method of factor analysis (PCA), rather than principal factor method (PFA), was used as the purpose of the analysis was to reduce the number of items in the measure.[5 6] No limitations were placed on the initial factor analysis, and the number of factors identified was determined by the Eigenvalue >1 rule.[8] The factors were then orthogonally rotated using the varimax procedure to simplify the factor structure of the measure.[8] Orthogonal, rather than oblique, rotation was used as the underlying factors were expected to be uncorrelated and measure unique areas of

need.[2 8 25 38] Factors which accounted for greater than 5% of the variance were considered important.[39] The number of factors to be retained was confirmed using a Cattell scree plot, with the number of factors above the "elbow" or break in the graph indicating the number of factors to retain.[2 8]

Subsequent factor analysis and rotation was performed, with the number of factors limited and reduced until each factor contained more than three items with unique loadings >0.40.[8] It is recommended that each factor be comprised of at least three items with strong loadings, as factors with less than three items can be weak or unstable.[8] The cut-off of 0.40 was chosen, as factor loadings greater than 0.40 are considered large.[39 40] Factors were also reviewed to confirm whether items on the same factor appeared to measure similar concepts, and whether items on different factors measured different concepts.[39] Once the final factor solution had been identified, the process of item reduction was performed. Item exclusion and inclusion were based on the criteria described below.

Exclusion criteria: The item has no loadings >0.40 on any factor *or* has a factor loading of >0.40 on two or more factors[8] and <20% of participants indicated having a high or very high need for that item. These criteria reflect recommendations that items with factor loadings <0.4 are unlikely to be related to other items in the measure, and items which cross-load on factors should be dropped if there are a number of other items which have strong and unique loadings on that factor.[8] The <20% rule was used to ensure that items which did not appear to correlate highly with other items, but had been identified as a high or very high area of need by a large proportion of young people, were not eliminated.[9]

Inclusion criteria: The item has a factor loading of >0.40 on only one factor[39 40] *or* >20% of participants indicated having a high or very high need for that item. When the latter occurred, items were assigned to the factor where they had the highest factor loading.

Due to the different number of observations for the six main domains (Cancer Treatment Staff, Cancer Treatment Centre, Information, Feelings, Relationships, Daily Life) and the additional domains completed by participant sub-samples (Education, Work, Partner and Siblings), only items from the six main domains were included in the initial factor analysis. Once the number of underlying factors had been identified, items from the remaining domains were added independently (by domain) to the analysis as additional factors. The procedure for retaining items for each of the additional domains followed the same inclusion and exclusion criteria outlined above.

Following factor analysis, an item-item correlation matrix for each of the identified factors and remaining items was created to identify whether any extra items within the factors could be excluded. Spearman's rank correlation coefficient rather than Pearson's product-moment correlation coefficient was used, as the item response scales were ordinal.[41] Items which were highly correlated with each other (correlations >0.90), and which had <20% of participants indicating a high or very high need, were examined. One of the correlated items was excluded if both items measured the same aspect of the factor.

Aim 2: Establish the internal consistency of items and identified factors

The internal consistency of items included in each identified factor was assessed using item-total correlations and Cronbach's coefficient alpha (α).[1 7 11] Cronbach's alpha, rather than the Kuder–Richardson formula 20 (KR–20), was used as the item response scales had more than two response choices.[1] Items which had an item-total correlation

with the total scale of <0.20 were discarded.[10] For each factor, an alpha value of >0.70 and <0.95 was considered acceptable.[7 13]

Aim 3: Assess the test-retest reliability of the measure

It is usual protocol to assess test-retest reliability and subsequently reduce the number of items in a measure, prior to conducting factor analysis.[39] However, given the small number of individuals who participated in the test-retest phase of the current study, the decision was made to perform factor analysis on the larger baseline sample prior to evaluating test-retest reliability. This allowed the number of items included in the test-retest analysis to be reduced, thereby limiting the likelihood of type I error. Cohen's kappa coefficient (κ), rather than the intra-class correlation coefficient (ICC), was used to measure the level of agreement between responses at baseline (time 1) and retest (time 2). Kappa was selected as the levels of need on the response scale were more akin to categorical (ordinal) than continuous data. As recommended by Ciccheti (1976), a weighted kappa was used as this is best suited for ordinal scales where the lowest value indicates the absence of an issue, and the remaining values measure the degree to which the issue is present.[7 42]

Quadratic weights were calculated as $1 - \left\{ \frac{(i - j)}{(k - 1)} \right\}^2$ where *i* and *j* index the row and column of the two ratings, and *k* is the maximum number of possible ratings.[12] Items which had a weighted kappa of >0.60 were considered to have excellent test-retest reliability and were retained.[7 22] To ensure items related to high unmet needs were not dismissed, items which did not obtain a weighted kappa of >0.60 but for which >20% of participants indicated having a high or very high need, were also kept.

Revised factor analysis and Cronbach's alpha

Following the assessment of test-retest reliability and subsequent removal of items, factor analysis was re-run to confirm the factor structure of the measure and to identify any further items that were eligible for exclusion. Internal consistency of the measure was also re-checked to ensure the item-total correlations and Cronbach's alpha had not significantly changed.

Creation of factor scores

Once the final factor structure of the measure had been confirmed, factor scores were calculated so that scores for different groups of participants could be compared.[43] Two main classes of calculating factor scores exist: non-refined methods; and refined methods.[43] Non-refined methods involve simple summation techniques such as sum of scores by factor, sum of scores above a cut-off value, sum of scores on standardised variables, and weighted sum of scores.[43] Refined methods use linear relationships between the item and the factor to create a factor score and include regression scores, Bartlett scores and Anderson–Rubin scores.[43]

The sum of scores by factor method was used in the current study as this method preserves the variation in the original responses and is recommended when a measure is newly developed and analysis is exploratory.[43 44] All items in the measure were worded and scored in the same positive direction; therefore, no reversing of response scores for items was required.[2] "No Need" responses were given a raw score of one, and scores increased by intervals of one up to the "Very High Need" response with a score of five. Observations with missing values for >50% of items within a factor were excluded from the analysis. Factor scores were then calculated for the remaining participants by summing all raw scores for items within the factor and dividing by the number of non-missing items. Averaging the raw score enabled a comparison of factor

scores among factors with differing numbers of items, as all factor scores ranged from one to five; this also allowed for observations with missing values to be included without the need for imputation.

Aim 4: Determine the discriminative validity of the measure

Based on previous quality of life research with AYA cancer survivors,[45-48] it was hypothesised that young people receiving treatment (i.e. newly diagnosed or receiving treatment) would have a higher level of need, and therefore a higher median factor score for all factors, compared with young people who had finished treatment (i.e. finished treatment and having check-ups, or in remission). As the responses for most items were not normally distributed, medians and quartiles for both groups were calculated and the non-parametric Wilcoxon rank-sum test was computed to determine if any significant differences in the factor scores of the two groups existed.

Aim 5: Determine the potential responsiveness of the measure

Assessment of the measure's potential responsiveness and ability to detect change was gauged using floor and ceiling effects. The proportion of AYAs who had a factor score equal to the minimum (factor score=1) or maximum (factor score=5) possible score for each factor was calculated to determine whether any floor or ceiling effects existed.[13] Factors where less than 5% of participants scored the lowest possible score or the highest possible score were considered acceptable.[25]

Aim 6: Reassess the acceptability of the measure

The acceptability of the CNQ–YP was described using frequencies, proportions and 95% confidence intervals (CIs).

Results

Demographic characteristics, consent bias and measure completion

Of the 577 eligible young people identified by clinicians at the participating hospitals, 280 (49%) consented to be contacted by the research team. Of the 280 young people contacted, 139 (50%) returned questionnaires (24% of the 577 young people identified). The majority of participants completed the measure in their own home (n=122, 88%), with a small proportion completing it at work (n=9, 6.5%) or another location (n=8, 5.8%). The time taken to return the completed measure ranged from 10 to 116 days, with a median of 30 days (Q1=18 days, Q3=46 days).

Of the 139 participants who completed the measure at baseline (time 1), 116 (83%) consented to be contacted a second time. Of these, 15 (13%) were not approached as their retest time-frame fell within the two-week Christmas holiday period and was considered an inappropriate time for contacting young people. The remaining 101 participants were sent a retest survey, with 34 (34%) completing the measure at time 2. This meant that approximately one quarter (24%) of the participants who completed the measure at time 1 also completed it at time 2.

The time between being sent the retest measure and completing and returning the retest measure ranged between 9 and 64 days, with a median of 24 days (Q1=16 days, Q3=30 days). This was greater than the recommended 14 days.[16] However, as the response time-frame for the measure was “in the last month” for most items, a median retest period of less than one month did not appear to be unreasonable.

The demographic characteristics of AYAs who consented to participate in the baseline study, and those who did not consent, can be seen in Table 5.1. Consenters were significantly younger than non-consenters, and females were over-represented in the

consent sample. There were no significant differences in the demographic details of young people who completed the baseline and retest survey and those who completed the baseline survey only (Table 5.2).

Table 5.1: Demographic characteristics of consenters and non-consenters for the baseline study

Demographic Characteristic		Non-consent (baseline) n=438	Consent (baseline) n=139	Test statistic		
		n (%)	n (%)	χ^2	df	*p*
Gender	Female	188 (43)	89 (64)	18.8	1	<0.00*
	Male	250 (57)	50 (36)			
Cancer type	Haematological	194 (44)	65 (47)	0.31	1	0.58
	Non-haematological	243 (56)	73 (53)			
Time since first diagnosis	< 2 years	139 (32)	40 (29)	0.56	1	0.45
	≥ 2 years	293 (68)	99 (71)			
		Median (Q1-Q3)	**Median (Q1-Q3)**	**z**		***P***
Age at diagnosis		22 (19-24)	21 (18-23)	2.24		0.03*

**p value <0.05*

Table 5.2: Demographic characteristics of young people who completed the baseline and retest survey, and those who completed the baseline survey only

Demographic Characteristic		Completed baseline only n=105	Completed baseline and retest n=34	Test statistic		
		n (%)	n (%)	χ^2	df	*p*
Gender	Female	66 (63)	23 (68)	0.26	1	0.61
	Male	39 (37)	11 (32)			
Cancer type	Haematological	45 (43)	20 (59)	2.49	1	0.12
	Non-haematological	59 (57)	14 (41)			
Time since first diagnosis	< 2 years	32 (30)	8 (24)	0.60	1	0.44
	≥ 2 years	73 (70)	26 (76)			
		Median (Q1-Q3)	**Median (Q1-Q3)**	**z**		***p***
Age at diagnosis		21 (18-23)	21 (18-23)	-0.97		0.33

Reported reasons for non-consent to participate

Fifteen AYAs provided reasons for not wishing to participate in the study. Of these, 11 (73%) reported they did not like to talk or be reminded about cancer, 3 (20%) considered themselves cured of cancer and 1 (6.7%) reported being too busy.

Item reduction

Proportions and missing values

The proportion of participants who responded at each level of need, for each item, can be seen in Appendix 5.20. No items had >90% of participants reporting the same level of need, indicating reasonable variability of responses within items. Only three items had a response option which was not utilised. These items were all from the Relationships domain: item 116 – "Coping with my parent/s not giving me enough attention"; item 126 – "Coping with my partner giving me too much attention"; and item 128 – "Coping with my partner being over-protective". For these items either the "High Need" or "Very High Need" response options were not used. Histograms for each item and their response options also revealed reasonable variability. The majority of histograms were skewed to the right with the highest proportions at the "No Need" level and a decrease in proportions as the level of need increased.

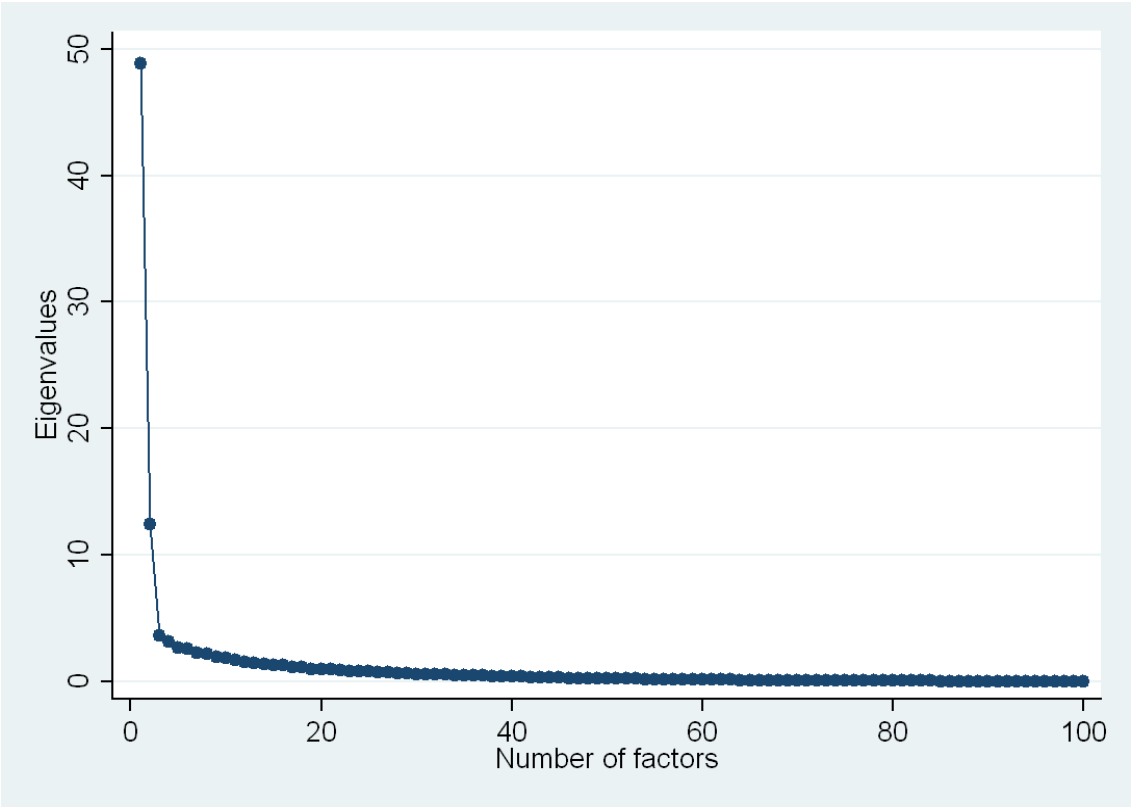
The proportion of missing values was low, ranging from 0% to 10.9% (Appendix 5.20). The highest proportion of missing values was seen in the Partner sub-scale of the Relationships domain. However, only a sub-sample of participants (those with partners) were required to answer these questions, and as a number of respondents did not answer the first screening question, "Do you have a spouse/partner or boyfriend/girlfriend?", subsequent questions had to be coded as missing. Of those who did report having partners, missing values ranged from 1.4% to 5.7%. Only two items in the measure had missing values greater than 10%. These were, once again, items

126 and 128 from the Relationships domain. Given their limited variability of endorsement on response options and high levels of missing values, these two items were removed from the measure prior to conducting the factor analysis.

