Follow-up of children with congenital clubfoot. Development of a new evaluation instrument.

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Development of a new evaluation instrument

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To my mother, Marie, for showing what patience, acceptance and tolerance means.

And

To my father, Co, for learning me to question everything and not to stagnate.

Contents

List of papers

This thesis is based on the following papers, which will be referred to in the text by their Roman numerals I–V.

I. Andriesse H, Hägglund G, Jarnlo G-B. The Clubfoot Assessment protocol (CAP); Description and reliability of a structured multi-level instrument for follow-up. BMC Musculoskeletal Disorders 2005, 6:40

II. Andriesse H, Roos EM, Hägglund G, Jarnlo G-B. Validity and responsiveness of the Clubfoot Assessment Protocol (CAP). A methodological study. BMC Musculoskeletal Disorders 2006, 7:28

III. Andriesse H, Hägglund G. Comparison of serial casting versus stretching technique in children with congenital idiopathic clubfoot. Evaluation of a new assessment system. Acta Orthopaedica (accepted for publication).

IV. Andriesse H, Westbom L, Jarnlo G-B, Hägglund G. Developmental disorders of motor function in children treated for idiopathic clubfoot? Submitted.

V. Andriesse H, Hägglund G, Isberg P-E. Reliability of motion analysis in children treated for congenital clubfoot. Submitted.

Abbreviations

ADHD	attention deficit hyperactivity disorder
AFO	ankle foot orthosis
CAP	clubfoot assessment protocol
CI	confidence interval
CPH	Copenhagen stretching and manipulation method
DCS	Dimeglio classification system
DMA	developmental motor ability
ES	effect size
ESISGC	Evaluation System of the International Clubfoot Study Group
FAO	foot abduction orthosis
HDJ	Hospital of Joint Diseases
ICF	international classification of functioning, disability and health
IQR	inter quartile range
κ	kappa value
KAFO	knee ankle foot orthosis
LOA	limits of agreement
MABC	movement assessment battery for children
PCT	Ponseti casting technique
Po	exact percentage agreement
Po-1	agreement within one level of difference
SD	standard deviation
SRM	standardized response mean
SSEP	somatosensory evoked potentials

Introduction

Congenital talipes equinovarus, better known as clubfoot, is a complex foot deformity with a high variability in severity. The incidence is about one in 1000 live births in a Caucasian population [1, 2]. The foot has a typical appearance (Figure 1) with the forefoot in adduction and the hind foot in varus and equinus.

Commonly a fourth component, excavatus, is included [2].

In general, tendons, muscles and ligaments on the posteromedial side of the foot are contracted. Imbalance is found in muscle function between eversion/inversion and dorsal flexion/plantar flexion [2].

It is still not known which factor(s) specifically cause the clubfoot deformity. Genetic factors seem to be of importance. Engell et al. [3] showed a clear genetic connection and also emphasized a multifactorial cause as did Dietz [4]. The studies by Heck et al. [5] and Ester et al. [6] continue to bring new evidence on the genetic aetiology. Environmental factors such as viral infections and seasonal variation, drugs and smoking [7, 8] have shown strong association with the development of clubfoot. One of the oldest explanations for the cause of clubfoot has been intrauterine narrowness. This cause was actualized in the nineties with a sudden increase of clubfoot incidence which could be explained by early amniocentesis [9, 10] disturbing acquisition of amnion fluid and causing decreased fetal movement during a key phase of foot and ankle development.

Figure 1. Child with bilateral clubfeet (Photo H. Andriesse)

Pathology

The most important deformity factors in clubfoot are the subluxation of os naviculare medially, medial deviation of the head and neck of talus and equinus of the calcaneus [11, 12] (Figure 2).

The cuboid bone may, as a result of the plantar flexed and internally–medially rotated os calcaneus, be displaced medially [13, 14].

Intrinsic primary growth disorder causing the formation of a small hypoplastic bone and, subsequently, a smaller foot may also be a part of the clubfoot pathology [15, 16].

Changes in muscle fibre histology with an increase of type II fibres have been shown [17, 18]. Loren et al. [19] found, in 50 percent of biopsies from clubfeet, abnormal muscle fibre morphology, classified as congenital fibre-type disproportion or fibre-size variation. A significantly greater incidence of recurrent equinovarus deformity requiring re-operation was also registered in these cases. In contrast Herceg et al. [20] found no evidence for the theory that a neuromuscular abnormality may be important in the aetiology of idiopathic clubfoot.

Several studies support the hypothesis that an increase of the cells and collagen fibres of the medial ankle ligaments of club feet appears to be the site of the earliest changes [21-23]. These connective tissue structures had lost their spatial orientation and become contracted. In stillborn children with clubfoot and before the third trimester of gestation, myofibroblast-like cells seemed to create a disorder of the ligaments resembling fibromatosis

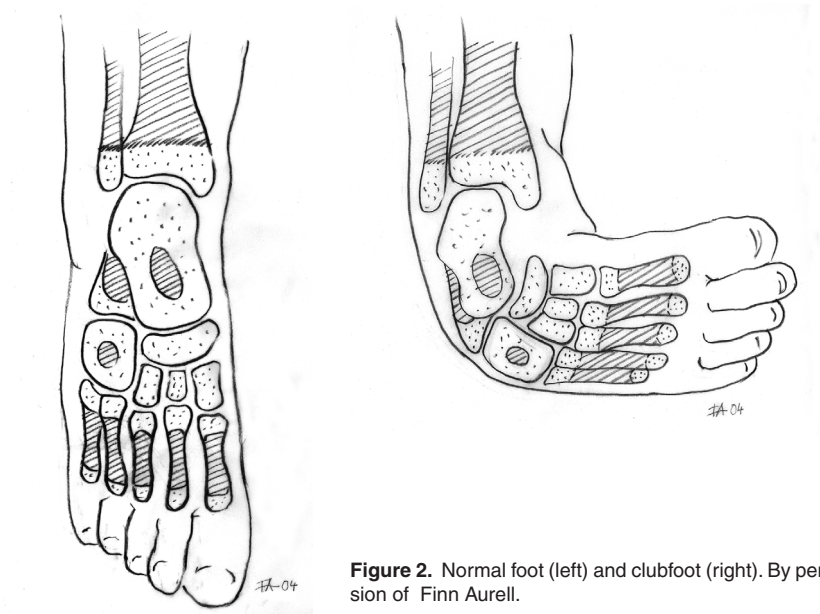

Figure 2. Normal foot (left) and clubfoot (right). By permission of Finn Aurell.

leading to contraction and resulting in typical clubfoot deformity [22, 23]. Furthermore, an increase of intercellular connective tissue and decreased non-collagen protein synthesis were found in feet classified as very severe [24].

The hypothesis of a neuromuscular disorder causing muscular imbalance is advocated by several authors. Increased reflex activity from the gastrocnemius muscle was shown by Trontelj and Pavlovcic [25], indicating an increased muscle tone. Naadem et al. [26] investigated SSEPs measuring the conduction pathways from the periphery to the brain in 44 children (95 feet) with surgically corrected club feet. Overall, 44 of 95 feet (46%) showed abnormal SSEPs or motor electrophysiological tests. Neurological abnormality was related both to the severity of the deformity and the surgical outcome. One opinion was that the changes found in muscular structure were secondary and that abnormal innervation was the primary factor causing clubfoot [27]. Feldbrin et al. [28] investigated 52 children with electrophysical tests. Only nine children showed normal values. They also found a clear relationship between pathological neurological findings and outcome results. Muscular imbalance seems to play an important role in the development and prognosis of clubfoot.

New born children with clubfoot have decreased joint mobility causing difficulties for the child to actively evert and dorsal flex its foot. Also extreme equinovarus–adduction position causes elongation of the muscles on the lateral side of the clubfoot weakening the contractility of the muscles [29].

Clubfoot treatment

The aim of the clubfoot treatment is a foot that in adult age will be well functioning and enabling participation in daily activities, outdoor and sport leisure, without troublesome pain and stiffness [2]. Furthermore the aim is a cosmetic foot that is acceptable for the patient. For obtaining these goals the foot needs a mobility that makes walking, running and jumping possible without excessive compensation mechanisms in the knee- and hip joints. Alignment of the forefoot in relation to the mid and hind foot, the hind foot in relation to the tibia and at least 10 degrees of foot dorsal flexion is of importance for proper distribution

of weight-bearing loading. A certain degree of motion at the subtalar joint is needed for neutralizing rotational forces on the foot, knee and hip joints [30]. Besides sufficient mobility, function of the muscles in and around the foot should be strong enough and well balanced/coordinated to enable stabilization, foot progression control and push-off in activity.

Treatment of the clubfoot can be divided into two phases. The first phase is correction of the deformity, which can be made by serial casting, stretching and manipulation methods and with soft tissue surgery. The second phase aims on maintenance of the obtained correction.

Correction phase

Serial casting

The most used technique is the one described by Ponseti [2]. The method consists of serial casting with above–knee casts preceded by gentle manipulations and correcting simultaneously the clubfoot components. If equinus deformity still exists after 5–10 casts a percutaneous Achilles tenotomy is performed. Good rapid initial results and long-term outcomes with minimal need of surgical correction have been shown. The need for an Achilles tendon lengthening varies around 85% and there is a minimal need (~ 2.5%) for extensive corrective surgery [31].

