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GENERAL ORTHOPAEDICS

Development of a patient-reported outcome measure in limb reconstruction

A PILOT STUDY ASSESSING FACE VALIDITY

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Aims

Patients undergoing limb reconstruction surgery often face a challenging and lengthy process to complete their treatment journey. The majority of existing outcome measures do not adequately capture the patient-reported outcomes relevant to this patient group in a single measure. Following a previous systematic review, the Stanmore Limb Reconstruction Score (SLRS) was designed with the intent to address this need for an effective instrument to measure patient-reported outcomes in limb reconstruction patients. We aim to assess the face validity of this score in a pilot study.

Methods

The SLRS was designed following structured interviews with several groups including patients who have undergone limb reconstruction surgery, limb reconstruction surgeons, specialist nurses, and physiotherapists. This has subsequently undergone further adjustment for language and clarity. The score was then trialled on ten patients who had undergone limb reconstruction surgery, with subsequent structured questioning to understand the perceived suitability of the score.

Results

Ten patients completed the score and the subsequent structured interview. Considering the tool as a whole, 100% of respondents felt the score to be comprehensible, relevant, and comprehensive regarding the areas that were important to a patient undergoing limb reconstruction surgery. For individual questions, on a five-point Likert scale, importance/relevance was reported as a mean of 4.78 (4.3 to 5.0), with ability to understand rated as 4.92 (4.7 to 5.0) suggesting high levels of relevance and comprehension. Flesch-Kincaid reading grade level was calculated as 5.2 (10 to 11 years old).

Conclusion

The current SLRS has been shown to have acceptable scores from a patient sample regarding relevance, comprehensibility, and comprehensiveness. This suggests face validity, however further testing required and is ongoing in a larger cohort of patients to determine the reliability, responsiveness, precision, and criterion validity of the score in this patient group.

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Introduction

Validated and effective outcome measures are an essential element of performing good quality research in orthopaedic surgery.[1] Increasingly, such measures are also being used in developing registry data as well as potential use in commissioning.[2] Patient-reported outcomes measures (PROMs) allow further understanding into the patient perspective of a disease and its treatment. Such information can guide effectiveness of treatment,[3] and help to counsel patient expectations for surgery.[4]

There are a wide range of PROMs that have been designed. These can include scores based on broader health-related quality of life (e.g. 36-Item Short Form Health Survey questionnaire (SF-36), EuroQol five-dimension

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questionnaire (EQ-5D)),[5,6] those specific to an anatomical area or joint (e.g. Oxford Hip Score, American Academy of Orthopedic Surgeons (AAOS) lower limb score),[7,8] or scores which are targeted at a particular condition (e.g. Boston Carpal Tunnel Syndrome Questionnaire, Western Ontario Shoulder Instability Index).[9,10] The patient undergoing limb reconstruction surgery has particular challenges, both due to the underlying condition (deformity, complex trauma, infection, nonunion) and the treatment process itself. As well as pain and functional limitations, psychological, cosmetic, and socioeconomic issues may affect the healthcare-related quality of life.[11-16] As such, existing anatomy specific scores, which focus on pain or function alone, may not capture the improvements that correction of deformity may have on social impairment or patient perception of cosmesis.

A recent systematic review demonstrated that no single PROM adequately captures all domains of importance to this patient group.[17] This results in existing research either using different collections of instruments to try to effectively understand patient-reported outcomes, or even neglecting patient-reported measures entirely to focus on the more easily attainable radiological outcomes and complication profile. Use of multiple measures not only limits comparisons with other published data, but also limits feasibility of data collection, due to "respondent fatigue" from both the patient and the collecting clinician.[18]

We have aimed to create and develop such an outcome measure in the hope that this could provide a tool for research and quality improvement by the limb reconstruction community. The first iteration of this has been previously published.[17] We present the initial introduction of this measure, along with assessment of face validity in the form of patient comprehension, comprehensiveness, and relevance.

