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Abstract

The adolescent and young adult (AYA) period is unique; individuals are faced with personal and developmental challenges, which are amplified by having a diagnosis of cancer. Understanding the unique challenges of AYA is critical and may be assessed through the use of clinically meaningful and psychometrically sound scales measuring the impact of cancer on health-related quality of life (HRQL). The purpose of this chapter is to identify patient-reported outcome (PRO) instruments used with AYA with cancer to develop a preliminary conceptual framework of the HRQL content deemed important for AYA. Findings from two previous systematic reviews and a search of Medline, PsycINFO, EMBASE, and CINAHL from January 2008 to December 2014 were conducted by our team to identify self-report cancer-specific PRO instruments for AYA. Twelve instruments developed for cancer patients and survivors were identified. A content analysis of 418 items from these instruments led to the identification of six major domains as follows: psychological, social, physical, general, sexual, and spiritual. Important differences in content were noted between PRO instruments designed for pediatric patients versus young adults. Specifically, pediatric tools lacked items to measure spirituality,

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goal setting/future plans, and sexual and reproductive health, while instruments designed specifically for AYA tended to measure the breadth of concerns of AYA. Using the most appropriate PRO instrument in clinical research and/or practice is crucial. Therefore, in selecting a PRO instrument to measure the HRQL of AYA, it is important to carefully consider how an instrument was developed and whether its content will appropriately answer the research question or clinical evaluative purpose.

30.1 Introduction

The cancer experience of adolescents and young adults (AYA) is unique [1]. The AYA period is a distinct developmental stage that is characterized by social, emotional, physical, and neuropsychological development [2]. During this time, AYA are faced with the challenges of gaining autonomy from parents, building personal values and identity, developing strong peer relationships (including intimate and sexual relationships), pursuing further education at college or university, and joining the workforce to become financially independent [3]. Cancer-specific issues, such as premature confrontation with mortality, changes in physical appearance, increased dependence on parents, disruptions in social life, education or employment due to treatment, loss of reproductive capacity, and health-related concerns about the future, may be particularly distressing for AYA [4]. Such concerns can have an important impact on the health-related quality of life (HRQL) of patients during active treatment and during survivorship [4]. As evidence-based medicine is rapidly setting a standard for clinical decision-making in the care of AYA cancer patients, the availability of clinically meaningful and psychometrically sound tools to measure the impact of cancer on HRQL is essential.

30.1.1 Definitions of HRQL

The World Health Organization's (WHO) definition of health "a state of complete physical, mental, and social well-being..." [5] has been the cornerstone for the definition of HRQL for many years. Beyond this definition, which categorizes

health into three broad aspects (physical, social, and psychological), there is a lack of consensus on what constitutes HRQL, and more than 100 definitions have been proposed, with a variety of terms (e.g., quality of life (QOL), functional status, health status) sometimes used interchangeably [5–7]. A helpful definition of HRQL by the USA Food and Drug Administration (FDA) is as follows: "HRQL is a multi-domain concept that represents the patient's general perception of the effect of illness and treatment on physical, psychological, and social aspects of life" [8].

Fayed and colleagues performed content analyses of the most common patient-reported outcome (PRO) instruments measuring health outcomes of children and adolescents (generic [9] and cancer-specific [10]). They found that the content of instruments that purport to measure HRQL, or the broader notion of QOL, is weighted toward measuring performance, capacity, frequency, severity, and presence or absence of life and/or health domains, rather than targeting enjoyment, satisfaction, expectations, standards, or concerns about life and/or health domains [9, 10]. Given the lack of conceptual consistency among instruments, these authors advise that, in choosing an instrument, the content of potentially relevant scales should be considered carefully relative to one's research or clinical evaluative purpose.

