

A systematic review of patient reported outcome measures (PROMs) used in child and adolescent burn research

Griffiths, C^{a*}, Armstrong-James, L^a, White, P^b, Rumsey, N^a, Pleat, J., & Harcourt, D^a

^a Centre for Appearance Research (CAR), University of the West of England, Bristol.

^b Department of Engineering Design and Mathematics, University of the West of England, Bristol.

^c Frenchay Burns Unit, North Bristol NHS Trust.

* Corresponding author at: Centre for Appearance Research (CAR), Department of Health and Social Sciences, University of the West of England, Frenchay Campus, Coldharbour Lane, Bristol, UK. BS16 1QY. Tel: +44117 32 83947.

Email addresses: catrin.griffiths@uwe.ac.uk (C. Griffiths; Research Associate), Laura2.Armstrong-James@uwe.ac.uk (L. Armstrong-James; PHD Researcher), paul.white@uwe.ac.uk (P. White; Lecturer in Quantitative Research Methods and Statistics), Nichola.rumsey@uwe.ac.uk (N. Rumsey; Co-director of CAR), j.pleat@btinternet.com (J. Pleat), diana2.harcourt@uwe.ac.uk (D. Harcourt; Co-director of CAR).

Abstract (200 words)

Introduction: Patient reported outcome measures (PROMs) can identify important information about patient needs and therapeutic progress. The aim of this review was to identify the PROMs that are being used in child and adolescent burn care and to determine the quality of such scales.

Methods: Computerised and manual bibliographic searches of Medline, Social Sciences Index, Cinahl, Psychinfo, Psycarticles, AMED, and HAPI, were used to identify English-language articles using English-language PROMs from January 2001 to March 2013. The psychometric quality of the PROMs was assessed.

Results: 23 studies met the entry criteria and identified 32 different PROMs (31 generic, 1 burns-specific). Overall, the psychometric quality of the PROMs was low; only two generic scales (the Perceived Stigmatization Questionnaire and the Social Comfort Scale) and only one burns-specific scale (the Children Burn Outcomes Questionnaire for children aged 5 -18) had psychometric evidence relevant to this population.

Conclusions: The majority of PROMs did not have psychometric evidence for their use with child or adolescent burn patients. To appropriately identify the needs and treatment progress of child and adolescent burn patients, new burns-specific PROMs need to be developed and validated to reflect issues that are of importance to this population.

Keywords: Systematic Review; Patient-Reported Outcome Measures; PROM; Pediatric; Adolescent; Burn

Introduction

Around 52,200 children and adolescents suffer a burn injury in the UK each year [1]. Historically such injuries have resulted in high mortality rates, however with advances in medical and surgical treatment, an increasing number of children and adolescents are living with their injuries.

Irrespective of a person's age at the time of injury, a burn can have a significant psychosocial impact, both for patients and the family members supporting them. Those affected can struggle with coming to terms with the traumatic event that caused their injuries, while changes in physical appearance, loss of independence, pain and restricted physical abilities can also be hard to cope with [2].

Late childhood and adolescence can be a challenging time, during which most young people experience physical, psychological and social changes. The stress of experiencing a burn injury and associated treatment during these developmental stages can add further pressure [3], increasing the likelihood of key developmental processes being disrupted [4]. A recent review found that a considerable proportion of young adults burnt as children still experienced anxiety and affective disorders in adulthood [4]. It is therefore important to identify the needs of children and adolescents with burn injuries, in order to ensure that they receive the most appropriate support, with a view to reducing the likelihood of them experiencing difficulties later on.

The National Burn Care Review (2001) [5] raised awareness of the need for improvements in the provision of care for people affected by burns. As well as highlighting the need for routine audits of mortality and morbidity rates and surgical outcomes, the review noted that a systematic assessment of patients' experiences of their care, treatment, scarring and psychosocial functioning after burns was lacking. The review emphasised that the development of new patient reported outcome measures for this population was a priority. Historically patient needs have been identified through the use of clinician reported measures; however these might miss key aspects of the patient experience. Patient-reported outcome measures (PROMs) are preferable, as unlike clinician reported measures, they provide information about patient experiences and satisfaction with care [6].

PROMs are clearly needed in order to appropriately measure outcomes in paediatric and adolescent burn care. However, careful attention must be given to the development and implementation of these measures. A recent review of the psychosocial consequences of burn scars highlighted the lack of PROMs that have been validated with burns patients [7]. Although PROMS may investigate relevant patient experiences, their reliability (e.g. ability to yield consistent scores over time), validity (e.g. ability to measure what it intends to measure) and responsiveness (e.g. capacity to identify changes in scores over time) cannot be assumed without their psychometric qualities being formally tested with child and adolescent burn patients [8].

When PROMS are used by clinicians and researchers in this and related fields, there has been a tendency to use generic (rather than condition specific) measures to assess patient needs. Although generic PROMs are useful for identifying general health outcomes, they do not capture data relating to experiences that are specific to a particular patient population – in this case, paediatric and adolescent burns patients, and may lack the sensitivity to identify changes resulting from treatment. Rumsey & Harcourt [9] have also argued that there is a need to develop psychometrically robust instruments that can be used to evaluate the psychosocial adjustment of children with visible differences such as burn injuries.

Whilst a number of reviews have summarised the types of outcome measures in research focussing on adults with burn injuries [10, 11, 12]; none of these reviews have assessed the psychometric qualities of these outcome measures and little attention has been given to outcome measures used

in paediatric burn research. Van Baar et al [13] reviewed the functional consequences of experiencing a burn injury in adult and paediatric burn care. Although this review included information about the types of measures used in the studies, the psychometric qualities of the measures were not assessed in relation to paediatric burn patients. Similarly Lawrence et al's review [7] did not assess or report the psychometric quality of the measures used in the studies included in their review. A more recent systematic review of burn scar rating scales assessed the validity, reliability, responsiveness, interpretability and feasibility of these measures [14]. However the findings relate only to scar assessment; this is only one element of burn outcome. In addition, the majority of the scales included in this review were objective ratings, with only the Patient and Observer Scale Assessment Scale (POSAS) [15] providing subjective ratings by patients themselves.

