

Idiopathic infantile asymmetry, proposal of a measurement scale

Heike Philippi^{a,*}, Andreas Faldum^b, Holger Bergmann^a,
Tatjana Jung^a, Bianka Pabst^a, Angela Schleupen^a

^aUniversity Children's Hospital, Johannes Gutenberg-University, Langenbeckstr. 1, 55101 Mainz, Germany

^bInstitute for Medical Biometry, Epidemiology and Informatics, Johannes Gutenberg-University, Mainz, Germany

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Abstract

To evaluate the development of idiopathic infantile asymmetry and the efficacy of therapeutic interventions, spinal scoliosis can be quantified on the basis of radiographs. For obvious reasons, use of this technique is limited. Here we present a clinical method to describe and quantify infantile asymmetry.

For item selection, spontaneous movements (SMs), reactive movements (RMs) and length differences were video recorded in 30 infants (median age 10 weeks, range 6–16) with variable degrees of asymmetry.

Within these three categories, reactive movements elicited by head turns to the right and left side in the prone and supine position emerged as reliable parameters reflecting trunk convexity and cervical rotation deficits. Six-point scales were developed for both measurements and added to form final scales.

Consistency and interobserver reliability were evaluated in another 20 infants (median age 9 weeks, range 6–15) with variable degrees of asymmetry. Statistical analysis indicated good reliability and consistency of the testing method with an intraclass correlation coefficient of 91.5% (Cronbach alpha 0.84).

* Corresponding author. Tel.: +49 6131 172443; fax: +49 6129 512545.

E-mail address: philippi@kinder.klinik.uni-mainz.de (H. Philippi).

Conclusion. During the first months of life, idiopathic infantile asymmetry can be clinically assessed using a highly consistent and reliable measurement scale describing degrees of trunk convexity and cervical rotations deficit.

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Infantile idiopathic scoliosis was first described as an entity by James [1]. “Infantile” was defined as under the age of 4 years. Scoliosis is often present at birth and becomes more prominent during the first months of life. It is often combined with secondary deformities including plagiocephaly [2–6], facial asymmetry [7–10], strabism [8], torticollis [7,8,11], subluxation or dysplasia of the hip [7,9,11,12], unilateral lumbodorsal kyphosis [11], and asymmetric foot position [9–11]. The terms “moulded baby syndrome” [3,9,13], “seventh syndrome” [14], “congenital predilection syndrome” [10,15], and “turned head-adducted hip-truncal curvature (TAC) syndrome” [16] have been proposed to describe the same condition. In some studies, infants with idiopathic scoliosis are subsumed under the term “torticollis” [8,9,17]. To cover all functional and deformational aspects, we prefer to the more common “infantile scoliosis”, the descriptive designation “idiopathic infantile asymmetry” which encompasses the entire spectrum of deformities associated with preferential position [18,19]. Both endogenous and exogenous factors are thought to cause idiopathic infantile asymmetry. Exogenous factors are intrauterine and/or postnatal constraint and birth injuries [3,6,9,10,13,17]. Genetic disposition, as shown by a family history of scoliosis, rapid growth, reduced muscle tone, and reduced motor activity are presumed to be endogenous factors [4–6,11]. Contractures, followed by asymmetric structural growth and weight-bearing deformities may produce a vicious cycle [6,11].

The prognosis of idiopathic infantile asymmetry varies considerably. In retrospective studies of patients with infantile scoliosis or torticollis, the rate of either persistent or progressive scoliosis ranged between 10% and 50% [2–8,20–22]. In these studies, the severity of torticollis was based on the clinical impression, except in the most severe form of torticollis. The latter was defined as a passive rotation deficit of the head of more than 30° [8,9,17]. The curvature of the scoliosis was evaluated by measuring the rib-vertebra angle difference in spine radiographs [23]. An angle above 20° and a passive rotation deficit of the head of more than 30° are risk factors for non-resolving scoliosis [2,7,8,20,23]. However, neither criterion reliably predicts persistent or progressive scoliosis [7,8,21,22]. Furthermore, X-ray exposure limits the use of spine radiographs for predictive and investigative purposes. As a consequence, studies on effectiveness of conservative, orthotic and operative interventions in idiopathic scoliosis have focused to children older than 1 year [24–26]. In contrast, there is little scientific evidence for the effectiveness of conservative therapeutic interventions below that age, such as stretching, contralateral posturing, handling, osteopathy, craniosacral therapy and physiotherapy.