1) Exploratory factor analysis

Of the 139 participants who completed the measure, 111 observations had no missing values for any items and were included in the analysis. Factor analysis of the six main domains (112 items) revealed 18 factors with Eigenvalues >1 (Appendix 5.21). When the factors were orthogonally rotated, three factors accounted for greater than 5% of the variance (Factor 1 − 26%, Factor 2 − 16%, Factor 3 − 8%).

Figure 5.1: Scree plot of Eigenvalues for the 112 factors from the initial analysis



The Cattell scree plot also suggested that there were three main factors which accounted for the majority of factor loadings (Figure 5.1). Based on these findings, a number of forced factor analyses (five, four, three and two factors) and orthogonal rotations were performed. The forced factor analyses and rotations confirmed that the three-factor structure was the simplest and clearest (Appendix 5.22).

In the three-factor rotation, each factor had more than three items with loadings of >0.40 on only one factor. Seventeen items had <20% of participants reporting a high or very high need and a factor loading of >0.40 on two or more factors, or no loadings of >0.40 on any factor, and were removed from the measure. Details of removed items are presented in Appendix 5.23. The remaining 95 items fell into the following three factors and accounted for 58% of the total variance: Factor 1 – "Treatment Environment and Care"; Factor 2 – "Feelings and Relationships"; and Factor 3 – "Daily Life". The three main factors and their corresponding items can be seen in Table 5.3.

Of the six main theoretical domains originally identified (Cancer Treatment Staff, Cancer Treatment Centre, Information, Feelings, Relationships and Daily Life), items primarily from the Cancer Treatment Staff, Cancer Treatment Centre and Information domains formed the first factor. Similarly, items from the Feelings and Relationships domains formed the second factor, and items from the Daily Life domain formed the third factor.

Table 5.3: Factor structure of the CNQ–YP from the initial factor analysis (n=111)

Item number		Description of item	Factor loading		
			Factor 1	Factor 2	Factor 3
Factor 1 – Treatment Environment and Care					
Cancer treatment staff telling me:	1	about my diagnosis	0.78		
	2	what might happen during treatment	0.78		
	3	about different treatment options	0.71		
	4	whether I had the option to decline treatment	0.59		
	5	about the short-term side-effects of treatment	0.75		
	6	about the long-term side-effects of treatment	0.68		
	7	my chances of a full recovery	0.79		
	8	what would happen when treatment finished	0.71		
	9	whether I would be able to have children	0.65		
	10	how treatment might affect my study/future career	0.66		
	11	what support services were available	0.60		
	12	whether my treatment was working	0.83		
	13	my test results as soon as possible	0.82		
	14	the way I felt was normal	0.76		
	15	how to manage my medication	0.81		
	16	what I could do to stay healthy	0.76		
	17	what to do if I noticed a particular side-effect	0.81		
Missing the:	19	support of the cancer treatment staff	0.61		
Having cancer treatment staff who:	20	listened to my concerns	0.87		
	21	treated me as an individual	0.84		
	22	were respectful	0.90		
	23	were approachable	0.89		
	24	were friendly	0.88		
	25	could have a laugh with me	0.86		
	26	explained what they were doing	0.88		
	27	spoke to me in a way that I could understand	0.87		
	28	let me talk about my feelings	0.79		
	29	let me ask questions	0.88		
	30	let me make decisions about my treatment	0.76		
	31	talked to me in private, without my family	0.72		
	32	talked to me and my family together	0.88		
Being able to:	33	get treatment in my local area	0.48		
	34	get transport to/from the cancer treatment centre	0.43		
	35	get overnight accommodation near the cancer treatment centre	0.47		
	36	see people I care about	0.80		
	37	spend time with people my own age	0.47		
	39	have time to myself	0.61		
	40	express my feelings	0.75		
Being able to have:	41	privacy	0.61		
	42	pleasant surroundings	0.59		
	43	good food	0.51		
	44	leisure spaces and activities	0.44		
	45	my family with me	0.89		
	46	a choice of cancer specialists	0.71		
	47	the same cancer treatment staff throughout treatment	0.68		
	48	a choice of times for appointments	0.68		
	49	enough time to make decisions about my treatment	0.71		
	50	access to professional counselling	0.64		
	51	opportunities to take part in research	0.62		
	52	someone to help me fill out forms	0.61		

	Item number	Description of item	Factor loading		
			Factor 1	Factor 2	Factor 3
Finding information that:	75	was specifically designed for me	0.54		
	76	was easy to understand	0.73		
	77	was easy to get hold of	0.70		
	78	I could trust	0.73		
	79	came in different forms (brochure, CD, DVD, online)	0.51		
	81	described relaxation techniques	0.46		
Factor 2 – Feelings and Relationships					
Feeling:	82	scared		0.73	
	84	frustrated		0.66	
	85	helpless		0.73	
	86	anxious or nervous		0.76	
	87	distressed		0.72	
	88	embarrassed		0.58	
Worrying about:	91	my cancer spreading		0.68	
	92	my cancer returning		0.60	
	93	whether my cancer treatment has worked		0.63	
	94	going to the cancer treatment centre		0.67	
	95	having cancer treatment		0.72	
	96	test results		0.73	
	97	how my family is coping		0.72	
Finding:	98	inner strength		0.75	
	99	hope		0.74	
	100	meaning in my experience		0.72	
Being able to:	108	focus on tasks		0.55	
	109	remember things		0.53	
	110	make plans or think about the future		0.63	
	112	accept my diagnosis		0.62	
	113	be independent		0.61	
Coping with:	114	changes in my relationships with my parent/s		0.41	
Knowing how to:	118	ask my parent/s for support		0.43	
	119	give support to my parent/s		0.60	
	123	give support to my friends		0.55	
Factor 3 – Daily Living					
Being able to:	38	talk to people my age who had been through a similar experience			0.41
Coping with:	102	changes in my physical ability			0.61
	103	changes in my appearance			0.68
Coping with:	107	not being able to do the same things as other people my age			0.71
	117	my parent/s being over-protective			0.49
Managing:	134	pain			0.53
	135	medication			0.43
	136	physical side-effects of treatment			0.73
	137	feeling tired			0.71
	138	loss of mobility			0.58
	140	to do chores/housework			0.58
	141	to eat			0.49
	142	to take part in social activities			0.72
	143	to travel to social events			0.71
% of Total Variance			**29%**	**17%**	**12%**

When items from both the Education and Work domains were independently added to the factor analysis (Appendix 5.24) two additional independent factors were identified, taking the total number of factors to five (Factor 4 – "Education"; and "Factor 5 – Work"). For the Relationship sub-domains (Partner and Siblings), five items had significant loadings >0.40 on a single factor and were added to Factor 2 (Appendix 5.25). Twelve items from the additional domains and sub-domains had <20% of participants reporting a high or very high need and a factor loading of >0.40 on two or more factors, or no loadings >0.40 on any factor, and were excluded from the measure (Appendix 5.26). The remaining items and their factor loadings are shown in Table 5.4.

Table 5.4: Factor structure of the items retained following factor analysis with the additional domains and sub-domains

Item number		Description of item	Factor loading		
			Factor 2	Factor 4	Factor 5
Factor 2 – Feelings and Relationships (Partner n=54)					
Coping with**:**	125	changes in my relationship with my partner	0.53		
Knowing how to:	130	give support to my partner	0.55		
Factor 2 – Feelings and Relationships (Siblings n=96)					
Coping with:	131	changes in my relationships with my sibling/s	0.59		
Knowing how to:	132	ask my sibling/s for support	0.51		
	133	give support to my sibling/s	0.52		
Factor 4 – Education (n=65)					
Being able to:	56	attend classes		0.69	
	58	get extensions or special consideration		0.74	
	59	get guidance about study options or future career paths		0.56	
Knowing:	61	how to ask teachers/students for support		0.79	
	63	that teachers/students had support to help them cope		0.64	
Factor 5 – Work (n=90)					
Knowing:	71	how much work I would miss			0.67
	72	how to ask managers/co-workers for support			0.78
	74	that managers/co-workers had support to help them cope			0.76

An item-item Spearman correlation matrix for each of the five factors revealed that two items from Factor 2 had correlations >0.90. These items both related to characteristics

of cancer treatment staff (item 22 – “Having cancer treatment staff who were respectful”; item 23 – “Having cancer treatment staff who were approachable”). However, members of the research team agreed that these items appeared to capture different aspects of the patient/staff relationship; therefore, neither item was removed from the measure.

2) Internal consistency

Item-total correlations for items within all factors were >0.20 and ranged from 0.33 to 0.88, with all items eligible to remain in the measure. All factors had Cronbach’s alphas of >0.70, indicating good internal consistency. The alpha values for all five factors can be seen in Table 5.5.

Table 5.5: Cronbach’s alpha for each factor of the CNQ–YP

Description of factor	Number of items	Cronbach’s alpha
Factor 1 – Treatment Environment and Care	56	0.99
Factor 2 – Feelings and Relationships	30	0.96
Factor 3 – Daily Life	14	0.93
Factor 4 – Education	5	0.88
Factor 5 – Work	3	0.89
Total Scale	**108**	**0.98**

3) Test-retest reliability

Weighted kappa values between responses at time 1 and time 2 ranged from 0.09 to 0.94. Twenty-four items did not have a weighted kappa of >0.60 and were excluded from the measure (Appendix 5.27). An additional two items had a weighted kappa of <0.60. However, >20% of participants indicated having a high or very high need, and therefore these items were retained. In Factor 4, “Education”, four items did not have

either a kappa of >0.60 or >20% of participants indicating a high or very high need. However, removing all four items would mean only one item remained in the factor. Therefore, the two items with both the largest proportions of participants reporting a high or very high need, and the largest kappa values (item 58 – "Being able to get extensions or special consideration", 12%, kappa=0.58; and item 59 – "Being able to get guidance about study options or future career paths", 17%, kappa=0.49) were retained. Items included in the final version of the measure and their kappa values are presented in Table 5.6.

Table 5.6: Weighted kappa values for items retained in the measure

	Item number	Description of item	Weighted Kappa
Factor 1 – Treatment Environment and Care			
Cancer treatment staff telling me:	1	about my diagnosis	0.73
	2	what might happen during treatment	0.79
	4	whether I had the option to decline treatment	0.68
	5	about the short-term side-effects of treatment	0.70
	6	about the long-term side-effects of treatment	0.67
	7	my chances of a full recovery	0.52
	8	what would happen when treatment finished	0.67
	9	whether I would be able to have children	0.83
	11	what support services were available	0.64
	12	whether my treatment was working	0.82
	13	my test results as soon as possible	0.66
	14	the way I felt was normal	0.76
	15	how to manage my medication	0.80
	16	what I could do to stay healthy	0.67
	17	what to do if I noticed a particular side-effect	0.74
Having cancer treatment staff who:	20	listened to my concerns	0.87
	21	treated me as an individual	0.75
	22	were respectful	0.75
	23	were approachable	0.82
	24	were friendly	0.68
	25	could have a laugh with me	0.61
	26	explained what they were doing	0.87
	27	spoke to me in a way that I could understand	0.85
	28	let me talk about my feelings	0.82
	29	let me ask questions	0.94
	30	let me make decisions about my treatment	0.83
	31	talked to me in private, without my family	0.66
Being able to:	33	get treatment in my local area	0.71
	35	get overnight accommodation near the cancer treatment centre	0.69
	37	spend time with people my own age	0.83
	39	have time to myself	0.70
	40	express my feelings	0.80
Being able to have:	41	privacy	0.65
	42	pleasant surroundings	0.70
	43	good food	0.88

	Item number	Description of item	Weighted Kappa
Being able to have:	44	leisure spaces and activities	0.59
	46	a choice of cancer specialists	0.61
	47	the same cancer treatment staff throughout treatment	0.68
	48	a choice of times for appointments	0.69
	50	access to professional counselling	0.62
	51	opportunities to take part in research	0.78
Finding information that:	75	was specifically designed for me	0.65
	76	was easy to understand	0.74
	77	was easy to get hold of	0.77
	78	I could trust	0.83
	81	described relaxation techniques	0.64
Factor 2 – Feelings and Relationships			
Feeling:	82	scared	0.82
	84	frustrated	0.69
	86	anxious or nervous	0.84
	87	distressed	0.61
	88	embarrassed	0.65
Worrying about:	91	my cancer spreading	0.65
	92	my cancer returning	0.90
Worrying about:	93	whether my cancer treatment has worked	0.72
	95	having cancer treatment	0.75
	97	how my family is coping	0.70
Finding:	98	inner strength	0.60
Being able to:	108	focus on tasks	0.72
	109	remember things	0.69
	110	make plans or think about the future	0.70
	112	accept my diagnosis	0.70
	113	be independent	0.69
Coping with**:**	125	changes in my relationship with my partner	0.81
	131	changes in my relationships with my sibling/s	0.70
Knowing how to:	132	ask my sibling/s for support	0.89
	133	give support to my sibling/s	0.90
Factor 3 – Daily Life			
Being able to:	38	talk to people my age who had been through a similar experience	0.71
Coping with:	102	changes in my physical ability	0.87
	103	changes in my appearance	0.85
	107	not being able to do the same things as other people my age	0.75
	117	my parent/s being over-protective	0.67
Managing:	134	pain	0.66
	135	medication	0.76
	136	physical side-effects of treatment	0.78
	137	feeling tired	0.87
	138	loss of mobility	0.83
	142	to take part in social activities	0.82
	143	to travel to social events	0.85
Factor 4 – Education			
Being able to:	56	attend classes	0.69
	58	get extensions or special consideration	0.58
	59	get guidance about study options or future career paths	0.49
Factor 5 – Work			
Knowing:	71	how much work I would miss	0.72
	72	how to ask managers/co-workers for support	0.81
	74	that managers/co-workers had support to help them cope	0.67

Revised factor analysis and Cronbach's alpha

Factor analysis on the 74 items from the three main factors was re-run to confirm the factor structure. One hundred and sixteen observations had no missing values for any items and were included in the analysis. The scree plot indicated that the number of important factors had increased from three to four (Figure 5.2). These four factors explained 63% of the variance (Factor 1 − 31%, Factor 2 − 13%, Factor 3 − 11%, Factor 4 − 8%). Fourteen items had <20% of participants reporting a high or very high need and a factor loading of >0.40 on two or more factors, or no loadings >0.40 on any factor, and were removed from the measure. Details of removed items are presented in Appendix 5.28. All but six items loaded on the same factor, as in the previous factor analysis. Five of the six items loaded on the new factor ("Information and Activities"), while one item (item 110 − "Being able to make plans or think about the future") moved from the "Feelings and Relationships" factor to the "Daily Life" factor (Table 5.7).

