Original Article

Validation of a modified Scoliosis Research Society instrument for patients with limb deformity: The limb deformity-Scoliosis Research Society (LD-SRS) score

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Abstract

Background: Despite the large negative effect of limb deformity on health-related quality of life (QoL), there exists no patient-reported instrument to quantify this impact. Rather, limb deformity research has been performed using global QoL measurements concurrently with joint-specific and/or arthritis outcome scales, thereby requiring the completion of multiple instruments. Furthermore, joint- and arthritis-specific instruments focus on the impact pain has on function, whereas limb deformities may be pain-free with greater social and functional impairment. The purpose of this study was to validate a patient-reported instrument to quantify limb deformity-related QoL.

Materials and Methods: Because of the similarities with regard to pain, function, and body image between limb deformity and scoliosis, the Scoliosis Research Society-30 (SRS-30) spine deformity instrument was modified such that the words "back" and "trunk" were replaced with "limb" to create a novel instrument: the limb deformity-SRS (LD-SRS). Testing for construct validity (both convergent and discriminant), reliability, floor and ceiling effects, and minimal clinically important difference (MCID) was performed in a validation cohort of 62 subjects aged 18 years or older with nonarthritic, unilateral lower extremity deformity.

Results: Scale reliability was excellent (test-retest reliability, intraclass correlation coefficient = 0.977; internal consistency, Cronbach's alpha = 0.906), scores were normally distributed, and there were no floor or ceiling effects. There was also robust construct validity: convergent validity testing revealed positive correlations between the LD-SRS and all short-form-36 domains, the American Academy of Orthopaedic Surgeons-Lower Limb Module, and higher scores in those who were postcorrection. Discriminant validity was demonstrated with no correlations between the LD-SRS and subject age, sex, body mass index, surgeon-scored Limb Lengthening and Reconstruction Society-AIM Index, or surgeon-generated deformity measurements. MCID was calculated to be 0.3 (on a 4.0-point scale).

Conclusions: The LD-SRS score is a reliable and valid instrument to measure limb deformity-related QoL in patients with nonarthritic lower extremity deformity. It is a valuable tool which allows clinicians

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to quantify patients' deformity-related QoL with a single instrument, rather than repurposing scales which have been validated for other conditions and have limited applicability to the unique challenges of treating patients with a lower limb deformity.
Level of Evidence: Diagnostic, Level 2.

Key Words: Angular, femur, length discrepancy, lower extremity, outcomes, quality of life, tibia

Introduction

Having simple and reliable validated outcome measures is vital to conducting high-quality outcomes research in the field of orthopedic surgery. Specifically, patient-reported outcomes (PROs) allow surgeons and researchers to understand the impact of disease from the patients' perspective, as well as guide treatment and counsel patients with appropriate expectations. By combining features of both disease-specific scales and global quality of life (QoL) PROs, disease-specific QoL instruments are uniquely able to capture and quantify the effect of a particular disease on the health-related QoL of a single patient or a population of subjects with that disease.

Despite the large negative effect of limb deformity on health-related QoL, to our knowledge, there exists no PRO which quantifies this impact. Rather, existing limb deformity research has been performed using global QoL measurements concurrently with joint-specific (e.g., hip, knee, ankle) and/or arthritis outcome scales.[1-6] This methodology is problematic because it requires the completion of several outcomes instruments, none of which adequately portray the effect of limb deformity on patient QoL, and additionally result in a large respondent burden and difficulty in obtaining adequate follow-up. Furthermore, joint- and arthritis-specific instruments focus on the impact pain has on function, whereas limb deformities may be pain-free with a larger component of social and functional impairment. A standardized limb-deformity QoL instrument would alleviate all of these problems, thereby greatly improving limb deformity clinical outcomes research.

