Clin Orthop Relat Res
DOI 10.1007/s11999-012-2609-8

Clinical Orthopaedics and Related Research®
A Publication of The Association of Bone and Joint Surgeons®

CLINICAL RESEARCH

Limb Lengthening and Reconstruction Society AIM Index Reliably Assesses Lower Limb Deformity

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Received: 19 March 2012 / Accepted: 7 September 2012

Abstract

Background Although several systems exist for classifying specific limb deformities, there currently are no validated rating scales for evaluating the complexity of general lower limb deformities. Accurate assessment of the complexity of a limb deformity is essential for successful treatment. A committee of the Limb Lengthening and Reconstruction Society (LLRS) therefore developed the LLRS AIM Index to quantify the severity of a broad range of lower extremity deformities in seven domains.

Questions/Purposes We addressed two questions: (1) Does the LLRS AIM Index show construct validity by correlating with rankings of case complexity? (2) Does the LLRS AIM Index show sufficient interrater and intrarater reliabilities?

Methods We had eight surgeons evaluate 10 fictionalized patients with various lower limb deformities. First, they ranked the cases from simplest to most complex, and then they rated the cases using the LLRS AIM Index. Two or more weeks later, they rated the cases again. We assessed reliability using the Kendall's W test.

Results Raters were consistent in their rankings of case complexity (W = 0.33). Patient rankings also correlated with both sets of LLRS AIM ratings ($r^2 = 0.25$; $r^2 = 0.23$). The LLRS AIM Index showed interrater reliability with an intraclass correlation (ICC) of 0.97 for Trial 1 and 0.98 for Trial 2 and intrarater reliability with an ICC of 0.94. The LLRS AIM Index ratings also were highly consistent between the attending surgeons and surgeons-in-training (ICC = 0.91).

Conclusions Our preliminarily observations suggest that the LLRS AIM Index reliably classifies the complexity of lower limb deformities in and between observers.

Each author certifies that he or she, or a member of his or her immediate family, has no funding or commercial associations (eg, consultancies, stock ownership, equity interest, patent/licensing arrangements, etc) that might pose a conflict of interest in connection with the submitted article.
All ICMJE Conflict of Interest Forms for authors and *Clinical Orthopaedics and Related Research* editors and board members are on file with the publication and can be viewed on request.
This work was performed at Cincinnati Children's Hospital Medical Center, Cincinnati, OH, USA.

Electronic supplementary material The online version of this article (doi:10.1007/s11999-012-2609-8) contains supplementary material, which is available to authorized users.

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Introduction

Limb length inequality and deformity are common in the general population with approximately 23% of people having a 1-cm or greater discrepancy [16] and many others with limb deformities that vary from mild varus to complex congenital deformities. Accurate assessment of the complexity of a limb deformity is essential for successful treatment [4, 7, 15, 20, 22, 23]. During the past two decades, several classification systems have been established and disseminated for assessing and categorizing specific joints, limb segments, and disease states (Table 1) [1–3, 5, 6, 8, 10–14, 17–19, 24, 25]. Paley [17] used a four-category classification system for femoral deficiency and deformity that is based on the factors that influence lengthening reconstruction of the congenitally deformed femur. He classified congenital femoral deficiency (CFD) into four groups, and although his scheme did allow for assessment of CFD, it failed to account for the broader clinical picture. Similarly, Jones et al. [13] proposed a classification scheme consisting of four morphologic and four radiographic groups in patients with congenital aplasia or dysplasia of the tibia with an intact fibula. Radiographic features were used to distinguish between anatomic variants, whereas morphologic classification was used to guide treatment. Before the treatment of fibular hemimelia, Catagni et al. [9] used a modified Dalmonte classification consisting of three types based on the structural features of the fibula. Birch et al. [5] proposed a new classification system for congenital fibular deficiency that was based on the functionality of the foot and leg length inequality. In a similar fashion, Pappas [19] proposed a nine-category classification system to cover the spectrum of femoral deficiency that later was modified by Paley [17]. The only rating system that has been developed evaluates femoral length discrepancies, but it has yet to be validated [18]. Currently, there are no validated rating systems for determining the severity of general lower limb deformities. A universal, valid, and reliable rating system for such assessment based on multiple criteria could be beneficial for comparing deformities within and between studies and for determining appropriate treatments.