30.1.2 Increase in Publications About QOL in AYA

Studies of QOL in AYA with cancer have increased dramatically over the past two decades. Figure 30.1 shows the yearly number of

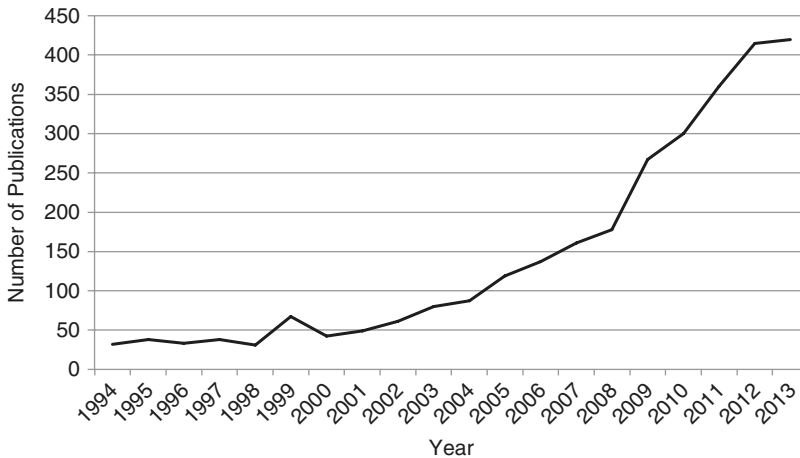


Fig. 30.1 Number of publications per year from January 1994 to December 2013

publications indexed in PUBMED from the following search: “Neoplasms” [MeSH] and “Quality of Life” [MeSH], with age limited to adolescents (13–18 years) and young adults (19–24 years). A total of 2,915 articles (of 22,670 without an age limitation) were retrieved through this search.

30.1.3 Patient-Reported Outcome Instruments

HRQL is typically measured using Clinical Outcome Assessment (COA) tools, i.e., instruments designed to measure concepts that include symptoms, overall mental state, or the effects of a disease or condition on how the patient functions and feels in their daily life [8]. The USA FDA has classified COA tools into four types: (1) clinician reported, (2) observer reported, (3) performance, and (4) patient reported [8]. PRO instruments are based on a report that comes from the patient about the status of his/her health condition without amendment or interpretation by a healthcare professional or anyone else [8]. The focus in this chapter is on PRO instruments developed for use with AYA cancer patients and survivors. We are particularly interested in self-report tools, as these are generally considered to be the preferred method for assessing a patient’s experience of a construct [11].

30.1.4 Generic Versus Cancer-Specific PRO Instruments

PRO instruments that measure HRQL can be generic or condition/disease specific. Generic instruments are those designed for use across many types of diseases, treatments, and populations [12]. Such broad-based tools can lack content validity for particular patient populations, e.g., fail to measure issues that matter [12]. Content validity is the measurement property that assesses whether items are comprehensive and adequately reflect the patient’s perspective for the concept of interest (COI) [13]. A range of generic instruments have been used with AYA cancer patients [14]. For example, in the younger cohort of AYA, our team conducted a systematic literature review valid through May 2011 and found that ten generic HRQL instruments had been used in 148 publications involving patients and survivors up to 25 years of age [14]. In the 148 publications, the most common measure used was the Pediatric Quality of Life Inventory (PedsQL) [15], which appeared in 58 publications [14]. The PedsQL is a 23-item PRO instrument that measures health problems within the following four domains: physical, emotional, social and school function [15]. The Health Utilities Index (HUI) [16, 17] (used in 26 publications) and the Child Health Questionnaire (CHQ) [18] (used in 25 publications) were the second and third most common generic HRQL instruments [14].

Another approach is the use of cancer-specific PRO instruments that were designed for use with various cancer subtypes. Since disease-specific PRO instruments address aspects specific to one disease (e.g., cancer), they may be more responsive to changes in health status [19]. The most common examples of such scales for cancer patients include the Functional Assessment of Cancer Therapy Scale (FACT-G) [20] and The European Organisation for Research and Treatment of Cancer Quality of Life Questionnaire (EORTC QLQ-C30) [21]. Both of these instruments, which were designed for adults (age range 27–89), also include a suite of scales for particular cancer subtypes. The FACT-G measures a range of domains, including physical, social, emotional, and functional well-being and relationship with doctor [20]. There are separate versions for certain cancer types prevalent among AYA (e.g., lymphoma, leukemia, and central nervous system (CNS) tumors) [20]. The EORTC QLQ-C30 measures a range of constructs, including function (physical, role, cognitive, emotional, and social), symptoms (fatigue, pain, and nausea/vomiting), global health, and QOL [21].

