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Systematic review: Measurement properties of patient reported outcome measures evaluated with childhood brain tumor survivors or other acquired brain injury

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Abstract

Background

Survivors of childhood brain tumors or other acquired brain injury (ABI) are at risk of poor health-related quality of life (HRQoL); its valid and reliable assessment is essential to evaluate the effect of their illness on their lives. The aim of this review was to critically appraise psychometric properties of patient-reported outcome measures (PROMs) of HRQoL for these children, to be able to make informed decisions around selection of the most suitable PROM for use in clinical practice.

Methods

We searched MEDLINE, EMBASE, and PsycINFO for studies evaluating measurement properties of HRQoL PROMs in children treated for brain tumors or other ABI. Methodological quality of relevant studies was evaluated using the COSMIN checklist.

Results

Eight papers reported measurement properties of four questionnaires: Health Utilities Index (HUI), PedsQL Core and Brain Tumor Modules, and Child and Family Follow-up Survey (CFFS). Only the CFFS had evidence of content and structural validity. It also demonstrated good internal consistency whereas both PedsQL modules had conflicting evidence regarding this. Conflicting evidence regarding test-retest reliability was reported for HUI and PedsQL Core Module only. Evidence of measurement error/precision was favorable for HUI and CFFS and absent for both PedsQL modules. All four PROMs had some evidence of construct validity/hypothesis testing but no evidence of responsiveness to change.

Conclusions

Valid and reliable assessment is essential to evaluate impact of ABI on young lives. However, measurement properties of PROMs evaluating HRQoL appropriate for this population require further evaluation, specifically construct validity, internal consistency, and responsiveness to change.

Keywords: systematic review, patient-reported outcomes, acquired brain injury, brain tumor, children

Introduction

One child in every 600 will develop some form of cancer by 16 years of age[1] and around 20% to 27% of these children will have a brain tumor[2]. Currently, 65.4% of children diagnosed with a brain tumor in Europe from 1999-2007 are reported to survive 5 or more years from diagnosis[3] and the majority should have prolonged survival and become adults. They often have multiple impairments and reduced health-related quality of life (HRQoL)[4-8]. Approximately 62% will be left with a life-altering long-term disability[9] comparable to the life-changing sequelae of severe traumatic or other acquired childhood brain injuries (ABI). ABI is post-natal injury to the brain that is sudden in onset and may be the result of head trauma or non-traumatic, following meningitis, stroke, metabolic derangement, sickle cell disease, or a brain tumor.

In children aged less than 16 years, the incidence of hospitalization for traumatic brain injury (TBI) has been reported to be between 280 and 500 per 100,000. This implies that the total number of children admitted to hospital for TBI per annum in the UK is at least 35,000. Of these, about 2,000 (5.7%) will have severe TBI, 3,000 (8.6%) moderate TBI, and 30,000 (85.7%) mild TBI. In addition, the total number of children who sustain non-traumatic coma associated with severe or moderate encephalopathy is around 4,000 per year[10]. Also, the Central Brain Tumor Registry of the United States reported an incidence rate of newly diagnosed cases of brain tumor in children to be 5.54 per 100,000, equating to 4,500 new cases annually[11] and the overall annual incidence of childhood stroke has been estimated to be around 1.2 to 13 cases per 100,000 children under the age of 18 years[12].

In the context of delivery of clinical care, doctors vary in their ability to explore, elicit, and respond to information about HRQoL[13] and discussion of the emotional, social, and cognitive issues impacting HRQoL after ABI or childhood cancer does not routinely take place in clinic consultations[14]. In addition, children and parents are often reluctant to raise psychosocial issues at clinic appointments[15,16] which they perceive to be more focused on medical issues such as monitoring tumor status and its response to anti-tumor treatments or complications of other types of ABI.

PROMs measure a patient's health status or health-related quality of life at a single point in time, and are collected through short, self-completed questionnaires[17] without any third party acting as an intermediary. In the context of clinical research, the use of PROMs, including those assessing HRQoL, has proved to be a practicable means of assessing quality of survival in multicenter treatment trials[18,19]. Individualized use of PROMs in the routine care of children with a long-term illness has the potential to add valuable information about the impact of the disease, inform treatment planning, provide clinicians with timely information about a patient's functional and emotional status and wellbeing[20], and enhance family-clinician communication[21]. This helps clinical staff to deliver care focused on the needs and choices of each individual child and family[22]. Such use of PROMs has been evaluated in large groups of typically developing children, adolescents and adults and in adult patients with cancer[23] and children with other long-term conditions[24-27] but not in child/adolescent survivors of brain tumor or other ABI.