Stretching and manipulation methods

The so called "French method" includes daily intensive physiotherapist-led stretching and manipulation supported by taping and the use of a continuous passive motion machine [32-34]. Outcome results are varying [34-36]. The need for extensive operative correction varies between 23% and 49% [34, 36].

The "Copenhagen method" is a combination of daily intensive physiotherapist-led stretching and manipulation supplemented with an adjustable splint during the first three months. Treatment starts within two weeks after birth. The aim is to decrease the need for excessive invasive surgery [37-39]. At the age of three months the need for complementary surgery is assessed. In most cases (about 80%) a posteromedial release and an Achilles lengthening is applied [36, 39].

Surgery

During the 70's surgical correction of the infant clubfoot developed rapidly as techniques were refined and casting or stretching results poor. These surgical interventions include various degrees of soft tissue release [40, 41] correcting the clubfoot deformity posteriorly, medially and sometimes also laterally. In the last 20 years, the approach to clubfoot surgery has changed [42]. To avoid excessive tissue scaring all necessary surgical correction should be done during one operation [43] and a stepwise procedure is advocated [44]. Particular attention has been paid to the relationship between the age at operation and the outcome more than four years later. The results were superior when operation was undertaken early [45, 46]. Short-term results were often good, but the long-term outcomes after extensive surgery are less positive and related to an increased risk for pain, stiffness and arthrosis later in life [47-50]. Nowadays surgery is advocated as the last alternative in correction treatment.

Maintenance phase

Orthosis treatment

For maintaining the attained correction, orthosis treatment is generally advocated. Many different kinds of orthosis concepts and treatment regimes are used [2, 34, 38, 45, 51]. The Ponseti method, for instance, advocates a foot abduction orthosis (FAO) which is worn continuously the first 2–3 months and thereafter at night until the age of 2–4 years. The Copenhagen method uses a dynamic Knee Ankle Foot Orthosis (KAFO) [38]. This is initially used the whole day and after walking debut night time until the age of 3 years. The French method uses so called flexible splints as long as necessary [34]. Studies on the effects of orthosis have shown that non-compliance to orthosis usage is one of the most important factors correlated with relapses [31, 52-55]. To my knowledge no outcome studies have compared the different orthoses in effectiveness (including application time), user friendliness and

compliance. The article by Miura et al. [56] is one of the rare studies on the effect of an orthosis construct on foot mobility.

Clubfoot relapse

Nowadays, with a relapse of the deformity, renewed serial casting is often advocated [57]. If a muscular imbalance is diagnosed as a cause of the relapse, transfer of the m. tibialis anterior is the primary intervention. Sometimes this is completed with an Achilles tendon lengthening and /or a posteromedial release. Outcome reports are mainly positive [58-60]. Furthermore, osteotomies of the os calcaneus (relapse of varus deformity) or os cuboideum (relapse of adductus deformity), such as Dwyer [58-60], can be needed when bone deformity is part of the relapse problem. In the resistant clubfoot deformity the Iliazarov method, with external fixation and gradual distraction, is reported as an alternative to conventional surgery. Varying outcome results are though reported: negative [61-63] and positive [64, 65] articles.

Measurement properties in clubfoot assessment

Focus is shifting from disease severity to assessments on impairments, disability and participation problems according to the International Classification of Function, Disabilities and Health (ICF) [66]. As treatment interventions and goals cannot be derived from only disease severity classification the need for assessment instruments based on the ICF guidelines is advocated.

Also the demand for evidence-based outcome have put pressure on the development of reliable and valid assessment instruments. Clinimetrics focus on the quality of measurement instruments [67, 68] based on clinical judgement and experience in relation to outcome and what is meaningful for the patient and the clinician. It includes also the quality of performance of the actual measurement such as the assessors experience and quality of study sample. Firstly the aim of the instrument should be stated as it concerns the constructs or aspects one wants to measure. Secondly, the purpose should be made clear such as if the instrument is used for evaluation or cross sectional study. A checklist has been developed facilitating the systematic evaluation of clinimetric properties in measurements instruments [69] It contains:

- *Validity:* Refers to the degree to which the instrument measures what it is intended to measure.
- *Reproducibility:* The extent to which an instrument is free of measurement error.
- *Responsiveness* (a form of longitudinal validity): Refers to an instruments ability to detect change over time.
- *Interpretability:* This is defined as the degree to which one can assign a qualitative meaning to a quantitative score.
- *Feasibility:* Refers to administration time and ease of scoring.

A large amount of assessment instruments can be found [70]. Most instruments aim towards classification or cross-sectional outcome. They often concentrate on variables belonging to the domains of body functions and structures [71-77]. Only one patient-based outcome instrument has been specifically developed for outcome [78]. Variables on activity and participation are sparsely used and addressed generally [76, 79-81].

In general these instruments are based on a variety of clinical and functional criteria, which are scored separately and then aggregated to a sum score. The aggregate score is then assigned a categorical ranking that ranges from e.g. excellent to poor. The developers of different clubfoot assessment systems have chosen different outcome criteria, assigned different weights to each criterion, and accorded different ranges of values to each categorical ranking. This precludes valid comparison between studies as categorical rankings cannot be relied on to provide meaningful comparisons either within or between cohorts of patient [82, 83] .

A Medline literature search (1987–2007) using the following keywords in different combinations: clubfoot, assessment, outcome, reliability, validity, classification and evaluation was done. Inclusion criteria were: studies only in English that were designed to specifically describe clubfoot instru-

Table 1. Summary of clubfoot instruments with documented studies on their clinimetric properties in Medline

Instrument	Reliability	Content validity	Construct validity	Responsive-ness	Floor/ceiling effect
Dimeglio (75)	Yes (84, 85)	Yes (75)	No	Yes (53, 84)	No
Pirani (77)	Yes (85) [a]	Yes (77) [a]	Yes (86)	Yes (53)	No
Laaveg–Ponseti (76)	Yes (87) [a]	No	Yes (82)	No	No
McKay (80)	No	No	Yes (82)	No	No
Magone (88)	No	No	Yes (82)	No	No
Ghanem (89)	No	Yes (89)	Yes (82)	No	No
Munshi (82)	Yes (82)	No	Yes (82)	No	No
ESICSG (90)	Yes (91) [a]	Yes (90)	No	No	No
Ponseti–Smoley (92)	Yes (84)	No	No	No	No
Harold–Walker (93)	Yes (84)	No	No	No	No
Catteral (73)	Yes (84)	No	No	No	No
Roye (78)	Yes (78)	Yes (78)	Yes (47, 78)	No	Yes (78)
Functional rating system HJD (53)	No	No	No	Yes (53)	No

[a] article title reflects specifically study purpose.

ments clinimetric properties. This search showed that studies focusing specifically on reliability and validation, reflected in the article title, of clubfoot instruments were rare. Four articles described six instruments (Table 1). An additional seven instruments were found if the criterion 'specifically' was excluded. The qualities of these studies were not included in this thesis.

There is a lack of methodological sound, developed (Table 2) assessment instruments in clubfoot treatment and the need for an instrument sensitive enough to discriminate between subjects and show change over time. Within the field of clubfoot assessment instruments this is still an area in need of further development.

Table 2. Overview of clubfoot assessment instruments found in Medline search (1987-2007) that had documented clinimetric properties

Instrument	Aim	Admini-stration	Manual	Domains	Scales	Items	Scoring levels (items)	Range of scores	Category
Dimeglio	C	CB	Yes	B	1	8	4 (4) 2 (4)	0–40	Benign < 6, moderate 6–10, severe 11–15, very severe > 15
Pirani	C	CB	Yes	B	1	6	3	0–6	None
Laaveg–Ponseti	O	CB	No	B, PS	1	13	5 (4), 4 (1), 3 (1), 2 (7)	0–100	Excellent 91–100, good 81–90, fair 71–80, poor < 70
McKay	O	CB	No	B	1	10	4 (4), 3 (4), 2 (2)	0–180	None
Magone	O	CB	No	B, A, PS	1	13	5 (1), 4 (3), 3 (2), 2 (7)	0–100	Excellent 91–100, good 81–90, fair 71–80, poor < 70
Ghanem	O	CB	No	B, A, PS	2	54	Weighted scores, 1 to 12	0–100	Very good 91–100, good 81–90, fair 71–80, poor < 70
Munshi	O	CB	No	B, A	3	26	4 (11), 3 (9), 2 (6)		None
ESICSG	O	CB	Yes	B, A	3	40	2 (19), 3 (20), 4 (1)	0–60	Excellent 0–5, good 6–15, fair 16–30, poor > 30.
Ponseti–Smoley	C	CB	No	B	1	4	None	None	Good, acceptable, poor
Harold–Walker	C	CB	No	B	1	1	None	None	Mild, moderate, severe
Catteral	C	CB	No	B	1	9	None	None	Resolving, tendon contracture, joint contracture, false correction
Roye	O	PB	Yes	B, A PS, P	5	9	4	0–36	None
Functional system HJD	O	CB	No	B, A, PS	1	6	3 (5), 2 (1)	0–60	Good > 30

Aim: C = classification, O = outcome
Administration: CB = clinician based, PB = patient based
Domains: B = body-structure and function, A = activity, P = participation, and PS = patient satisfaction.