30.1.5 Longitudinal Follow-Up of QOL

To understand the impact of childhood cancer on HRQOL, a longitudinal frame of reference is often necessary [22]. HRQOL effects may change substantially throughout the course of the illness. Unfortunately, our systematic review primarily identified cross-sectional studies, which cannot detect the effects of illness that evolve with time. The QOL for AYA patients can change dramatically during their treatment course. For example, teenagers with Hodgkin disease had a significant improvement in their PedsQL of more than ten points when compared between the radiation and chemotherapy phase of treatment, which further improved off treatment [23]. In general, only short-term follow-up studies have been done in these patients, with long-term studies sorely needed. Future research should seek to incorporate longitudinal assessments of HRQOL in order to capture the evolving effects of childhood cancer for AYAs.

30.1.6 The Call for an AYA Cancer-Specific PRO Instrument

In order to include the voice of AYA in the assessment of treatment outcome, well-defined, valid, reliable and responsive PRO instruments that measure the concepts of importance to AYA are needed. The choice of which PRO instrument to use in clinical research or clinical practice is a crucial decision. If the wrong scale is used, it may appear that an effective treatment has little or no benefit. The use of a generic or even cancer-specific scale that was not designed specifically for the AYA population may not provide evidence that a treatment works or may not adequately assess HRQL. The right scale to use in a clinical trial is the one that has content validity for the context of use [8, 13, 24].

There have been recent calls for the development of AYA-specific tools. Nightingale and colleagues reviewed 16 qualitative studies of young adult survivors of childhood cancer and suggested that existing HRQL instruments do not comprehensively cover the concerns of AYA, which they found to include the following six domains: physical, social, psychological, spiritual, fertility/sexual, resilience, and body appearance [25]. Quinn et al. took a different approach by interviewing 30 young adult survivors of childhood cancer to identify content limitations in two adult-onset cancer survivor-specific HRQL instruments, i.e., Quality of Life in Adult Cancer Survivors (QLACS), Quality of Life – Cancer Survivors (QOL-CS), and a generic HRQL instrument (SF-36) [26]. The authors report three areas where content was lacking, including perceived sense of self, relationships, and parenthood [24]. Kuhlthau and colleagues [27] conducted focus groups with 19 AYA survivors of CNS tumors and identified the following seven key survivorship domains: physical health and well-being, mental health and well-being, cognitive functioning, social health and well-being, health behaviors, sexual and reproductive health, and support systems. These authors suggest that there are aspects of HRQL important to patients that are not covered by currently available HRQL tools [27]. The

common theme across these three qualitative studies is that HRQL tools not developed specifically for AYA may lack content validity [25–27].

30.1.7 Study Aim

In order to conceptualize the most important health concerns of AYA patients with cancer and survivors, our team performed a content analysis of HRQL instruments used with AYA to date. Our specific aims were as follows: (1) to identify cancer-specific PRO instruments measuring the HRQL concerns of AYA and (2) to perform a content analysis that involved coding and categorizing the items of each identified PRO instrument. Our overall goal was to develop a preliminary conceptual framework of HRQL content deemed important to AYA by PRO instrument developers.

30.2 Methods

We aimed to identify self-report cancer-specific PRO instruments for AYA, which were available in English and have published evidence of a development and/or validation process. We used the findings from two previous systematic reviews [14, 28] and performed an additional literature search of our own. In the first review, Anthony et al. sought to identify generic and cancer-specific PRO instruments that measured HRQL in cancer patients and/or survivors aged up to 25 years. The methods and results are described in detail elsewhere [14]. In the second review, Clinton-McHarg et al. [28] sought to identify cancer-specific multidimensional PRO instruments that measure psychosocial outcomes, including HRQL, in AYA cancer survivors. Finally, to ensure all possible HRQL instruments were identified, we performed an updated search of Medline, PsycINFO, EMBASE, and CINAHL databases from January 2008 to December 2014 for English language articles, replicating the search strategy outlined by Clinton-McHarg et al. [28]. For all PRO instruments identified, we

obtained a paper copy and transferred the content into an Excel spreadsheet for coding. We then used the content analysis method described by Anthony et al. [14], which classified content according to the broad structure of the Patient-Reported Outcomes Measurement Information Systems (PROMIS), a health framework consisting of domains, subdomains, and identifying concepts [14].