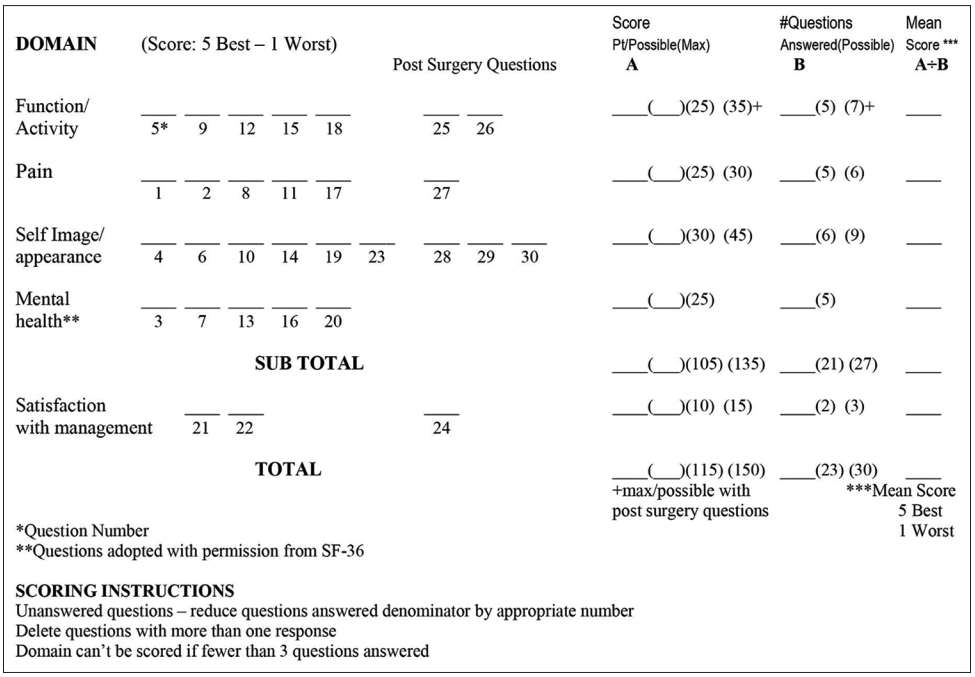
The Scoliosis Research Society (SRS) outcomes instrument was developed in 1999[7] and has been successfully translated into 17 languages and is universally used to evaluate the impact of spinal deformity on patient QoL. A similar instrument is vital for limb deformity research as well. Because of the noted similarities between limb deformity and scoliosis with regard to pain, function, and body image, the purpose of the current study was to attempt to validate a modified SRS-30 questionnaire,[8] the limb deformity-modified SRS Score (LD-SRS score) for use by patients with limb deformity.

Materials and Methods

This study was approved by the hospital Institutional Review Board (IRB) and consent or refusal did not affect patient care in any way.

In preparation instrument development and scale validation, a formal search of PubMed and Ovid/Medline was performed to locate any existing pediatric or adolescent activity scales. A search strategy was employed using the following terms and Boolean operators: "(scale OR questionnaire OR outcome OR instrument) AND (limb OR deformity)." In addition, medical subject headings ("MeSH terms") were included in the search to include hierarchically-organized search terms. After eliminating joint- and arthritis-specific scales, only the SRS instrument (and its derivations) remained. Because of the ubiquity of the SRS outcomes instrument for spinal deformity and the noted similarities with regard to pain, function, and body image between limb deformity and scoliosis, the decision was made to modify the SRS-30[8] to be applicable to limb deformity after ensuring that the scale was in the public domain.[9] For instance, the words "back" and "trunk" were replaced with "limb;" the final scale is presented in [Supplement 1 - The LD SRS Score is available online as a supplementary material on the website www.jlimblengthrecon.org]. The modified scale (LD-SRS) was scored in the same manner as the SRS-30 [Figure 1]. The senior authors experienced limb deformity surgeons, felt that this score captured relevant issues including pain, function, and body image for limb deformity patients.