In order to critically evaluate these early intervention methods, we developed a clinical procedure to quantify idiopathic infantile asymmetry on the basis of standardized examination and video recording of movements. Further studies with repeated measure-

ments will give an insight into the spontaneous development of idiopathic infantile asymmetry. With this, the method will possibly provide a noninvasive predictive tool to differentiate at an early stage between progressive and resolving idiopathic infantile asymmetry. Because of developmental changes during infancy, we had to focus on infants between 6 and 16 weeks old. The lower limit of 6 weeks was chosen because many asymmetric movement patterns disappear spontaneously during the first 6 weeks of life [3,14]. The upper limit of 16 weeks was chosen because beyond that age infants develop the ability to voluntarily turn into a stable side position by full body rotation, thus masking their torticollis and scoliosis. The chosen age period allows for early intervention [2,7,21].

1. Methods

1.1. Selection of items and composition of the scale

1.1.1. Subjects

From March 1999 to September 2000, 31 infants (16 female) at a median post term age of 10 weeks (range 6 to 16 weeks) were recruited from three paediatric practices in Mainz, Germany. The paediatricians were asked to refer infants with three different movement patterns: (a) symmetric, (b) slightly asymmetric, (c) asymmetric in whom the clinical course (progressive or resolving) was uncertain. These infants were judged to be otherwise healthy as judged on the three first routine examinations (after birth, at 3 to 10 days, and at 4 to 6 weeks of age). All infants were neurologically examined by the first author and two experienced physiotherapists (co-authors). The neurological examination included the evaluation of spontaneous movements, orientation responses, positional reactions, and reflexology according to Vojta [27–29]. Inclusion criteria were term birth, absence of a neurological disease, notably hemiplegia, at outset and at 10–12 months of life, a prompt orienting response to optic and acoustic stimuli and informed parental consent. The study protocol was approved by our ethic committee. The final study group consisted of 30 infants (16 male).

1.1.2. Procedures

Out of a list of conventional screening methods for infantile asymmetry [27–31], the following were selected as diagnostic candidates: spontaneous movements (SMs), reactive movements (RMs), and metric asymmetry by inspection of the superior spinae iliacae (SIAS) and the malleoli mediales. SMs were evaluated in an initial 60 s period in the prone and supine position, respectively. RMs were stimulated by an orienting head turn to the right and left side (RMs-right/left) in the prone and supine position, and by the Landau, Galant, and Piper-Isbert manoeuvres (RMs-Landau, RMs-Galant, RMs-Piper-Isbert). The SMs and RMs were scored for five movement patterns: “trunk convexity, cervical rotation deficit, cervical lateral flexion, preferential head position and oblique trunk position”. Movement patterns and their application to the procedures are shown in Table 1.

In order to videotape these procedures, the infant, together with a physiotherapist, was placed supine and prone on a prewarmed mattress with the video camera above

Table 1
Movement patterns and procedures

Manoeuvres	Criteria				
	<u>Trunk convexity</u>	<u>Cervical rotation deficit</u>	<u>Cervical lateral flexion</u>	<u>Preferential head position</u>	<u>Oblique trunk position</u>
	Curvature formed by the thoracolumbar spine	Movement deficit of the vertical plane of the head (face) in relation to the vertical plane of the trunk (breast, back)	Deviation of the vertical axis of the head and cervical spine on the vertical axis of the thoracolumbar spine	Predominance of a head turn to one side which looks comfortable for the infant	Torsion of the thoracolumbar spine which is indicated by a preferential turn of the pelvis to one side (see Fig. 3b)
SMs	×	×	×	×	×
RMs-right/left	×	×	×	×	×
RMs-Landau	×		×		
RMs-Galant	×				
RMs-Piper-Isbert	×		×		×

SMs=spontaneous movements, RMs=reactive movements.

them at a distance of 2 m (Fig. 1a). For the Piper-Isbert manoeuvre, the infant was vertically suspended with the camera in a horizontal position at a distance of 3 m at the same level (Fig. 1b). Observers and parents stood behind the camera. The environment was kept monotonous and the light intensity was uniform and moderate. For the assessment of left–right differences of the SIAS and the malleoli, the infant was placed in the supine position with both legs extended. Thereafter, we waited until the infants had reached a state of active wakefulness without crying [32]. Recording of the SMs and RMs was started with the infant's head held shortly in the middle supine position. For the next 60 s, the SMs were recorded. Then a head turn was induced by presenting noises, toys, or the observer's face, and moving them from one side to the other. After at least two turns to each side, the infant's head was held shortly in the middle prone position and the same procedures were repeated. Finally, the Landau and Galant and Piper-Isbert manoeuvres were performed. The videos were recorded with a digital camcorder (Digital Video Camera DCR-PC100E, Sony, Oehling Mainz, Germany) and stored.