30.3 Results

Our search identified a total of 12 instruments for use with AYA cancer patients and survivors. Table 30.1 outlines the characteristics of each instrument. The age range of participants for whom the scales were developed ranges from 8 to 39 years. The number of items included in each instrument varied ranging from 16 to 90, and the number of domains ranged between four and nine. Seven instruments were designed for use with cancer patients both on and off treatment, and five were designed for childhood cancer survivors.

30.3.1 Brief Description of PRO Instruments for AYA

30.3.1.1 Adolescent Quality of Life Instrument (AQoL)

The AQoL [29, 30] is a 16-item instrument for assessment of HRQL in adolescents with cancer. Items for this measure were generated from previously established QOL instruments and did not involve patient, parent, or expert opinion. High scores on the AQoL are associated with better QoL. Item reduction was conducted using feedback from researchers and by piloting the survey with seven volunteers who highlighted the items of most and least concern. Acceptable reliability was reported in a population of 75 participants with cancer aged 9–20 years [29], and test–retest reliability was adequate with overall scores ranged from 0.75 to 0.90 in three administrations [30]. No other psychometric results for the AQoL were described.

Table 30.1 PRO instruments used to measure HRQL in AYA cancer patients

Measure	Patient group	Versions	Age range (years)	No. items	Domains
Adolescent Quality of Life instrument (AQoL)	On and off treatment	Child and adolescent	9–20	16	Normal activities, social/family interactions, health status, mood, and meaning of being ill
Bone tumor DUX (Bt-DUX)	On and off treatment	Child and young adults	8–25	20	Social, emotional, cosmetic, physical
Cancer Assessment for Young Adults – Testicular (CAYA-T)	On and off treatment	Young adults	18–29	90	Physical, sexual, intrapersonal, social-relational, educational/vocational/ avocational, spiritual
Impact of Cancer for Childhood Cancer Survivors (IOC-CS)	Survivors	Young adults	18–39	45	Life challenges, body/health, talking with parents, personal growth, thinking/memory problems, health literacy, socializing, financial problems
Minneapolis–Manchester Quality of Life Instrument – Adolescent Form (MMQL)	Survivors	Adolescent	13–20	46	Physical functioning, cognitive functioning, psychological functioning, social functioning, body image, outlook on life, intimate relations
Pediatric Functional Assessment of Cancer Therapy – Childhood Brain Tumor Survivor (Peds-FACT-Brs)	Survivors	Adolescent	13–18	37	Physical well-being, emotional well-being and illness experience, social/family well-being, survivor-specific concerns
Pediatric Quality of Life Brain Tumor Module (PedsQL-BT)	On and off treatment	Adolescent	13–18	24	Cognitive problems, pain and hurt, movement and balance, procedural anxiety, nausea, and worry
Pediatric Quality of Life Cancer Module (PedsQL-C)	On and off treatment	Adolescent	13–18	27	Pain and hurt, nausea, procedural anxiety, treatment anxiety, worry, cognitive problems, perceived physical appearance, communication
Quality of Life – Cancer Survivors (QOL-CS)	Survivors	Adolescents and young adults	16–29	41	Physical well-being, psychological well-being (distress and fear), social well-being, spiritual well-being
Quality of Life for Children with Cancer Scale (QOLCC)	On and off treatment	Adolescent	13–18	34	Physical function, psychological function, peer/school function, treatment/disease symptoms, cognitive function, plus 2 subscales of communication and understanding
Quality of Life in Children and Adolescents with Cancer (PEDQOL)	Survivors	Child	8–18	34	Physical functioning, autonomy, emotional functioning, cognitive functioning, social functioning peers/family, body image
Perceived Illness Experience Scale (PIE)	On and off treatment	Child and young adults	8–24	34	Interference with activity, disclosure of illness, school/work, peer rejection, parental behavior, manipulation, preoccupation with illness, treatment, physical appearance