Figure 5.2: Scree plot of Eigenvalues for the 74 factors from the revised analysis

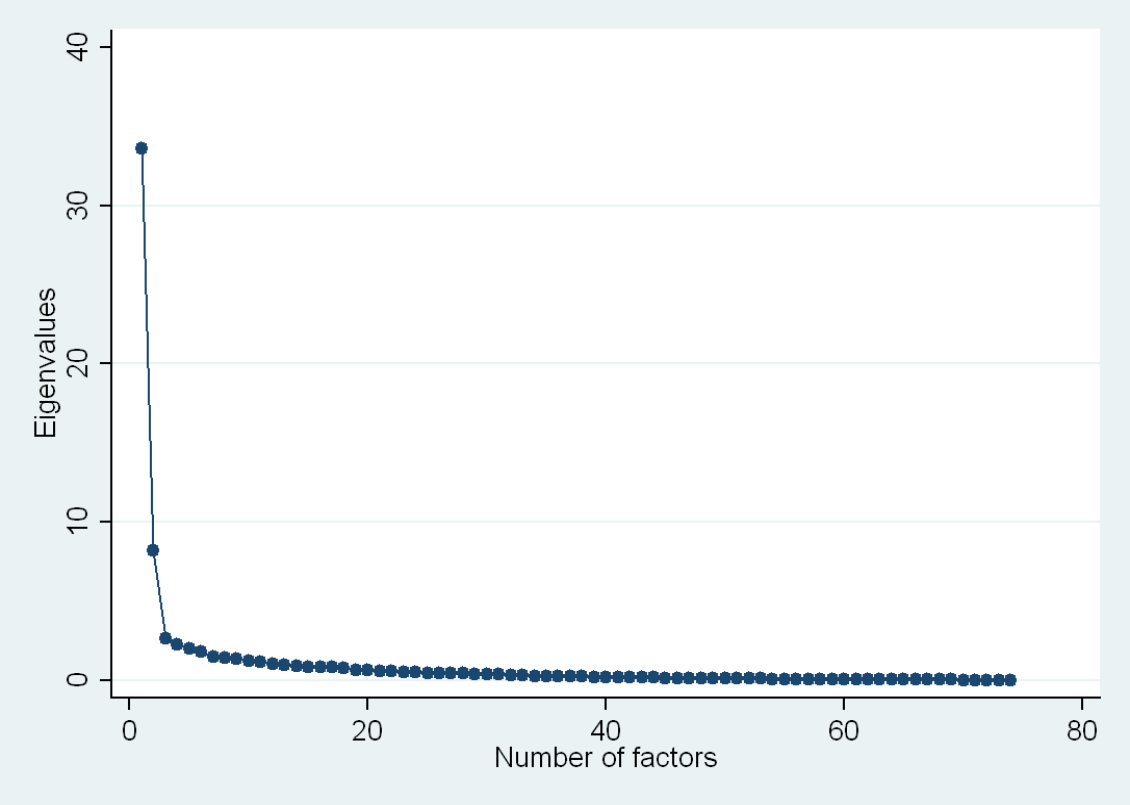


Table 5.7: Factor structure of the CNQ–YP from the revised factor analysis (n=116)

Item number		Description of item	Factor loading			
			Factor 1	Factor 2	Factor 3	Factor 4
Factor 1 – Treatment Environment and Care						
Cancer treatment staff telling me**:**	1	about my diagnosis	0.76			
	2	what might happen during treatment	0.77			
	4	whether I had the option to decline treatment	0.54			
	5	about the short-term side-effects of treatment	0.73			
	6	about the long-term side-effects of treatment	0.65			
	7	my chances of a full recovery	0.78			
	8	what would happen when treatment finished	0.71			
	9	whether I would be able to have children	0.60			
	12	whether my treatment was working	0.87			
	13	my test results as soon as possible	0.87			
	14	the way I felt was normal	0.80			
	15	how to manage my medication	0.82			
	16	what I could do to stay healthy	0.67			
	17	what to do if I noticed a particular side-effect	0.74			
Having cancer treatment staff who:	20	listened to my concerns	0.89			
	21	treated me as an individual	0.87			
	22	were respectful	0.93			
	23	were approachable	0.91			
	24	were friendly	0.91			
	25	could have a laugh with me	0.90			
	26	explained what they were doing	0.89			
	27	spoke to me in a way that I could understand	0.90			
	28	let me talk about my feelings	0.77			
	29	let me ask questions	0.90			
	30	let me make decisions about my treatment	0.74			
	31	talked to me in private, without my family	0.66			
Being able to have:	39	time to myself	0.55			
	41	privacy	0.51			
	42	pleasant surroundings	0.52			
	43	good food	0.45			
	46	a choice of cancer specialists	0.64			
	47	the same cancer staff throughout treatment	0.63			
	48	a choice of times for appointments	0.64			
Factor 2 – Daily Life						
Being able to:	110	make plans or think about the future		0.51		
Coping with:	102	changes in my physical ability		0.68		
	103	changes in my appearance		0.69		
	107	not being able to do the same things as other people my age		0.75		
	117	my parent/s being over-protective		0.49		
Managing:	134	pain		0.65		
	135	medication		0.48		
	136	physical side-effects of treatment		0.76		
	137	feeling tired		0.72		
	138	loss of mobility		0.65		
	142	to take part in social activities		0.71		
	143	to travel to social events		0.67		
Factor 3 – Feelings and Relationships						
Feeling:	84	frustrated			0.56	

	Item number	Description of item	Factor loading			
			Factor 1	Factor 2	Factor 3	Factor 4
Feeling:	86	anxious or nervous			0.73	
Worrying about:	91	my cancer spreading			0.76	
	92	my cancer returning			0.66	
	93	whether my cancer treatment has worked			0.68	
	95	having cancer treatment			0.70	
	97	how my family is coping			0.68	
Finding:	98	inner strength			0.66	
Being able to:	112	accept my diagnosis			0.60	
	113	be independent			0.51	
Factor 4 – Information and Activities						
Being able to:	37	spend time with people my own age				0.44
	38	talk to people my age who had been through a similar experience				0.69
Being able to have:	44	leisure spaces and activities				0.45
Finding information that:	75	was specifically designed for me				0.61
	81	described relaxation techniques				0.54
% of Total Variance			**31%**	**13%**	**11%**	**8%**

Following the removal of these items the internal consistency of the measure was checked to ensure that the item-total correlations and Cronbach's alpha had not significantly changed. Item-total correlations for the 70 items from all six factors were still >0.20 and ranged from 0.32 to 0.90. All factors maintained alphas >0.70 (Table 5.8).

Table 5.8: Revised Cronbach's alpha for each Factor of the CNQ–YP

Description of factor	Number of items	Cronbach's alpha
Factor 1 − Treatment Environment/Care	33	0.98
Factor 2 − Daily Life	12	0.94
Factor 3 − Feelings/Relationships	14	0.92
Factor 4 − Information /Activities	5	0.83
Factor 5 − Education	3	0.82
Factor 6 − Work	3	0.89
Total Scale	**70**	**0.98**

Summary of item reduction for the measure

The psychometric inclusion criteria for the measure and the number of items removed at each stage of analysis are summarised in Table 5.9. The final measure had six factors and 75 items, including five screening items: 1) Treatment Environment and Care (33 items); 2) Daily Life (12 items); 3) Feelings and Relationships (15 items including 1 screening item); 4) Information and Activities (5 items); 5) Education (5 items including 2 screening items); and 6) Work (5 items including 2 screening items).

Table 5.9: Summary of item reduction for the measure

Item inclusion criteria	Number of items removed from measure	Total number of items remaining
Original number of items (less 5 screening questions)		139
Proportions and missing values <90% of participants indicate the same level of need and <10% are missing values	2	137
Exploratory Factor Analysis Factor loading of >0.40 on only one factor or >20% of participants indicate high/very high need	29	108
Spearman correlation Item-item correlation <0.90	0	108
Test-retest reliability Weighted kappa >0.60 or >20% of participants indicate high/very high need	24	84
Revised Exploratory Factor Analysis Factor loading of >0.40 on only one factor or >20% of participants indicate high/very high need	14	70
Inclusion of 5 screening questions		75

4) Discriminative validity

Of the 139 AYAs who completed the measure, six young people were unsure of their treatment status and were excluded from the known-groups comparison. Median factor scores and quartiles for all six factors and each known group are presented in Table 5.10. Participants receiving treatment had higher median factor scores than those who

had finished treatment for all factors except Factors 5 and 6 (“Education and Work”). However, these differences were not statistically significant.

Table 5.10: Comparison of factor scores between AYAs receiving treatment and AYAs who had finished treatment

Factor	Receiving Treatment				Finished Treatment				Wilcoxon rank sum	
	n	median	Q1	Q3	n	median	Q1	Q3	*z*	*p*
1 − Treatment Environment/Care	17	1.8	1.1	2.1	116	1.5	1.2	2.2	0.32	0.75
2 − Daily Life	17	2.3	1.1	3.2	115	1.4	1.0	2.2	1.46	0.14
3 − Feelings/Relationships	17	2.4	1.6	2.6	115	1.5	1.2	2.3	1.81	0.07
4 − Information /Activities	17	3.6	1.6	3.8	116	2.2	1.5	2.8	1.58	0.11
5 − Education	11	1.3	1.0	2.0	67	1.7	1.0	2.7	-0.92	0.36
6 − Work	13	1.3	1.0	3.0	94	1.3	1.0	2.0	0.53	0.60

5) Responsiveness

The proportion of participants who scored the minimum and maximum scores for each factor can be seen in Table 5.11. The proportion of participants ranged from 0% to 5.1% for the maximum score to 8.3% to 43% for the minimum score, with large proportions of participants having floor effects in the “Education” and “Work” factors (42% and 43% respectively).

6) Measure acceptability

When asked about the acceptability of the measure, 80% of participants agreed or strongly agreed that the instructions were easy to follow (n=111, 95% CI 72-86%) and 73% agreed or strongly agreed that the questions were clear (n=102, 95% CI 65-80%). Seventy-eight percent (n=108, 95% CI 71-84%) of AYAs disagreed or strongly disagreed that the questions were distressing. Just over half of the participants (56%,

n=78, 95% CI 48-64%) agreed or strongly agreed that the answer choices were easy to understand, while 12% (n=16, 95% CI 7.1-18%) were unsure, and 32% (n=45, 95% CI 25-41%) disagreed or strongly disagreed.

Table 5.11: Floor and ceiling effects per factor

Factor		Lowest possible score	Highest possible score
	n	n (%)	n (%)
Factor 1 − Treatment Environment/Care	133	11 (8.3)	1 (0.8)
Factor 2 − Daily Life	132	36 (27)	0 (0.0)
Factor 3 − Feelings/Relationships	132	19 (14)	0 (0.0)
Factor 4 − Information /Activities	133	16 (12)	6 (4.5)
Factor 5 − Education	78	33 (42)	4 (5.1)
Factor 6 − Work	107	46 (43)	4 (3.7)

Discussion

This research has attempted to establish the internal consistency, test-retest reliability, discriminative validity and potential responsiveness of the CNQ–YP using the most rigorous psychometric criteria possible. Item reduction following factor analysis and the assessment of test-retest reliability resulted in a final measure with six factors and 75 items. The four main factors in the measure accounted for 63% of the variance, and all factors achieved alpha values greater than 0.80. Test-retest reliability was also high, with the majority of items reaching weighted kappa values above 0.60. There are a number of limitations related to the study sample and methodology which should be considered when interpreting these results.

Limitations

A primary limitation of the research was the size of the sample achieved. Only 139 young people were able to be recruited through the seven treatment centres involved. This has implications for the statistical analysis performed, particularly the exploratory factor analysis. When performing factor analysis it is recommended that the number of participants in the sample be at least five times the number of items in the measure.[1] As there were 144 items in the original measure, only a 1:1 item-to-participant ratio was actually achieved. This small sample size means that some items may not have been highly correlated with other items on the measure.[8] However, as the inclusion criteria allowed items which had a large proportion of participants reporting a high or very high level of need (>20%) to be retained, it is unlikely that items considered important by a large proportion of AYAs were excluded.

Furthermore, the method of recruiting the sample was not population-based, and therefore the sample achieved may not have been representative of all groups of AYA cancer patients and survivors. However, recruitment of young people through a state-based cancer registry during the pilot phase of the study showed that, due to low clinician consent rates and the high mobility of AYAs, this methodology was not feasible. The sample recruited for the present study was national (from five states) and included both large- and small-volume treatment centres for AYA cancer patients. Therefore, it is likely that a wide range of young people were involved. Recruitment of patients through treatment centres is a commonly accepted method and has been used in previous studies which describe the development of quality of life measures for AYA cancer survivors.[45-51] Those who consented to take part in the study were slightly younger and more likely to be female than those who did not consent. However, participation by young people with a range of cancer types and at different stages since

their cancer diagnoses increased the probability that the items identified as important in the measure represent the views of the larger AYA cancer population.

Finally, the test-retest sample was self-selected, not randomised. However, this could not be avoided due to the low number of participants who completed the baseline measure and agreed to be contacted again (n=116, 83%). The median time to return the retest measure was also greater than the recommended 14 days. Consequently, responses to the retest measure may have reflected a change in participants' needs.[16] Despite the longer than recommended retest period, the majority of items had acceptable kappa values (>0.60), and as the time-frame for the response scale was either "any time since your cancer diagnosis" or "in the last month", it is unlikely that the longer period of retest would have greatly affected the overall responses obtained. The inclusion criteria also allowed items which had a low kappa value but a large proportion of participants (>20%) reporting a high or very high level of need to remain in the measure, further ensuring that important items were not excluded.