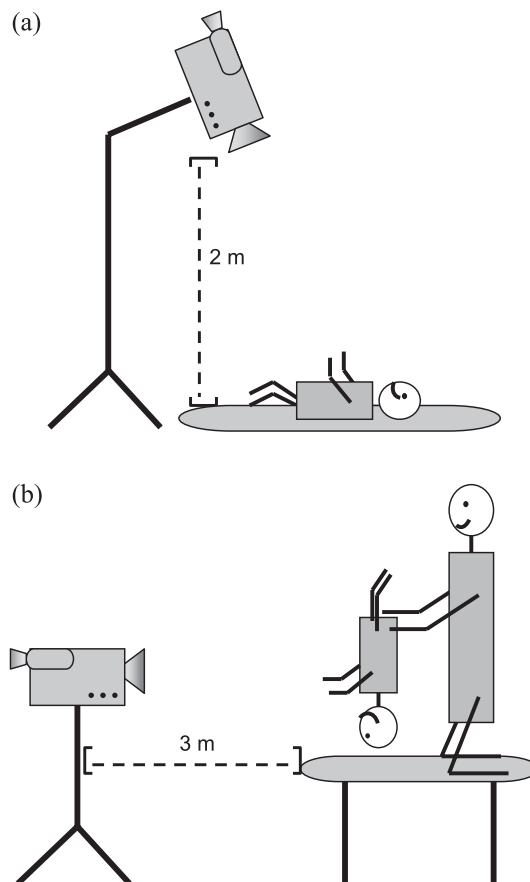


Fig. 1. (a) Vertical camera position. (b) Horizontal camera position.

The scoring was performed by five independent observers. Following the recommendations of Streiner and Norman [33], three to eight point numerical scales without description of the different categories were tested. Finally, scoring difficulties were analysed to optimise videotaping, examination techniques and scoring protocol. Once the scoring technique had been elaborated, the final items were categorized (Fig. 2).

a

Categories		Spine Pictograms
1 Point	No convexity or equal convexity of the spine.	or)(
2 Points	Slightly differing convexity of the spine.) (
3 points	Clearly differing convexity of the spine, resolution possible.) (
4 points	Convexity of the spine can be resolved to a straight line.	(
5 Points	Convexity of the spine can be resolved to a flat curve.	((
6 Points	Convexity of the spine can not be resolved.	((

b


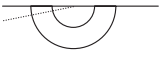




Categories		Rotation pictograms
1 Point	Free rotation	
2 Points	Slight head rotation deficit, with a slight resistance during rotation.	
3 Points	Clear head rotation deficit, preferential head position.	
4 Points	Restricted head rotation (inner arrow), which may be intermittently overcome, working area = external arrow.	
5 Points	Restricted head rotation (inner arrow), which may be intermittently overcome (dotted arrow), working area = external arrow.	
6 Points	Restricted head rotation (inner arrow), which may barely be overcome, working area = external arrow.	

Fig. 2. (a) Definition of the six categories of trunk convexity. (b) Definition of the six categories of cervical rotation deficits.

1.2. Interobserver reliability and homogeneity of the items

Interobserver reliability and consistency of the final scale were tested in another 20 infants (9 female) at a median post term age of 9 weeks (range 6 to 15 weeks). These infants were recruited from January to June 2001 using the same selection criteria as in Section 1.1.

1.2.1. Statistics

Interobserver reliability and consistency of this multi-item score were assessed using intraclass correlation and Cronbach alpha. In order to prove that the intraclass correlation coefficient is larger than 0.6 at a significance level of 5%, 20 patients were necessary to be examined by five raters achieving a power of 80% while expecting an intraclass correlation coefficient of 0.80 [34]. The estimation of the intraclass correlation coefficient is based on the maximum likelihood method with patients and raters figuring as random factors. The interpretation of Cronbach alpha according to DeVellis [35] is as follows: <0.60 unacceptable, 0.60–0.65 undesirable, 0.65–0.70 minimally acceptable, 0.70–0.80 respectable, 0.80–0.90 very good, and >0.90 redundant. All analyses were done using SPSS 10.0.