30.3.1.2 Bone Tumor DUX (Bt-DUX)

The Bt-DUX [31] is a 20-item HRQL measure for children and young adults aged 8–25 years with malignant bone tumors. The Bt-DUX was created from the generic DUX-25 QOL questionnaire, which is a short form of the Dutch Children TNO-AZL Quality of Life Questionnaire. The DUX-25 contains 25 items that measure four domains, i.e., emotional, social, familiar, and physical. Ten items for the Bt-DUX were taken directly from the DUX-25, with the remaining items generated from interviews with ten patients and four healthcare experts. Item reduction involved input from four experts. Psychometric validation revealed good internal consistency reliability for all domains and the total score (Cronbach's $\alpha \geq 0.73$).

30.3.1.3 Cancer Assessment for Young Adults: Testicular (CAYA-T)

The CAYA-T [32] is a 90-item measure used to assess HRQL in young adults with testicular cancer, aged 18–29 years. Items for this measure were generated from a literature review, 21 patient interviews, and input from healthcare providers. Items were refined according to participant feedback and clinical applicability. A modern psychometric approach called Rasch Measurement Theory (RMT) analysis was used for item reduction to ensure that the observed data fit the responses of the predicted Rasch model. Psychometric validation showed adequate internal consistency reliability and test–retest reliability, with a Cronbach's $\alpha \geq 0.70$ reported for all scales, and intraclass correlation coefficients that ranged from 0.49 to 0.91.

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

The IOC-CS [33, 34] is a 45-item HRQL instrument for childhood cancer survivors aged 18–39 years. Items were generated through 64 patient interviews and refined in a focus group with 13 healthcare professionals and researchers and 17 patient advocates. Item reduction involved cognitive interviews with 13 young adult survivors

and factor analysis. Higher scores on this instrument indicate greater impact. The IOC-CS evidenced adequate internal consistency reliability, with Cronbach's $\alpha \geq 0.70$ for all scales. Test–retest reliability from 136 respondents was good, with an overall ICC of 0.75 [34]. The IOC-CS was able to differentiate between cancer types.

30.3.1.5 Minneapolis–Manchester Quality of Life Instrument: Adolescent Form (MMQL)

The MMQL-Adolescent Form [35, 36] is a 46-item HRQL instrument for adolescent survivors of cancer aged 13–20 years. Item generation involved input from patients (focus group), parents, and expert healthcare professionals. Higher scores on the MMQL indicate better HRQL. A psychometric analysis of the MMQL-Adolescent Form showed adequate overall internal consistency reliability (Cronbach's $\alpha = 0.92$, range 0.67–0.89), with a test–retest reliability of 0.71 (ranged from 0.60 to 0.90) [35].

30.3.1.6 Pediatric Functional Assessment of Cancer Therapy: Childhood Brain Tumor Survivor (Peds-FACT-Brs)

The Peds-FACT-Brs [37] is a 37-item HRQL instrument for survivors of brain tumors. Item generation involved interviews with 20 survivors, 20 parents, 5 clinicians, and 7 teachers. Input from clinicians, QOL researchers, and children aged 7–15 years was used to refine a set of items, and the instrument was tested in 46 brain tumor survivors. RMT analysis was used to ensure that all items within the domains could be scaled together. Adequate internal consistency reliability was reported for three of four domains (Cronbach's α ranged from 0.51 to 0.82). No further psychometric analyses were reported.

30.3.1.7 Pediatric Quality of Life Brain Tumor Module (PedsQL™-BT)

The PedsQL™-BT [38] is a 24-item instrument that measures HRQL in children aged 2–18 years on or off treatment for a brain tumor.

Item generation for this instrument was developed from focus groups with patients, parents, and healthcare professionals. This module is identical to the PedsQL™ Cancer Module in the layout and instructions. Higher scores indicate better HRQL. A psychometric analysis of the PedsQL™-BT in adolescents aged 13–18 years showed adequate internal consistency reliability (Cronbach's $\alpha \geq 0.70$, ranged from 0.69 to 0.93).