In order to provide essential information to underpin the future choice of PROMs by researchers and clinicians working with children and young people affected by burns, the purpose of this review was firstly, to identify PROMs currently being used in paediatric and adolescent burn research to assess health and well-being (e.g. anxiety, depression, pain, post-traumatic stress disorder, mobility) and secondly, to conduct quality assessments to identify the psychometric properties of these measures.

Methods

The protocol for this review was accepted on the PROSPERO systematic review database (<http://www.crd.york.ac.uk/prospere>) on 8th November 2013 and is reported using the PRISMA checklist for reporting systematic reviews [16] and guidance published by the Cochrane Collaboration [17].

Search strategy

A computer literature search was conducted to identify studies in the burn care literature which have used PROMs with patients with a burn injury. Articles obtained were analysed using the systematic review technique.

Computerised and manual bibliographic searches were conducted using 7 databases (Medline, Social Sciences Index, Cinahl, Psychinfo, Psycharticles, AMED, HAPI). Articles published since the publication of the National Burn Care Review (2001) [5] were considered in order to assess whether the conclusions of this review remain current - thus a publication window of January 2001 – March 2013 was imposed on the search strategy. The overall strategy was to search for articles related to outcomes and/or measures assessing the effects of treatment in burns care. Once the articles had been identified, they were then split according to whether they reported using PROMs with adult or child/adolescent patients. This paper reports those used with children and adolescents; the adult patient PROMs are reported elsewhere [18]. 'Childhood' and 'adolescence' are flexible terms in which the age ranges vary socially and culturally. In the present study 'childhood' was defined with an age range of 8 – 11 years. By the time children reach the age of 8, they are considered to have the skills to be able to express more complex concepts such as their own thoughts and feelings [19]. An upper limit of 11 years was chosen in line with the definition of childhood outlined by the Centres for Disease Control and Prevention [20]. 'Adolescence' was defined as an age range of 12 – 17 years, since this is consistent within most definitions of adolescence [20] and as the authors are reporting studies relating to adults (aged 18 and older) elsewhere. In instances where an article also included patients that were slightly younger or older than the specified age range (e.g. aged 6 or aged 19), authors used the average age of participants in the study as the exclusion criteria (e.g. the mean age of participants had to be between 8 and 17 years).

The search terms used were:

- Scale OR score OR instrument OR research instruments OR questionnaire OR inventory OR survey OR measure OR form OR patient reported outcome measure OR pro OR prom
- AND burn OR burns.

Reference follow-up was conducted to identify relevant articles which were not detected in the online bibliographic search. Unpublished articles were also included to reduce the risk of publication bias in the overall findings.

Articles were screened based on the following criteria:

Inclusion criteria

- Articles which used a PROM/s.
- Articles written in the English language.
- Articles which used an English language PROM.
- Articles which used the PROM with children (aged 8 – 11) or adolescents (aged 12-17) with a burn injury, or the mean age of participants in the study was between 8 and 17.
- Articles in which there is published psychometric evidence of measurement reliability, validity or responsiveness of PROM used in the article.
- Articles that used PROMs with more than one item.
- Articles that were published from January 2001 – March 2013.

Exclusion criteria

- Articles which used instruments that were not patient reported (e.g. parent or clinician reported)
- Articles written in a language other than English
- Articles which used PROMs written in a language other than English.
- Articles which reported data from participants under the age of 8 or over the age of 17 with a burn injury or the mean age of participants in the study was under the age of 8 or over the age of 17.
- Articles which include data on other patient groups in addition to burn patients.
- Articles in which no published psychometric evidence of measurement reliability, validity or responsiveness of the PROM used in the article.
- Articles that used a single item PROM.
- Articles that reported data from patients who have not had a burn injury.
- Articles that had been published before January 2001.

Data extraction procedure

Two reviewers (CG and LA-J) independently extracted data which included study design and the country in which the study was conducted in, participants (e.g. number and characteristics); PROM type and characteristics (e.g. number of items). Unpublished or missing information was requested from the study's corresponding author when necessary. Any discrepancies in extracted data was discussed between reviewers and resolved through consensus and the double checking of papers by both authors.

Quality assessment procedure

The quality of the PROMs used in the studies identified in the systematic review was assessed in relation to their adherence to the international guidelines for the development and validation of health outcome measures. Cano et al (2004) [21] reports a three-stage development and validation process based on the guidelines outlined by the Scientific Advisory Committee of the Medical Outcomes Trust for the development of health outcome measures [22]. This involves a step-by-step process for item generation, item reduction and psychometric evaluation. The first stage involves creating a conceptual model based on the aims of the instrument. A pool of items is then developed to ensure that all important aspects of the health outcomes are considered for the final scale. Initial versions of the measure are developed using three approaches: a literature review, qualitative interviews with patients and expert opinion. The item pool is then evaluated in a small sample of patients to confirm appropriateness, readability, and wording and to determine acceptability and completion time [8]. In stage 2, the measure is evaluated in a larger sample of patients. The final items are then identified according to item redundancy, missing data, endorsement frequencies, factor analysis, and tests of scaling assumptions. In stage 3, the final measure is administered to large samples of the specified population to evaluate the psychometric properties of the instrument, including acceptability, internal consistency reliability, test-retest reliability, validity within scale, validity with other measures, responsiveness and item total correlations [23]. The current review uses these guidelines to assess the quality of the development and validation evidence for each of the reviewed PROMs in relation to children and/or adolescents with a burn injury.

Two review authors (CG, PW) independently assessed the quality of the included articles by considering the presence or absence of the above characteristics in the reported measures. Disagreements over the quality of particular studies were resolved through discussion and the double checking of papers by both authors.