Possible reasons for the small sample size and low response rate

There are a number of possible reasons for the smaller than expected sample recruited for this study. It is possible that mailing consent forms and questionnaires to participants to complete at home, rather than having patients complete them at the hospital or treatment centre, may have contributed. However, patient data collection at hospitals, while accessing the young person directly, introduces difficulties of working around treatment appointments and can add to clinician workloads.[52] Also, as many young people may have completed the active or intense phases of their treatments, visits to hospitals or treatment centres may be infrequent. Mailing questionnaires to young people at home allows more efficient data collection which is not dependent on hospital variables.[52] Given the large proportion of AYAs in the study who had

completed treatment (n=116), compared with those currently receiving treatment (n=17), recruitment and data collection *via* post seems appropriate.

The overall response rate for completing and returning the measure in the present study was 50%, the same as the response rate achieved in the pilot study. Other studies describing the development of measures for AYA cancer patients have reported response rates of about 90%.[46 48 50] However, the age range of these samples (8-20 years) was lower than in the current study (16-30 years). A study describing the development of a measure for a similar age group (16-28 years) only achieved a response rate of 53%.[51] Reasons for lower response rates with older AYA samples, compared with younger samples, can only be speculated. It is possible, however, that as described in Chapter 4, AYAs in this age group are highly mobile.[53 54] Therefore, a large proportion of AYAs may not have received the questionnaire because of incorrect contact details.

It is also possible that some young people were not interested in participating in this type of research. Reasons given for not wishing to participate in the study focussed mostly on AYAs not wishing to be reminded of or talk about their cancer. This suggests that at least some young people do not like to focus or dwell on their emotions or experiences related to cancer, but rather want to put the experience behind them. This may be a mechanism for coping with the cancer experience, or it may be that this group of non-participants had few or no needs. Low participation may be especially applicable to psychosocial research studies where personal issues such as feelings and emotions related to cancer are discussed. Similar results have been found with adult breast cancer survivors,[55] indicating that participants in psychosocial cancer research may be self-selected and are only representative of a sub-population of survivors who wish to talk about their experiences.

Psychometric strengths of the CNQ–YP

Despite the difficulties with recruitment, the current study had a number of strengths related to the psychometric development of the CNQ–YP, and the measure compared favourably with the psychometric criteria outlined in Chapter 2 (Appendix 5.29). First, reliability and validity of the CNQ–YP was examined using the most applicable psychometric methods, with analysis adhering to the highest recommended statistical levels. The >0.40 cut-off stipulated in the factor analysis ensured that each factor comprised large and unique factor loadings. The final factor structure of the CNQ–YP showed that the four main factors accounted for 63% of the variance. This was considered good, as the average variance accounted for by exploratory factor analysis is about 60%.[8] This outcome also compares well with other quality of life measures developed for AYA cancer survivors, such as the Adolescent Quality of Life Instrument (AQoL) which reported having six factors representing 67% of the variance,[50] and the Quality of Life – Cancer Survivors (QOL–CS) instrument which also has six factors accounting for 56% of the variance.[51]

In addition, the CNQ–YP achieved high Cronbach's alphas, with all six factors reporting alphas greater than 0.80. It is possible that some alpha values may have been artificially high due to the large number of items in the factors.[7 13] However, no items had correlations <0.20, and alphas >0.80 were also reached in Factors 5 and 6 ("Work" and "Education"), both of which had only three items. These findings compare favourably with the seven quality of life measures developed for AYA cancer survivors identified in Chapter 2. Although all of these scales had at least some domains with alphas >0.70, no scales achieved alphas greater than 0.70 for all domains. In the case of the Perceived Illness Experience Scale (PIE), only two out of nine domains had alphas >0.70, showing variability in the internal consistency of these measures.

A further strength of this study is that it assessed test-retest reliability, unlike many other studies reporting the development of measures for cancer patients and survivors.[56 57] As the response scale of the CNQ–YP was scored from one to five, it could have been interpreted as being either continuous or categorical. Therefore, either the intra-class correlation coefficient (ICC) or kappa could have been used to compare retest agreement. However, as the responses themselves represented "levels of need" which may not be linear, and a score of one represented the "absence of need", item responses were considered to be categorical, and therefore kappa was the more appropriate statistic to use. Test-retest reliability was also performed after, rather than before, factor analysis in the current study. However, given the small number of individuals in the test-retest sample, it was decided that reducing the number of items through factor analysis prior to calculating test-retest reliability would help reduce the probability of type one error. All but four items in the measure had weighted kappa values >0.60, and these four items all had weighted kappas >0.49. In comparison, only one instrument measuring quality of life in AYA cancer survivors, the Minneapolis–Manchester Quality of Life Instrument − Adolescent Form (MMQL Adolescent form), reported agreement values for test-retest reliability. However, ICCs were only reported at the domain level. This can be misleading, as although total agreement levels for the domain may be high, agreement for individual items may vary.[58]

Recommended improvements for the CNQ–YP

The known-groups comparison in the current study was unable to distinguish between the median factor scores of young people currently receiving treatment and young people who had completed treatment. For Factors 1, 4, 5 and 6 ("Treatment Environment and Care", "Information /Activities", "Education" and "Work") this may have been because the response time-frame was "any time since diagnosis". Therefore, patients who had completed treatment may have been reflecting a need

level they had while receiving treatment. The small sample size in the receiving treatment group (n=17) may have also limited the power of the hypotheses testing. For Factor 2 (“Feelings and Relationships”), the significance level was close to 5% (p=0.07), suggesting AYAs receiving treatment may have higher needs in this factor than patients who have completed treatment, and this should potentially be explored with a larger sample.

The CNQ–YP did not appear to have a ceiling effect (<5.1% of participants scored the highest possible score in each domain). However, there was a large floor effect for all domains. This may have implications for intervention studies where researchers wish to measure a reduction in needs, as a large proportion of participants (between 8.3% and 43%) are already scoring the minimum possible scores for each factor. However, factors with the largest floor effects (“Education” 42% and “Work” 43%) were only completed by a sub-group of participants. Therefore, a larger sample of AYAs may produce different results. These floor effects may also indicate that the majority of young people do not experience high levels of need in these areas. However, the “Education” factor also had the highest ceiling effect (5.1%), suggesting that this is probably not the case.

Compared to the pilot study, the proportion of AYAs who agreed or strongly agreed that the measure was acceptable increased for the item “I found the questions clear” (from 66% to 73%). The proportion of young people who disagreed or strongly disagreed with the item “I found the questions distressing” remained relatively stable (from 80% to 78%). However, despite a revision of the five-point response scale only 56% of participants in the study agreed or strongly agreed with the statement “I found the answer choices easy to understand”, suggesting further modification of the response scale is required.

Testing of convergent and divergent validity, responsiveness and predictive validity was beyond the scope of the present study. It is recommended that future psychometric testing of the measure be undertaken to explore these issues further. The small sample size means that the factor structure achieved in the current study may not be reproducible. Therefore, it is recommended that confirmatory factor analysis with a larger sample of AYA cancer survivors be conducted prior to using the measure in clinical practice.[5 8]

Conclusion

The CNQ–YP is the first multi-dimensional measure of unmet need which has been developed specifically for AYA cancer patients and survivors. The measure displays a strong factor structure, and good internal consistency and test-retest reliability. Future studies with a larger sample size are recommended to determine the discriminative validity and floor and ceiling effects of the measure. Longitudinal studies to establish responsiveness and predictive validity should also be undertaken.

CHAPTER 6

Prevalence of and factors associated with high unmet needs reported by adolescent and young adult (AYA) cancer survivors

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Chapter 6

Prevalence of and factors associated with high unmet needs reported by adolescent and young adult (AYA) cancer survivors

Introduction

Chapter 5 described the psychometric evaluation of a newly developed measure, the Cancer Needs Questionnaire – Young People (CNQ–YP), to assess the unmet needs of adolescent and young adult (AYA) cancer patients and survivors. The measure satisfied recognised criteria for internal consistency, test-retest reliability, construct validity and acceptability. The following chapter examines the prevalence of unmet needs in young people with cancer, and factors associated with high levels of unmet need measured by the CNQ–YP.

Assessing the prevalence of unmet needs at the population level

Prevalence refers to the proportion of individuals in the population who are currently experiencing a disease or illness.[1 2] Identifying prevalence allows an estimation of the burden of illness caused by the disease and its treatment and assists in the planning of health services.[2] Assessing the prevalence of unmet needs among cancer survivors at the population level can: provide an overview of which issues or needs are most common; and aid in prioritising issues for which individuals would most like help.[3] Sub-groups of patients within the population who are at risk of experiencing high levels of need can also be identified.[4] In addition to the collection of cross-sectional data, examining unmet needs at the population level creates an opportunity to discover how the needs of patients and survivors change over time. By examining levels of unmet need at different points along the disease trajectory (e.g. newly diagnosed, in treatment, off treatment and long-term survival), important information regarding the natural history of needs can be obtained.[5-7] Such information is useful for determining

where service improvements are needed, which groups should be targeted and when is the most appropriate time to intervene.[4 8]

Assessing the prevalence of unmet needs at the individual level

At an individual level, needs assessment measures can be implemented as part of the routine care provided to cancer patients and potentially form part of a two-step approach.[9] First, the unmet needs measure can be used for initial screening to identify any issues that are important to the patient. Next, a health care provider can follow-up with the patient using the measure to see whether or not there are extenuating circumstances which may explain, or not explain, the unmet needs reported by the individual.[10] A screening and follow-up process such as this can help to initiate and facilitate communication regarding needs between patients and providers,[11 12] identify patient preferences[13] and inform and guide the planning of treatment and care.[13] It also attempts to ensure that patients are not inappropriately labelled as having an issue or problem based on responses on the measure alone. Importantly, routine screening can provide a mechanism for recognising patients who have a high index of suspicion of experiencing greater levels of unmet need.[8] In this way, patients who are at most risk may be identified for early intervention in order to prevent poor psychosocial outcomes in the future.[9 14]

Examining predictors of unmet need in AYA cancer survivors

To accurately estimate the prevalence and predictors of unmet need in AYA cancer survivors the outcome measure used should meet accepted standards for psychometric rigour.[15-17] However, prior to the development of the CNQ–YP, no psychometrically robust measures of unmet need for this population existed (see Chapter 2).

There are a number of studies which have identified predictors of quality of life (QOL) among AYAs with cancer, and these may help to inform the types of patient characteristics that could be associated with high levels of unmet need. These predictors cover three main areas: 1) demographic characteristics and personality traits; 2) disease and treatment characteristics; and 3) social environment characteristics.

Demographic characteristics and personality traits

Both demographic characteristics (e.g. age, gender, education level, socio-economic status and non-cancer-related health status) and personality traits (e.g. coping style, self-esteem and outlook) have been associated with QOL outcomes. For example, Stam and colleagues assessed the health-related quality of life (HRQOL) of young adult survivors of childhood cancer in The Netherlands.[18] Three hundred and fifty-three AYAs aged 18 to 30 years who were at least five years post-treatment participated in the study.[18] Gender and age were found to be associated with lower mean scores of HRQOL in survivors, with female participants and those who were diagnosed with cancer at an older age reporting significantly worse physical and mental HRQOL than AYAs who were male or who were diagnosed at a younger age.[18] Cognitive coping styles and the current health status of the young person were also associated with differences in HRQOL.[18]

Langeveld and colleagues investigated the QOL of 400 long-term survivors of childhood cancer aged 16-49 years of age.[19] Survivors who were unemployed or had low self-esteem had poorer QOL and higher levels of concern.[19] Female gender and lower levels of education were associated with poorer physical functioning and increased levels of pain.[19] Wu and colleagues also found that female AYAs aged 13-20 years, both on and off therapy, were significantly more likely to report poor QOL

compared with males, particularly for body image and physical, psychological and cognitive functioning.[20] A study by Zebrack and Chesler of cancer survivors aged 16-28 years, at least three years post-diagnosis, revealed that being diagnosed with cancer at an older age or reporting a medical condition were associated with poorer QOL in AYAs.[21] However, female survivors reported significantly higher QOL than males for the spiritual domain.[21]

These studies highlight that a variety of demographic characteristics may be associated with poor QOL in AYA cancer survivors. Female AYAs appear to report lower functioning across almost all dimensions of QOL.[18-20] However, these findings may or may not be a direct result of having experienced cancer. For example, compared with males, female AYAs reported having worse physical functioning and higher levels of pain.[19] This may indicate that cancer and its subsequent treatment has a more detrimental effect on this group. However, it may also be a reflection of an inherent gender bias. Male AYAs may simply be less likely to report physical symptoms compared to female AYAs due to feeling greater pressure to conform to certain social norms, such as not wishing to be perceived as weak.[22 23]

Similarly, other associations between demographic characteristics and poor QOL may not be specific to AYAs with cancer, but rather to AYAs more generally. Associations between demographic characteristics, such as education and self-esteem, and perceived QOL have been reported among healthy populations of AYAs.[24] Young women are also commonly reported to have higher levels of anxiety[25] and depression[26] and lower self-esteem related to body image[27] than males, regardless of their disease status. However, a diagnosis of cancer may potentially add to these already existing discrepancies in psychological functioning. For example, hair loss as a result of chemotherapy could potentially be much more distressing for females than males. For

male cancer patients having a shaved head or no hair may be perceived as reasonably socially acceptable,[28] whereas females may find that the loss of hair is more incongruous with their self-perception and identity which in turn could contribute to lower self-esteem and poorer QOL.[29]

Disease and treatment characteristics

In addition to the demographic and personality-related predictors described above, the same studies established associations between the QOL of AYA cancer survivors and types of diseases and treatments. Wu and colleagues reported that AYAs who were diagnosed with leukaemia had significantly lower QOL than AYAs diagnosed with lymphoma or solid tumours, for individuals both on and off therapy.[20] While Zebrack and Chesler established that childhood cancer survivors diagnosed with cancer of the central nervous system (CNS) or brain tumours reported significantly lower QOL than AYAs with all other cancer types combined.[21]

The type and number of cancer treatments received were found to be important variables as well. In their study of Dutch AYAs, Stam and colleagues found that AYAs who had been treated with surgery alone had worse mental HRQOL than AYAs who were treated with radiotherapy, chemotherapy or combination therapy.[18] In contrast, Langeveld and colleagues determined that AYAs who received combination treatment (both radiotherapy and chemotherapy, with or without surgery) had poorer mental QOL than AYAs treated with radiotherapy or chemotherapy alone (with or without surgery).[19]

Treatment stage and time since diagnosis have also been associated with poor QOL in some AYA survivors. Ward-Smith and colleagues assessed the QOL of 75 patients aged 9-20 years with various types of cancer.[30] Those who were currently receiving treatment reported significantly lower QOL than AYAs who had finished treatment.[30]

Furthermore, Wu and colleagues found that being off therapy for more than seven years was associated with higher QOL in AYA cancer survivors, compared with AYAs who had been off treatment for seven years or less.[20]

Due to differences in the disease course, level of impairment and treatment regimes that accompany different cancer diagnoses,[31-35] it is not surprising that some types of cancer are associated with higher levels of QOL in AYAs compared to others. Furthermore, the stress and significant long-term consequences (e.g. risk of secondary cancers or infertility) of particular treatment modalities such as radiotherapy could explain why poorer QOL is highly associated with some treatment types more than others (e.g. surgery alone).[36]

However, findings related to the impact of disease and treatment characteristics on QOL in the literature appear to be somewhat inconsistent, making it difficult to determine which variables have the strongest association with QOL.[18 19] It is possible that these conflicting findings could be explained by the concept of "response shift".[37] According to this theory, experiencing poor cancer-related QOL in the past may result in a shift in the health expectations of an AYA. Consequently, AYAs who have experienced worse QOL may have lower health expectations and therefore rate their current QOL as better than AYAs who have experienced less cancer-related morbidity and therefore have comparatively higher health expectations.[38-40] It is possible that expectations may play a similar role in determining unmet needs, however, this has not yet been examined.