Three groups of subjects were used to complete the study, all of whom met inclusion criteria of our target cohort: (1) 18–70-years-old, (2) seeking or undergoing treatment for a primary diagnosis of unilateral congenital or acquired lower limb length and/or angular deformity (e.g., not deformity secondary to arthritis), (3) fluent in English, and (4) willing to participate in the study. Exclusion criteria were: (1) primary diagnosis of osteoarthritis, (2) diagnosis of inflammatory arthropathy, and (3) primary deformity distal to the ankle (e.g., brachymetatarsia). These three groups consisted of the pilot testing group ($n = 20$), the validation group ($n = 62$), and the reliability group ($n = 42$) which was a subset of the validation group who responded that they experienced "no change in condition" between two



DOMAIN	(Score: 5 Best – 1 Worst)	Post Surgery Questions	Score Pt/Possible(Max) A	#Questions Answered(Possible) B	Mean Score *** A÷B
Function/ Activity	___ ___ ___ ___ ___ 5* 9 12 15 18	___ ___ 25 26	____(___)(25) (35)+	____(5) (7)+	____
Pain	___ ___ ___ ___ ___ 1 2 8 11 17	___ 27	____(___)(25) (30)	____(5) (6)	____
Self Image/ appearance	___ ___ ___ ___ ___ ___ 4 6 10 14 19 23	___ ___ ___ 28 29 30	____(___)(30) (45)	____(6) (9)	____
Mental health**	___ ___ ___ ___ ___ 3 7 13 16 20		____(___)(25)	____(5)	____
SUB TOTAL			____(___)(105) (135)	____(21) (27)	____
Satisfaction with management	___ ___ 21 22	___ 24	____(___)(10) (15)	____(2) (3)	____
TOTAL			____(___)(115) (150) +max/possible with post surgery questions	____(23) (30)	____ ***Mean Score 5 Best 1 Worst

*Question Number
**Questions adopted with permission from SF-36

SCORING INSTRUCTIONS
Unanswered questions – reduce questions answered denominator by appropriate number
Delete questions with more than one response
Domain can't be scored if fewer than 3 questions answered

Figure 1: The limb deformity-Scoliosis Research Society Score and scoring rubric. Adapted from the Scoliosis Research Society-30[8]

administrations of the questionnaire. Demographics of the study subjects are presented in Table 1.

Pilot testing

The modified scale was administered to the pilot testing group ($n = 20$), followed by cognitive interviews to ascertain subject comprehension and face validity. The subjects reported no difficulty in understanding the items and completing the scale.

Validation

Sixty-two English-speaking subjects who met study inclusion criteria were enrolled in the validation group. Consent or refusal was solely the decision of the patient-subject and did not affect care in any way. Construct validity was assessed through evaluation of the pediatric sports activity rating scale for both convergent and discriminant validity using widely accepted methodology.[10,11] In doing so, existing scales and demographic variables were considered *a priori* for either convergent or discriminant validity testing. Construct validity is assured in the event that there are significant correlations between the LD-SRS score and those measures of convergent validity, and there is an absence of significant correlation with those variables tested for discriminant validity,[10-13] for at least 75% of the tested hypotheses in at least 50 subjects.[14]

Convergent validity was evaluated by comparing scores on the LD-SRS to the American Academy of Orthopaedic Surgeons-Lower Limb Module (AAOS-LLM)[15] and short-form-36 (SF-36) health-related QoL outcome instrument (8 domains, mental component score, and physical component score).[16-18] While these scales are not validated specifically for limb deformity, they are helpful in providing construct validity by comparing scores with the LD-SRS through regression analysis.[10-13] Scales that were specifically designed for patients with osteoarthritis (e.g., WOMAC) were intentionally excluded from this study. In addition, stage of treatment (before/during correction vs. postcorrection) was investigated for convergent validity, as it is known that deformity correction improves health status and QoL.[19-22]

Discriminant (divergent) validity was assessed between the LD-SRS score and those variables which were hypothesized *a priori* to lack significant associations. In patients with lower limb

Table 1: Demographics and clinical characteristics of patients in the validation cohort (*n*=62 subjects)