Results

Study selection

Figure 1

Flow diagram of systematic selection of articles in the review

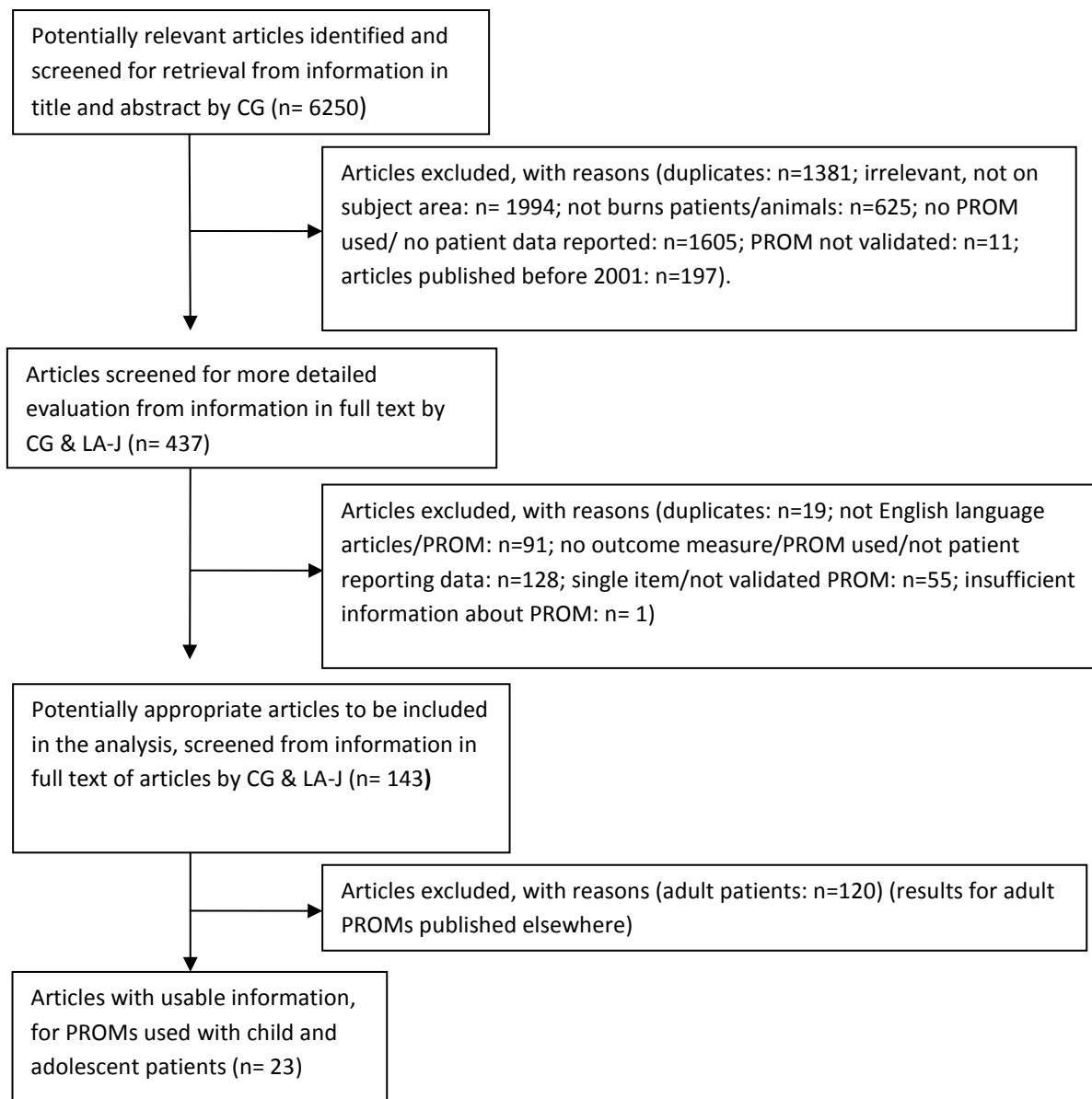


Figure 1 shows the flow chart of the data screening process. A total of 6250 articles were identified. The first author (CG) screened the articles by applying the inclusion/exclusion criteria. Articles were excluded for the following reasons: duplicates (n=1381), irrelevant (n= 1994), not burn patients/animals (n=625), no PROM used/no patient data reported (n=1605), PROM not validated (n=11) and articles published before 2001 (n= 197).

Of the 437 articles that remained, articles were screened by CG and LA-J. Articles were excluded for the following reasons: duplicates (n=19), not an English language article/PROM (n=91), no PROM used/ no patient data reported (n=128), single item /not validated PROM (n=55) and insufficient information about the PROM (n=1).

Of the 143 remaining articles, 120 reported data from adult patients so were excluded (results from these articles are discussed in Griffiths, Harcourt, Rumsey & Pleat, in preparation) [18].

A total of 23 articles met the inclusion criteria of using one or more PROM with children or adolescents with a burn injury, and were included in the review.

Study design

Table 1 lists details of the 32 PROMs used in the 23 articles identified by the systematic search.

The majority of studies were conducted in the USA (n=16), with the remainder being from the UK (n=4), Australia (n=2) and South Africa (n=1). The studies were intervention evaluations (n=8) or cross sectional (n=9) and longitudinal studies (n=6).

The majority of articles (n=19) had less than 100 participants. Only four studies had more than 100 participants, with the largest sample size comprising of 202 young people [24]. Patients completing the PROMs ranged between 6 and 19 years of age, the mean age of participants ranged from 11.2 – 15.1 years old.

Types of measures

Two main types of PROMs were identified: generic and burns-specific. Of the 32 measures reviewed, 31 were generic measures, and only 1 was burns-specific (κ). Table 1 shows the domains (e.g. areas of health) assessed by the reviewed measures.

Generic and burns-specific measures assessed a variety of outcomes. All measures assessed psychological characteristics/outcomes. The most commonly measured domains was appearance (β, C, D, K, N, O, P), self-esteem (A, B, C, D, M, Zb) and social skills/competence (Q, R, S, U, V), followed by anxiety (E, F, G, H), depression (I, J) and quality of life (L, M). The remaining studies each measured broadly one domain: post-traumatic stress disorder (Zf), perceived stigmatisation from others (w), social support (v), attitudes to illness (z), self-control (za), resilience (zb), sense of community alienation (zc), personal development (zd), and personality (ze). Please see Table 1 for further information relating to the subscales in each construct.