Social environment characteristics

There are some studies in the AYA cancer literature which have proposed links between the QOL of AYA cancer survivors and social environment characteristics,

such as living arrangements and relationships. Zebrack and Chesler found that survivors of childhood cancer who were living alone had significantly lower QOL scores for the psychological domain, compared with survivors who were living with others.[21] Sawyer and colleagues investigated HRQOL in 70 AYAs aged 10-18 years.[41] Parents reported that AYAs from single-parent families had significantly lower HRQOL than AYAs from families with two parents.[41] However, as these findings relied on the observations of parent proxies they should be interpreted with caution.[41]

As with other characteristics associated with poor QOL, the existing social environment and its impact on the QOL of AYAs may be further complicated by the diagnosis of cancer. For example, a relationship between single parent families and poorer QOL has been observed in the general population, primarily related to financial stress.[42] This stress could potentially increase for AYAs who are diagnosed with cancer, as parents may no longer be able to work in order to take care of the young person.[43]

Based on the findings regarding predictors of QOL in AYAs with cancer, it is likely that similar associations might be found between demographic, personality, disease, treatment and social environment variables, and high levels of unmet need. However, investigating personality traits requires the use of psychometrically robust instruments which can accurately capture these difficult to measure attributes.[15] Given the length and comprehensive nature of the unmet needs measure used in the current study, a decision was made not to further burden respondents by asking them to complete an additional measure of personality traits or coping styles. Instead, a small number of items regarding demographic, disease and treatment characteristics and aspects of the AYAs social environment were included as part of the survey instrument so that associations between these variables and reported levels of high unmet need could be explored in greater depth.

Aims

The aims of this study were to identify: 1) the ten most prevalent unmet needs endorsed by AYA cancer survivors at any level of need (Low to Very High); 2) the ten most prevalent unmet needs endorsed by AYA cancer survivors at the High to Very High level of need; 3) the domain with the highest median level of need; 4) associations between the demographic, disease and treatment and social environment characteristics of AYAs, and the item which had the highest proportion of AYAs endorsing a High to Very High need for each domain; and 5) preferences for the format of the CNQ–YP, mechanisms for feedback and the preferred location for completion.

Hypotheses

Based on previous studies of AYA cancer survivors and their reported QOL, it was expected that AYAs who were older, female, had lower levels of education, were diagnosed with a haematological cancer, received more than two types of treatment, were currently receiving treatment, were less than two years post-diagnosis, had experienced a cancer recurrence or who did not have a partner would have higher needs that those without these characteristics

Methods

The setting, sample and procedure used were the same as described in Chapter 5.

Measure

Cancer Needs Questionnaire – Young People (CNQ–YP)

Adolescents and young adults indicated their levels of unmet need using the Cancer Needs Questionnaire – Young People (CNQ–YP). As discussed in Chapter 5, factor analysis revealed the CNQ–YP has six domains and 70 items which capture aspects of

need related to Treatment Environment and Care, Education, Work, Feelings and Relationships, Daily Life and Information and Activities. Five response choices range from “No Need” to ”Very High Need” with scores on each item ranging from one to five.

Demographic characteristics

Adolescents and young adults provided demographic information including their age, gender, cancer type, time since diagnosis, types of treatment received, partner status, language preferences, living arrangements and level of education.

Measure format preferences

As described in Chapter 5, the initial design of the study had included randomisation of both paper and online versions of the measure. This was based on previous research which suggested that web-based surveys may be more user-friendly for the AYA population than paper surveys.[44 45] Low recruitment rates *via* the treatment centres prevented this randomisation. However, questions regarding AYA levels of computer use and views regarding paper surveys compared with online surveys were asked as part of the current study to determine whether any preference for web-based surveys existed among this group.

Feedback of results and preferred location for completion

Information was also collected about choices related to the feedback of results to health professionals, treatment centres, researchers and other organisations, as well as preferred locations for questionnaire completion.

All of the above questions regarding measure format, feedback and completion were answered using a five-point Likert scale with responses that ranged from “Strongly Disagree” to “Strongly Agree”.

Statistical analysis

Stata Version 11 software was used to perform statistical analysis.[46]

Demographic characteristics

The demographic characteristics of participants were reported using descriptive statistics (e.g. frequencies, percentages, means and standard deviations).

Aims 1-2: Identify the most prevalent unmet needs at any level of need (Low to Very High) and at the High to Very High level of need

Descriptive statistics including frequencies, percentages and 95% confidence intervals (CIs) were used to identify the ten most prevalent items endorsed by AYAs for any level of need (Low to Very High) and the ten most prevalent items endorsed by AYAs at the High to Very High level of need.

Aim 3: Identify the domain with the highest median level of need

Participant responses for each item were given a raw score of one to five ("No Need" to "Very High Need"). Factor scores were then calculated using the sum score by factor method (described in Chapter 5).[47] For each participant, the raw scores for all items within a domain were summed, and then divided by the number of non-missing items. Observations with missing values for >50% of items within the domain were excluded from the analysis. The factor scores for all participants by each domain were then analysed. As the distribution of factor scores within each domain were skewed, medians and quartiles were used to determine which domain had the highest level of need.

Aim 4: Identify associations between AYA characteristics and items with the highest proportion of High to Very High need per domain

The item with the highest proportion of AYAs endorsing a High to Very High level of need for each of the four main domains (Treatment Environment and Care, Daily Life, Feelings and Relationships, and Information and Activities) was determined. Chi-square tests were then performed to investigate associations between participant demographic, disease and treatment and social environment characteristics, and the highest need items from each domain. To ensure a sufficient expected frequency per cell in the Chi-square analysis ($n \geq 5$)[48], participant characteristics were dichotomised into the following variables: gender (male, female); partner status (partner, no partner); time since first diagnosis (<2 years, ≥2 years); cancer type (haematological, non-haematological); cancer recurrence (yes, no); treatment status (in treatment, finished treatment); education completed (primary/secondary, trade/tertiary); and number of treatments (≤2 treatments, >2 treatments). The response scale was also divided into two dichotomous outcomes: No/Low/Moderate need and High/Very High need, so that associations with High to Very High levels of need could be identified. Demographic variables which had a p value of <0.25 on univariate analysis for each of the four highest need items were included in multivariable logistic regression. Age was also included as a continuous variable in all logistic regression models. The standard error was adjusted for clustering of participants within the seven treatment centres using the Huber–White formula and "vce(cluster)" command in Stata.[49-51] Variables with $p>0.1$ on the Wald test were removed from the final regression analysis.

Aim 5: Identify preferences for format, mechanisms for feedback, and the preferred location for completion

Adolescent and young adult preferences regarding the format of the CNQ–YP and feedback of results were described using frequencies, percentages and 95% CIs.

Sample size and power

It was aimed to recruit a sample of 200 respondents. Assuming a prevalence of need of 50% and allowing for a design effect of 1.1 due to clustering of individuals within centres, this would allow estimation of prevalence of needs with 95% confidence intervals within +/- 7.5% of the point estimate and detection of difference in characteristics of those with and without need of approximately 21% for binary factors and 0.45 of a standard deviation for continuous factors, with a 5% significance level and 80% power. These differences were considered large enough to be clinically meaningful.

Results

The response rate for the study has been reported in Chapter 5.

Demographic characteristics

A summary of all demographic characteristics for the 139 participants is presented in Table 6.1. English was the preferred language for 99% (n=137) of participants. Ninety-one percent (n=124) of AYAs had received their cancer treatments at an adult treatment centre, and 8% (n=11) had received treatment at both adult and children's hospitals.

Since being diagnosed, 11% (n=15) of AYAs reported having difficulty enrolling in secondary, tertiary, or other forms of education, and 22% (n=20) had problems finding work. Approximately half of all young people had undertaken some form of study, or had been employed full-time since their diagnoses (n=60, 51%; and n=52, 49% respectively). With regard to relationships, 84% (n=111) of AYAs reported having at least one sibling, and just over half (n=70, 53%) reported having a spouse, partner or boyfriend/girlfriend.

Table 6.1: Participant demographic characteristics (n=139)

Demographic Characteristic		n (%)
Gender	Female	89 (64)
Cancer type	Lymphoma	35 (25)
	Leukaemia	30 (22)
	Sarcoma	18 (13)
	Melanoma	14 (10)
	Testicular	8 (5.8)
	Bone	7 (5.1)
	Thyroid/Endocrine	6 (4.3)
	Brain	4 (2.9)
	Other	16 (11.6)
Time since first diagnosis	< 12 months	10 (7.2)
	1 to < 2 year	30 (22)
	2 to < 5 year	58 (42)
	≥ 5 years	41 (29)
Recurrence	Same cancer	38 (27)
	Different cancer	5 (3.6)
	Never	96 (69)
*Treatment type**	Chemotherapy	86 (80)
	Radiotherapy	73 (68)
	Surgery	71 (66)
	Stem cell transplant	28 (26)
	Hormone treatment	16 (15)
	Bone marrow	12 (11)
	Other	18 (17)
Treatment stage	Receiving treatment	17 (12)
	Finished treatment and having check-ups	51 (37)
	Cured or in remission	65 (47)
	Other	6 (4.3)
Education completed	University degree	45 (33)
	Trade certificate/diploma	35 (26)
	Secondary (to Grade 12)	46 (34)
	Secondary (to Grade 10)	8 (5.8)
	Primary school	1 (0.7)
	Other	2 (1.5)
Marital status	Single	71 (51)
	Married/De facto	47 (34)
	Separated/Divorced/Widowed	20 (14)
*Living arrangements**	With parent(s)	53 (43)
	With spouse or partner	43 (35)
	With other family	3 (2.4)
	With flatmates	17 (14)
	Alone	7 (5.7)
		Mean (SD)
Current age of young person		25 (3.2)

**Participants were able to select multiple treatment types*

1) Prevalence of unmet needs for any level of need (Low to Very High)

The ten most prevalent items endorsed by AYA cancer survivors for any level of need are presented in Table 6.2. Five items were endorsed by >60% of participants: "Worrying about my cancer returning"; "Finding information that was specifically designed for me"; "Being able to have good food at the treatment centre"; "Being able to talk to people my age who had been through a similar experience"; and "Being able to have leisure spaces and activities at the treatment centre".

Table 6.2: Ten most prevalent items endorsed for any level of need

Item description	n (%)	95% CI
Worrying about my cancer returning	101 (73)	65-80
Finding information that was specifically designed for me	98 (72)	63-79
Being able to have good food at the cancer treatment centre	96 (70)	61-77
Being able to talk to people my age who had been through a similar experience	95 (68)	60-76
Being able to have leisure spaces and activities at the cancer treatment centre	83 (61)	52-69
Cancer treatment staff telling me about the long-term side-effects of treatment	83 (60)	52-68
Being able to have pleasant surroundings at the cancer treatment centre	83 (60)	51-68
Finding information that was easy to get hold of	79 (58)	50-66
Cancer treatment staff telling me what would happen when treatment finished	78 (57)	48-65
Feeling tired	77 (56)	47-64

2) Prevalence of unmet needs at the High to Very High level of need

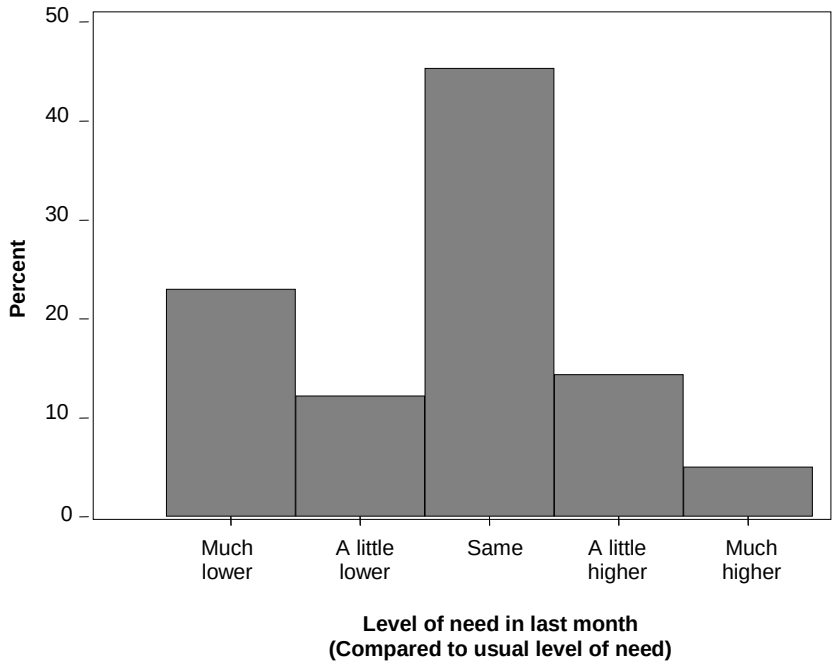
Items with the highest prevalence of endorsement by AYAs at the High to Very High level of need can be seen in Table 6.3. The five items which had the highest proportions at the High to Very High need were the same five items that had the highest proportions at any level of need, although their ranking varied. "Being able to have good food at the treatment centre" became the highest endorsed need, with 38% (n=52) of AYAs indicating they had a High to Very High need for help with this issue.