Variable	Value
Age, mean±SD	38.9±13.9
BMI, mean±SD	27.2±5.5
Sex, *n* (%)	
Male	31 (50)
Female	31 (50)
Time of enrollment, *n* (%)	
Pre/during treatment	52 (83.9)
Post-treatment	10 (16.1)
Deformity type† (%)	
Angular only	7 (11.3)
Length only	9 (14.5)
Combined	36 (58.1)
Deformity measurements†	
Major angular deformity (°)	16.7±11.5
Leg length discrepancy (mm)	28.6±25.6
Mechanical axis deviation (mm)	30.6±23.8

†For those patients pre/during treatment. BMI: Body mass index, SD: Standard deviation

deformity, these included age, sex, and body mass index (BMI). Furthermore, the Limb Lengthening and Reconstruction Society-AIM (LLRS-AIM) Index,[23] a physician-rated measure of the treatment complexity of lower limb deformity, was evaluated for discriminant validity as well. Because the purpose of the LD-SRS score was to quantify the patient experienced deformity-related QoL, the investigators felt it was important to ensure the instrument was measuring a patient-related construct rather than a physician-related construct. To that end, scores on the LD-SRS were compared with quantitative measurements of the degree of deformity (major angular deformity, leg length discrepancy, and mechanical axis deviation) to ensure the LD-SRS construct scores were not influenced by the degree of lower limb deformity, and rather reflected the patient experienced deformity-related QoL.

Face validity of the final LD-SRS instrument was ensured by the clinician-investigators prior to initiation of the validation phase. Criterion validity could not be evaluated because there is currently no accepted reference standard measurement tool for limb deformity-related QoL.

Reliability

Reliability of the LD-SRS score was evaluated using two complementary methods: test-retest reliability and internal consistency. Sample size calculation in the previous methodology indicated that at least forty subjects are required for reliability testing.[24] In the current study, all 62 subjects who participated in the validation phase (the validation group) were included in internal consistency reliability testing. Four to twenty-one days after the first completion, this group of subjects were asked to complete the LD-SRS again.[25,26] They were also asked if their condition had changed in any way. Those who replied "no change" (42 subjects) were included in the final test-retest reliability analysis.

Floor and ceiling effects

Responses to the LD-SRS instrument was evaluated for floor and ceiling effects in the validation group ($n = 62$). A floor or ceiling effect was considered to be present if >15% of respondents scored the lowest or highest possible score, respectively.[14,27]

Statistical methods

Statistical analyses were performed by members of the research team with advanced training in epidemiology and biostatistics using SPSS statistics version 22 (IBM Corporation, Armonk, NY, USA). The sample size was derived from the previously validated methodology.[24,28,29] After ensuring data normality using skewness and kurtosis thresholds,[30] Pearson correlations were calculated to evaluate associations between the LD-SRS and the continuous variables used for convergent and discriminant validity (e.g., age, BMI, radiographic deformity measurements LLRS-AIM, AAOS-LLM, and SF-36 domains). The binary validation variables (e.g., sex, treatment stage) were evaluated using independent samples Student's *t*-tests. Test-retest reliability was assessed using the intraclass correlation coefficient (ICC) (2, 1). Internal consistency was evaluated using Cronbach's alpha. Minimal clinically important difference (MCID) was determined using one-half standard deviation of the cohort,[31,32] and was compared to the MCID of the original SRS instruments. All analyses were two-tailed and used $P = 0.05$ as the threshold for statistical significance.

Results

Demographic, clinical, and outcome instrument data on the study cohort is recorded in [Tables 1 and 2]. Subject diagnoses for those who were pre/during treatment ($n = 52$) included: angular deformity only ($n = 7$; 11.3%), limb length discrepancy only ($n = 9$; 14.5%), and combined angular and length deformities ($n = 36$; 58.1%). Ten patients (16%) were posttreatment whereas 52 patients (84%) were pretreatment or undergoing treatment at the time of the study.