The majority of the generic measures were psychometrically validated with children and/or adolescents in the general population, although a few had only been validated with adults (E, H, I, O). Only two of the generic PROMs, the Perceived Stigmatization Questionnaire (w, Lawrence, et al) [25, 26] and the Social Comfort Questionnaire (u, Lawrence, et al) [25, 26] had evidence of validation data with children and adolescents with burn injuries.

Only one measure (the Children Burn Outcomes Questionnaire for children aged 5 -18) (κ, Daltroy et al) [27] was burn-specific and had been validated with adolescents with burns.

Quality assessment

All PROMs included in the review were subjected to a quality assessment based on the psychometric criteria specified in the method. Of the 32 reviewed measures, only three (two generic and one burns-specific) demonstrated any evidence of validity with children or adolescents with burn injuries. The two generic PROMs were the Perceived Stigmatization Questionnaire (PSQ) and the Social Comfort Questionnaire (SCQ) which were originally developed for adult burns patients [25] and then validated in a paediatric and adolescent population (aged 8 - 18) with burns [26]. The burns-specific PROM was the Children Burn Outcomes Questionnaire (CBOQ) for children aged 11 - 18 [27] which was developed specifically for adolescents with a burn injury. The quality assessment of these three measures is shown in table 2.

The PSQ measures stigmatising behaviours from others commonly reported by people with a visible difference such as burn injuries [25]. It has 21 items which require participants to rate how often they experience specific behaviours from others (e.g. *People call me names*) on a 5-point Likert scale (never, almost never, sometimes, often, always). The SCQ measures the extent to which people feel comfortable in social situations [25]. It has 8 items which ask participants to rate the extent to which they think and feel in certain ways about social situations (e.g. *I feel like I don't fit in with other people*). The Likert scales used in the SCQ have the same response categories as the PSQ. Although originally developed for use with adults with a burn injury, the PSQ and the SCQ are generic PROMs (e.g. they are intended to be used in any condition/disease population) and do not specifically refer to a person's burn injury or scar.

The items in the PSQ and the SCQ were originally generated by reviewing published literature and eliciting expert clinician opinion. Patient interviews were not conducted and a conceptual framework was not developed, as per Cano et al's (2004) [21] guidelines when generating instrument items. However, 10 adult burn survivors and 10 clinician experts reviewed drafts of the measures and the items were amended in line with their feedback. Since the original measure was only intended for adults, the opinions of child and adolescent burn patients were not obtained in the measure development phase. In the item reduction phase, items were excluded based on item redundancy, endorsement frequencies, factor analysis and testing scale assumptions. This is consistent with Cano et al's (2004) [21] measurement development guidelines.

Psychometric evidence for the use of the PSQ and SCQ with paediatric and adolescents burn patients is demonstrated in a validation study which involved 361 patients aged 8 to 18 [26]. The study reported less than 1% missing data for both measures, which indicates that they were acceptable to paediatric and adolescent patients. However no data was reported relating to the time it took paediatric patients to complete the measures. The scales had also not undergone a formal acceptability or feasibility study with paediatric burns patients during the development or re-validation stage.

Confirmatory factor analysis showed that both scales had adequate evidence of internal consistency reliability (.78 - .89) and construct validity (.46 - .69) [26]. When comparing the fit indices of nested models, both measures demonstrated measurement invariance (equal latent form, factor loadings and indicator thresholds) across paediatric and adult groups. However, as yet, there is no published evidence of item total correlations, test-retest reliability, responsiveness or validity hypothesis testing with paediatric burn patients.

The only burns-specific scale identified in this review was the Children Burn Outcomes Questionnaire (CBOQ) for adolescents aged 11 -18 [27]. It has 52 items and 12 different subscales which assess

various physical, psychological and social health outcomes (e.g. upper extremity function, physical function and sports, transfers and mobility, pain, itch, appearance, compliance, satisfaction with current state, emotional health, family disruption, parental concern and school re-entry). This scale used a range of different response categories.

The items in the CBOQ were generated from a review of the instrument literature and by expert clinician input. However interviews with child and adolescent patients were not conducted and no conceptual framework was developed. Item reduction was conducted using expert opinion, factor analysis and tests of scaling assumptions. However tests of item redundancy, endorsement frequencies and missing data were not used to exclude items.

Evidence to support the psychometric properties of the CBOQ was obtained in the instrument validation study with 86 adolescent burn patients (aged 11 – 18). There were few missing data. On average, adolescents took 33 minutes to complete the questionnaire. This suggests an acceptable degree of burden for paediatric burn patients; however formal acceptability and feasibility studies were not conducted. The scale demonstrated fair to excellent internal reliability ($\alpha = .75$ to $.92$) and it correlated with the Child Health Questionnaire (a validated measure assessing similar domains) indicating adequate construct validity. The scale demonstrated satisfactory to excellent test-retest reliability ($r = 0.67 - 0.99$). However there was no published evidence of the item total correlations, validity hypothesis testing or responsiveness.

Table 2 The psychometric quality assessment of the three PROMs with some evidence of validity in target population

Method/Evaluation	Generic PROMs		Burns-specific PROM
	PSQ (Lawrence et al, 2006)	SCQ (Lawrence et al, 2006)	CBOQ (Daltroy et al, 2000)
Item generation			
Literature review	✓	✓	✓
Expert opinion	✓	✓	✓
Patient interview	X	X	X
Develop conceptual framework	X	X	X
Item reduction			
Expert opinion	X	X	✓
Item redundancy	✓	✓	✓
Endorsement frequencies	✓	✓	X
Missing data	X	X	X
Factor analysis	✓	✓	✓
Tests of scaling assumptions	✓	✓	✓
Psychometric analyses			
Acceptability	✓	✓	✓
Internal consistency reliability	✓	✓	✓
Item total correlations	X	X	X
Test-retest reliability	X	X	✓
Validity within scale	✓	✓	✓
Validity comparison with other measures	X	X	✓
Validity hypothesis testing	X	X	X
Responsiveness	X	X	X

Discussion

The current study aimed to identify PROMs that have been used in paediatric and adolescent burn care research and to assess their psychometric properties for their use with these patient groups. An extensive systematic review identified that a variety of PROMs have been used to assess a range of outcomes in this population. The majority of the reviewed PROMs were generic rather than burns-specific, and covered a broad spectrum of psychological and physical health domains including social skills, post-traumatic stress symptoms, anxiety/depression, appearance and quality of life. Evidence to assess the psychometric properties of the majority of measures was lacking.