Table 6.3: Ten most prevalent items endorsed at the High to Very High level of need

Item number and description	n (%)*	95% CI
Being able to have good food at the cancer treatment centre	52 (38)	30-46
Finding information that was specifically designed for me	41 (30)	23-38
Being able to talk to people my age who had been through a similar experience	36 (26)	19-34
Being able to have leisure spaces and activities at the cancer treatment centre	35 (26)	19-34
Worrying about my cancer returning	34 (25)	18-33
Being able to spend time with people my own age during treatment	34 (25)	18-33
Cancer treatment staff telling me whether I would be able to have children	32 (24)	17-31
Coping with changes in my appearance	33 (24)	17-32
Coping with not being able to do the same things as other people my age	33 (24)	18-32
Feeling frustrated	31 (23)	16-30

Almost half the participants stated that their overall level of need in the previous month was the same as in other months since their cancer diagnosis (n=63, 45%). However, Figure 6.1 reveals that 23% (n=32) of AYAs reported that their level of need in the last month was much lower than usual.

Figure 6.1: Overall level of need in the last month compared to other months



3) Domain with the highest median level of need

The domain with the highest median factor score was the Information and Activities domain (Table 6.4). This is not surprising as four of the five items in this domain appeared in the list of the ten most prevalent High to Very High needs. The Education domain had the next highest median level of need. This domain contains three items: "Being able to attend classes"; "Being able to get extensions or special consideration"; and "Being able to receive guidance about study options or future career paths".

Table 6.4: Median factor scores for each domain

			Factor score	
Rank	**Domain**	**n**	**Median**	**Q1-Q3***
1	Information and Activities	139	2.2	1.6 – 3.2
2	Education	81	1.7	1.0 – 2.3
3	Feelings and Relationships	138	1.6	1.2 – 2.4
4	Treatment Environment and Care	139	1.5	1.2 – 2.2
5	Daily Life	138	1.4	1.0 – 2.3
6	Work	111	1.3	1.0 – 2.0

**Q1-Q3 reports the first and third quartile*

4) Associations between AYA characteristics and items with the highest need

The items endorsed by the highest proportion of AYAs at the High to Very High level of need from each of the four main domains were identified as: "Being able to have good food at the cancer treatment centre" (Treatment Environment and Care); "Finding information that was specifically designed for me" (Information and Activities); "Worrying about my cancer returning" (Feelings and Relationships); and "Coping with changes in my appearance" (Daily Life).

Being able to have good food at the cancer treatment centre

Results for the Chi square tests examining differences in AYA characteristics and levels of need for the item “Being able to have good food at the treatment centre”, are presented in Table 6.5. Three variables had p values <0.25 and were eligible for inclusion in the initial logistic regression. “Cancer type” and “Time since first diagnosis” had p<0.1 on the Wald test and were included in the final model (Table 6.6).

Participants diagnosed with haematological cancers had 3.2 times higher odds of endorsing a High to Very High level of need for “Being able to have good food at the treatment centre” compared with AYAs diagnosed with non-haematological cancers (p<0.05). There was also a trend towards AYAs <2 years since their first diagnosis having higher odds of endorsing this item compared to AYAs who were ≥2 years post-diagnosis. However this was not significant at the 0.05 level (p=0.06), possibly due to the small sample size in the former group (n=14).

Finding information that was specifically designed for me

Chi square analysis revealed two AYA characteristic variables which had values of p<0.25 for the item “Finding information that was specifically designed for me” (Table 6.7). The variables “Treatment stage” and “Recurrence” were added to the initial logistic regression model, with only “Recurrence” having a value of p<0.1 on the Wald test (Table 6.8).

Young people who had experienced a recurrence had 2.9 times higher odds of endorsing a High to Very high need for the item “Finding information that was specifically designed for me” compared with AYAs whose cancer had not returned.

Table 6.5: Chi-square tests for the item "Being able to have good food at the cancer treatment centre"

		High to Very High	Chi-square test		
Variable	**Group**	**n (%)**	χ^2	***df***	***p***
Gender	Female	33 (38	0.00	1	0.95
	Male	19 (38)			
Cancer type	Haematological	34 (52)	10.82	1	<0.01*
	Non-haematological	18 (25)			
Number of treatment types	≤ 2	28 (33)	2.39	1	0.12*
	> 2	24 (46)			
Treatment stage	Receiving treatment	4 (25)	1.02	1	0.31
	Finished treatment	44 (38)			
Time since first diagnosis	< 2 years	14 (56)	4.36	1	0.04*
	≥ 2 years	38 (34)			
Recurrence	No	34 (36)	0.62	1	0.43
	Yes	18 (43)			
Partner status	Partner	22 (34)	0.60	1	0.44
	No partner	29 (40)			
Education completed	Tertiary/ trade	31 (39)	0.04	1	0.84
	Primary/ secondary	20 (37)			

**p value <0.25*

Table 6.6: Final logistic regression model for the item "Being able to have good food at the cancer treatment centre"

						Wald test	
Variable	**Group**	**n (%)****	**Odds ratio**	**SE**	**95% CI**	***z***	***p***
Cancer type	Haematological	34 (52)	3.2	0.44	2.5-4.2	8.52	<0.01
	Non-haematological	18 (25)	*				
Time since first diagnosis	< 2 years	14 (56)	2.4	1.09	0.95-5.8	1.85	0.06
	≥ 2 years	38 (34)	*				

**Reference group*

***Proportion of young people with a High to Very High level of need*

Table 6.7: Chi-square tests for the item "Finding information that was specifically designed for me"

		High to Very High	Chi-square test		
Variable	**Group**	**n (%)**	χ^2	***df***	***p***
Gender	Female	29 (33)	1.08	1	0.30
	Male	12 (24)			
Cancer type	Haematological	21 (32)	0.28	1	0.60
	Non-haematological	20 (28)			
Number of treatment types	≤ 2	24 (29)	0.15	1	0.70
	> 2	17 (32)			
Treatment stage	Receiving treatment	8 (47)	3.09	1	0.08*
	Finished treatment	30 (26)			
Time since first diagnosis	< 2 years	9 (36)	0.54	1	0.46
	≥ 2 years	32 (29)			
Recurrence	No	22 (23)	7.31	1	0.01*
	Yes	19 (46)			
Partner status	Partner	18 (28)	0.23	1	0.63
	No partner	23 (32)			
Education completed	Tertiary/ trade	25 (32)	0.23	1	0.63
	Primary/ secondary	15 (28)			

**p value <0.25*

Table 6.8: Final logistic regression model for the item "Finding information that was specifically designed for me"

						Wald test	
Variable	**Group**	**n (%)****	**Odds ratio**	**SE**	**95% CI**	***z***	***p***
Recurrence	No	22 (23)	*				
	Yes	19 (46)	2.9	0.75	1.7-4.8	4.02	<0.01

**Reference group*
***Proportion of young people with a High to Very High level of need*

Worrying about my cancer returning

Table 6.9 reports the results of Chi square tests to detect differences between AYA characteristics and responses to the item "Worrying about my cancer returning". Five variables had values of $p<0.25$ and were added to the initial logistic regression analysis. Two variables had values of $p<0.1$ on the Wald test and were included in the final regression model (Table 6.10). Participants who had experienced >2 different types of treatment (Odds ratio=2.1) or who were <2 years post-diagnosis (Odds ratio=4.0) had significantly higher odds of endorsing a High to Very High level of need for the item "Worrying about my cancer returning" compared with young people who had ≤ 2 treatment types or who were ≥2 years post-diagnosis.

Coping with changes in my appearance

Chi square tests to determine differences between characteristics of young people and levels of need for the item "Coping with changes in my appearance" showed that all variables except "Education completed" had p values <0.25 (Table 6.11). Following the initial logistic regression, "Gender", "Cancer type", "Number of treatment types" and "Recurrence" had values of $p<0.1$ on the Wald test and were included in the final model (Table 6.12).

Female AYAs had almost five times higher odds of endorsing a High to Very High level of need for the item "Coping with changes in my appearance" compared with males (Odds ratio = 4.8). Those who were diagnosed with haematological cancer, had >2 treatment types or had experienced a recurrence had approximately two times higher odds of endorsing a High to Very High level of need for this item (Odds ratios=2.1, 2.4, and 2.2 respectively), compared with young people who were diagnosed with non-haematological cancer, had ≤2 treatment types or had not experienced a recurrence.

Table 6.9: Chi-square tests for the item "Worrying about my cancer returning"

		High to Very High	Chi-square test		
Variable	**Group**	**n (%)**	χ^2	***df***	***p***
Gender	Female	24 (27)	0.91	1	0.34
	Male	10 (20)			
Cancer type	Haematological	19 (30)	1.53	1	0.22*
	Non-haematological	15 (21)			
Number of treatment types	≤ 2	16 (19)	3.87	1	0.05*
	> 2	18 (34)			
Treatment stage	Receiving treatment	6 (38)	1.74	1	0.19*
	Finished treatment	26 (22)			
Time since first diagnosis	< 2 years	12 (48)	8.97	1	<0.01*
	≥ 2 years	22 (19)			
Recurrence	No	20 (21)	2.46	1	0.12*
	Yes	14 (33)			
Partner status	Partner	14 (22)	0.44	1	0.51
	No partner	19 (26)			
Education completed	Tertiary/ trade	19 (24)	0.18	1	0.67
	Primary/ secondary	15 (27)			

**p value <0.25*

Table 6.10: Final logistic regression model for the item "Worrying about my cancer returning"

						Wald test	
Variable	**Group**	**n (%)****	**Odds ratio**	**SE**	**95% CI**	***z***	***p***
Number of treatment types	≤ 2	16 (19)	*				
	> 2	18 (34)	2.1	0.74	1.1-4.2	2.15	0.03
Time since first diagnosis	< 2 years	12 (48)	4.0	1.69	1.8-9.2	3.34	<0.01
	≥ 2 years	22 (19)	*				

**Reference group*
***Proportion of young people with a High to Very High level of need*

Table 6.11: Chi-square tests for the item "Coping with changes in my appearance"

		High to Very High	Chi-square test		
Variable	**Group**	**n (%)**	χ^2	***df***	***p***
Gender	Female	28 (32)	8.34	1	<0.01*
	Male	5 (10)			
Cancer type	Haematological	22 (34)	6.44	1	0.01*
	Non-haematological	11 (15)			
Number of treatment types	≤ 2	13 (15)	9.47	1	<0.01*
	> 2	20 (38)			
Treatment stage	Receiving treatment	7 (41)	3.78	1	0.05*
	Finished treatment	23 (20)			
Time since first diagnosis	< 2 years	10 (40)	4.34	1	0.04*
	≥ 2 years	23 (20)			
Recurrence	No	18 (19)	4.48	1	0.03*
	Yes	15 (36)			
Partner status	Partner	11 (17)	3.18	1	0.07*
	No partner	21 (30)			
Education completed	Tertiary/ trade	19 (24)	0.03	1	0.85
	Primary/ secondary	14 (25)			

**p value <0.25*

Table 6.12: Final logistic regression model for the item "Coping with changes in my appearance"

						Wald test	
Variable	**Group**	**n (%)****	**Odds ratio**	**SE**	**95% CI**	***z***	***p***
Gender	Female	28 (32)	4.8	0.81	3.4-6.7	9.21	<0.01
	Male	5 (10)	*				
Cancer type	Haematological	22 (34)	2.1	0.51	1.3-3.4	2.86	<0.01
	Non-haematological	11 (15)	*				
Number of treatment types	≤ 2	13 (15)	*				
	> 2	20 (38)	2.4	0.78	1.3-4.6	2.73	0.01
Recurrence	No	18 (19)	*				
	Yes	15 (36)	2.2	0.88	1.0-4.8	1.97	0.05

**Reference group*
***Proportion of young people with a High to Very High level of need*

5) Preferences for format, mechanisms for feedback, and preferred location for completion

Measure format preferences

More than 80% of AYAs spent two or more hours on a computer each week, with almost 60% reporting spending more than eight hours. However, less than half of all AYAs agreed or strongly agreed that they would prefer to complete the measure online (43%, n=60, 95% CI 35-52%), with 36% (n=50, 95% CI 28-44%) disagreeing or strongly disagreeing and 21% (n=29, 95% CI 15-29%) unsure. Furthermore, a large proportion of participants also disagreed or strongly disagreed that web-based surveys provided greater privacy than paper-based surveys (45%, n=63, 95% CI 37-54%) and were easier to restart following interruptions (46%, n=64, 95% CI 38-55%). However, young people did agree or strongly agree that web-based surveys were easier to complete (47%, n=65, 95% CI 39-55%), more convenient (63%, n=87, 95% CI 54-70%) and less likely to get lost (60%, n=83, 95% CI 51-68%) than paper-based surveys.

Feedback of results

When asked about utilising the data from the measure, over 90% of AYAs agreed or strongly agreed that the results could be provided to treatment staff (93%, n=128, 95% CI 87-96%), treatment centres (95%, n=131, 95% CI 90-98%), researchers (94%, n=130, 95% CI 89-97%) and funding organisations (94%, n=130, 95% CI 89-97%) to inform research, interventions and better care for young people with cancer.

Preferred location for completion

The majority of participants indicated that they would prefer to complete the measure at home (90%, n=124, 95% CI 84-94%), with most young people disagreeing or strongly disagreeing that appropriate locations were their general practitioners' surgeries (83%, n=114, 95% CI 75-88%), the treatment centres while they were receiving treatment

(70%, n=96, 95% CI 61-77%) or the treatment centres while they were having check-ups (67%, n=93, 95% CI 59-74%).