When selecting PROMs for a specific purpose, it is necessary to examine how robust (valid and reliable) is the measurement of HRQoL produced by such questionnaires. A number of methodological approaches are available to determine aspects of reliability and validity[28]. The aim of the present systematic review was to critically appraise the psychometric properties of patient-reported outcome measures (PROMs) of HRQoL for these children, in order to be able to make informed decisions around selection of the most suitable PROM for use in clinical practice.

Materials and Methods

Systematic Review

We undertook a systematic review of published evidence relating to the measurement properties of PROMs in children with brain tumors and other ABI and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement[29]. A protocol was written which specified, a priori, the inclusion criteria and methods to be used. We also used methods recommended for appraising measurement properties and for assessing the methodological quality of papers that evaluate PROMs[30], including the consensus-based standards for the selection of health status measurement instruments (COSMIN) checklist for evaluation of publications [31].

Search strategy

The search strategy was designed by an experienced information specialist (see acknowledgements) in discussion with topic experts (KB, CK, and CM) and an experienced systematic reviewer (JS). Blocks of search terms were combined including variants of 'brain tumor/acquired brain injury', 'child/adolescent', 'patient reported outcome measure', 'psychometric' and the titles of generic PROMs suitable for use in all children or in all children with long-term health conditions, as listed in the most recent systematic review focusing on HRQoL in children with disabilities[32].

MEDLINE, EMBASE, and PsycINFO were searched for studies published from 1992 onwards in peer-reviewed journals whose purpose was to evaluate measurement properties of PROMs. An example from MEDLINE of this search strategy is shown in Appendix 1. The electronic searches were completed on 7th February 2017 and updated on 28th May 2019. Publication details were uploaded into an Endnote reference management database and duplicates removed. Backwards citation chasing (one generation) from the reference lists of included papers was conducted by CM. Forwards citation chasing for each included study using all databases in the Web of Science cited reference search resource was conducted by SH.

Inclusion and exclusion criteria

We sought published papers reporting evaluations of the measurement properties of multi-dimensional child (aged 5 to 18 years) self-report and/or parent-proxy report PROMs assessing health and wellbeing in children receiving care either for a brain tumor or other ABI of any kind (rather than for specific types of brain tumors or ABI). Evaluation of an English language version of the PROM was a requirement for inclusion. Studies in which only part of the sample were eligible for review were included only if psychometric analyses had been conducted on the eligible sub-groups within the sample. Instruments administered by an interviewer and single domain-specific questionnaires (e.g. to assess only depression, fatigue, or pain) were excluded.

Study selection

An inclusion/exclusion criteria decision chart was used to aid the selection of articles likely to yield relevant results from their titles and abstracts. The use of this chart was piloted by SH and KB who screened the first 10 articles together to test agreement over inclusion of articles. All remaining titles and abstracts were screened in batches of 40 by SH and, independently, by KB. The evaluations of each batch of 40 by the two reviewers were then compared and any disagreements discussed and resolved. Full texts were then retrieved from this list of potential studies by SH. KB then checked the list of included and excluded studies to confirm agreement. Disagreements were discussed and resolved between the reviewers.

Data extraction, appraisal, and synthesis of included studies

Descriptive characteristics of included studies and measurement properties of the PROMs were extracted by SH. These extracted data were checked by KB and the final extracted data set was agreed in discussion with CM. The criteria of Fitzpatrick et al. (1998)[33] were adopted for evaluation of the patient-based outcome measures within the extracted data set.

The COSMIN Risk of Bias checklist of Consensus-based standards for the selection of health status measurement instruments (COSMIN) was used to assess the methodological quality of the included studies. The checklist is comprised of 12 boxes which together cover three domains: content validity, internal structure, and remaining measurement properties – namely reliability, measurement error, criterion validity, hypothesis testing for construct validity, and responsiveness to change[31]. Ten of the 12 boxes can be used to assess whether a study meets standards for good methodological quality and 9 of them contain standards for the included measurement properties. These are each scored on a four-point rating scale of the way in which each measurement property was assessed.