The majority of the generic measures reviewed had only been validated with child and/or adolescents in the general population. Generic PROMs that have not been developed and/or validated with paediatric and adolescent burn patients are unlikely to have adequate responsiveness to identify changes in this population. For example, Gaskell (2007) [33] found that qualitative interview data from paediatric patients identified psychological benefits of attending burns camps, but no significant improvements were identified on the standardised generic measures completed by participants at the same time.

Only three PROMs had undergone some level of psychometric testing with child or adolescent burn patients. The two generic scales, the PSQ and SCQ had evidence of validity within the scale, internal consistency reliability and acceptability with paediatric and adolescent burn patients (aged 8 – 18 years). However there is currently no evidence of item total correlations, test-retest reliability, validity with other measures, validity hypothesis testing and responsiveness of these scales. The only burns-specific scale, the CBOQ, had evidence of internal consistency, test-retest reliability, validity within the scale and with other measures, validity hypothesis testing and acceptability with adolescent burn patients (aged 11- 18 years old). However it lacked evidence of item total correlations and responsiveness, so it is unclear whether the instrument can detect clinical change over time.

These three PROMs show promise, however they are not without limitations. Each scale was written in American English and was validated with American paediatric/adolescent burns patients; there might therefore be cultural differences in the relevancy of the scales to paediatric/adolescent burn populations in the UK and elsewhere. For a PROM to be effective and useful in identifying patients' needs and experiences, it must be applicable to each cultural and linguistic group being investigated [76]. PROMs require intra-lingual validation if they are intended to be used with a population that speaks a different form of the language from which it was originally developed (e.g. US English and UK English) [77]. Further research should explore their relevance and validity with paediatric and adolescent burn patients in the UK.

Increasingly researchers have highlighted the importance of conducting qualitative research with patients when developing PROMs, in order to ensure that the items are well understood, wide-ranging and appropriately reflect the experiences of the specified population [78]. Guidance offered by the Scientific Advisory Committee of the Medical Outcomes Trust [22] and Cano et al (2004) [21] indicate that patient interviews are an important information source when generating items during the development of a PROM. Generating items from patient interviews increases the likelihood that questions will adequately reflect patient experiences [79]. During the development of all three PROMs, burns patients reviewed drafts of the measures and provided feedback (namely children in Daltroy et al's PROM [27] and adult burns survivors in Lawrence et al's PROMs [27]). However none of the three reviewed PROMs had been developed from patient interview data. Instead, PROM items were based on expert opinion and literature reviews. Recent guidelines from the Patient Reported Outcomes (PRO) Content Validity Good Research Practices Task Force [80, 81] have

highlighted the importance of gaining target population input during item generation and for reviewing draft versions of the PROM to provide evidence of content validity. Developing questionnaire items without patient input may mean that important aspects of the patient experience are missed [8].

Additionally these three measures were psychometrically tested using classical test theory. Classical test theory (CTT) is the most common psychometric method to develop patient reported outcome measures [82]. However there are a number of limitations to developing PROMs using this method. The first is that collected data are ordinal rather than interval, therefore measurement invariance (e.g. the rule that the relationship between the latent variables and items needs to remain the same across groups) is not confirmed [82]. Accordingly, it is not clear whether the observed differences in scores can be attributed to actual differences in the latent variable measured or for example, differences in the way different patients interpret the questions.

Secondly the scale properties such as validity or reliability are sample dependent. Therefore the probability distribution of scale scores can vary between population subgroups, making it difficult to accurately compare subgroups of the same population [82]. Thirdly, the total scores can only be used for group comparisons and not for individual patient measurement [8]. This means that PROMs developed using CTT may lack validity in the context of measuring progress in individual patients. As this is the purpose of patient reported outcome measures when used in a clinical context, this may pose problems for burn care health professional and researchers.

An alternative psychometric measurement model called Rasch item analysis [83] was established to overcome some of the limitations associated with classical test theory. For example, Rasch analysis creates interval measurements from ordinal-level scale data, therefore measurement invariance can be tested and items fitting the Rasch model can be summed to produce valid total scores. This would facilitate consistency in the measurement of the full range of impact of burn injuries [82] and would increase the accuracy of the PROM in identifying clinical change. Secondly, Rasch analysis develops items that are independent of the sampling distribution of items (and patients). This allows measurement to be accurately conducted at both individual and subsample levels [8]. Currently very few PROMs used in burn care have been developed or validated using Rasch analysis. Research by van de Wal et al (2012) [84] is the only study to date that has applied the Rasch analysis to a burns PROM. This involved subjecting the Patient and Observer Scar Assessment Scale (POSAS) [15] (which was originally developed using CTT) to a Rasch analysis. The results showed it is a reliable and valid scale for assessing burn scarring based on the Rasch principles. Researchers and clinicians that are developing and validating new PROMs for paediatric and adolescent burn patients should consider using Rasch methodology to ensure that the new scales are appropriate for assessing the experience of both individual patients and population subgroups.

Limitations

The current study aimed to include all relevant articles reporting the use of standardised PROMs with child and adolescent burn patients. However our search was limited to articles that had been written up for research purposes. It is possible that clinicians or researchers may be using additional PROMs in their clinical work that have been missed in this review. To overcome this limitation, the authors are conducting interviews with burn care professionals to identify any further PROMs currently in use in the clinical context.

Conclusions

Although a variety of different PROMs have been used with child and adolescent burn patients, only three have some level of psychometric evidence derived from this population. Additionally, these three PROMs lack evidence of their responsiveness and thus it is not clear how effective they are at identifying clinical change in this group of patients.