30.3.1.8 Pediatric Quality of Life Cancer Module (PedsQL™-C)

The PedsQL™-C [39] is a 27-item instrument of HRQL for children aged 2–18 years on or off treatment for cancer. The PedsQL™-C was derived from an earlier instrument called the Pediatric Cancer Quality of Life Inventory (PCQL) [40–42]. Item generation and reduction techniques for the PCQL are described by the authors; however it is unclear how these data were used to develop the eight domains that comprise the PedsQL™-C. Specifically, it is not clear whether literature review, patient interviews, or parent input were used to develop the PedsQL™-C. Higher scores on this instrument indicate better HRQL. Psychometric analysis for the PedsQL™-C demonstrated adequate internal consistency reliability for all domains (Cronbach's $\alpha \geq 0.70$) in individuals aged 13–18 years. Construct validity was also established [43].

30.3.1.9 Quality of Life: Cancer Survivors (QOL-CS)

The QOL-CS [44] is a 41-item HRQL instrument for cancer survivors aged 16–29 years. Items for the QOL-CS were derived from a literature review and qualitative interviews with five cancer survivors. The authors do not report how many participants were young adults, and thus it is unclear whether there was any input from this patient age group. Factor analysis was conducted to inform item reduction. Other psychometric analysis revealed adequate internal consistency reliability, with Cronbach's $\alpha \geq 0.76$ for five of six scales (with the exception of the distress scale with a Cronbach's $\alpha = 0.54$), and test–retest reliability ($r \geq 0.81$) high for all domains. Zebrack

and colleagues [45] later undertook a validation study of the QOL-CS in a childhood cancer survivor population.

30.3.1.10 Quality of Life for Children with Cancer Scale (QOLCC)

The QOLCC [46–48] is a 34-item HRQL instrument for adolescents aged 13–18 years on or off treatment. Items were generated through a literature review and interviews with patients and their caregivers. Higher scores represent poorer HRQL. Psychometric analysis revealed adequate internal consistency reliability, with Cronbach's $\alpha \geq 0.74$ for four of five domains. The QOLCC appears to differentiate well between patients on versus off treatment in three of five domains.

30.3.1.11 Quality of Life in Children and Adolescents with Cancer (PEDQOL)

The PEDQOL [49] is a 34-item HRQL measure for children aged 8–18 years who have completed cancer treatment. Items were generated from existing HRQL measures for children and expert opinion. Item reduction involved a factor analysis. Further psychometric analysis highlighted problems with internal consistency reliability, i.e., Cronbach's $\alpha \leq 0.64$ for six of seven domains. This instrument does not appear to discriminate well between children with cancer and healthy controls.

30.3.1.12 Perceived Illness Experience Scale (PIE)

The PIE Scale [50] is a 34-item measure of perceived illness experience in children with cancer and long-term survivors aged 8–24 years. Item generation for the PIE involved input from 15 children and adolescents who had undergone or recently completed cancer treatment. Formal item reduction strategies were not described. Higher scores on the PIE indicate more negative illness experience. Psychometric analysis highlighted problems with internal consistency reliability, with Cronbach's $\alpha \leq 0.68$ for six of nine domains. Test–retest reliability was acceptable for the total score ($r = 0.92$).

30.3.2 Content Analysis

The 12 instruments reviewed above provided a total of 489 items that we included in our concept sort. A total of 71 items (14.5%) were deemed to be a determinant, rather than an outcome (e.g., family social support or techniques for coping with illness). The remaining 418 items were classified as outcomes and were assigned a major domain, a subdomain, and (if relevant) an identifying concept. A total of six major domains were identified as follows: Psychological (207 items), Social (88 items), Physical (76 items), General (23 items), Sexual (18 items), and Spiritual (6). We also identified 21 subdomains and 51 unique health concepts (see Fig. 30.2).