Methods

This project was assessed and approved as a service evaluation project by the local research and ethics panel (Reg No. SE20.46). The methodology of initial score design, current pilot study, and plans for future validation are based on the methods as described by Fizpatrick et al,[19] and the more recent COnsensus based Standards for the selection of health status Measurement INstruments (COSMIN) methodology.[20,21]

A combination of patient and clinician involvement was used throughout the design of the score. Patients currently undergoing limb reconstruction surgery were recruited to take part in a structured interview following attendance at the outpatient clinic. Inclusion criteria at each stage was for patients aged > 18 years who are currently undergoing treatment with external fixator or lengthening nail. Exclusion criteria included limited understanding of English, the need for a translator, or any patients who were unwilling or unable to consent to take part.

Score design. Structured interviews were performed in 20 patients using described techniques to allow exploration of general and then subsequently specific areas relating to their underlying condition and limb reconstruction treatment.[22] These interviews were transcribed and conventional content analysis used to identify issues and themes of relevance.[23] This was used as a basis to determine the domains of relevance for the structure of the score. Subsequently, interviews with limb reconstruction surgeons (4), clinical nurse specialists (3), and therapists (2) were performed to further guide design of the score and ensure that the content was also thought to be comprehensive and relevant from the clinician perspective. The pain domain was based on a modification of the Brief Pain Inventory.[24] The remaining domains, as determined by the categories and themes identified from the content analysis, were assessed using a five-point Likert scale. The preliminary score has been published previously as an appendix to a systematic review of PROMs in limb reconstruction;[17] this paper shall focus on the pilot assessment of face validity.

Pilot and refinement. The score was trialled in 11 patients recruited in clinic with the same criteria as the design interviews, but these were new patients not previously involved in the design. They completed a further written questionnaire and short structured interview to assess the comprehensibility and relevance of each individual question, along with comprehensiveness of the score as a whole.

These comments were then used in two iterative group discussions involving limb reconstruction surgeons, clinical nurse specialists, and physiotherapists to further refine the language, appearance, and question structure of the score. The modified score (see Supplementary Material) was then trialled again in ten patients, once again using short structured interviews and a questionnaire to assess the comprehensibility and relevance of each individual question, along with comprehensiveness of the score as a whole. The patients were asked about the acceptability of the test and whether they would be willing to complete this while waiting in clinic. The score was tested with the use of Flesch-Kincaid readability tests (mathematical assessment of structure, sentence length, and word length in a document)[25] for objective assessment of comprehension level.

Statistical analysis was performed using SPSS 27 (IBM, USA) for Mac. Unless otherwise stated, categorical variables are expressed as frequency (percentage) and continuous variables are expressed as mean (range) with $p < 0.05$ considered as statistically significant. Non-parametric group comparisons of data were made with the Mann-Whitney U test.

Table I. Demographic details of patients undergoing pilot testing of the score.

Variable	First pilot group	Second pilot group
Sex, n		
Male	5	2
Female	6	8
Mean age, yrs (range)	33 (17 to 68)	42 (18 to 78)
Indication for treatment, n		
Deformity correction/ lengthening	6	7
Nonunion/infection	4	3
Trauma	1	0

Demographic details. The demographic details of the initial pilot group, and the second pilot after refinement of the score, are shown in Table I.

Results

Comprehensibility. The first test group showed that 3/11 patients found at least one question difficult to understand. Clarifications were made to the questions on use of public transport and employment, along with general adjustment for readability. The subsequent test group had no patients (0/10) who found any of the questions difficult to understand. The mean understandability of individual questions was rated as 4.92/5 (4.8 to 5).

The Flesch-Kincaid readability tests demonstrated a Flesch reading ease index of 58.6 for the initial version and 78.2 for the modified version (this index is out of 100, with a higher value representing an easier to read text). The Flesch-Kincaid reading grade level improved from 15.6 initially (equivalent to 18 years old) to 5.2 on the modified version (equivalent to a reading age of 10 to 11 years old).