Aims

General

The overall purpose was to develop an assessment instrument for short- and long-term follow-up of children with congenital clubfoot containing variables from both body structure / body function and activity level according to the International Classification System.

Specific

- To describe the development of the new instrument (Paper I)
- To evaluate the instrument reliability, validity and responsiveness (Papers I, II, III, IV and V)
- To test the instrument in a cross sectional and longitudinal study (Papers III and IV)
- To evaluate different treatment concepts for children born with clubfoot (Paper III)
- To investigate neuromotor ability in children treated for idiopathic congenital clubfoot and its relation to the child's foot status (Paper IV)

Methods

Participants

Patients

All patient data were consecutively and prospectively collected from 1994 to 2003. Catchment area was related to the University Hospital of Lund with about 300,000 inhabitants in southern Sweden. A total of 77 children born with clubfoot were eligible. Two children had cerebral palsy, one child had an unknown brain disease and one child had myelomeningocele. These children were excluded in the total study material. Included were five children who also had arthrogryposis, two children with heart diseases, and two children with hip dislocation and hyperextension of the knee. Two children were preterm and two children had a syndrome. Written informed consent was obtained from these 73 childrens' parents. In Paper IV and V only children with idiopathic clubfeet and without learning disabilities were included.

Assessors

The CAP and DCS measurements in the clinical and methodological studies were done by the same experienced assessor well known to the children. In Paper V, inexperienced CAP observers were used with no previous knowledge of the children.

The MABC assessments were done by an experienced child health and rehabilitation physiotherapist with no previous knowledge of the children or their foot status.

Study designs

The designs of the studies are shown in Table 3.

Assessment instruments

The original instrument Clubfoot Assessment Protocol (CAP) will be described in Results.

The Dimeglio Classification System (DCS) (Papers II and III)

The DCS [75] is one if the most cited instruments and is used both for classification and in follow-up studies in children with clubfoot [34, 52-54, 94-

Table 3. A summary of the aims of the studies I–V, design, sample size, age and gender

Study	Aim	Study design	Sample size	Age at assessment	Gender M/F
I	Description and reliability of the CAP	Methodological, daily clinical practice. 2 experienced CAP observers	48 children/ 69 clubfeet	Median 2.1 (0–6.7) years	35/13
II	Validity, responsiveness of the CAP	Methodological, longitudinal cohort	32 children/ 45 clubfeet	New-born, 1, 2, 4 months and 2 years	10/22
III	Evaluation of treatments and assessment instruments	Clinical and methodological. Longitudinal and cross-sectional cohort	16 + 16 children / 23 + 22 clubfeet	New-born, 1, 2, 4 months and 2 years	4/12 + 6/12
IV	Neuromotor ability in children treated for clubfoot	Clinical and methodological. Cross-sectional cohort	20 children / 30 clubfeet	Mean 7.5 (±0.25) years	14/6
V	Reliability of the domain CAPMotion quality	Methodological, video analysis. 4 unexperienced CAP observers	11 children/ 22 feet	Median 5.5 (4–7) years	8/3

Table 4. A summary of the Dimeglio classification system (DCS)

Rating	4	3	2	1	0
1. Equinus	90–45° plf	45–20° plf	20° plf–0 °	0°– +20° dsx	>+20°dsx
2. Varus	90–45° var	45–20° var	20° var–0°	0–20° vlg	>20° vlg
3. Supination	90–45° sup	20–45° sup	20° sup–0°	0–20° prn	>20°prn
4. Adductus	90–45° add	20–45° add	20° add–0°	0°>–<20 abd	>20°abd
5. Posterior crease				yes	no
6. Medial crease				yes	no
7. Cavus				yes	no
8. Deviant muscle function				yes	no

plf = plantarflexion, dsx = dorsalflexion, var = varus, vlg = valgus, sup = supination, prn = pronation, add = adduction, abd = abduction.

96] (Table 4). This instrument assesses primarily the mobility of the clubfoot. One item concerns muscle function.

The DCS consists of eight items. Scorings for four items range from 0–4 (best to worst). Four items can only score zero or one. Total score ranges between 0 and 20; very severe 16–20, severe 11–15, moderate 6–10, and postural 0–5. Focus is on total score and classification.

Good reliability has been shown for the DCS with kappa (κ) varying between 0.4 and 0.77 [84], mean difference scores of 1.4 points [85] and correlation coefficients of 0.83 ($p = 0.0001$) [85]. Content validity was described in the article of Dimeglio et al. [75]. No further studies on validity have been found. The DCS is comparable with the first 5 items in the subgroup mobility of the CAP.

The Movement Assessment Battery for Children (MABC) (Paper IV)

The MABC [97] is a standardized screening instrument integrating cognitive-, attention- and motor functions. The MABC has been proven to be a valid and reliable instrument [97–100]. The instrument contains eight items that represent main motor skills of children between the ages of 4 and 12 years. These items are divided into three subgroups of manual dexterity, ball skills and static and dynamic balance (Table 5). Four age bands are formed with different items but covering similar skills which are age adjusted (4–6, 7–8 9–10, and 11–12 years). Total score can vary between 0–40 (best to worst). A large sample of norm-reference was studied and the raw scores were transformed into percentiles provided in the manual. A MABC result below the 5th percentile (MABC ≥ 13.5) indicates definite motor problems. Results between the 5th and the 15th percentile (MABC 13.4–10) indicate borderline problems. The motor performance in normally developing children, above the 15th percentile, corresponds to a MABC score below 10.0. The MABC sub scores and their cut-off percentiles are mainly used for creating a profile on the child's motor difficulties. Factors such as how the child carries out the task (motor qual-

Table 5. A summary of the Movement ABC (MABC) test age band 2 (7–8 years)

Items	Scores	Cut-off 15th percentile
Manual dexterity	Total: 0–15	≥ 5
Placing pegs in a peg board	0–5	
Threading a lace	0–5	
Drawing a continuos line into a trail	0–5	
Ball skills	Total: 0–10	≥ 2.5
Bouncing and catching ball with one hand	0–5	
Throwing bean bag into a box	0–5	
Balance	Total: 0–15	≥ 5
Stork balance	0–5	
Jumping i squares	0–5	
Heel–to–toe walk on a line	0–5	
MABC	Total 0–40	≥10

ity), its behaviour (e.g. concentration, impulsivity) and physical deficiencies (vision, neurological and orthopedic problems) are to be incorporated in the evaluation for the need of intervention and treatment planning. In this study only the quantitative data from the motor test was used and was compared to the expected distribution according to the MABC standardization. The children were tested with age band 2 (7–8 years) (Table 5) [97].

The expected distribution according to the MABC standardization based on 868 healthy children between the ages 6 and 12 years was used as a reference [97].

Clinimetric methods and related statistics

In general non-parametric statistics were used as the underlying data are based on ordinal and interval scores.

Reliability measurements (Papers I and V)

As the CAP items have an ordinal scale construct the unweighted kappa (κ) (Paper I) and weigthed κ (Paper V) statistics for agreement (inter- and intra-rater reliability) were used [101-103] with 95% confidence interval (CI). It calculates agreement beyond chance. As κ values can become unstable under certain conditions [103, 104], the observed percentage agreement (Po) (Papers I and V) and the percentage agreement within one-level difference (Po-1) was calculated. In cases with limited distribution of cell frequency, the Po was preferred instead of κ. The amount of categories is also regarded as κ values decrease when categories increase [104]. According to Altman [101] the κ values are to be interpreted as follows: < 0.20 as poor agreement, 0.21–0.40 as fair, 0.41–0.60 as moderate, 0.61–0.80 as good and > 0.80 as very good agreement.

In Paper I, item reliability for each 22 CAP items was evaluated. Two experienced CAP examiners assessed 69 clubfeet in 48 children (range 0–6.7 years). Both treated and untreated feet with different severity grades were included. Three age groups were constructed for studying the influence of age on agreement. The intra-rater study included 32 feet in 20 children (range 4 months – 6.8 years). The unweigthed κ statistics, the exact percentage observer agreement (Po > 75% was regarded as good) and the amount of categories defined how reliability was to be interpreted. A good reliability was considered when the κ value was high, or a low κ value combined with a high Po. A sufficient reliability was considered in cases with fair to moderate κ values and good percentage agreement.

To keep observation phenomena stabile between several observers the reliability testing in Paper V used video recorded motor performances according to the items in CAP**Motion quality.** These recordings contained 11 children treated for idiopathic clubfoot with a median age of 5.5 (range 4 to 7) years. The clubfoot severity distribution at newborn and the functional outcome results at the time of motion analysis were varying. Four inexperienced CAP raters (two experienced paediatric orthopedic surgeons and two physiotherapists) assessed the children's motion at two different occasions. As the CAP**Motion quality** domain exists out of five scoring possibilities we regarded a Po ≥ 50% or a Po-1 ≥ 80% as good.

Good item reliability was considered when more than half of the κs had high value and a good percentage agreement. Sufficient item reliability was considered when the κ values ranged between fair and moderate for more than half of the inter-/intra-ratings and had good percentage agreement. The reliability analyses were supplemented with calculating the median differences and interquartile ranges (IQR) for each item and the mean difference and limits of agreement (LOA)[105] for the domain CAP**Motion quality** for the inter- and intra-rater testing.