Generic outcome measures are unlikely to address the specific issues commonly experienced by child and adolescent burn patients and may not have the ability to detect changes associated with rehabilitation over time. In order to ensure that PROMs are specific to the needs of this group, in-depth patient interviews should inform item generation and scale development. Patient input is also vital to ensure that questions are relevant and phrased appropriately for this population.

In order to address the recommendations of the National Burn Care Review (2001) [5] and to improve the provision of care for patients with burns, it is essential for burn care professionals and researchers to have access to age appropriate PROMs that have been developed and validated in line with expert guidance from Cano et al (2004) [21] and the Medical Outcomes Trust [22]. The development of such measures and the involvement of patients in this process remain a priority.

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References

- [1] Making the Link (2012) *The Costs of Burns*. Retrieved on 8th May 2013 from <http://www.makingthelink.net/tools/costs-child-accidents/costs-burns>. Number of A&E attendances compiled by Ken Dunn from IBID and HES data. Percentage of children attending A&E identified from HASS and LASS data.
- [2] Lawrence JW, Rosenberg, L, Mason, S, Fauerbach, JA. Comparing parent and child perceptions of stigmatizing behavior experienced by children with burn scars. *Body image* 2011; 8 (1): 70-3.
- [3] Dise-Lewis, JE. A developmental perspective on psychological principles of burn care. *J Burn Care Rehabil* 2001; 22(3): 255-60.
- [4] Bakker, A., Maertens, K. J., Van Son, M. J., & Van Loey, N. E. (2013). Psychological consequences of pediatric burns from a child and family perspective: A review of the empirical literature. *Clin Psychol Rev* 2013; 33 (3): 361-71.
- [5] National Burns Care Review. Committee Report: Standards and Strategy for Burn Care: A Review of Burn Care in the British Isles; 2001.
- [6] Matza LS, Patrick DL, Riley AW, Alexander JJ, Rajmil L, Pleil AM, Bullinger M. Pediatric patient-reported outcome instruments for research to support medical product labeling: Report of the ISPOR PRO good research practices for the assessment of children and adolescents task force. *Value Health* 2013; 16 (4): 461-79.
- [7] Lawrence JW, Mason ST, Schomer K, Klein MB. Epidemiology and impact of scarring after burn injury: a systematic review of the literature. *J Burn Care Res* 2012; 33 (1): 136-46.
- [8] Pusic A, Liu JC, Chen CM, Cano S, Davidge K, Klassen A, et al. A systematic review of patient-reported outcome measures in head and neck cancer surgery. *Otolaryng Head Neck* 2007; 136 (4): 525-35.
- [9] Rumsey N, Harcourt D. Visible difference amongst children and adolescents: issues and interventions. *Dev Neurorehabil* 2007; 10 (2): 113-23.
- [10] Mahar PD, Wasiak J, O'Loughlin CJ, Christelis N, Arnold CA, Spinks AB, Danilla S. Frequency and use of pain assessment tools implemented in randomized controlled trials in the adult burns population: a systematic review. *Burns* 2012; 38 (2): 147-54.
- [11] Falder S, Browne A, Edgar D, Staples E, Fong J, Rea S, Wood F. Core outcomes for adult burn survivors: a clinical overview. *Burns* 2009; 35 (5): 618-41.
- [12] Wasiak J, McMahon M, Danilla S, Spinks A, Cleland H, Gabbe B. Measuring common outcome measures and their concepts using the International Classification of Functioning Disability and Health (ICF) in adults with burn injury: A systematic review. *Burns* 2011; 37 (6): 913-24.
- [13] Van Baar ME, Essink-Bot ML, Oen, IMM, Dokter J, Boxma H, van Beeck EF. Functional outcome after burns: a review. *Burns* 2006; 32 (1): 1-9.
- [14] Tyack Z, Simons M, Spinks A, Wasiak J. A systematic review of the quality of burn scar rating scales for clinical and research use. *Burns* 2012; 38 (1): 6-18.

- [15] Draaijers LJ, Tempelman FR, Botman YA, Tuinebreijer WE, Middelkoop E, Kreis RW, van Zuijlen PP. The patient and observer scar assessment scale: a reliable and feasible tool for scar evaluation. *Plast Reconstr Surg* 2004; 113 (7): 1960 - 65.
- [16] Moher D, Liberati A, Tetzlaff J, Altman DG. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Ann Intern Med* 2009; 151 (4): 264-69.
- [17] Higgins JPT, Green S. *Cochrane Handbook for Systematic Reviews of Interventions Version 5.1.0*; 2011.
- [18] Griffiths C, Harcourt D, Rumsey N, Pleat J (in preparation). A systematic review of patient reported outcome measures (PROMs) used in adult burn research.
- [19] Bevans KB, Riley AW, Moon J, Forrest CB. Conceptual and methodological advances in child-reported outcomes measurement. *Expert Rev Pharmacoecon Outcomes Res* 2010; 10 (4): 385-96.
- [20] Centres for Disease Control and Prevention. *Child Development* 2012. Retrieved from <http://www.cdc.gov/ncbddd/childdevelopment/positiveparenting/middle2.html>
- [21] Cano SJ, Browne JP, Lamping DL. Patient-based measures of outcome in plastic surgery: current approaches and future directions. *Brit J Plast Surg* 2004; 57 (1): 1–11.
- [22] Aaronson N, Alonso J, Burnam A, Lohr KN, Patrick DL, Perrin E, et al. Assessing health status and quality-of-life instruments: attributes and review criteria. *Qual Life Res* 2002; 11 (3): 193-05.
- [23] Pusic AL, Klassen AF, Scott AM, Klok JA, Cordeiro PG, Cano SJ. Development of a new patient-reported outcome measure for breast surgery: the BREAST-Q. *Plast Reconstr Surg* 2009; 124 (2), 345-53.
- [24] Lawrence JW, Rosenberg LE, Fauerbach JA. Comparing the body esteem of pediatric survivors of burn injury with the body esteem of an age-matched comparison group without burns. *Rehabil Psychol* 2007; 52 (4): 370 - 79.
- [25] Lawrence JW, Fauerbach JA, Heinberg LJ, Doctor M, Thombs BD. The reliability and validity of the Perceived Stigmatization Questionnaire (PSQ) and the Social Comfort Questionnaire (SCQ) among an adult burn survivor sample. *Psychol Assessment* 2006; 18 (1): 106 - 11.
- [26] Lawrence JW, Rosenberg L, Rimmer RB, Thombs BD, Fauerbach, JA. Perceived stigmatization and social comfort: Validating the constructs and their measurement among pediatric burn survivors. *Rehabil Psychol* 2010; 55 (4): 360 – 71.
- [27] Daltroy LH, Liang MH, Phillips CB, Daugherty MB, Hinson M, Jenkins M, et al. American Burn Association/Shriners Hospitals for Children burn outcomes questionnaire: construction and psychometric properties. *J Burn Care Res* 2000; 21 (1): 29-39.
- [28] Rosenberg, M. *Conceiving the self*. New York: Basic Books; 1979.
- [29] Arnoldo BD, Crump D, Burris AM, Hunt JL, Purdue GF (2006). Self-esteem measurement before and after summer burn camp in pediatric burn patients. *J Burn Care Res* 2006; 27 (6): 786-89.