Relevance. For the initial group, the mean score for relevance of the questions was 4.34/5 (3.8 to 5). The second group showed reported a mean relevance of 4.78 (4.3 to 5) for the modified score, which is a significant improvement ($p < 0.001$). When taking the score as a whole, 10/10 patients in the second group felt the score to be relevant to their needs. Similarly, 10/10 patients felt that the score was comprehensive of their situation as a limb reconstruction patient.

Feasibility. All patients using the modified score (10/10) felt the score was acceptable to complete while waiting in clinic.

Discussion

This pilot study has demonstrated that the updated version of the SLRS shows good levels of comprehensibility, comprehensiveness, relevance, and acceptability to a patient cohort undergoing limb reconstruction surgery. This is an indication of face validity from the patient perspective, which can be taken as an initial stage of the validation process.

By the nature of a pilot study, the numbers are small, however we feel the results are adequate to consider continuing with further validation. This has included patients with a variety of indications but limited to a single centre. The score has been designed using only adult patients and further work would be required if a paediatric limb reconstruction PROM was to be considered. While a combination of trauma patients and elective deformity/nonunion patients were included in the design of the score, this pilot did not include trauma patients, which is a limitation. For the ongoing further validation of this score, both trauma and elective patients will be included.

The previous systematic review from this unit has suggested that there is at present no adequate score which covers all the needs of the limb reconstruction patient.[17] This score has been created with the limb reconstruction patient at its centre from the beginning, to address their specific requirements. These methods have allowed us to endeavour to ensure that all domains of importance to the patient have been captured. Beyond the assessment of pain and physical function, the score includes evaluation of social function, cosmesis, and emotional state, to allow for a more nuanced assessment of the experience of the limb reconstruction patient within a single score. We believe this allows for better understanding of the outcomes of this patient group, both during and after treatment, which may help improve their holistic surgical care. Multiple existing scores using joint-specific and generic quality of life outcome measures could be used to capture elements of these data over the range of domains required. However, this would potentially risk both respondent fatigue[18] and, if too time-consuming, could also decrease feasibility of use of the score in a clinical setting.[26] A single specific outcome score can avoid these limitations.

At the time of writing, there is a UK group investigating the current state of PROMs in limb reconstruction. They have published their protocol for the theory of design of a new score,[27] although they are yet to report on the outcomes of a design process. There is also a proposed paediatric limb reconstruction score from Canada,[28] which is currently undergoing validation, although not yet available for general use. As our score has been designed for the adult patient group, this paediatric score may prove complementary for outcomes data collection in limb reconstruction and we look forward to further results as their score is developed.

Further work is underway that will be required to fully validate the SLRS. The requirements for developing an effective and valid patient-reported measure were initially described by Fitzpatrick et al.[19] The COSMIN methodology, however, has built further on this and gives a clear

and reproducible framework with which to consider the design and validation of such a score. Criterion validity can be tested considering both convergent and divergent validity in comparison to existing (albeit not comprehensive) scores. Reliability to repeated testing, sensitivity to change, internal consistency, and floor and ceiling effects will all need to be examined.

These methods of validation will require a larger number of patients to use the score and take part in the validation process. Due to the complex and subspecialized nature of limb reconstruction, the number of patients in any single unit are small, so ideally this testing process will be through a multicentre collaboration. At the time of writing we have a number of limb reconstruction units working together on the validation project, with an aim to further expand this. Once more centres are using a similar standardized score, more effective comparison will be allowed between published results for techniques, leading to easier collaboration for research, and this may pave the way for further registry data collection of outcomes to everyone's benefit.

In conclusion, this pilot study has demonstrated the SLRS has acceptable relevance, comprehension and comprehensiveness for the needs of a cohort of limb reconstruction patients. Further work is now ongoing in order to ensure the score is fully validate.

Take home message

- The Stanmore Limb Reconstruction Score has shown face validity in the pilot testing phase.
- Further validation is ongoing in a larger patient cohort.

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Supplementary material

Patient questionnaire.

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- P. Calder: Conceptualization, Writing – review & editing.

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