The LLRS AIM Index was developed by a committee of the Limb Lengthening and Reconstruction Society through review of the literature and integration of concepts from multiple classification systems for disease-specific limb malformations (Table 1). The LLRS AIM Index measures the severity and complexity of a lower limb deformity through seven domains: location of the deformity, the length of the leg inequality, risk factors, soft tissue injury, angular deformity, infection or bone quality, and motion or stability of the joint. This index provides a uniform assessment of all deformities in a single limb and allows for pretreatment assessment of a broad range of lower extremity disorders. However, it is unclear whether this index is valid and reliable.

We therefore determined (1) whether the LLRS AIM Index shows construct validity by correlating with rankings of case complexity and (2) whether the LLRS AIM Index shows sufficient interrater and intrarater reliabilities.

Materials and Methods

First, members of the Limb Length Reconstruction Society (LLRS) performed a literature review outlining previously established classification systems (Table 1) and complications related to lower limb malformations to guide development of an index for rating the complexity of lower limb deformities. This index was greatly influenced by the scale published by Paley et al. [18] for rating the level of difficulty of femoral lengthening procedures, which accounted for angulation of the deformity, tibial lengthening, joint instability, knee flexion and deformity, joint osteoarthrosis, bone quality, soft tissue quality, and associated medical problems. The reliability and validity of their rating scale has not been evaluated, but many of the components have been incorporated into this index. The small study group of experts met on multiple occasions to determine the relevance and importance of specific items and features of this index. After repeated modification, the index was presented to the entire LLRS during a special session of the annual meeting. The final version of LLRS AIM Index contains seven pretreatment domains that are rated on a scale from 0 to 4 with increasing severity.

The domains are assessed through history and physical examination and include the Location and number of deformities, the Length of the leg inequality at maturity, Risk factors, Soft tissue coverage, Angular deformity, Infection and bone quality, and the Motion and/or subluxation of joints above and below the deformity (LLRS AIM). The scores are combined into a single index of complexity ranging from normal to high complexity (Table 2). The minimal LLRS AIM Index is 0 and the maximum is 28. An index of 0 is considered normal, an index 1 to 5 is considered to be of minimal complexity, 6 to 10 moderate complexity, 10 to 15 substantial complexity, and 16 to 28 high complexity. The relative weights of the score and overall level of complexity were based on summation of the literature and vetted through expert consensus opinion from the LLRS.

In this study, eight physicians (six attending orthopaedic surgeons and two orthopaedic surgeons-in-training) evaluated 10 fictional patients with various lower limb deformities (Table 3) (Appendix 1. Supplemental material is available with the online version of CORR®). The 10