All of the above properties were assessed (Table 1) excepting cross-cultural validity which was not relevant as our search only included English language reports. Criterion validity was not applicable as in the case of HRQoL there is no criterion against which HRQoL measures can be judged (except for the purpose of comparing long versions of an instrument and shortened forms of the same instrument).

An overall score for the methodological quality of a study was determined by CM for each measurement property separately as a single rating[34], arrived at by taking the lowest rating of any of the items in a box[35]. The review team then considered the evidence for each PROM and summarized in a single rating for each measurement property following methods commonly used for presentation of findings against the COSMIN criteria (Table 2). From these ratings conclusions were drawn on the extent to which each PROM could be considered robust for measuring HRQoL in children treated for brain tumors or other ABI.

Table 1 about here**.**

Table 2 about here

Results

The electronic searches resulted in 472 articles after the removal of duplicates. Of these, 374 were excluded leaving 98 potentially relevant studies whose full text articles were retrieved. Screening of these led to the exclusion of a further 90 papers leaving eight studies remaining for evaluation (Fig. 1). Backwards citation chasing identified two potentially relevant papers and forward citation chasing identified six potentially relevant papers, all of which were subsequently excluded due to inappropriate population (n=4), inappropriate instrument (n=3), or lack of relevant data (n=1).

Four self-report and/or parent-proxy report PROMs – the Health Utilities Index (HUI), the Pediatric Quality of Life Inventory Core Module (PedsQL), the PedsQL Brain Tumor Module, and the Child and Family Follow-Up Survey (CFFS) - were evaluated and appraised in the eight included studies (Tables 3&4) and these are briefly described here.

The HUI and PedsQL are generic measures of HRQoL whereas the PedsQL Brain Tumor Module and the CFFS are disease-specific. The HUI is a rating scale used to measure general health status with one question relating to HRQoL. Health utility values are commonly produced using HUI as a component

of the quality-adjusted life years (QALY) calculation used in population health and economics. Answers to 15 questions about health state, scored at 3 to 6 health status levels, can be grouped in two different ways to produce either HUI2 or HUI3 scores across 7 or 8 'attributes' of health. HUI3, for example, groups health status levels to create attribute scores for Vision, Hearing, Speech, Ambulation, Dexterity, Emotion, Cognition, and Pain.

The PedsQL is a measure of HRQoL with 23 questions across four core scales: Physical, Emotional, Social, and School. The 24-item PedsQL Brain Tumor Module was designed to measure HRQoL in children undergoing treatment for a brain tumor. The questions are divided between six subscales: Cognitive problems, pain and hurt, movement and balance, procedural anxiety, nausea, and worry.

The CFFS was developed as a parent report measure to monitor needs and outcomes of children and youth with acquired brain injury and their families. It consists of five sections with a total of 71 closed or open-ended questions. Section 1 asks about the child's physical and emotional health and well-being, primary way of moving around and communicating, and medical problems or hospitalizations within the last year or since leaving the rehabilitation program. Section 2 includes the Child and Adolescent Scale of Participation (CASP) and three subsequent open-ended questions about equipment, modifications or strategies that are used to promote the child's participation. Section 3 includes the Child and Adolescent Factors Inventory (CAFI) and Child and Adolescent Scale of Environment (CASE) and a question about health or medical restrictions on the child's daily activities. Section 4 enquires about the child's current educational placement, rehabilitation and health services, satisfaction with services, the family's quality of life, and current services and needs. Finally, Section 5 seeks suggestions to improve services at the program from where the child was discharged to better address the needs of the child and family and additional information that was not addressed in the CFFS.

Completion time for the HUI and the PedsQL (core or brain tumor module) is about 5 minutes and for the CFFS about 30 minutes. Child self-report is available from 5 years old for the PedsQL

modules and from 12 years old for the HUI while the CFFS is available as parent-report only (Table 4). None of the studies had assessed all psychometric properties of the PROM in question.

Content validity: this had been assessed only for the CFFS and in this case the evidence for its validity was good.

Internal structure: only the CFFS had been assessed for evidence of structural validity and there was good evidence that it possessed this property. Internal consistency had been evaluated for the CFFS (good evidence) and for the PedsQL Core and PedsQL Brain Tumor Modules (equivocal evidence) but not for the HUI (Supplementary Table S1and Table 5).