Discussion

The results of this research have revealed that a large proportion of AYA cancer patients and survivors perceive that they have needs which remain unmet. Seventy-three percent of young people endorsed an item at any level of need, and 38% endorsed an item at the High to Very High level of need. Of the six domains explored, the highest median factor score was observed in the Information and Activities domain, with four of the five items in this domain appearing among the ten most prevalent needs at the High to Very High level. For AYAs who were currently studying, the median level of need for the Education domain was also high.

Although the greatest proportion of participants (45%) reported that their level of need in the last month was the same as usual, almost a quarter (23%) of participants reported that their needs in the last month were much lower than usual. The variability in these results may reflect the differences in the time since diagnoses for AYAs included in the current sample. These results also highlight that needs are dynamic and can vary over time, an attribute which may need to be accounted for when considering the prevalence of unmet needs at the population level.[8] For this reason regular assessment of the unmet needs of AYA cancer survivors might be required in order to identify the emergence of any new needs.

Prevalence of unmet need in AYAs compared with other cancer populations

A number of the unmet needs reported by young people in this study appear to be universal to both AYAs and older adult cancer survivors. Seventy-three percent of

young people in the current study reported having some level of need for "Worrying about the cancer returning", with 25% of AYAs endorsing a High to Very High level of need for this item. Similar findings have emerged in the adult unmet needs literature, with between 26-39% of cancer patients and survivors reporting unmet needs with this issue.[4 52] Over half of all AYAs (56%) reported wanting assistance with "Feeling tired", another need frequently reported by adult cancer survivors of all ages (25-52%).[52-55] Information needs related to cancer treatment and potential long-term side-effects are also shared among both groups.[56] Therefore, unmet needs concerned with the fear of the cancer returning and managing to live with the long-term physical side-effects of cancer treatment are recognised as being common to AYAs and older adults alike.

However, differences between the needs of AYAs and older adult cancer survivors were observed in the area of psychological distress. Common themes in the adult unmet needs literature are the high levels of reported anxiety and depression. For older adult cancer patients and survivors, needs for assistance with anxiety have been reported to range from 9-37%, while needs related to depression range between 11-36%.[7 53-55] Although a large proportion of AYAs in the current study reported having a High to Very High level of need for help with "Feeling frustrated" (23%), unmet needs related to anxiety and depression were notably absent. This may indicate that psychological needs such as anxiety and depression for AYAs are being adequately met. However, it may also be a product of the developmental stage that AYAs are experiencing. For example, changes in mood such as feeling down or anxious are a common feature of adolescence,[57] therefore AYAs may not perceive that their levels of anxiety or depression have changed significantly post-diagnosis. Alternatively, AYAs may not be aware that they are experiencing symptoms of anxiety or depression. For this reason, the two step screening approach (as outlined earlier in this chapter) is recommended.[9] This process provides an opportunity for health care providers to

identify additional needs following the initial screening, such as requiring help for depression, that the individual themself may be unable to recognise .

A further essential difference in the needs of AYAs and older adults was observed in the emphasis placed on peers compared to close family members. Older adult cancer survivors expressed high levels of concern about the ability of family members to care for them,[52] or their own ability to care for others.[55] In contrast, AYAs placed a greater level of importance on their relationships with peers. “Spending time with people their own age during treatment” was reported as a High to Very High level of need by 22% of participants. Differences in the importance of relationships with family members compared to peers is possibly another reflection of the life stages of each group. Adolescence and young adulthood is a time of establishing independence and for identity creation in young people.[58] It is also the time in their life where they begin to move away from their family unit, and acceptance by peers takes on a new level of significance.[59] For AYAs, relying on their parents to care for them may interfere with their achievement of independence.[60] However, AYAs may perceive that the care provided to them by their parents is just an extension of their parents’ usual role. This, or simply the fact that their family have gathered close around them, may explain why AYAs do not appear to have high needs related to the ability of others to care for them. Similarly, AYAs did not report any needs related to caring for others, possibly due to the small proportion of participants who live with a spouse or partner (35%), or who potentially have children of their own.

The issue of one day having children however, is of great concern to AYAs with almost a quarter of all young people (24%) reporting that they had a High to Very High level of need to be told “Whether I would be able to have children” by cancer treatment staff. The issue of fertility is a common concern for cancer patients and survivors in their

reproductive years. It has been estimated that around 50% of women under the age of 40 who undergo treatment for cancer will lose their reproductive functioning.[61] For post-pubertal men under 55 years of age, the option of sperm-banking may be available, however, future conception will not be natural and the reported success rates are low.[62] In a study of 577 breast cancer survivors who were under 50 years of age at diagnosis, Ganz and colleagues found that treatment related menopause was associated with poorer health perceptions and emotional functioning, particularly for women between 25 and 34 years of age.[63] Given that available information on the topic of fertility for young adult cancer survivors is limited,[64] it is not surprising that this remains a high area of unmet need.

Some of the most prevalent High to Very High unmet needs reported by AYAs appear to be related to the treatment centre environment itself. The most prevalent unmet need reported by participants at the High to Very High level was “Being able to have good food at the treatment centre” (38%). The experience of receiving treatment for cancer has the potential to alter a young person’s relationship with food in a number of ways. For example, side-effects of chemotherapy and radiotherapy such as nausea, vomiting, diarrhoea, constipation, changes in taste perception and mucositis may significantly limit the types of foods which a young person can tolerate or digest.[65] The inability to access food at the treatment centre which is nutritional, appealing and suitable for AYAs undergoing treatment may not only be an unmet need, but could also potentially lead to more serious outcomes such as the reduced effectiveness of cancer treatment.[65]

“Being able to have leisure spaces and activities” and “Being able to have pleasant surroundings” at the cancer treatment were also among the most prevalent High to Very High unmet needs reported by AYAs. While there has been limited research

assessing the role of treatment centre characteristics on the psychosocial well-being of cancer patients and survivors,[66] it is plausible that the physical environment, together with the processes and facilities within the treatment centre, may influence such outcomes. In response to reported needs related to the treatment centre environment, purpose built Teenage and Young Adult (TYA) cancer centres have been established in the United Kingdom (UK).[67] The specific features of these centres include pleasant, age appropriate surroundings and leisure spaces, kitchens, and music.[67] The centres ensure that AYAs are placed in units with other young people who are going through a similar experience, rather than in general hospital wards with possibly very young children or much older adults. There is some evidence which suggests that AYAs who receive their cancer treatment at a purpose built TYA cancer centre may have better psychosocial outcomes than those who have been treated at either a paediatric or adult hospital.[68]

Associations between high unmet needs and characteristics of AYAs

Of the three demographic variables (i.e. age, gender and education completed) that were examined in the logistic regression analyses, only one variable was associated with High to Very High levels of need. Being female significantly increased the odds (between 3 and 7 times) of an AYA endorsing a High to Very High level of need for the item “Coping with changes in my appearance”. This result mirrors similar findings in the AYA QOL literature indicating that female AYAs report significantly worse quality of life related to body image compared to males.[20] As discussed in Chapter 1, the degree to which a young person perceives themselves to be sexually attractive can have a considerable impact on their overall self-esteem.[27] Concerns related to body image in adolescent girls are frequently cited in the AYA literature and may be a result of increasing social pressure to conform to the feminine ideal.[27] Adolescence and young adulthood is the time of life when a young person is already experiencing physical

changes related to the onset of puberty. It is no wonder that the addition of other treatment-related physical changes, such as surgical scars, hair loss or weight loss or gain can lead to high levels of distress.[69]

All of the remaining variables associated with High to Very High levels of need were related to AYA disease and treatment characteristics. Adolescents and young adults diagnosed with haematological malignancies had more than three times higher odds of endorsing a High to Very High level of need for "Being able to have good food at the treatment centre" compared to AYAs with non-haematological cancers. The experience of being diagnosed with a haematological malignancy may be unique to other cancers in terms of the type, length and severity of the treatment required.[70] Adolescents and young adults with haematological cancers may remain at the treatment centre as inpatients for extended periods of time.[70] As the AYA's primary source of food would more than likely be provided by the hospital it is understandable that the perceived quality and choice of available foods may be an important need for this sub-group.

Receiving more than two different types of treatment and being less than two years post-diagnosis were associated with AYAs having higher odds of "Worrying about my cancer returning". This finding once again aligns with the AYA QOL literature finding that AYAs who were treated with combination therapy or who were still receiving treatment reported worse quality of life.[19] Combinations of treatment may result in significantly more severe short- and long-term side-effects for AYAs which may lead to a more negative cancer experience compared with AYAs who only receive one type of treatment, or a less severe type of treatment such a surgery. The greater levels of psychosocial morbidity generated by the receipt of multiple treatment types may make AYAs more fearful of cancer and its treatment and therefore lead to increased worries about the cancer returning. For AYAs who are less than two years post-diagnosis the

memory of the cancer is potentially still very vivid. It is also possible that some AYAs who are less than two years post-diagnosis are still receiving treatment. As time passes, the likelihood that the cancer will return diminishes. Therefore, it is not surprising that for AYAs being two or more years post-diagnosis was not associated with a high level of need for this item.

For AYAs who had experienced a recurrence, high levels of unmet need were identified for the item "Finding information that was specifically designed for me". Information which was suitable around the time of an initial cancer diagnosis may not be appropriate for AYAs who experience a second cancer or recurrence. For this sub-group it is possible that the chances of remission or survival may be reduced, and that alternatives to treatment such as palliation may need to be considered.[71] Given the prevalence of High to Very High levels of need endorsed for this item, it appears that information targeted to AYAs who experience a recurrence may be potentially lacking.

Finally, it is interesting to note that characteristics of the social environment (i.e. partner status) were not associated with any of the most prevalent unmet needs. This may simply be because no associations exist. However, the low number of AYAs who reported having a partner could also mean that there was insufficient power to detect small differences between these two groups.

Potential for routine screening of unmet needs

Over 90% of participants in this study agreed that it would be suitable and acceptable to provide feed back to health care providers, treatment centres, researchers and funding organisations regarding their reported unmet needs. This implies that the CNQ–YP has the potential to be used routinely by these groups in order to inform research funding, intervention development, service delivery and individual care.

However, 90% of participants also indicated that they would they would prefer to complete the measure at home, with most AYAs indicating that the treatment centre was an inappropriate setting for administering the survey.

This finding raises issues for the potential use of the CNQ–YP for routine screening in the clinical setting. Reasons as to why AYAs would prefer to complete the measure at home can only be speculated, however may be related to a lack of time and privacy while at the treatment centre. Adolescents and young adults may also feel more comfortable reporting unmet needs which could potentially criticise their treatment and care away from their health care provider. If routine screening was to occur in the patients home, issues related to cost, data collection and adherence would need to be considered.

Potential for an online version of the CNQ-YP

Web-based unmet needs assessment may be a practical alternative to conducting routine screening within a treatment centre or clinic.[44] Not only does an online format allow participants the flexibility to complete the measure in their own home, it also overcomes problems of data entry as responses can be exported directly into an existing database.[72] A number of previous studies have successfully assessed the psychosocial well-being and the information and service needs of AYA cancer patients and survivors using online surveys.[44 45] However, although 80% of AYAs in the present study reported spending at least two hours per week on a computer, less than half of all AYAs said they would prefer to complete the CNQ–YP in an online format, with 36% of AYAs disagreeing or strongly disagreeing that they would prefer to complete the measure online. These results suggest that barriers to using online needs assessment measures with AYA cancer survivors may need to be further explored.

Despite this, a large proportion of AYAs in the current study agreed or strongly agreed that online surveys were more convenient (63%) than paper-based surveys. Furthermore, web-based assessment has other advantages such as the potential to access individuals who may be either socially or geographically isolated.[73] They are also reasonably cost-efficient as, once developed, maintenance only requires the costs associated with online hosting, which may compare favourably with the costs of printing survey materials or postage.[73] Therefore, it would seem appropriate to develop and validate the psychometric properties of an online version of the CNQ– YP.

Limitations

The primary limitations of this study are related to the small sample size achieved (n=139) and problems with the potential representativeness of the sample. Potential reasons for the small sample size have been discussed in detail (Chapter 5) and may be related to the high mobility of AYAs leading to difficulties in obtaining up-to-date contact details. In the current study, the small sample size may have limited the power to detect differences in levels of unmet need between different characteristics of AYAs. It is possible that more significant associations between individual variables and high levels of unmet need may have emerged with a larger sample.

The rationale for recruiting AYAs to the study using patient databases from treatment centres, rather than through a population-based source such as a cancer registry has also been explained (Chapters 4 and 5). A disadvantage of treatment centre recruitment is that the sample achieved may not be representative of all groups of AYA cancer patients and survivors. However, the demographic characteristics of participants (Table 6.1) show a wide range of cancer types and a variety of times since diagnosis. Therefore, it is likely that the results represent the overall needs of the larger AYA cancer population.

A further limitation of this study may be the way in which associations between AYA characteristics and High to Very High levels of unmet need were assessed. The approach employed in this study was to use a highly prevalent single item of unmet need as the outcome and then undertake a logistic regression to that one item. Another approach may have been to explore associations between individual characteristics and factor scores for each of the six domains. However, the factor scores were highly skewed, which meant that the assumption of normality of residuals for linear regression would likely be invalid. In addition, interpretation of the clinical importance of any differences in factors scores associated with explanatory variables may be difficult, particularly given the early development phase of the instrument.

An alternative method could be to dichotomise the factor scores into “High” versus “Other” levels of need. However, determining what cut-off value for a factor score represents a high level of unmet need is potentially problematic. A moderate factor score may actually be the result of primarily low levels of need for the majority of items, averaged with very high levels of unmet need for one or two items. For this reason logistic regression to an entire domain would fail to separate out which items are most relevant to which sub-groups of patients. For example, knowing that female AYAs have higher odds of reporting high levels of unmet need for the Feelings and Relationships domain provides a vague guide at best to informing the types of interventions or services that may be necessary to meet these needs. In contrast, regression to an individual item provides a detailed description of the exact need and sub-group of patients who require assistance. In this way specific and tailored interventions can be designed in an attempt to reduce the most prevalent needs within the population. Future research into the CNQ–YP should focus on consideration of the clinical relevance of factors scores and cut points.

Despite these limitations, the study also has a number of strengths. Information regarding the unmet needs of AYAs was collected using a self-report measure specifically developed for this population, which provided young people with the opportunity to directly voice their concerns. Furthermore, the measure used to assess these needs (the CNQ–YP) has displayed good reliability and validity, increasing the likelihood that the scale was able to accurately capture the reported needs of this group. To our knowledge this is the first assessment of the psychosocial needs of AYA cancer survivors which has been undertaken with a psychometrically robust measure .