2. Results

2.1. Selection of the items and composition of the scale

The tested preliminary examination techniques are listed in Table 1. From these, the items “preferential head position” and “cervical rotation deficit” yielded similar results in 25 of 30 cases. Since, in addition, the criterion “preferential head position” was felt to be covered by assessing the RMs-right/left, it was deleted. “Oblique trunk position” was demonstrated in only 7 of 22 asymmetric infants and was also deleted. “Cervical lateral flexion” turned out to be an inconstant movement pattern that compensated for better cervical rotation and was abandoned. The responses of the infants to the Landau, Galant and Piper-Isbert manoeuvres are complex three-dimensional movements and could not be reliably scored on a two-dimensional plane. They were excluded from the final protocol.

Thus “trunk convexity” and “cervical rotation deficit” emerged as the only items, which allowed standardized videotaping and scoring in the prone and supine position.

Morphometric items, i.e. measuring SIAS and malleolar differences in asymmetric infants, also proved to be ineffectual and were discarded. They require measurements in the millimeter range and were compounded by the inability of very young infants to completely extend their knees.

To quantify the degree of asymmetry, the two remaining criteria “trunk convexity” and “cervical rotation deficit”, both in the supine and prone position, a six-point scale ranging from 1 (symmetric) to 6 (asymmetric) was employed (Fig. 2). This six-point scale avoided overly coarse categorization without exceeding the discriminatory abilities of the five individual observers.

The final asymmetry scale given to an individual infant consists of the sum of one to six points accrued from the above four items: “trunk convexity supine”, “trunk convexity

Supine			
Trunk convexity	1	-	6 points
Cervical rotation deficit	1	-	6 points
Prone			
Trunk convexity	1	-	6 points
Cervical rotation deficit	1	-	6 points
Total score	<hr/>		4 - 24 points
	symmetric - asymmetric		

Fig. 3. Composition of the final measurement scale for idiopathic infantile asymmetry in infants at a post term age of 6 to 16 weeks.

prone”, “cervical rotation deficit supine”, “cervical rotation deficit prone”. A final score of four points represents the “symmetric”, and a score of 24 points the “asymmetric” end of the scale (Fig. 3). For better illustration, a picture series of category 6 of “cervical rotation deficit supine” and of category 5 of “trunk convexity prone” is shown in Fig. 4.

Once the measurement scale was composed, a detailed description (Fig. 2) and demonstration video film for each of the 2 six-point categories for “trunk convexity” and

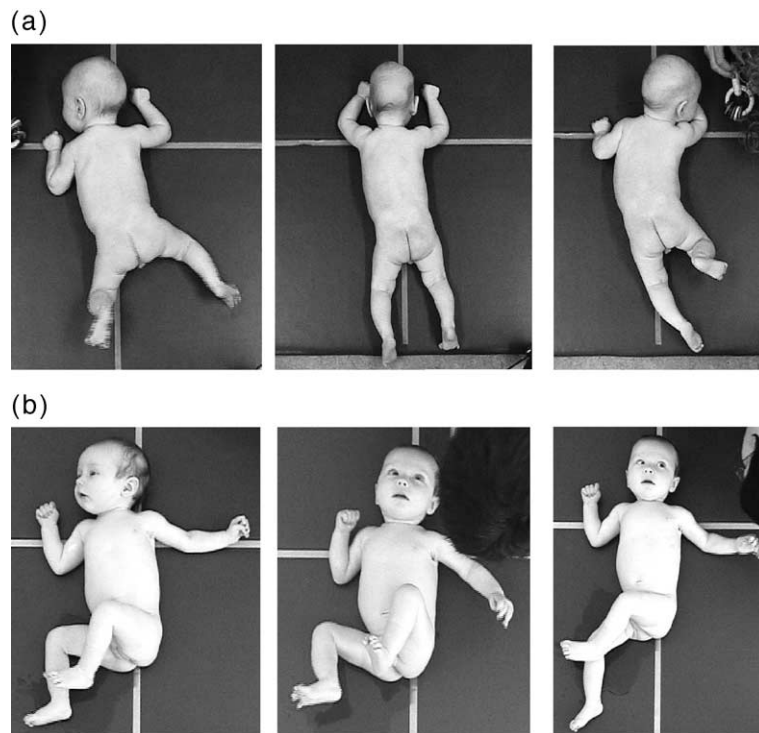


Fig. 4. (a) Picture series of an infant with a trunk convexity of category 5. (b) Picture series of an infant with a cervical rotation deficit of category 6.