Other measurement properties: evidence for test/re-test reliability and proxy reliability was available but conflicting for the HUI and PedsQL Core module and absent for the PedsQL Brain Tumor module and the CFFS. Favorable evidence of precision was available for the HUI but absent for the PedsQL Core and Brain Tumor Modules or the CFFS. Favorable evidence of hypothesis testing/construct validity was available for all measures. There was no evidence of responsiveness to change over time for any of the PROMs.

The methodological quality of the included studies varied from adequate to very good (Supplementary Table S2). The CFFS had had the most measurement properties evaluated and these studies were of high quality (Supplementary Table S2).

Table 3 about here

Table 4 about here

Table 5 about here

Discussion

This is the first systematic review of evaluations of the psychometric properties of PROMs in survivors of childhood brain tumors and other ABI of childhood. It identified only eight papers describing four

PROMs with relevant information about their measurement properties in children treated for brain tumors or ABI. Some evidence in favor of each instrument was found with respect to those properties that had been examined but caution is needed with respect to those properties that have not been evaluated: notably content and structural validity for the HUI and the PedsQL; test/retest reliability and precision / measurement error for the PedsQL; and responsiveness to change over time for all measures. In contrast to the HUI and the CFFS, the self-report versions of the two PedsQL modules had been specifically designed for the pediatric age group.

The PedsQL Core Module has previously been reported, in the setting of orthopedic, and rheumatology clinics, to be sensitive to increasing disease severity, responsive to clinical change over time, and to demonstrate impact on clinical decision-making resulting in increases in HRQoL[36]. The developer of the PedsQL has recommended it as a screening instrument to use in conjunction with disease-specific modules to target symptoms for interventions[37].

Our strict selection criteria did not reveal any longitudinal/follow-up studies in which responsiveness to change may have been assessed incidentally but the present study does not rule out their existence. Assessing the size of meaningful change above measurement error of the scores from PROMs is desperately needed from further research. It therefore behoves the user to design validation steps when adopting one of the questionnaires for clinical or research use to plug this evidence gap, for example when interpreting studies that have used these questionnaires to measure change.

The validity of the use of a PROM to communicate with families and better focus their care to improve their HRQoL depends on the method by which it was developed. This method of development of a PROM is to an extent separate from its measurement properties although may be reflected in measures of content validity. These methods have been highly variable and are often not clearly specified. Thus, there would be merit in discussing further with survivors of brain tumor or other ABI and their caregivers the salience and relevance of the individual questions within questionnaires and relying on responses to individual questions rather than questionnaire scores as a means to enhance communication between care-providers and service users about HRQoL. Such discussion with survivors of brain tumors or other ABI in

childhood would also help to identify whether or not there is a need to develop a condition-specific PROM for use in child and adult survivors of brain tumor or other ABI in childhood.

Two systematic reviews of HRQoL measures in children with long-term conditions other than ABI seem to have particular relevance to selection for use in child survivors of brain tumors or other ABI. The first conducted was a systematic review of the psychometric properties of measures for use in children with neurodisability[32,38]. It found evidence relating to measurement properties of seven generic PROMs (The Child Health and Illness Profile, The Child Health Questionnaire, the Child Quality of Life questionnaire, KIDSCREEN, the PedsQL, the Student Life Satisfaction Scales, and the Youth Quality of Life Instrument), two chronic-generic PROMs (the DISABKIDS and the Neurology Quality of Life Measurement System) and three preference-based measures (HUI, the EQ-5D-Y, and the Comprehensive Health Status Classification system – Preschool). In the instance of preference-based measures, they noted a dearth of evidence of face, content, and construct validity, or test-retest reliability and for all measures, a lack of evidence for responsiveness and measurement error.