Table 1. Review of classification systems for lower limb deformities

Study	Type	Characteristics
Congenital femoral deformity		
Aitken, 1969 [2]	A	Present femur head, shortened femur, normal acetabulum abnormal connection of femur head and neck to shaft, severe subtrochanteric varus
	B	Present femur head, shortened femur, normal acetabulum, small bony proximal tuft, no connection of femur head and neck to shaft
	C	Severely dysplastic acetabulum, shortened femur, bony proximal tuft, no connection of femur head and neck to shaft
	D	Absent femoral head and acetabulum, deformed, shortened femur, no proximal tuft
Pappas, 1983 [19]	I	Congenital absence of femur
Hefti et al., 2007 [12]	II	Proximal femoral and pelvic deficiency
	III	Proximal femoral deficiency with no osseous connection between femoral shaft and head
	IV	Proximal femoral deficiency with disorganized fibroosseous disconnection between femoral shaft and head
	V	Midfemoral deficiency with hypoplastic proximal and distal femur
	VI	Distal femoral deficiency
	VII	Hypoplastic femur with coxa vara and sclerosed diaphysis
	VIII	Hypoplastic femur with coxa valga
	IX	Hypoplastic femur with normal proportions
Paley, 1998 [17]	1A	Intact femur, mobile hip and knee, normal proximal femur ossification
	1B	Intact femur, mobile hip and knee, delayed proximal femur ossification
	2A	Mobile pseudarthrosis and knee, femoral head mobile in acetabulum
	2B	Mobile pseudarthrosis and knee, femoral head absent or stiff in acetabulum
	3A	Diaphyseal deficiency of femur, knee motion 45° or more
	3B	Diaphyseal deficiency of femur, knee motion less than 45°
	3C	Complete absence of femur
	4	Distal deficiency of femur
Torode & Gillespie, 1991 [25]	I	Congenital short femur, 20%–30% leg length discrepancy, valgus and hypoplastic knee with laxity, coxa vara, lateral bowing shaft
	II	Proximal femoral focal deficiency, 35%–50% leg length discrepancy, ankle at knee level, absent or deficient femoral head, neck, or shaft
Congenital tibial deformity		
Andersen, 1973 [3]	Cystic	Cysts in tibia with little anterior angulation or lateral bowing
	Dysplastic	Segmental dysplasia and hourglass constriction in bowed area of tibia
	Clubfoot	Clubfoot with anterior angulation of tibia and fibula; no bone constriction
	Sclerotic	Sclerosis in distal tibia with anterior angulation, little lateral bowing, and possible fracture in distal fibula
Jones et al., 1978 [13]	1A	Tibia not seen, hypoplastic lower femoral epiphysis
	1B	Tibia not seen, normal lower femoral epiphysis
	2	Distal tibia not seen
	3	Proximal tibia not seen
	4	Diastasis
Boyd, 1982 [6]	I	Pseudarthrosis with anterior bowing and tibial defects
	II	Pseudarthrosis with anterior bowing and tibial hourglass constriction
	III	Pseudarthrosis with congenital bone cyst, possible anterior bowing
	IV	Pseudarthrosis in sclerotic bone segment without tibial narrowing
	V	Pseudarthrosis of tibia with dysplastic fibula
	VI	Pseudarthrosis as intraosseous neurofibroma or schwannoma

Table 1. continued

Study	Type	Characteristics
Kalamchi & Dawe, 1985 [14]	I	Total absence of tibia, proximal migration of fibula head, marked hypoplasia of distal femur, reduction in femur distal metaphysis width and distal epiphysis ossification
	II	Distal tibial aplasia, small and cartilaginous proximal tibial fragment, slight proximal migration of fibula head
	III	Dysplasia of distal tibia with diastasis of tibiofibular syndesmosis, distal fibula prominence, foot in varus
Crawford, 1986 [10]	I	Medullary canal is preserved, cortical thickening possible
	II	Thinned medullary canal, cortical thickening, tabulation defect
	III	Cystic lesion with possible fracturing
	IV	Pseudarthrosis is present with tibial and fibular nonunion
Congenital fibular deformity		
Achterman & Kalamchi, 1979 [1]	IA	Fibular hypoplasia, proximal fibular epiphysis distal to tibial growth plate, distal fibular growth plate proximal to dome of talus
	IB	Fibular hypoplasia, proximal fibula absent for 30%–50% of its length
	II	Complete absence of fibula, only distal, vestigial fragment present
Stanitski & Stanitski, 2003 [24]	I	Nearly normal fibula
	II	Small or miniature fibula
	III	Complete absence of fibula
	H	Horizontal tibiotalar joint and distal tibial epiphysis
	V	Valgus (triangular distal tibial epiphysis)
	S	Spherical (ball-and-socket) tibiotalar joint and distal tibial epiphysis
	c	Presence of tarsal coalition
	1–5	Number of foot rays (medial to lateral)
Birch et al., 2011 [5]	1A	$<$ 6% inequality
	1B	6%–10% inequality
	1C	11%–30% inequality
	1D	$>$ 30% inequality
	2A	Functional upper extremity
	2B	Nonfunctional upper extremity
Congenital ankle and foot deformity		
Hamanishi, 1984 [11]	1	Neural tube defects or spinal anomalies
	2	Neuromuscular disorders
	3	Malformation syndromes
	4	Chromosomal aberrations
	5	Idiopathic congenital vertical talus
	5A	Intrauterine molding or deformation
	5B	Digitotalar dysmorphism
	5C	Familial occurrence of congenital vertical talus or oblique talus
	5D	Sporadic and unassociated
Catagni et al., 2000 [8]	1	Equinus varus, alteration in foot-to-tibia relationship
	2	Cavus foot, foot deformity without alteration in relation to tibia
	3	Fibular hemimelia, foot deformity with alteration in relation to tibia
	4	Posttibial pylon fracture deformity, foot deformity secondary to supramalleolar deformity and osteotomy
	5	Bone loss or absence in foot