Validity measurements of the CAP (Papers I–IV)

Content validity (Paper I). The selection of important items to be included in the protocol and scoring system was an act of balance between considerations of clinical utility and scientific interest. Literature studies, expert opinions and clinical

experience on what patients/parents present as important factors formed the platform for the CAP prototype.

Concurrent validity demonstrates if a test correlates well with a measure that has previously been validated. Two experienced raters defined in the reliability study of the domain CAP**Motion quality** the correct scoring for each of the children's item performance used in the video recordings. These scorings were then used as a gold standard for comparison with the untrained CAP observers. The Po and the Po-1 were used for evaluating validity.

Construct validity examines the theoretical construct underlying the test [106].

Convergent construct validity (Papers II and IV) implies that the items and/or domains analyzed assess the same construct. Divergent construct validity implies that the items and/or domains analyzed assess different constructs and show none or poor correlation. These were specified a priori describing the expected correlation between the CAP domains (Paper II and IV) and items (Paper IV) and the comparing measurement instruments (DCS in Paper I and MABC in Paper IV). The non-parametric Spearman correlation coefficient was used.

The floor and ceiling effects (Paper II) for the CAP and the DCS were assessed at two occasions; at baseline/newborn (untreated clubfeet) and at the age of two years (treated clubfeet).

Discriminant validity (Papers II and III) demonstrates the ability to show variation (that is being sensitive for difference). In paper II the ability of the CAP and the DCS to show variation was assessed by comparing their ability to differ clubfoot severity in 13 bilateral clubfeet. The right and left foot were compared at new-born and preoperatively. In paper III the ability of the CAP and the DCS to assess differences between two different treatment groups was evaluated. The Mann-Whitney U test was used.

Responsiveness measurements (Papers II and III)

Responsiveness refers to the instrument's ability to detect important change over time in the concept being measured [107, 108]. In Paper II the CAP and the DCS were applied in 32 children with clubfeet at the age new-born (the pre-treatment phase), 1 month, 2 months (pre-operative), 4 months (post-operative) and at 2 years of age. In Paper II responsiveness was calculated for both instruments by the use of effect size (ES) [102]. Effect size was defined as the mean change scores divided by the standard deviation of the baseline score, which in this case is the score in new-born. Finally we assessed if changes had occurred across the whole follow-up period with Friedman's test for change. Thereafter change between measurement and its preceding assessment was calculated by using Wilcoxon's signed rank test.

Effect sizes of 0.2 are defined as small, 0.5 as medium and 0.8 as large [109]. In Paper III, the developments of two different treatment groups were followed during two years using the CAP and the DCS. Responsiveness was calculated using Wilcoxon's signed rank test.

Clinical studies and related statistics

Paper III

Two intervention groups were compared with the CAP and the DCS (Figure 3).

Consecutively, 16 children were treated with intensive stretching according to the Copenhagen method and 16 children with casting according to the Ponseti casting technique during their first two months of age. The need for surgery was then assessed. At four months of age all children used a dynamic Knee Ankle Foot Orthosis (KAFO). Two months after walking debut all children used an Ankle Foot Orthosis (AFO). At baseline the two intervention groups showed no statistical significant differences in foot status with both assessment instruments, except for CAP**Mobility II.**

The Mann-Whitney U test was used for between-group comparisons.

Paper IV

We studied the prevalence of neuromotor ability problems in children treated previously for idio-

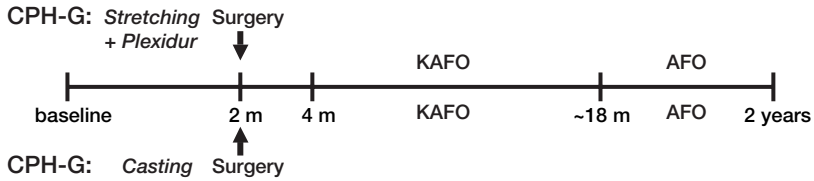

Figure 3. Study design of comparison study between two treatment groups. CPH-G is the group (n= 22 clubfeet) treated with the Copenhagen stretching method and PCT-G is the group (n=23 clubfeet) treated according to the Ponseti casting technique.

Table 6. A summary of the Clubfoot Assessment Protocol (CAP) and its cut-off points

Items	Scores	Cut-off
Mobility I	Total: 0–20	≤ 14
Ankle dorsal extension, plantar flexion, heel varus/valgus, eversion/inversion and forefoot adduction/abduction (5)	Item level: 0, 1, 2, 3 and 4	≤ 2
Mobility II	Total: 0–8	≤ 6
Length of toe flexors (2)	Item level: 0, 2 and 4	≤ 2
Muscle function	Total: 0–8	≤ 6
Strength of foot extension and eversion (2)	Item level: 0, 2 and 4	≤ 2
Morphology	Total: 0–16	≤12
Tibial torsion, heel and forefoot position, cavus or planus. (4)	Item level: 0, 2 and 4	≤ 2
Motion quality I	Total: 0–16	≤ 12
Walking, running, toe walking, heel walking (4)	Item level: 0, 1, 2, 3 and 4	≤ 2
Motion quality II	Total: 0–8	≤ 5
One-leg stance, one-leg hop (2)	Item level: 0, 1, 2, 3 and 4	≤ 2

Item level scores:
0 = cannot or ++ poor,
1 = very deviant or + poor,
2 = deviant or poor,
3 = slightly deviant or slightly poor,
4 = within normal.

pathic clubfoot and its relation to the child's clinical foot status and parental observation. Twenty children (mean age 7.5 years, SD 3.2 months) from a consecutively born cohort were assessed with the MABC (Table 5), a neuromotor test, and the CAP (Table 6), a disease specific test on foot function.

Cut-off points were established for the scores of the CAP (Table 6). A score below "slightly deviant to normal" for item scores or less than 75% of the maximal subgroup scores was chosen as cut-off points. Scores below these cut-offs were assessed as deviant outcome. Parental observations of their child's daily activity were categorized into two groups: non/sometimes and regular problems.

The Fisher exact t-test using a multinomial distribution was used to check if there was a significant statistical difference between the study group

and the expected distribution of motor performance problems according to the MABC test. Cut-off points used were the 15th and 5th percentile [97]. The Mann-Whitney U-test was used for analyzing differences between motor ability in children with uni- and bilateral idiopathic clubfoot. The mean values from the right and left foot of CAP assessments were used for correlation between the MABC and the CAP.

In all the statistical analysis a $P < 0.05$ was considered significant.

The Statistical Package for Social Science (SPPS Inc., Chicago, IL) version 11.0, 12.0 and 12.1 and StatXact 3.0 were used.

Results

Description of the CAP (Paper I) (content validity)

The purpose of the CAP is to provide an overall profile of the clubfoot child's functional status within the domains of body function/structure and activity on single assessment occasions and over time. Furthermore, the CAP aims to provide structure and standardization for follow-up procedures from 0 to11 years of age in daily clinical decision making. It is an observer administered test.

The original CAP (Table 7) contains 22 items in four sub-groups: mobility (eight items), muscle function (three items), morphology (four items), and motion quality (seven items). The first three sub-groups relate to body function/structure and the last to activity according to ICF [66]. Questions about pain, stiffness and daily activity/sport participation are routinely asked in a standardized way.

Each item is described in a manual along with the criteria for scoring. The scoring is divided systematically in proportion to what is regarded as normal variation and its supposed impact on perceived physical function ranging from 0 (severe reduction/no capacity) to 4 (normal). Score grading can vary between 3 to 5 levels. For sub-groups the sum of the items scores are calculated and can be visualized as profiles (transformed to a 0–100 scale score, with 0 = extremely deviant and 100 within normal variance; sub-group transformation score = actual score/maximal possible score × 100). The CAP is not intended for total scores.

Administration time varies between 10–15 minutes dependent on the child's cooperation. The items in the sub-group motion quality are age dependent. At the age of three years all children are presumed to be able to perform Motion Quality part I. At the age of four years all children are also expected to be able to perform Motion Quality part II.

Comments

The CAP initially included 22 items (Table 7). These were chosen on the basis of what Feinstein [68) calls "clinical sensibility ". Changes have been made during the years of practical usage and testing of the CAP. The Appendix shows the current version (CAP1.2). Three items have been deleted. The items tightness (no 6) and squatting (no 19) were found to be too subjective and/or too similar to another item, thereby not producing useful information to the construct. The item strength of the soleus-gastrocnemius muscle (no 11) showed problems in proper assessment in the younger children, implying low feasibility. In older children the ability to tiptoe, and their endurance in heel lifting, makes a proper estimation of the plantar flexion capacity possible. As the item toe walking is highly related and easier to assess also item 11 was distracted. The questions commonly used have now been structured and incorporated in the protocol. Furthermore a specification of the motion quality items was included illustrating more specific the main problems.

Reliability (Papers I and V)

In paper I, using two experienced CAP raters and items with 3, 4 or 5 point scales, the inter-rater reliability was assessed as moderate to good for all CAP items except for one (running). Eighteen items had $\kappa > 0.40$. Three items varied from 0.35 to 0.38. The mean percentage observed agreement was 82% (range, 62–95%). Different age groups showed sufficient agreement. The intra-rater reliability was good to excellent; all items had $\kappa > 0.40$ (range, 0.54–1.00) and a mean percentage agreement of 90%.