- [30] Rimmer RB, Fornaciari GM, Foster KN, Bay CR, Wadsworth MM, Wood M, Caruso DM (2007). Impact of a pediatric residential burn camp experience on burn survivors' perceptions of self and attitudes regarding the camp community. *J Burn Care Res*; 28 (2): 334-41.
- [31] Wiens B. The Relationship Between Injury-specific Teasing and Adjustment in Children with Burn Injuries. Unpublished dissertation; 2004.
- [32] Harter S. Manual for the self-perception profile for children. University of Denver; 1985.
- [33] Gaskell SL. The challenge of evaluating rehabilitative activity holidays for burn-injured children: qualitative and quantitative outcome data from a Burns Camp over a five-year period. *Dev Neurorehabil* 2007; 10 (2): 149-60.
- [34] Girolami, P. The impact of a school reentry program on the psychosocial adjustment of children with burn injuries. Unpublished dissertation; 2004. Retrieved from Google Scholar.
- [35] Harter S. Manual for the Adolescent Self-Perception Profile. University of Denver; 1988.
- [36] Piers EV, Herzberg DS. Piers-Harris Children's Self Concept Scale Manual. Los Angeles: Western Psychological Services; 2002.
- [37] Maskell J, Newcombe P, Martin G, Kimble R. Psychosocial functioning differences in pediatric burn survivors compared with healthy norms. *J Burn Care Res* 2013; 34 (4): 465-76.
- [38] Bendig AW. The development of a short form of the Manifest Anxiety Scale. *J Consult Clin Psychol* 1956; 20 (5): 384.
- [39] Rivlin E, Faragher EB. The psychological sequelae of thermal injury on children and adolescents: Part 1. *Dev Neurorehabil* 2007; 10 (2): 161-72.
- [40] Reynolds CR, Paget KD. Factor analysis of the revised Children's Manifest Anxiety Scale for blacks, whites, males, and females with a national normative sample. *J Consult Clin Psychol*, 1981; 49 (3): 352-59.
- [41] Stokes DJ, Dritschel BH, Bekerian DA. The effect of burn injury on adolescents autobiographical memory. *Behav Res Ther* 2004; 42 (11): 1357-365.
- [42] March JS, Parker JD, Sullivan K, Stallings P, Conners CK. The Multidimensional Anxiety Scale for Children (MASC): factor structure, reliability, and validity. *J Am Acad Child Adolesc Psychiatry* 1997; 36 (4): 554-65.
- [43] Saxe GN, Stoddard F, Hall E, Chawla N, Lopez C, Sheridan R, et al. Pathways to PTSD, part I: children with burns. *Am J Psych* 2005; 162 (7): 1299-304.
- [44] Saxe G, Geary M, Bedard K, Bosquet M, Miller A, Koenen et al. Separation anxiety as a mediator between acute morphine administration and PTSD symptoms in injured children. *Ann N Y Acad Sci* 2006; 1071 (1): 41-45.
- [45] Conte HR, Weiner MB, Plutchik R. Measuring death anxiety: conceptual, psychometric, and factor-analytic aspects. *J Pers Soc Psychol* 1982; 43 (4): 775-85.
- [46] Beck AT, Steer RA, Ball R, Ranieri WF. Comparison of Beck Depression Inventories-IA and-II in psychiatric outpatients. *J Pers Assess* 1996; 67 (3), 588-97.

- [47] Pope SJ, Solomons WR, Done DJ, Cohn N, Possamai AM. Body image, mood and quality of life in young burn survivors. *Burns* 2007; 33 (6): 747-55.
- [48] Birleson P. The validity of depressive disorder in childhood and the development of a self-rating scale: A research report. *J Child Psychol Psychiatry* 1981; 22 (1): 73-88.
- [49] Dodd AR, Nelson-Mooney K, Greenhalgh DG, Beckett LA, Li Y, Palmieri TL. The effect of hand burns on quality of life in children. *J Burn Care Res* 2010; 31; (3): 414-22.
- [50] Stubbs TK, James LE, Daugherty MB, Epperson K, Barajaz KA, Blakeney P, et al. Psychosocial impact of childhood face burns: A multicenter, prospective, longitudinal study of 390 children and adolescents. *Burns* 2011; 37 (3): 387-94.
- [51] Varni JW, Seid M, Rode CA. The PedsQL (TM): Measurement model for the pediatric quality of life inventory. *Med Care* 1999; 37 (2): 126-13.
- [52] Powers SE. Risk versus resilience: an exploratory study of factors influencing the development of posttraumatic stress symptoms in pediatric burn patients. Unpublished dissertation; 2011.
- [53] Weedon M, Potterton J. Socio-economic and clinical factors predictive of paediatric quality of life post burn. *Burns* 2011; 37 (4): 572-79.
- [54] Topolski TD, Edwards TC, Patrick DL. User's Manual and Interpretation Guide for the youth quality of life (YQOL) instruments. Seattle, WA: University of Washington, Department of Health Services; 2002.
- [55] Mendelson BK, Mendelson MJ, White DR. Body-esteem scale for adolescents and adults. *J Pers Assess* 2001; 76 (1): 90-106.
- [56] Lawrence JW, Heinberg LJ, Roca R, Munster A, Spence R, Fauerbach JA. Development and validation of the Satisfaction with Appearance Scale: Assessing body image among burn-injured patients. *Psychol Assessment* 1998; 10 (1), 64 -70.
- [57] Varni JW, Burwinkle TM, Katz ER, Meeske K, Dickinson P. The PedsQL in pediatric cancer: reliability and validity of the Pediatric Quality of Life Inventory Generic Core Scales, Multidimensional Fatigue Scale, and Cancer Module. *Cancer*, 2002; 94 (7): 2090–106.
- [58] Spence, S. *Social Skills Training: Enhancing Social Competence with Children and Adults*. London: NFER-Nelson; 1995.
- [59] Gill M (2008). Psychosocial benefits for child burn survivors who attend summer camp. Doctoral dissertation; 2008. Retrieved from ProQuest.
- [60] Gresham FM, Elliott SN. *Social skills rating system: Manual*. American Guidance Service; 1990.
- [61] Miller SM, Roussi P, Caputo GC, Kruus L. Patterns of children's coping with an aversive dental treatment. *Health Psychol* 1995; 14 (3): 236–246.
- [62] Young SR. The potential mediating effects of learned resourcefulness and coping disposition on post-burn adjustment in severely burned adolescents. Doctoral dissertation; 2003. Retrieved from ProQuest.