fictional cases shown in the appendix were developed by the senior authors (JJM, SRR, SS) of this manuscript. All cases were based on real patients whose data were de-identified. The goal was to have a variety of diagnoses with varying degrees of complexity. The physicians were first asked to review the 10 cases and rank them from 1 to 10 in

Table 2. LLRS AIM Index for Limb Deformity*

Location (number of deformities per limb $\geq$ 10° angulation in separate planes and rotation all count as separate deformities)	
No deformity	0
One deformity	1
Two deformities	2
Three deformities	3
More than three deformities	4
Leg length inequality (estimate at skeletal maturity)	
0–2 cm	0
> 2–5 cm	1
> 5–10 cm	2
> 10–15 cm	3
> 15 cm	4
Risk factors (assess clinically)	
None	0
Age < 5 or > 40 years	Add 1 point
Smoker	Add 1 point
Obese	Add 1 point
Other disease (eg, diabetes)	Add 1 point
Soft tissue coverage	
Normal	0
Bruising/contusion	1
Scarring/open grade I	2
Poor coverage/open grade II	3
Inadequate coverage/open grade III	4
Angular deformity (measure and assign greatest primary deformity)	
0°–10°	0
> 10°–20°	1
> 20°–40°	2
> 40°–60°	3
> 60°	4
Infection/bone quality (select most severe)	
Normal	0
Osteoporotic	1
Dysplastic	2
Infection	3
Combination	4
Motion/stability of the joints above and below	
Normal	0
Decreased motion (< 60% of normal)	1
Subluxation of joint	2
Dislocation of joint	3
> 1 joint affected	4
LLRS AIM Index scoring	
Scores range from a minimum of 0 to a maximum of 28	
0 = normal	
1–5 = minimal complexity	
6–10 = moderate complexity	
11–15 = substantial complexity	
16–28 = high complexity	

* LLRS AIM is a mnemonic of the seven criteria that are required to determine the index.

Table 3. The 10 cases of lower limb deformities presented to raters

Case number	Diagnosis
1	Limb length inequality with infection
2	Blount's disease
3	Limb length inequality
4	Tibial hemimelia
5	Ollier's disease with femoral bowing and limb length inequality
6	Anterior lateral bowing of the tibia
7	Congenital femoral deficiency
8	Blount's disease
9	Congenital femoral deficiency with limb length inequality
10	Valgus in transplant

order of apparent complexity, as determined by their experience, with 1 being the easiest and 10 being the most complex. Rankings were performed without use of the LLRS AIM Index. After ranking the cases, they were asked to evaluate them using the LLRS AIM Index and to score each case individually. At least 2 weeks later, the same 10 cases were presented to the same eight raters for reassessment with the LLRS AIM Index.