The second systematic review was of PROMs of 'cancer-specific' HRQoL measures for use in children with cancer and identified nine measures for proxy completion, of which six had parallel measures for self-completion by children[39]. This review did not consider generic scales that had been applied in children with cancer (e.g. the PedsQL Core Module) but did note that the MMQL-UK child and parent versions have been validated as generic measures of QoL that can be used with healthy children and those with chronic conditions other than cancer. Adequate detail about how questionnaire items were generated from qualitative interviews was provided for only four questionnaires and most did not combine this with literature review or expert opinion. Some questionnaires required further psychometric evaluation before they could be recommended leaving just five recommendable measures: the Miami Pediatric Quality of Life Questionnaire (MPQS), the Minneapolis-Manchester Quality of Life Instrument (MMQL), the PedsQL Cancer Module, the Pediatric Functional Assessment of Cancer Therapy-Childhood: Brain Tumor Survivor (PFACT-BT), and the Pediatric Oncology Quality of Life Scale (POQOLS) (ibid.). These questionnaires may be suitable for clinical use in children receiving care

for a brain tumor or other ABI but, with the exception of the PFACT-BT, their measurement properties and performance have not been evaluated in either of these groups. The PFACT-BT is administered by an interviewer. This was an exclusion criterion for the present review and unfortunately also greatly limits the applicability of this measure.

Advantages of self-administered questionnaires include the reduction in burden associated with respondents of being able to answer at their own convenience and in their own time, the obviation of any need for a trained administrator, and, when done on line, the avoidance of transcription errors and greater efficiency and of data being entered at the moment that it is self-administered. However, the development of the questionnaires needs to be robust since measurement error may be made more likely by the absence of a trained administrator, if questions are poorly worded or formatted.

However, other considerations relating to the constraints of health care systems, including time and resources, need to be taken into account. Not all the PROMs we identified are suitable for systematic use in an outpatient clinical health care setting. PROMs with costly licensing fees are not feasible to use in public health care systems where funds are limited. Also PROMs which are lengthy to discuss will not be adopted due to clinical time constraints. PROMs also need to be relevant and suitable for follow-up consultations after treatment has ended. The CFFS appears to be the most thoroughly developed and comprehensive measure in this population but it is lengthy, at 71 questions, and the absence of any self-report version is a limitation of its use as a measure of quality of life. For these reasons, the PedsQL – Core Module, which is being widely used in childhood cancer research, may be the most suitable PROM for use in a clinical setting, notwithstanding the gaps in evidence regarding some of its psychometric properties.

Strengths of the present review include a comprehensive and systematic search strategy, use of standard criteria for the evaluation of the measurement properties of each PROM, and use of defined criteria to measure the quality of the studies that had been undertaken to assess these properties in participants with brain tumors or acquired brain injury in childhood. Synthesis of the findings of this review with the findings of previous reviews relating to children with other long-term conditions is also a

strength. The restriction of the systematic review to evaluations of questionnaires in the English language is both a limitation of this study, in that it restricts its relevance to English speaking service users, and a strength in that issues of cross-cultural validity apply to a much smaller extent than would be the case for an evaluation of instruments in more than one language[40].

In summary, both the present systematic review of measurement properties of PROMs when used in child survivors of brain tumors or other ABI and the preceding systematic reviews of PROMs when used in survivors of childhood cancer and in children with neurodisability indicate lack of evidence regarding measurement error or responsiveness to change and, in the case of preference-based measures, lack of evidence of content or construct validity, or test-retest reliability. Factors contributing to this lack of evidence may include the assumption by investigators that psychometric properties shown in healthy populations also apply to survivors of brain tumors, difficulty of accessing study populations of sufficient size to reach reliable conclusions about the validity of measures used, and/or limited awareness of investigators about the importance of validating psychometric properties of those measures.

To conclude, the four PROMs that were identified in our systematic review and a handful of other PROMs identified in previous systematic reviews of child survivors of non-CNS cancers and of children with neuro-disability had some evidence of favorable measurement properties but this was limited and insufficient to enable selection of PROMs suitable for use in survivors of childhood brain tumors or other ABI, particularly for the measurement of change. For communication about HRQoL, the paucity of evidence of content validity in these groups suggests the need for further discussion with these patient groups to inform selection of questions that address their concerns and we are, to that end, currently engaged in a qualitative study of the expressed views of brain tumor survivors. In the meantime there is clearly a need for studies that evaluate the measurement properties of those generic PROMs of HRQoL when used in these patients whether the purpose is to inform the care of individuals or to describe the HRQoL of groups of patients.

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Figure Legend

Figure 1. PRISMA flowchart for the identification and selection of studies evaluating psychometric properties of PROMs in children treated for brain tumors or acquired brain injury.

PRISMA=Preferred Reporting Items for Systematic Reviews and Meta-Analyses; PROMs=Patient-reported outcome measures