Of the 12 identified instruments, three (i.e., CAYA-T [32], IOC-CS [34], and QOL-CS [44]) had content that covered all six domains of our working framework. Two domains were measured by only a few instruments (e.g., spirituality and sexuality). Some subdomains were included in most instruments (e.g., emotional distress), while others were rare and were assessed by only a few instruments (e.g., behavior). Of the 51 unique health constructs pertaining to AYA cancer patients and survivors, the most commonly measured identifying concepts were anxiety/fear (22 items), worry (21 items), relationships with people (21 items), and relationships with peers (20 items).

An important difference in content was noted between the various PRO instruments designed for adolescents versus young adult patients and survivors. Specifically, the pediatric tools relevant to adolescents lacked items to measure spirituality, goal setting/future plans, and sexual and reproductive health. The PRO instruments designed specifically for AYA patients and survivors, on the other hand, were more likely to include the breadth of AYA-specific concerns.

30.4 Discussion

Though initially designed for use in academic and industry research, PRO instruments are increasingly being used in clinical care, patient/consumer education, benchmarking, and quality

improvement. Such data facilitate comparative effectiveness research, inform discussions with regulatory bodies, and support an evidence-based approach to treatment [19, 51, 52]. It is thus important that clinically meaningful and psychometrically sound AYA-specific PRO instruments are available.

In order to carefully assess HRQL in AYA cancer patients, reliable, valid, and responsive PRO instruments are needed [8]. Best practice guidelines for PRO instrument development (e.g., those outlined by the USA FDA [8]) suggest that the combination of a literature review, qualitative interviews, and expert opinion together optimize the development of a comprehensive PRO instrument. This chapter outlined 12 unique PRO instruments designed to measure HRQL for adolescent and/or young adults. We found that interviews with AYA were part of the development in most of the identified instruments, with between 5 and 64 patients involved. Exceptions were the AQoL [29, 30] and PEDQOL [49], which did not involve any patient input, but were instead developed from existing HRQL instruments.

Most of the current PRO instruments were designed using a Classical Test Theory approach (CTT) [53]. Exceptions identified are the PedsFACT-Brs [37], and the CAYA-T [32], which used RMT analysis, a modern psychometric method. Although CTT methods are widely used, they have limitations that have important consequences for the use of PRO instruments. These limitations include the following: (1) data generated are ordinal rather than interval; (2) scores for people and samples are scale dependent; (3) scale properties, such as reliability and validity, are sample dependent; and (4) data are suitable for group studies rather than individual patient assessment. The increasingly popular use of modern psychometric approaches in scale design offers certain advantages, including the possibility for item banking, scale equating, computerized scale administration, and methods for handling missing data [54, 55]. A modern psychometric approach can also provide scales that can be used with individual patients in clinical practice.