The interrater reliability, or agreement between raters, of the initial complexity rankings of patients was evaluated with Kendall's W (coefficient of concordance) as a result of the ranked nature of the data. Linear regression was performed to determine whether patient rankings correlated with LLRS AIM scores, when controlling for rater. The interrater reliability, or agreement between raters of their LLRS AIM indices (for the first and second evaluations separately), was assessed by the intraclass correlation ($ICC_{2,k}$) from a two-way random effects ANOVA with absolute agreement [21]. The intrarater reliability, or agreement between the first and second evaluations, was assessed with the $ICC_{1,k}$ from a one-way ANOVA [21]. Kendall's W and ICC range from 0 (no agreement) to 1 (complete agreement). Statistical analyses were performed using SPSS software (Version 19.0; SPSS Inc, Chicago, IL, USA).

Results

Raters agreed on their rankings of the complexity of the patient cases ($W = 0.33$, $X^2_9 = 23.81$, $p = 0.005$). Patient rankings correlated with the LLRS scores for Trial 1 ($r^2 = 0.25$, $p < 0.001$) and Trial 2 ($r^2 = 0.23$, $p < 0.001$), when controlling for rater. The reliability between raters on LLRS AIM Index scores was shown by an $ICC_{2,k}$ of 0.97

Table 4. Intrarater reliability of the LLRS AIM Index with time

Rater	Number of patients	LLRS Trial 1	LLRS Trial 2	Intrarater reliability	
		Mean ± SD	Mean ± SD	$ICC_{1,k}$ (95% CI)	% perfect agreement
1	10	8.80 ± 4.87	8.20 ± 3.99	0.89 (0.59–0.97)	20%
2	10	7.10 ± 3.99	6.90 ± 2.92	0.94 (0.77–0.99)	30%
3	10	7.70 ± 3.92	8.60 ± 3.98	0.95 (0.80–0.99)	50%
4	10	7.10 ± 3.96	7.00 ± 3.20	0.95 (0.79–0.99)	0%
5	10	5.60 ± 2.76	6.40 ± 3.50	0.93 (0.74–0.98)	50%
6	10	6.80 ± 4.29	6.80 ± 4.29	1.00 (1.00–1.00)	100%
7	10	7.00 ± 3.77	6.40 ± 3.69	0.95 (0.80–0.99)	40%
8	10	7.60 ± 4.40	7.00 ± 3.71	0.96 (0.86–0.99)	40%
All raters	80	7.21 ± 3.95	7.16 ± 3.60	0.94 (0.91–0.96)	41%

(95% CI, 0.93–0.99) for Trial 1 and an $ICC_{2,k}$ of 0.98 (95% CI, 0.94–0.99) for Trial 2. The reliability with time of the LLRS AIM Index scores also was shown by a combined $ICC_{1,k} = 0.94$ (95% CI, 0.91–0.96) for all raters and $ICC_{1,k}$ values ranging from 0.89 to 1.00 for individual raters (Table 4). On average, raters gave the same score for both trials 41% of the time (Table 4).

When comparing levels of experience, the LLRS AIM Index ratings were highly consistent between the attending surgeons and surgeons-in-training ($ICC_{2,k} = 0.91$). Additionally, the agreement between the two surgeons-in-training ($ICC_{2,k} = 0.96$) was better than the average agreement between the six attending surgeons ($ICC_{2,k} = 0.86$). The surgeons-in-training ($ICC_{1,k} = 0.96$) also had slightly better intrarater reliability than the attending surgeons ($ICC_{1,k} = 0.94$).

Discussion

Accurate assessment of the complexity of a limb deformity is essential for successful treatment, and a validated rating scale for evaluating the complexity of general lower limb deformities currently does not exist. The purpose of this study was to describe the development and perform a preliminary assessment of the validity and reliability of a general limb deformity index for systematic pretreatment assessment of a broad range of lower extremity disorders (the LLRS AIM Index). The index accounts for seven domains, including the number of locations of deformity, leg length inequality, risk factors, soft tissue assessment, angular deformity, infection and bone quality, and motion and stability of the joints.