In paper V, using four inexperienced CAP raters and items with a 5-point scale, the κ values for the items of domain CAP**Motion quality** varied

Table 7. The CAP original

Name:			Date of birth:		
Date of assessment:			Assessment number:		
Side: O Left O Right					
Rating	0	1	2	3	4
Passive mobility I					
1. Dorsiflexion	< -10°	-10°– < 0°	0°– < +10°	+10° – +20°	> +20°
2. Plantar flexion	0°– < 10°	10° – < 20°	20° – < 30°	30° – 40°	> 40°
3. Varus/valgus	> 20° varus	20° – > 10° varus	10° – > 0° varus	0° – neutral	> 0° valgus
4. Inversion/eversion	> 20° inver.	20° – > 10° inver.	10° – > 0° inver.	0° – 10° evers.	>10° evers.
5. Adduction/abduction	> 20° add.	20° – > 10° add.	10° – > 0° add.	0° – neutral	> 0° abd.
Passive mobility II					
6. Tightness	+ tight	tight		soft–tight	soft
7. Flex. digiti longus	+ reduced		reduced		normal
8. Flex. hallucis longus	+ reduced		reduced		normal
Muscle function (strength)					
9. M. peroneus	absent/poor		reduced		normal
10. M. ext. dig. longus	absent/poor		reduced		normal
11. M. soleus/gastroc.	absent/poor		reduced		normal
Morphology					
12. Tibial rotation	+ inward		inward		normal
13. Calcaneus position	> 10° varus		10° – > 0° varus		neutral/valgus
14. Forefoot position	> 20° add.		20° – 10° add.		< 10° add.
15. Foot arch	+ cavus		cavus		normal
Motion quality I					
16. Walking	+ deviant	deviant		slightly deviant	normal
17. Toe walking	cannot	deviant		slightly deviant	normal
18. Heel walking	cannot	deviant		slightly deviant	normal
19. Squatting	cannot	deviant		slightly deviant	normal
20. Running	+ deviant	deviant		slightly deviant	normal
Motion quality II					
21. One-leg stance	cannot	deviant		slightly deviant	normal
22. One-leg hop	cannot	deviant		slightly deviant	normal

Extra notes: Questions about pain, stiffness, shoe problems, physical condition, activity level, sports and social participation and patient/parent satisfaction.
+ = Pronounced / very, inver. = inversion, evers. =eversion, add. = adduction, abd. = abduction, flex. digit. longus = length of m. flexor digitorum longus, flex. hallucis longus = length of m. flexor hallucis longus

between 0.25 and 0.79 and a mean Po/Po-1 of 48/88%. According to the reliability criteria set a priori five out of six items showed sufficient inter-rater reliability. The item heel walking showed lower, though fair, reliability. Intra-rater κ values varied between 0.22–0.81 and had a mean Po/Po-1 of 63/96% and assessed as good to sufficient reliability for all items. The median inter- and intra-raters item score differences and the mean difference on domain levels were relatively small (0.00, range 1 to 1 and -1.10, LOA -1.86–1.66, respectively). No learning effects could be found on the Po level and the Po-1 between the first and second session.

Comments

The CAP contains more detailed information than previous protocols. It is a multi-dimensional observer administered standardized measurement instrument with the focus on item and subgroup level. It can be used with sufficient reliability, independent of age, during the first seven years of childhood by examiners with good clinical expe-

Table 8. Inter-raters Po and Po-1, between each rater and the criterion from session one. All items were assessed as having sufficient reliability according to the criteria

Item	A	B	C	D
Running	45/ 100	73/ 95	59/ 82	54 / 91
Walking	59/ 100	59/ 100	41/ 86	59 / 96
Toe walking	32/ 64	59/ 100	64/ 100	55 / 100
Heel walking	41/ 86	50/ 86	59/ 95	52 / 96
One-leg stance	32/ 82	48/ 86	46/ 73	46 / 82
One-leg hop	55/ 86	50/ 82	72/ 100	46 / 82

Table 9. Convergent and divergent construct validity. The Spearman correlation coefficient (rs) was used. N = 45 clubfeet

CAP	Expected correlation	Dimeglio-newborn	p-value
Mobility I	High	0.77	0.000
Mobility II	Low	0.09	0.55
Muscle function	Low	0.20	0.20
Morphology	Moderate	0.44	0.002
Motion Quality I+II	Moderate	0.06	0.72 [a]

[a] assessed at age 2 years

rience of the instrument. As expected reliability with inexperienced CAP raters, as shown in paper V, was lower but still acceptable in relation to the testing circumstances. A few items showed low reliability, probably dependent on the child's age and/or varying professional backgrounds between the examiners. No items have been distracted from the protocol because of poor reliability.

Future studies of the effects of regular usage of the CAP or special CAP education on the reliability are needed.

Validity

Concurrent validity (Unpublished data)

The item inter-rater agreement for each rater using the criterion is presented in Table 8. For all items together the mean exact Po/Po-1 was 51/ 90%.

Item one-leg stance seems to give most problems in assessing correctly. All observers had Po < 50%. All other items had each at least three out of four observers with Po ≥ 50%.

Comments

The relatively sufficient percentage agreement found shows, in general, good standardization of the domain CAPMotion quality, confirming acceptable validity. Item one-leg stance may need more clarification in its description. It is planned to evaluate the effects of specific education and training in the CAP and the effects of learning through regular usage of the CAP.

Construct validity—convergent and divergent (Papers II and IV)

In Paper II, high to moderate significant correlation was found between CAP**Mobility I** and CAP**Morphology** and the DCS (rs = 0.77 and 0.44, respectively) indicating good convergent construct validity (Table 9). Low correlation was found between CAP**Muscle function**, CAP**Mobility II** and CAP-**Motion quality** and the DCS (rs = 0.20, 0.09 and 0.06, respectively) indicating divergent construct validity and implying that different constructs are assessed.

Paper IV showed, as expected a priori, the item CAP**one-leg stand** to be the only variable that correlated moderately and significant with the MABC (rs= -0.53, p=0.02). No or low correlations were found between the subgroups CAP**Mobility**, CAP-**Morphology** and the remaining items of the CAP-**Motion quality** and MABC (Table 10).

Floor and ceiling effects (Paper II)

No floor effects and low ceiling effects were found in the untreated clubfeet for both the CAP and DCS instruments (Table 11). High ceiling effects were found in the CAP for the treated children and low for the DCS.

Comments

It is of importance to have in mind what the instruments aims for usage are, the construct and which group of patients that are assessed. The scoring construct of the CAP concentrates on smaller intervals in the middle of the scale, where changes

Table 10. Expected correlation and the Spearman correlation coefficients between the CAP domains, CAPMotion quality items and the MABC in 20 children treated for idiopathic clubfoot. N = 30 clubfeet

CAP	Expected correlation	MABC	p-value
Mobility I	Low	0.11	0.65
Mobility II	Poor	-0.36	0.12
Muscle function	Low	0.52 *	0.02 [a]
Morphology	Low	-0.21	0.39
Motion quality I	Low	0.14	0.57
Running	Poor	0.08	0.72
Walking	Poor	-0.15	0.53
Heel walking	Moderate	-0.26	0.26
Toe walking	Moderate	-0.37	0.11
Motion quality II	Moderate	-0.15	0.54
One-leg stance	High	-0.53 *	0.02
One-leg hop	Low	0.23	0.33

*= $p < 0.05$
[a] skewed distribution

Table 11. Floor/ceiling effects in percentage of patients assessed with the CAP and the DCS at two treatment phases, untreated and treated clubfeet. N= 45 clubfeet.

	Floor / Ceiling Newborn	Floor / Ceiling 2 years
CAP		
Mobility I	0 / 0	0 / 35
Mobility II	0 / 37	0 / 84
Muscle function	0 / 4	0 / 86
Morphology	0 / 0	0 / 42
Motion	–	0 / 38
DCS	0 / 0	0 / 4

are of most clinical importance. The CAP is not intended to measure changes above normal or extreme abnormal. In the untreated group of children with clubfoot, both instruments showed no floor effects in this population. Moderate ceiling effects where found for CAP**Mobility II** (37%) which indicates that about 60% of the children have problems with this item, which is of clinical importance. In the follow-up, at two years of age most of the children should have reached a functional level within normal variation. As the CAP has its end levels within normal variation and the DCS has its on more extreme levels (e.g. forefoot abduction > 20º or valgus > 20º), the CAP will sooner reach its ceiling levels. Furthermore, with usage of only three scoring levels such as in domain CAP**Muscle function** there is less room for discrimination which gives higher ceiling or floor effects.

We conclude that both the CAP and the DCS floor and ceiling effects are in accordance with the concept and construct of the instruments.

Discriminant validity (Papers II and III)

In Paper II, 11 out of 13 children with bilateral clubfeet showed different CAP**Mobility I** scores between right and left foot at baseline (untreated) compared with 5 with the DCS. At the following assessment occasions the CAP**Mobility I** continued to show higher discriminate ability than the DCS, indicating a better sensitivity for the CAP.