Conclusion

High levels of unmet need are experienced by a large proportion of AYA cancer patients and survivors. In particular, unmet needs related to the treatment centre environment such as the availability of good food and leisure spaces, as well as needs related to body image, fertility, peer interaction, physical functioning, and tailored information, are prevalent. These areas should be the focus of any future interventions which aim to improve psychosocial outcomes for this population. AYAs who are female, have been diagnosed with haematological cancer, have experienced a recurrence, received more than two types of treatment, or who are less than two years post-diagnosis may have higher odds of experiencing high levels of unmet need for a number of issues. Therefore, the development of targeted services and support for these AYA sub-groups appears to be warranted.

The CNQ–YP is a potentially feasible way of providing feedback to health care providers, treatment centres, and researchers regarding the concerns of this population. Given that completion of the measure at home is the overwhelming preference of AYAs, flexible methods of administration such as web-based assessment should be explored.

CHAPTER 7

Addressing the unmet needs of adolescent and young adult (AYA) cancer survivors: the way forward

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Chapter 7

Addressing the unmet needs of adolescent and young adult (AYA) cancer survivors: the way forward

Why is this research needed?

Adolescent and young adult (AYA) cancer survivors are a vulnerable population who may experience unique psychosocial needs

As discussed in Chapter 1, the life-stages of adolescence and young adulthood represent critical phases of human development when the physical, psychological, cognitive, sexual and social aspects of an individual reach new levels of maturation. The realisation of identity, intimacy, independence and autonomy are key milestones which need to be achieved in order to make a successful transition into full adulthood.[1] For adolescents and young adults (AYAs) a diagnosis of cancer and the impact of its treatment can potentially interfere with the attainment of these milestones, resulting in short- and long-term psychosocial morbidity.[2] Although the survival rates for many AYA cancer types are high, the young age at which the diagnosis occurs can mean that a large proportion of AYAs may live for many years with the late effects of their disease and treatment.[3]

To better understand the context of psychosocial outcomes for AYA cancer survivors, an examination of the health care system and the way it responds to the needs of young people, should be performed.[4] Assessing the prevalence and predictors of unmet need in AYAs is the first step towards developing interventions and targeting resources aimed at improving the psychosocial outcomes of this population.[5] Given that the diagnosis of cancer in AYAs occurs at a distinct time of life, AYAs may experience unique psychosocial needs which may not occur in children or older

adults.[6] Therefore, psychosocial health measures specifically developed and validated with AYA cancer patients and survivors may be required to enable accurate assessment of the diverse physical, psychological and social needs of this group.

Existing measures of psychosocial health for AYAs may be limited

A critical review of the literature was undertaken in Chapter 2 to identify multi-dimensional, self-report measures which had been developed to assess the psychosocial health of AYA cancer survivors. The psychometric characteristics of all scales were reviewed to determine how well each measure met predetermined criteria considered important for measure development.[7-9] The review concluded that the internal consistency and face, content and construct validity for all seven measures were psychometrically adequate. However, the test-retest reliability, predictive validity, responsiveness, acceptability and feasibility of the scales were rarely examined. The results of this review raised doubts about the ability of existing measures to accurately estimate the prevalence of psychosocial morbidity among AYA cancer survivors. Furthermore, the capacity of these measures to reliably and validly: identify individuals at risk of experiencing poor psychosocial health outcomes; detect clinically important changes in psychosocial well-being; or predict future psychosocial health outcomes, is dimished.[10] As such, the adoption and utilisation of these measures at either the population or individual screening level may be limited.

The critical review also revealed that no existing measures of unmet need had been developed specifically for AYA cancer patients or survivors. Measures of unmet need may have some advantages over other types of multidimensional scales, such as quality of life (QOL) measures, as they remove the assumptions made by health care providers or researchers regarding the issues a patient would like to receive help

with.[11] The relative importance of an item compared with other unmet needs can also be determined.[12]

What has this research contributed?

Development of a new measure of unmet need specifically designed for AYA cancer patients and survivors

Chapters 3 and 4 described the preliminary development of a measure specifically designed to assess the unmet needs of AYA cancer patients and survivors. A draft measure of 108 items and seven domains thought to be relevant to AYAs was generated from existing measures of unmet need for adult cancer patients and survivors. The face and content validity of the draft measure were established *via* feedback from AYA cancer patients and survivors, parents of AYAs, health care providers, researchers, and consumers and professionals who had not experienced cancer. Advice from these groups led to a revised measure with 36 new items and one additional domain.

The draft version of the Cancer Needs Questionnaire – Young People (CNQ–YP) was then pilot tested to determine its acceptability. Adolescent and young adult cancer survivors were recruited through a population-based, state cancer registry using an active clinician and patient consent protocol. The length, reading age, and clarity of the CNQ–YP were determined to be acceptable by AYAs who participated in the study.

The recruitment protocols used to generate both the focus groups in Chapter 3 (*via* CanTeen) and the population-based sample in Chapter 4 (*via* the New South Wales Central Cancer Registry) resulted in smaller than anticipated sample sizes. However, the methods used to establish the face and content validity of the CNQ–YP, as well as

its acceptability, also had a number of strengths. First, qualitative feedback from not only health professionals but also consumers contributed to the development and assessment of items and domains in the measure. Second, although small, the sample of AYAs recruited through the cancer registry that assessed the acceptability of the measure did not significantly differ to the population of non-consenters, except by age. Third, the response rate for measure completion in the pilot study was also reasonable, providing further evidence that the CNQ–YP was perceived to be acceptable to AYA cancer survivors.

Psychometric evaluation of the reliability and validity of the CNQ–YP

Revision of the CNQ–YP based on feedback from the pilot study resulted in an extension of the recall time-frame from "in the last month" to "any time since your cancer diagnosis" for five of the domains. The response scale was also modified and screening questions were added so that AYAs were only required to answer questions for domains which were relevant to their situation. Following these revisions, the psychometric properties of the measure were examined in Chapter 5, including its factor structure, internal consistency, test-retest reliability, discriminative validity, potential responsiveness and acceptability.

A sample of 139 AYAs recruited through seven treatment centres across five states in Australia completed the measure at baseline, with 34 AYAs completing the measure a second time to establish test-retest reliability. Factor analysis of the CNQ–YP identified six underlying factors related to the AYAs Treatment Environment and Care, Daily Life, Feelings and Relationships, Information and Activities, Education and Work. The four main factors of the CNQ–YP accounted for 63% of the variance. Internal consistency of the measure was high, with all factors achieving Cronbach's alpha values greater than 0.80. Test-retest reliability was also acceptable, with all but four items achieving a

weighted kappa value greater than 0.60. As in the pilot study, the CNQ–YP was perceived to be acceptable to AYAs who had been diagnosed with cancer. Adolescents and young adults who were “receiving treatment” displayed higher median factor scores for the four main factors compared with AYAs who had “finished treatment”. However, no significant differences between the factor scores of the two groups were detected. One explanation for the non-significant differences may have been the recall time-frame used for some of the factors in the measure (“any time since diagnosis”). The phrasing of this stem question effectively allowed AYAs who had finished treatment to answer items in a way that reflected the level of unmet need they had experienced while receiving treatment. It is also possible that the small sample size in the “receiving treatment” group (n=17) may have limited the power of the statistical tests by only allowing the detection of large differences.

The potential responsiveness of the CNQ–YP may be restricted by the large floor effects achieved. The presence of floor effects in a measure can have implications for assessing the effectiveness of intervention studies, as researchers may be unable to detect a reduction in unmet need due to the large proportion of participants who report having no needs.[13] Despite this, the distribution of participant responses across different levels of need for all items in the measure suggests that the CNQ–YP is able to capture variations in the levels of unmet need experienced by AYAs.

Although the analysis performed in this chapter was limited by the sample size, the CNQ–YP displayed psychometrically acceptable reliability and validity and conformed to other recommended psychometric criteria. Assessing and achieving high test-retest reliability for the measure at the item level was a particular strength, as test-retest reliability in other multi-dimensional (QOL) scales developed for AYAs with cancer has rarely been examined. Analysis of the CNQ–YP’s factor structure and test-retest

reliability also resulted in a shorter version of the measure (a reduction from 144 items to 75 items) which will significantly reduce the burden placed on future respondents (Appendix 7.1).

Description of the prevalence of unmet needs and associations between high unmet needs and characteristics of AYAs

In Chapter 6, the prevalence of unmet needs reported by the 139 AYAs in the study were explored at both the item and domain level. Seventy-three percent of AYAs endorsed at least one item at any level of need, with 38% endorsing an item at the High to Very High level. The most prevalent items endorsed at the High to Very High level of need included: “Being able to have good food at the cancer treatment centre”; “Finding information that was specifically designed for me”; “Being able to talk to people my age who had been through a similar experience”; “Being able to have leisure spaces and activities at the cancer treatment centre”; and “Worrying about my cancer returning”. The Information and Activities domain had the highest median factor score.

The most prevalent need for each of the four main domains was then identified and multivariable logistic regression was performed to identify associations between high unmet needs and demographic, disease, treatment and social environment characteristics of AYAs. Participants who were female, diagnosed with haematological cancer, had experienced a recurrence, received more than two types of treatment, or who were less than two years post-diagnosis had significantly higher odds of endorsing high levels of unmet need for a number of issues. Identifying these sub-groups of AYAs who are at risk of experiencing high levels of unmet need is important for the reducing the increased psychosocial burden experienced by these groups.

Alternative methods for investigating associations between AYA characteristics and High to Very High levels of unmet need could have been used in the study. For example, linear regression to identify associations between AYA characteristics and median factor scores for each of the six domains could have been examined. However, in the current study this method of analysis was not considered suitable, as the assumption of the normality of residuals would have been invalid due to the highly skewed distribution of the factor scores.

The method of regression to an individual item rather than a domain has the advantage of providing specific and detailed information about particularly high unmet needs which should be addressed. Regression to a domain can only allude to a general area of need, which would subsequently require further exploration before any tailored interventions could be designed. Future research could, however, focus on the possible clinical relevance of certain cut-off values of factor scores, and how these might be interpreted.

The way forward

Refine and redevelop the measure with a larger sample

A common thread throughout the studies reported in this thesis has been the enormous challenges involved in recruiting large and representative samples of AYA cancer survivors. These challenges became apparent from the very initial stages of research planning and design for the study. Due to their young age, a number of barriers to accessing AYAs for participation in psychosocial research exist. Young people with cancer are highly protected by external moderators such as ethics committees, cancer registries and clinicians. This protection, in addition to the already low incidence of cancer diagnosed in AYAs, severely limits the potential to recruit sufficiently large

numbers of this vulnerable population to allow meaningful data collection and statistical interpretation of results.

Additional characteristics of the population, such as the high mobility of AYAs further decreases the likelihood of being able to recruit an adequate sample of the population. Current protocols and procedures in place to access other cancer populations, such as older adults, may be inappropriate for survivors in this age group. Instead, a nationally coordinated effort involving the commitment of a wide range of healthcare providers and a large number of treatment centres may be necessary for undertaking research with this population.

Recruitment of an adequately large and representative sample is necessary to allow further development and refinement of the CNQ–YP. Although the acceptability, reliability and face and content validity of the measure has been demonstrated, there are a number of psychometric properties of the measure which require further evaluation. First, the test-retest reliability of the measure should be re-examined over a shorter time-frame. A time-frame of 2 to 14 days is recommended as the most appropriate interval to assess test-retest reliability.[14] In the current study however, the median time taken to complete and return the measure was closer to one month. This delayed response in returning the measure may have been attributable to the paper and pencil format of the CNQ–YP. Alternative formats and methods of administration, such as through the use of an online survey or via computer assisted telephone interviews may assist in decreasing the time taken for young people to respond.

Second, the predictive validity of the CNQ–YP should be evaluated to identify possible future morbidity. However, there are a number of methodological issues which would need to be considered. Determining possible future outcomes of AYA cancer survivors

who are currently experiencing high levels of unmet need, may be difficult to establish. Although there is a large body of epidemiological data which has explored the predictive nature of disease and treatment characteristics on the long-term QOL of AYA cancer survivors, associations between unmet needs and long-term outcomes have yet to be investigated.

Conducting longitudinal studies which explore the predictive nature of unmet needs on future health outcomes may be one method of obtaining this data. However, longitudinal research with cancer populations is often difficult to conduct due to problems of sample attrition, and high rates of mortality and morbidity. These methodological issues may be further complicated when studying AYAs due their high level of geographical mobility.

Develop interventions to address the unmet needs of AYA cancer survivors

Once the reliability and validity of the CNQ–YP has been established, it could be used as an outcome measure to test the effectiveness of interventions designed to improve the psychosocial well-being of AYAs. As the unmet needs of this population are multi-dimensional, a broad range of factors may contribute to the unmet needs of a young person with cancer. Interventions may therefore need to target factors associated with the characteristics of the individual, as well as the broader system in which health care is delivered.[15] The design of any such interventions may also need to be considered, given the low incidence and potentially limited participant samples of AYAs available at treatment centres.

Conclusion

The diagnosis of cancer in AYAs can be an unexpected, frightening and highly emotional experience for both the young person and their family. The impact of the

disease and its treatment can have a detrimental effect on almost all aspects of a young person's life. At a time when AYAs should be exerting their independence and autonomy from their parents, they can suddenly find themselves thrown back into a position of high dependence for not only practical assistance, but also emotional and financial support. Other developmental milestones, such as the formation of intimate relationships with peers, are also affected with young people often experiencing feelings of isolation. Regardless of the prognosis of their disease, the impact of cancer will have a profound effect upon the life of the individual and those around them, possibly for many years. Plans for the young person's future, such as being able to study, follow a particular career path or have their own family may be altered by the cancer experience. Understanding the psychosocial well-being, and in particular, the unmet needs of this population is therefore critical. Managing to access sufficiently large samples of AYA participants is the key to being able to investigate the needs of this group. Without this access, the further development of a psychometrically robust measure of unmet needs will be difficult and the prevalence of needs in this population will remain unknown. There are many challenges involved with attempting to undertake research with AYAs, however understanding of the needs of this vulnerable population is obviously worthy of investigation. Although hampered by the challenges of accessing AYAs, this thesis has made a contribution to furthering our knowledge of, and ability to assess, psychosocial well-being in this important group.

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Chapter 1

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