We caution readers of the limitations of our study. First, this study did not thoroughly assess the validity, the predictive value, or the usefulness of the LLRS AIM Index in guiding treatment. Future studies are necessary to determine whether the LLRS AIM ratings correlate with patient outcomes. Second, all patients, although based on true patient encounters, were fictionalized to protect patient identity among a unique group of patients (who may have seen many of the limb deformity experts) and for greater ease of the study. Third, the LLRS AIM Index does not differentiate between acquired and congenital deformities, although it can be used to evaluate both. Surgical correction for congenital deformities often is more difficult than for acquired deformities, with a higher complication rate [15, 22]. Fourth, development of the score was performed primarily through consensus and expert opinion. Relative weights of the score were extrapolated from the literature. Although the score was vetted on several occasions, including an open forum at the LLRS, there is an inherent difficulty in exactly determining the weighted components of each domain. Further analysis of the specific contribution of the seven domains to complexity scores in a larger study is necessary to validate the LLRS AIM Index and to determine whether specific scores can be used to effectively predict and guide treatment decision-making.

The first aim of this study was to perform a simplistic evaluation of the construct validity of the LLRS AIM Index by determining whether LLRS AIM scores correlate with rankings of case complexity. The raters showed statistically significant agreement in their rankings of the complexity of the patient cases. The patient rankings also correlated with their LLRS AIM scores, suggesting that higher LLRS AIM scores are indicative of more complex cases. There are currently no validated rating systems for evaluating the complexity of general lower limb deformities to compare the validity of the LLRS AIM Index. One study by Paley et al. [18] rated 29 patients who had undergone femoral lengthening over an intramedullary nail using a similar rating scale of complexity to guide treatment choice. Unfortunately, the variability of their patient scores was not reported, and the rating scale has not been validated.

Although further validation is necessary for the LLRS AIM Index, preliminary evaluation suggests that it is a valid measure of the complexity of nonspecific lower limb deformities.

The second aim of this study was to evaluate the interrater and intrarater reliabilities of the LLRS AIM Index. The LLRS AIM Index showed near perfect reliability between the eight raters and with time for assessing the complexity of the 10 cases. The raters also were more consistent at evaluating the complexity of patients with lower limb deformities when using the LLRS AIM Index than when simply ranking complexity. Although the LLRS AIM Index is a more complex rating scale, it was capable of producing much more repeatable results. Furthermore, this reliability was obtained despite the varying experience of our raters, which suggests that the LLRS AIM Index can provide for a simple, reproducible, common language of limb deformity.

The LLRS AIM Index is capable of assessing the entire clinical picture of a patient with any lower limb deformity through a common language that is simple enough for referring providers to use when discussing the complexity of a case with a specialist. A complete clinical examination is necessary, especially in a patient with a congenital limb deformity [23]. The LLRS AIM Index not only measures the amount of deformity present with uniform methodology in the affected limb, but also considers other factors, such as infection, soft tissue coverage, and chronic medical conditions, that are known to affect treatment. The LLRS AIM index allows for stratification of patients with higher versus lower complexity deformities in a reliable (repeatable) context, and potentially will be the basis for incorporating risk-adjusted complications in future research. To our knowledge, a rating system that can be used to assess the complexity of general lower limb deformities, whether acquired or congenital, does not exist. The LLRS AIM Index was developed to fulfill this need, and this study showed that the LLRS AIM Index is a highly reliable tool, consistent between raters and with time, for assessing the complexity of various lower limb deformities.

Acknowledgments We thank the members of the Limb Lengthening and Reconstruction Society and John G. Birch, MD, David J. Klimaski MD, Viral Jain MD, Nathan Faulkner MD, and Esther Cheng BS for their guidance and assistance with this research project and article.

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