In Paper III, the development of foot status was compared in two groups with two different treatment procedures. The CAP and the DCS were used to assess the clubfeet statuses. At baseline the two intervention groups showed no statistically significant differences in foot status with both assessment instruments. The CAP but not the DCS could show statistical significant differences in foot status development between the two intervention groups. According to the CAP but not the DCS, the casting technique according to Ponseti was superior in clubfoot correction shown as better mobility and motion quality at the children's age of two years.

Comments

As a result of different scaling intervals and distribution of scores, the use of different instruments can result in different conclusions. Because of its multidimensional and narrower scoring interval construct the CAP was able to elucidate and evaluate different clinical functions and not the DCS. This can have consequences for design and treatment outcome results in studies. The results also verified the clinical experience that children with bilateral clubfeet generally have a difference in severity between left and right feet. With the DCS the conclusion would be that most children with

Table 12. Effect Sizes (ES) from baseline(treatment starting within 8 days after birth to 1 month, 2 months, 4 months and 2 years of age for all clubfeet (n = 45)

	1 month	2 months, preop.	4 months, postop.	2 years
CAP				
Mobility I	2.70	3.20	4.21	4.35
Mobility II	0.60	0.80	1.00	0.80
Muscle function	1.28	1.60	1.87	2.10
Morphology	1.55	1.84	2.01	1.66
DCS	2.42	2.82	4.00	4.68

bilateral clubfeet have similar severity. In such case, it would not be appropriate to use both feet in bilateral children as independent observations.

Responsiveness (Papers II and III)

In Paper II the responsiveness was good for both the CAP and the DCS. Effect sizes of treatment at the age of 2 years, from untreated to treated, ranged from 0.80 to 4.35 for the CAP subgroups and was 4.68 for the DCS (Table 12). The first four treatment months, the CAP**Mobility I** had generally higher ES compared with the DCS. Both instruments showed significant changes ($p < 0.05$) between the scoring occasions except for the pre- and postoperative measurements for the CAP-**Mobility II**, CAP**Muscle function** and CAP**Morphology** and between postoperative and 2 years of age for the CAP**Mobility I and II**. From baseline to the age of the 2 years all the CAP subgroups scores and the DCS showed significant improvement (Friedman's test, $p < 0.0001$).

Paper III showed statistically significant progress or regress over time for the CAP domains at several different assessment occasions. The DCS also showed significant change over time.

Comments

The ES of the CAP domains should not be compared with each other or the DCS as they assess different entities except for the CAP**Mobility I** and DCS. These two assess generally the same mobility construct. The CAP**Mobility I** is more responsive when severity ranges between mild and moderate to severe as its scoring intervals are less coarse than the DCS. This tendency is maintained until the postoperative phase where the ES for DCS increases between 4 months and age 2 years compared with the CAP**Mobility I** (with 1.10–4.68 and 0.26–4.35, respectively). This is caused by the fact that the best possible score is reached earlier with the CAP**Mobility I** than the Dimeglio. Furthermore both instruments contain slight different items such as plantar flexion for the CAP**Mobility I** and integration of muscle function in the Dimeglio score, increasing or decreasing the subgroup score.

The remaining CAP subgroups showed change over time, though with lower ES as less scoring levels are used.

As data distribution was not normal in Paper III we could not use the ES. This statistical method would have made more clear which instrument was most responsive in showing treatment efficacy.

Comparison of serial casting versus stretching in children with congenital idiopathic clubfoot (Paper III)

According to the CAP but not the DCS, the casting technique according to Ponseti was superior in clubfoot correction shown as better mobility and better motion quality at age two years. These children also needed less surgery. The orthotic management functioned well in both groups, with high compliance and maintenance or slight improvement of the clinical status except for morphology.

Comments

Outcome results and the possibility to evaluate interventions depend on the instruments used and their construct. According to the CAP the casting technique according to Ponseti seems to be the method of choice in clubfoot correction concerning mobility and motion quality.

Table 13. Individual patient data: The total and subtest scores of Movement Assessment Battery for Children (MABC) and Clubfoot Assessment Protocol (CAP).

Case no Side Gender			Surgery [a]	ABC-Total 0–40	ABC-Hand 0–15	ABC-Ball 0–10	ABC-Balance 0–15	CAP-mob. I 0–20	CAP-mob. II 0–8	CAP-muscle 0–8	CAP-morph. 0–16	CAP-motion I (0–16)	CAP-motion II (0–8)	PDA [b]
1.	R	M	AL	**11**	4.5	1	**5.5**	20	8	8	**12**	14	6	N
2.	R	F	PMR	**10.5**	4	**3.5**	3	**14**	8	8	14	15	**5**	S
	L		PMR					**13**	8	8	**12**	**11**	**3**	
3.	R	M	PMR	**10**	1	**4**	**5**	15	**4**	8	**12**	12	6	N
	L		PMR					**14**	**4**	8	**12**	12	**5**	
4.	L	M	PMR	6	3	0	3	16	8	8	16	13	7	S
5.	R	F	PMR	3.5	0	0	3.5	**11**	8	8	**12**	12	**5**	S
	L		PMR					**11**	8	8	14	13	6	
6.	R	M	PMR	0.5	0	0	0.5	15	**4**	8	14	13	7	N
	L							16	**4**	8	16	13	4	
7.	R	M	PMR	1	0	0	1	17	8	8	16	16	7	N
	L		PMR					18	8	8	16	16	8	
8.	R	M	PMR	4	0	0	4	18	8	8	**12**	**7**	6	N
	L		PMR					16	8	8	**10**	**7**	6	
9.	R	M	AL	8.5	1.5	2	**5**	**14**	8	8	**12**	13	**4**	N
10.	L	M	PMR	7	4	0	3	17	8	8	16	16	8	N
11.	R	F	None+TT	3.5	0	**2.5**	1	19	8	8	**12**	13	7	S
12.	R	M	PMR +TT	**11.5**	1.5	**4.5**	**5.5**	17	6	8	**8**	13	7	R
	L		PMR +TT					16	6	8	**10**	13	7	
13.	R	M	PMR +TT	0	0	0	0	**14**	8	8	14	14	8	N
	L		PMR +TT					16	8	8	14	14	7	
14.	R	F	PMR +TT	**10.5**	0	8	2.5	15	**6**	8	14	12	6	R
15.	R	M	PMR +TT	4	1	0	3	16	**6**	8	14	12	6	S
16.	R	F†	None	5.5	2.5	0	3	16	8	8	16	15	8	N
17.	L	M	None	6.5	0	**3**	3.5	19	8	8	16	16	8	N
18.	R	M [c]	PMR	**21.5**	**5**	**7.5**	**9**	15	8	**6**	**6**	12	6	N
	L		PMR +TT					**14**	6	8	**8**	12	6	
19.	R	M	PMR	6.5	4	0	2.5	19	8	8	16	16	8	N
	L		PMR					19	8	8	16	16	8	
20.	R	F [c]	None	**13.5**	**5.5**	1.5	**6.5**	18	**4**	**6**	16	13	**5**	R

[a] Surgery: AL = Achilles lengthening, PMR = postero-medial release including Achilles lengthening, TT = secondary operation with m. tibialis anterior transposition.
[b] PDA = Problems in daily activity; N = none, S = sometimes, R = regular.
[c] Vision problems.
Scores below cut-off points are bold.

Motor ability in children treated for idiopathic congenital clubfoot (Paper IV)

An increased prevalence of motor ability problems was found both regarding the total score for MABC ($p = 0.037$) and for the subtest ABC-Ball skills (0.037). Seven children (three unilateral and four bilateral) had scores ≥ 10.0. No statistically significant differences of the MABC scores between children with uni- or bilateral clubfoot were found (MABC total $p = 0.49$). Surgery did generally not influence the motor ability results negative. Only five out of 14 children with extensive surgery had MABC scores ≥ 10.0. Four of these five children had bilateral clubfeet.

The IQR of the CAP subgroups were equal to or better than the chosen cut-off points. Six feet from six children (two unilateral and four bilateral) had unsatisfactory foot status according to the CAP. Four of these six children also showed developmental motor problems according to the MABC (one from unilateral and three from children with bilateral clubfoot). No statistical significant correlations could be found between the scores of the CAP body structure/body function subgroups and the MABC scores. On activity level only the item CAP**one-leg stance** showed moderate and statistically significant correlation with the MABC.

Three out of seven parents with children that showed to have motor problems reported concern on their childs daily functioning. All parents with children without motor problems according to the MABC showed no specific concern (Table 13).

Comments

This study showed that children treated for idiopathic congenital clubfoot had an increased prevalence of motor ability problems which did not correlate to the foot status of the child, nor could it be explained by the amount of surgery. This indicates that neurodevelopment deficiencies might be involved in the child with clubfoot. Deeper studies are though needed. The ability to keep balance on one leg combined with parental observations on their child's physical abilities may be a sufficient tool for determining which children in the orthopedic setting should be more thoroughly evaluated regarding their neuromotor function.