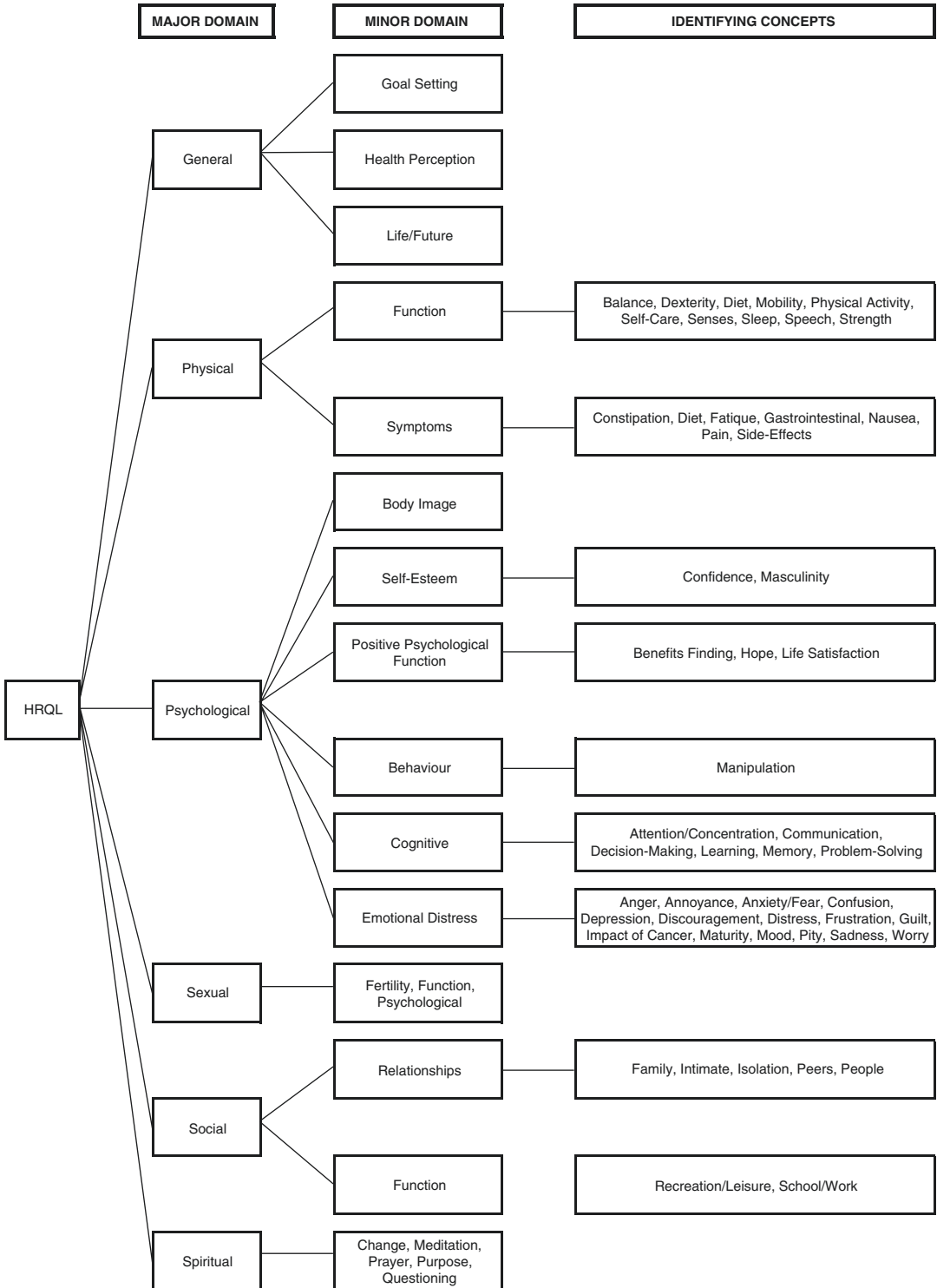


Fig. 30.2 Preliminary conceptual framework of HRQL in AYA

The AYA period of life is a unique stage, characterized by changes in many areas of development [1, 2]. AYA are faced with multiple transitions in this life stage, and challenges to this transition period arise as a result of having a cancer diagnosis [4]. Our team found that most of the identified HRQL instruments were developed for pediatric patients (that included adolescents), rather than AYA specifically, thus limiting their applicability to AYA as a distinct group. The pediatric measures fail to address some of the concepts important to AYA identified in recent qualitative studies [25, 27], including spirituality, sexual and reproductive health, goal setting, and body appearance. Newer measures such as the QOL-CS [44], IOC-CS [33, 34], and CAYA-T [32] were designed with AYA patients and cover their unique concerns, such as sexual and spiritual health needs. As these various PRO instruments are taken up and used in longitudinal studies and in clinical trials with AYA patients and survivors, we will begin to understand and interpret what the scores mean and to identify clinically important change.

Our team suggests that in choosing a PRO instrument to measure HRQL in AYA, one must consider the developers' approach (traditional versus modern) and adherence to international guidelines for PRO instrument development and validation. In addition, choosing the most appropriate PRO instrument for use in clinical research and/or practice is crucial, as results generated from an inappropriate scale may skew or provide false results, i.e., it may appear that an effective treatment has little or no benefit. Given the differences that exist between the 12 measures identified in our study, we highly recommend that, in addition to considering how an instrument was developed and validated, one must also closely consider the relevance of the content within the instrument in relation to the research question or clinical need. Finally, given that the conceptual framework of HRQL content, which we developed from AYA concerns by PRO instrument developers to date, is preliminary, future research in scale development for AYA cancer patients could build on this framework and enhance the validity of their importance to AYA.

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