variance of the score was 0.6% with a 95% confidence interval of 0.0–2.0%. Moreover, a Cronbach alpha of 0.84 signifies a very good internal consistency of the items without redundancy.

3. Discussion

The goal of our investigations was to develop an easy to perform, reliable clinical measurement scale for idiopathic infantile asymmetry. The assessment of spontaneous movements has been regarded as the gold standard for the examination of infantile development [36]. However, extended video recording is necessary to reliably assess general movement patterns of infants at risk for cerebral palsy [36], and the same is true for children with idiopathic infantile asymmetry. Furthermore, the main diagnostic feature of idiopathic infantile asymmetry is the asymmetric limitation of mobility, which is more easily detected after exogenous stimulation. We thus had to rely on reactive movements. RMs-Galant, RMs-Landau, and RMs-Piper-Isbert turned out to be inappropriate for precise videoscoring and were discarded. We rather focused on a scale composed of RM-right/left items, which allowed visualization of movement restrictions and were not disturbingly invasive. The final selection of the two relevant criteria “trunk convexity” and “cervical rotation deficit” and of a six-point numerical scale resulted from a critical discussion among all observers after repeated evaluation of the 30 videos.

Following the recommendations of Streiner and Norman [33], the validity of this scale ought to be checked by correlation analysis with similar scales. However, comparison with the two currently available approaches to measure “trunk convexity” and “cervical rotation deficit” is not possible. “Trunk convexity” is the clinical counterpart of “scoliosis” which is radiologically determined by the rib-vertebra angle, but obtaining radiographic for a comparative study is not possible for ethical reasons. The same holds true for scoliosis measurement by computer tomography and magnet resonance tomography, which requires sedation of infants and toddlers. Surface topography and laser scanned 3D torso topography are promising non-radiographic methods which will allow for a quantification of deformities. Nevertheless, these techniques depend on cooperation to minimize the postural sway and are therefore not suitable for infants and toddlers [37].

The elicited head rotation deficit in our study corresponds to the orthopaedic assessment of a passive head rotation deficit. Since the latter was categorized as “below or above 30°” [8,9,17], it does not allow for comparison with a graded continuum.

In view of this limitation, we had to rely on face and content validity. Statistical evaluation showed high Cronbach alpha and intraclass correlations proving consistency of the scale and interobserver reliability.

The proposed infantile asymmetry scale is easy and save to perform and precise enough to be used for epidemiological studies with long-term follow-up to determine whether idiopathic infantile asymmetry has functional sequelae later in life. Cut points in the asymmetry scale will emerge to predict the need for therapeutic intervention. Not until these studies are performed will we be able to judge the predictive power of the measurement scale to differentiate between progressive and nonprogressive forms of idiopathic asymmetry. If progression becomes evident, radiographs must be obtained to

visualize a segmental spine deformity and to calculate the rib-vertebra angle difference or Cobb's angle. The latter were shown to be currently the best predictors for progressive scoliosis [24,38,39]. An age-modified clinical video scale for older infants and toddlers is currently elaborated by our study group.

The scale opens the way for interventional studies, i.e. to prove or disprove the efficacy of physiotherapeutic, osteopathic, positional, or other therapies. As an extra benefit, the stored videotapes will document the presence and development of dysmorphic sequelae such as plagiocephaly, facial and thoracic asymmetry, preferential foot position, preferential head position, unilateral hip adduction, cervical lateral flexion, and oblique body position.

In conclusion, we present a standardized and practical measurement scale to quantitate idiopathic infantile asymmetry by assessing defined reactive movements in infants aged 6 to 16 weeks. The scale may serve as a basis for long-term follow-up studies for infants with idiopathic infantile asymmetry and for the evaluation of treatment effects in children with idiopathic infantile asymmetry. Repeated measurements will possibly identify progressive lesions that need more invasive diagnostic methods and appropriate orthopaedic intervention.

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