General discussion

Only a few assessment instruments of clubfoot children are tested for clinimetrics properties. In this thesis an assessment instrument was developed for the use in follow-up of children with clubfoot. The studies on the clinimetric properties showed overall sufficient reliability of the individual items and appropriate validity of the five domains of the CAP. Good responsiveness was found for four domains (Mobilty I and II, Muscle function and Morphology). CAP**Motion quality** responsiveness has not been evaluated in this thesis. In a clinical study the CAP showed ability to evaluate treatment on different functional levels both between treatment groups and within treatment group. Another study showed that neither the clinical foot status according to the CAP nor the extent of surgery related to the childrens neuromotor ability, and that children treated for idiopathic clubfoot had more neuromotor deficiencies than expected. Thus, the CAP is an important contribution to clubfoot evaluation both in clinical and in research settings.

Methodological issues

The samples

One strength of this thesis is that the age variance of the children used in the clinical studies was low compared to many other clinical studies [45, 48, 110]. Also in the reliability studies the effect of age was controlled. Furthermore the samples included consecutive (Papers II, III and IV) and prospectively (Papers II and II) assessed cohorts from a defined catchment area. All samples had been checked for their severity distributions.

Including data from two limbs without appropriate design or statistical approaches in clinical studies may contribute to outcome bias [111]. For instance, in studies where variables such as pain, satisfaction and overall functional ability are included, the effect of laterality must be accounted for. In Paper III the factor that could influence outcome was compliance to the orthosis treatment. This was checked for both groups and its laterality distribution and found to be equal. In Paper IV the mean for the right and left leg were used when evaluating the correlation between the CAP and the MABC. Differences in DMA between children with uni- and bilateral clubfeet were also checked. In the remaining papers the feet from children with bilateral clubfeet were used as independent observations in the statistical analysis. The study designs were such that laterality was not expected to influence the results.

The low sample sizes are due to the population of our catchment area. The risk for type II-errors should be considered.

Assessment instruments

There were problems finding appropriate instruments to validate the CAP with. No previous clubfoot instruments had been developed for longitudinal follow-up. Most previous instruments use a total score construct with mixed functional dimensions. Only a few clubfoot instruments had documented reliability and validity at the time our studies took place; Pirani, the DCS and Roye (Table 1). This last instrument is a patient based outcome instrument developed for use in older children. The first two instruments had primarily been developed for classification of clubfoot severity and mainly contain items on mobility. They have also shown sensitivity for change in a couple of studies [53, 84]. The DCS was chosen as being the most cited instrument.

Procedures

Reliability studies in children are difficult to perform. Children's co-operation and task understanding may vary from day to day and between different examiners. A child friendly environment and familiarity with the examiners are important factors to enhance reliability. Also a testing situation comparable with a normal clinical setting was

attained. These are the reasons why the investigation in Paper I was unblinded and no more than two examiners were involved. In Paper V, where the children had been video recorded, it was possible using several blinded raters. In the remaining studies the CAP assessments were done by an experienced assessor who was not blinded and who was familiar with the children. This could cause bias but procedures were standardized. In paper V as a control group the expected distribution based on healthy children in the MABC standardization procedure [97] was used. It would have increased the quality of the study if a control sample of healthy Swedish children in the same age had been used.

The CAP clinimetric properties

Reliability

The reliability studies in this thesis showed a sufficient inter-and intra rater reliability on item levels for the CAP in the hands of experienced CAP users. Dimeglio et al. [75], noticed the same tendency of reliability increase (discrepancy decreased from 40 to 6%) when raters became more experienced as did Flynn et al. [85]. Inexperienced CAP users were able to reproduce their own assessments with sufficient agreement but had lower, though acceptable, inter-rater reliability for the items of CAP-**Motion quality**.

Weighted κ statistics take into account the degree of disagreement [101]. This κ statistic was used in the second reliability study (Paper V). These weighted κ are usually higher. When recalculating the unweighted κ in Paper I to weighted, the values increased between 0.01 and 0.20. Also, in Paper V, the Po-1 was significant higher than the Po (88% and 48%, respectively). This indicates that reliability of the CAP could be increased by combining categories.

A recent study of Munshi et al. [82], which was the only known study comparable with paper I, showed ordinal items inter-rater κ between 0.35 and 0.87 (14 items). The CAP items varied between 0.35 and 0.94 (22 items). Their instrument items have, in general, broader intervals and less scoring levels. Celebi et al. [91] also used the κ statistics using the outcome categories defined by the total scores of the evaluation system designed by the International Clubfoot Study Group (ESISGC) [90]. They found a substantial degree of agreement (κ = 0.73).

Wainwright et al. [84] evaluated the reliability of different outcome systems as shown by Ponseti and Smoley [92], Harrold and Walker [93], Catterall [73], and the DCS.

The DCS gave the best outcome agreement, κ = 0.7, compared with κ below 0.4 for the other instruments. These reliability studies do not give any information on the item reliability.

Summarizing item scores and comparing mean scores/outcome categories gives a risk that item scores are weighed out against each other giving a better outcome when there actually exist differences [112, 113]. In Paper V the reliability study was completed with calculating the mean observer differences. It showed that on item level the median score differences for all four observers in general were good. On total score level the mean difference was low caused by the effect of out weighing. Celebi et al. [91] found mean difference scores of 0.17, 0.63 and 0.80 (LOA around -2.00 to 3.00, max domain score 36 and 2 or 3 point scale) with three experienced observers for their functional domain of the ISGC.

It is impossible to calculate the exact reliability of an instrument. It is sooner a question about estimation of relative reliability while keeping as many internal and external factors as possible under control. Having this in mind the conclusion is that in clinical practice, the CAP can be used with sufficient reliability with the same assessor. In research studies the users should be experienced with the CAP, and the observer differences should be checked.

There is no indication to combine scoring levels in the CAP, even though this would enhance the reliability. One reason for developing the CAP was that other clubfoot instruments have item constructs that are too coarse. This may be adequate/satisfying in cross sectional outcome studies where

the aim is differentiation between good and poor categories. The CAP though is intended to be used in longitudinal follow up, which demands sensitivity for change.

Content validity

No factor analyses were used for item reduction as in psychometrics [114], where one seeks a high internal consistency. In clinimetrics though, one tries to describe the "construct" or phenomena as varied as possible with a small amount of items [68]. The use of regression analysis is then more appropriate in selecting those items that associate most with the defined phenomena. This is a necessary next step in the developmental procedure of the CAP.

Construct validity

In comparison with the DCS most of the CAP domains showed good validity in terms of convergent and divergent construct validity (Paper II) and assumptions set a priori to testing. An exception was found for the CAP**Motion quality** and the DCS. We had expected a moderate correlation as foot mobility generally is a prerequisite for proper walking, running and other foot-leg activities. A poor correlation (rs = 0.06, p = 0.72) was found in contrast to the moderate correlation of 0.41 (p = 0.01) shown between the CAP**Mobility I** and CAP**Motion quality.** This difference was due to the coarse scale construct of the DCS. The low correlation found between the MABC and the CAP, except for the item CAP**one-leg stance**, confirmed the expected differences in the instruments content and construct. The MABC aims on diagnosing children with motor ability problems caused by neurogical deficiencies while the CAP**Motion quality** aims on motor ability related to musculoskeletal problems (Table 10).

In general the CAP showed better discriminating abilities than the DCS showing that the items interval score had sufficient distribution.

Responsiveness

The scoring construct of the CAP**Mobilty I** focus on smaller intervals in the middle of the scale, where changes are clinically most important. Therefore the CAP**Mobility I** shows higher ES but reaches sooner its ceiling levels than the DCS. Furthermore the remaining CAP domains showed ability in providing information, sensitive to change, on different functional levels compared with the DCS though with lower ES. Besides calculating ES as a mean for responsiveness it is also common to use the Standardized Response Mean (SRM) [115-117] which instead of using the standard deviation of the baseline uses the standard deviation of the mean change score. Using different indices makes it possible to illustrate responsiveness from different perspectives and therefore makes it more appropriate when evaluating instruments responsiveness [117].

Even though the CAP showed good responsiveness on group level through statistical detectable change it is more and more emphasized to study responsiveness in relation to patient/parents perceived clinical important change [118] or so called anchor- based approach [117]. Liang [118] actually defines responsiveness (or longitudinal construct validity) as the ability of an instrument to measure a meaningful or important change in a clinical state versus sensitivity as the ability of an instrument to measure change irrespective whether it is meaningful. This implies, according to this definition, that the thesis only has evaluated the CAP's sensitivity to change and not responsiveness. The CAP scoring levels and intervals are, however, based on the expected clinical impact on the child physical functioning.

Clubfoot treatment and outcome

It is known that factors as degree of deformity at birth [34, 36, 95], non-compliance with the use of orthosis and parental educational level influence outcome [31, 54]. Also factors as the age at follow-up, treatment experience and criteria for surgery should be taken into account. Comparisons of outcome studies are difficult as many different evaluation instruments are used and the baseline statuses of the study groups are in most studies unknown.

A search in Medline (keywords: clubfoot , treatment, outcome) revealed nine studies [48, 56, 96, 110, 119-123] of a total of 232 that included a baseline measurement and a control group. Only three out of these nine included assessment instruments with documented reliability [96, 110, 123]. No study accounted for the effects of laterality on outcome. In spite of the enormous amount of outcome studies made, there is still no treatment method that has been evaluated by prospective controlled studies. The use of the same assessment instruments tested for reliability and validity, control group, baseline severity, treatment experience, same orthosis treatment concept and orthosis compliance are all important factors to be included.