Validity results

Sixty-two subjects were included in the validation analyses [Table 3]. Statistically significant positive correlations were noted between the LD-SRS score and all eight SF-36 domains, mental component score, and physical component score ($r = 0.11$–0.77, $P \leq 0.001$ for all). Furthermore,

Table 2: Score distributions of the validation cohort for instruments investigated in the current study (*n*=62 subjects)

Instrument and subscales	Score (mean±SD)
LD-SRS	
Total/composite score	3.4±0.6
Function/activity subscale	3.3±0.8
Pain subscale	3.5±1.0
Self-image subscale	3.1±0.8
Mental health subscale	3.8±0.8
Satisfaction with management subscale†	4.0±0.9†
SF-36 domains	
General health subscale	51.4±9.6
Physical function subscale	37.2±11.8
Role-physical subscale	37.0±12.1
Bodily pain subscale	41.5±10.7
Vitality subscale	48.0±9.6
Social functioning subscale	41.7±10.7
Mental health subscale	47.7±10.9
Role-emotional subscale	41.6±14.9
Mental component score	48.0±12.4
Physical component score	39.7±10.7
AAOS-LLM (normalized), range	39.5±11.4 (10-57)
AAOS-LLM (standardized), range	76.0±15.8 (35-100)
LLRS-AIM (range)	5.0±3.1 (0-14)

†For those patients who had undergone treatment. LLRS-AIM: Limb Lengthening and Reconstruction Society-AIM Index, AAOS: American Academy of Orthopedic Surgeons, LD-SRS: Limb deformity-Scoliosis Research Society, SD: Standard deviation, SF: Short-form

statistically significant positive correlations were observed between the LD-SRS score and the AAOS-LLM ($r = 0.77$, $P < 0.001$). Furthermore, LD-SRS scores of postdeformity treatment subjects were significantly higher than those subjects who were pretreatment or undergoing treatment at the time of the study (mean difference = 0.47, $P = 0.02$). Therefore, all of the *a priori* convergent validation hypotheses were demonstrated.

Of the variables evaluated for discriminant validity, no significant associations were noted between the LD-SRS and subject age, sex, BMI, or LLRS-AIM index. Furthermore, no significant correlations were noted between LD-SRS score and any of the three absolute measures of physician-measured deformity severity (major angular deformity, leg length discrepancy, mechanical axis deviation). Therefore, all of the *a priori* discriminant validation hypotheses were demonstrated.

Reliability results

All 62 subjects in the validation cohort were used to evaluate the LD-SRS score for internal consistency reliability, resulting in a Cronbach's alpha of 0.906. The test-retest interval averaged 10 days (range: 4–21 days). Forty-two subjects of the original validation group answered "no change" when asked if there were any changes in their condition at the time of retest and were, therefore, eligible for inclusion in test-retest reliability calculation. Test-retest reliability of the LD-SRS score was excellent (ICC = 0.977).

Table 3: Validation testing results of the limb deformity-Scoliosis Research Society composite score against convergent and discriminant comparison variables (*n*=62 subjects)

Comparison variable	Statistic†	*P*	Hypothesis upheld?
Convergent validity testing			
SF-36 domains			
General health	*r*=0.41	<0.001*	✓
Physical function	*r*=0.66	<0.001*	✓
Role-physical	*r*=0.70	<0.001*	✓
Bodily pain	*r*=0.64	<0.001*	✓
Vitality	*r*=0.57	<0.001*	✓
Social functioning	*r*=0.71	<0.001*	✓
Mental health	*r*=0.61	<0.001*	✓
Role-emotional	*r*=0.51	<0.001*	✓
Mental component score	*r*=0.53	<0.001*	✓
Physical component score	*r*=0.62	<0.001*	✓
AAOS-LLM	*r*=0.77	<0.001*	✓
Stage of treatment‡	Mean difference=0.47	0.02*	✓
Discriminant validity testing			
Age	*r*=−0.23	0.08	✓
Sex	Mean difference=0.20	0.21	✓
BMI	*r*=−0.12	0.34	✓
LLRS-AIM	*r*=−0.06	0.64	✓
Deformity measurements			
Major angular deformity (°)	*r*=0.13	0.38	✓
Leg length discrepancy (mm)	*r*=0.05	0.72	✓
Mechanical axis deviation (mm)	*r*=−0.07	0.65	✓