- [63] Goodman R. Psychometric properties of the strengths and difficulties questionnaire. *J Am Acad Child Adolesc Psychiatry* 2001; 40 (11): 1337-345.
- [64] Martin G, Swannell S, Mill J, Mott J, Evans J, Frederiksen, N, et al. Spray on skin improves psychosocial functioning in pediatric burns patients: a randomized controlled trial. *Burn* 2008; 34 (4): 498-04.
- [65] Achenbach TM, Rescorla L. *ASEBA School-Age Forms & Profiles*. Burlington: Aseba; 2001.
- [66] Blakeney P, Thomas C, Holzer III C, Rose M, Bernige, F, Meyer III WJ. Efficacy of a short-term, intensive social skills training program for burned adolescents. *J Burn Care Res* 2005; 26 (6): 546-55.
- [67] Harter S. *Social support scale for children: Manual and Questionnaires*. University of Denver; 1985.
- [68] Austin JK, Huberty TJ. Development of the child attitude toward illness scale. *J Pediatr Psychol* 1993; 18(4): 467-80.
- [69] Rosenbaum M. A schedule for assessing self-control behaviors: Preliminary findings. *Behav Ther* 1980; 11 (1): 109-21.
- [70] Prince-Embury S. *Resiliency Scales for Children and Adolescents: Profiles of personal strengths*. San Antonio, TX: Harcourt Assessments; 2007.
- [71] Napier TL, Wright CJ. An evaluation of forced relocation of population due to rural community development. *Research Bulletin*. Ohio Agricultural Research and Development Center; 1974.
- [72] Hansen DM, Larson R. *The youth experience survey 2.0: Instrument revision and validity testing*, Unpublished manuscript, University of Illinois at Urbana-Champaign; 2005.
- [73] Rimmer RB, Pressman MS, Takach OP, Bay RC, Croteau R, Hansen LD, et al. Burn-injured adolescents report gaining multiple developmental benefits and improved life skills as a result of burn camp attendance. *J Burn Care Res* 2012; 33 (4): 552-60.
- [74] Eysenck SBC, Eysenck HJ. Test-Retest reliabilities of a new personality questionnaire for children. *Brit J Educ Psychol* 1973; 43: 126–30.
- [75] Horowitz MJ, Wilner N, Alvarez W. Impact of event scale: a measure of subjective stress. *Psychosom Med* 1979; 41 (3): 209–18.
- [76] Beaton DE, Bombardier C, Guillemin F, Ferraz MB. Guidelines for the process of cross-cultural adaptation of self-report measures. *Spine* 2000; 25 (24): 3186 – 91.
- [77] MAPI Institute. *Linguistic Validation Manual for Health Outcome Assessments*, 2nd Ed. edn. Lyon: MAPI Research Trust; 2012.
- [78] Brédart A, Marrel A, Abetz-Webb L, Lasch K, Acquadro C. Interviewing to develop Patient-Reported Outcome (PRO) measures for clinical research: eliciting patients' experience. *Health Qual Life Out* 2014; 12 (1): 15-25.
- [79] Lasch KE, Marquis P, Vigneux M, Abetz L, Arnould B, Bayliss M, et al. PRO development: rigorous qualitative research as the crucial foundation. *Qual Life Res* 2010; 19 (8): 1087-096.

[80] Patrick DL, Burke LB, Gwaltney CJ, Leidy NK, Martin ML, Molsen E, et al. Content validity—establishing and reporting the evidence in newly developed patient-reported outcomes (PRO) instruments for medical product evaluation: ISPOR PRO good research practices task force report: part 1—eliciting concepts for a new PRO instrument. *Value Health* 2011a; 14 (8): 967-77.

[81] Patrick DL, Burke LB, Gwaltney CJ, Leidy NK, Martin ML, Molsen E, et al. Content validity—establishing and reporting the evidence in newly developed patient-reported outcomes (PRO) instruments for medical product evaluation: ISPOR PRO Good Research Practices Task Force report: part 2—assessing respondent understanding. *Value Health* 2011b; 14 (8): 978-88.

[82] Cano S, Hobart J. The problem with health measurement, *Patient Preference and Adherence* 2011; 5: 279-90.

[83] Rasch G. Probabilistic models for some intelligence and attainment tests. Reprinted Chicago: MESA Press; 1993.

[84] van der Wal MB, Tuinebreijer WE, Bloemen MC, Verhaegen PD, Middelkoop E, van Zuijlen PP. Rasch analysis of the Patient and Observer Scar Assessment Scale (POSAS) in burn scars. *Qual Life Res* 2012; 21 (1): 13-23.