*$P \leq 0.05$, †Statistical comparisons for dichotomous variables ("Stage of treatment" and "Sex") were performed using an independent samples Student's *t*-test; all other continuous variables were compared using the Pearson correlation, ‡Stage of treatment comparisons were made between subjects who were postdeformity correction versus pretreatment or undergoing treatment. BMI: Body mass index, AAOS-LLM: American Academy of Orthopaedic Surgeons-Lower Limb Module (patient-scored), LLRS-AIM: Limb Lengthening and Reconstruction Society AIM Index (surgeon-scored), SF: Short-form

Floor and ceiling effects

LD-SRS total/composite scores for the validation cohort ($n = 62$) were normally distributed (skewness = −0.39; kurtosis = 0.001). LD-SRS total/composite scores for the validation group ranged from 1.6 to 4.6 (minimum and maximum possible scores are 1.0 and 5.0, respectively). No subject scored the minimum or maximum score; therefore, no floor or ceiling effects were present [Figure 2].

Minimal clinically important difference

Using the one-half standard deviation method,[31,32] the MCID of the LD-SRS total score is 0.3 (derived from a standard deviation of 0.6 for the validation cohort, [Table 1]).

Discussion

The current study is the first to our knowledge to develop and validate a PROs measure specific to lower extremity deformity. Use of this LD-SRS score will allow clinicians and investigators to quantify and understand patients' lower extremity deformity-related QoL, including pain, function, and body image, which is critical when providing care to these patients as well as performing clinical outcomes and comparative effectiveness research. This instrument can replace the practice of repurposing scales validated for nondeformity conditions, and that have limited applicability to the unique challenges of treating patients with limb deformity.

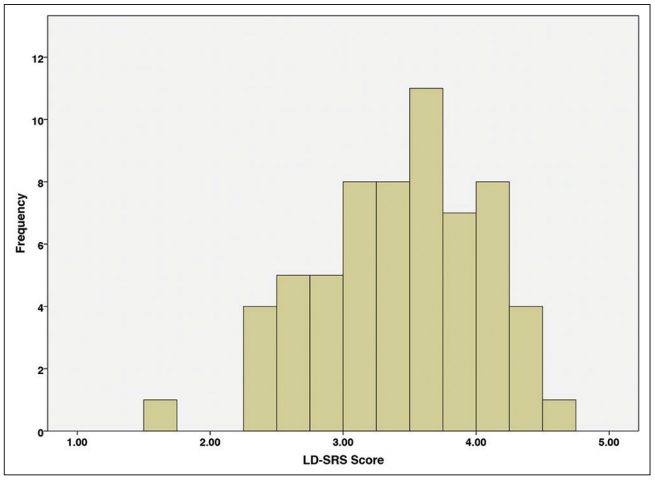


Figure 2: Score distribution of the limb deformity-Scoliosis Research Society in the validation cohort ($n = 62$ subjects) which indicates a normal distribution (skewness = −0.39; kurtosis = 0.001) and the absence of both floor and ceiling effects

In preparing to conduct this study, a formal literature search yielded a dearth of potential limb deformity-specific questionnaires, despite the high frequency with which lower extremity deformity is diagnosed and treated. Existing measures were designed for joint-specific (e.g., hip, knee, ankle) and/or arthritis. Using these scales requires the completion of several outcomes instruments, none of which adequately measure the effect of limb deformity on patient QoL. Furthermore, requiring the completion of multiple questionnaires results in a large respondent burden and creates difficulty with follow-up in clinical research studies. Moreover, joint specific outcome instruments tend to focus on the impact joint pain has on limb function and activities of daily living. When a limb is deformed, however, the condition is often painless (especially in congenital and developmental deformities), and the individual has adapted reasonably well to activities of daily living. Nevertheless, a heavy social and interpersonal burden often accompanies a limb deformity, one that should be relieved by a successful surgery. An outcome instrument specific for limb deformities must quantify this improvement. To do so, the questionnaire must include inquiries about social embarrassment, self-consciousness, and related issues that accompany a deformed limb. Treatment that reduces or eliminates such self-consciousness must be substantiated by better scores generated by the outcome instrument after treatment when compared to the pretreatment condition. The SRS recognized this imperative and created an outcome instrument for spinal deformity that measures such factors. Rather than developing a deformity-related outcome score from scratch, we have adapted their instrument to quantify the concerns of patients with limb deformities.

In evaluating the LD-SRS score, we found excellent reliability and validity. The LD-SRS performed well with both test-retest and internal consistency reliability testing. Furthermore, in addition to demonstrating both convergent and discriminant validity, the positive correlations in the 0.3–0.7 range [Table 3] indicate that while significant correlations do exist, the construct measured by the LD-SRS differs from that of existing scales. The lack of any significant associations with subject age, sex, BMI, or LLRS-AIM index indicated that this patient-reported instrument measures a patient-centered outcome, rather than one that is influenced by demographic or surgeon-related factors. Construct validity was excellent, as all the tested hypotheses for convergent and discriminant validity were upheld [Table 3].[10-14] The development of the questionnaire through modification of the SRS-30 was advantageous in that it allowed the scale to leverage the benefits of an existing, frequently-utilized scale. There were no floor or ceiling effects, and the MCID was comparable to previous studies of the original SRS instrument.[33,34]

MCID of the LD-SRS score was calculated using the one-half standard deviation method and was noted to be 0.3.[31,32] To corroborate this finding, the value can be compared to the calculated MCID of the original SRS-22 and SRS-30 instruments, which have been reported in the range of 0.4–0.5.[33,34] Future prospectively designed longitudinal research is required to determine responsiveness and confirm the value for MCID.

There are limitations to the current study. The LD-SRS was validated in subjects ages 18 and older; further validation is required to use the scale in children and adolescents, which is particularly important given the significant proportion of limb deformity in this demographic. Unfortunately, institutional IRB requirements for this initial investigation disallowed the enrollment of patients under age 18. However, the SRS instrument upon which the LD-SRS is based is frequently used in children and adolescents.[35-40] Therefore, the same will likely be true of the LD-SRS. Second, five questions from the mental health domain of the original SRS instrument (and therefore, this LD-SRS instrument) [Figure 1] were adapted from items originally in the SF-36, which can theoretically violate the independence assumption. This would have been problematic if all domains were combined for analysis, but was avoided in the current study by testing LD-SRS scores against each SF-36 domain individually [Table 3]. All domains showed moderate to high-moderate correlations with highly significant *P*-values, indicating that the LD-SRS is a sufficiently different construct than the SF-36 which shows excellent convergent validity as hypothesized *a priori*. Third, further longitudinal investigation will be required to determine the responsiveness of the SD-SRS as well as to confirm the MCID threshold established in the current study. Finally, this study validates the LD-SRS score in a cohort of subjects with lower extremity deformity in the absence of significant osteoarthritis or inflammatory arthropathy. Future validation studies will be required before the instrument's use in patients with upper extremity deformity and/or deformity caused by osteoarthritis or inflammatory arthritis.

Conclusions

The LD-SRS score is a reliable and valid instrument to measure lower limb deformity-related QoL in patients with nonarthritic lower extremity deformity. It is a valuable tool which allows clinicians to quantify patients' deformity-related QoL with a single instrument. Future research can expand the use of this instrument for use in children and adolescents as well as those with upper extremity deformity.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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