Currently non-operative treatment is advocated in the initial phase of correction [124-127] as outcome results after extensive surgical interventions has shown an increased risk for pain, stiffness and artrosis [48-50]. Several studies have shown the Ponseti casting method to be the most promising method with good rapid initial correction of clubfoot deformity. The French method [32] also gives good results, but the method is more time consuming and therefore more costly.

The comparison of two methods of treatment in paper III was designed with the aim of controlling as many factors as possible that could influence outcome. As a consequence, the Foot Abduction Orthosis (FAO) normally used in the Ponseti treatment concept was not used. These two treatment groups are still under follow-up. Most of clubfoot research has been concentrated on the initial correction of the clubfoot and its outcome. The next interesting step in clubfoot treatment development lies in evaluating the effects of orthosis treatment.

Motor ability in children treated for clubfoot

The study in Paper IV was initiated by the experience that some children with clubfoot seem to have problems with coordination, concentration and balance which could not be connected to the child's clubfoot function. It is, to our knowledge, the first study done on developmental motor ability (DMA) in a group of children of the same age previously treated for clubfoot. The high prevalence of DMA problems without clear relation to the foot function confirmed the observation. The only item in the CAP that showed statistically significant correlation with MABC was CAP**one-leg stand**. Three out of the seven children with motor problems according to the MABC also had unsatisfactory foot function outcome according to the established criteria's using the CAP. Another interesting, and positive, finding was that the amount of surgery did not seem to be related to increased DMA problems.

The results of this study indicated that factors causing nervous system dysfunction, giving rise both to development of the clubfoot deformity and to central motor performance problems, are plausible in at least a proportion of children with so-called idiopathic clubfoot. Max et al. [128] showed a surprisingly high rate of ADHD/traits among male children with idiopathic clubfoot which may support part of our results and our observation in daily clinical practice. Furthermore, some of the children with idiopathic clubfoot may, besides having musculo-skeletal problems, also have developed secondary problems with motor processes (neuromuscular), sensory processes and higher-level integrative processes (perception and cognition). There is an obvious risk that developmental disorders of motor function in children treated for idiopathic clubfoot might not be recognized. Instead the child's motor problems are blamed on their clubfoot.

We suggest that children with clubfoot and obvious problems with balance, irrespective of their clinical foot status, combined with parental observations, should be offered a thorough neurodevelopment assessment in addition to their orthopaedic follow-up. Experiencing less success in movement skills, in sports, playground and other daily activities, may cause avoidance of these situations, decreasing their opportunities to gain the practice and experience necessary to develop both motor skill competence and social interactions required for well-being [129, 130].

General conclusions

- The CAP can be used with sufficient reliability in clinical practice. In research studies the users should be experienced with the CAP, and the observer differences should be checked.

- The CAP has shown to have sufficient validity and a stronger discriminative validity than the DCS. It is more sensitive to change regarding severity in the moderate to severe range, while the DCS focuses on the extremes. The CAP provides information on the development of different functional domains over time within clubfoot diagnosis.

- Clubfoot outcome studies are in need of quality control. Most research has been concentrated on the initial correction phase without accounting for other influencing factors. The Ponseti casting technique is an effective and cheap correction procedure confirmed both by paper III and the literature. An important step in clubfoot treatment development lies in evaluating the effects of orthosis treatment.

- A significant higher prevalence of DMA problems was found in children treated for idiopathic clubfoot. Children with clubfoot showing obvious problems with balance should be offered a thorough neurodevelopment assessment in addition to their orthopaedic follow-up.

- This thesis contributed to the basics in the complex process of developing a measurement instrument. The results of the CAP clinimetric properties and clinical usefulness are promising. This justifies continuing simultaneous usage and evaluation of the CAP in different clinical and methodological study designs.

Summary in Swedish

Klumpfot är en medfödd fotfelställning, som drabbar 1 av 1000 barn. Behandlingen består initialt av korrektion av felställningen, genom töjning och eventuellt operation. Det finns flera alternativa metoder att genomföra töjningar, och flera alternativa operationsmetoder. När felställningen korrigerats övergår behandlingen till att bibehålla det uppnådda läget med hjälp av ortos (skena). Även här finns flera olika alternativa metoder. Det finns få vetenskapliga studier som utvärderat klumpfotsbehandling med instrument som visats var tillförlitliga och valida, och där man använt kontrollgrupp för jämförelse.

Denna avhandling belyser utvecklingen av ett bedömningsinstrument, Clubfoot Assessment Protocol (CAP), för uppföljning av barn med klumpfot. CAP ger information om flera olika aspekter på klumpfot (morfologi, rörlighet, muskelfunktion, och rörelsekvalitet). Instrumentets reliabilitet, validitet, förmåga att upptäcka förändring och dess kliniska användbarhet studerades prospektivt på 73 konsekutiva barn med klumpfot. Den kliniska användbarheten utvärderades genom att jämföra gipsteknik (enligt Ponseti) med töjningar (enligt Köpenhamnsmetod) och ortosbehandlingen, och genom att studera utvecklingsbetingade motoriska problem hos barn med klumpfot.

Metodstudierna visade att CAP kan användas med tillräcklig reliabilitet och validitet. CAP uppvisade en bättre förmåga att beskriva skillnader i fotens status jämfört med det vanligast förekommande instrumentet (Dimeglio Classification System). CAP var också mer känsligt för förändring jämfört Dimeglio's system.

Gipsteknik enligt Ponseti gav, enligt CAP, bättre resultat än töjningar enligt Köpenhamnsmetoden vad gäller rörlighet och rörelsekvalitet vid två års ålder. Dessutom krävdes färre kirurgiska ingrepp. Behandlingen med dynamiska Knä Ankel Fot Ortos (KAFO) eller Ankel Fot Ortos (AFO) gav bibehållet eller något förbättrat kliniskt status och visade god följsamhet.

Utvecklingsbetingade motoriska problem sågs oftare än förväntat hos sjuåriga barn som behandlats för klumpfot. Detta fynd kunde inte förklaras som en konsekvens av kirurgiska ingrepp eller av fotens funktion. Förmågan att stå på ett ben korrelerade bäst med motoriska problem och skulle kunna vara en screeningmetod i kliniskt arbete med klumpfot.

Sammanfattningsvis visar undersökningar att CAP förefaller vara ett värdefullt instrument vid bedömning och uppföljning av klumpfot.

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Appendix

Manual

Clubfoot Assessment Protocol

CAP (version 1.2)

Description of the assessment methods and criteria used in the clubfoot follow-up protocol.

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Introduction

The purpose of the CAP is to provide an overall profile of the clubfoot child's functional status within the domains of body function/structure and activity on single assessment occasions and over time. Furthermore, the CAP aims to provide structure and standardization for follow-up procedures from 0 to 11 years of age in daily clinical decision making. It is an observer administered test. The selection of important items to be included in the protocol and scoring system was an act of balance between considerations of clinical utility and scientific interest. Literature studies, expert opinions and clinical experience on what patients /parents present as important factors formed the platform for the CAP prototype.

The CAP contains 19 items in four sub-groups: mobility (seven items), muscle function (two items), morphology (four items), and motion quality (six items). The first three sub-groups relate to body function/structures and the last to activity according to ICF-2001. Questions about pain, stiffness and daily activity/sport participation are routine and asked in a standardized way.

Each item is described in the manual along with the criteria for scoring. The scoring is divided systematically in proportion to what is regarded as normal variation and its supposed impact on perceived physical function ranging from 0 (severe reduction/ no capacity) to 4 (normal). Score grading can vary between 3 to 5 levels. For sub-groups the sum of the items scores are calculated and can be visualized as profiles (transformed to a 0–100 scale score, with 0 = extremely deviant and 100 within normal variance; sub-group transformation score = actual score/maximal possible score × 100).

In general non-parametric statistics are advocated in data analysis as the underlying data have an ordinal/interval construct.

Administration time varies between 10 and 15 minutes dependent on the child's cooperation. Six items assess motion quality and are age dependent. At the age of three years all children are presumed to be able to perform CAP**Motion quality part I**. At the age of four years all children are expected to be able to perform CAP**Motion quality part II**. Knowledge and experience on normal child neuromotor development is a prerequisite for enabling proper assessment of the sub-groups muscle function and movement quality.

Reliability and validity of the CAP items

The CAP items can be used with sufficient reliability, independent of age, by examiners with good clinical experience of the instrument.[1] Inexperienced CAP users showed sufficient intra-rater reliability though inter-rater reliability was between

fair and moderate (unpublished data). If the instrument is to be used in research studies, training and a learning period is necessary before starting. The observers involved should synchronize their assessments and check their mean difference score in a small sample size reliability test.

Validity studies [1-4] showed appropriate face, content, construct, discriminate and longitudinal construct (responsiveness) validity.

The CAP is still under further development and more clinical and methodological testing is needed before its methodological properties are fully confirmed.

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The CAP protocol and its assessment procedure

The examination is done in a child friendly environment and with possibilities for observing activities. It is important that the child feels at ease to encourage co-operation.

For left and right different colours can be used when filling in the protocol (See Table). With uncertainty between two scores chose the lower one. A mark, e.g. ↔, can be placed beside that category as an observation in future assessment.

Scoring and interpretation

Scoring is made using a 3 or 5 point ordinal scale. Higher scoring indicates better clinical/functional status.

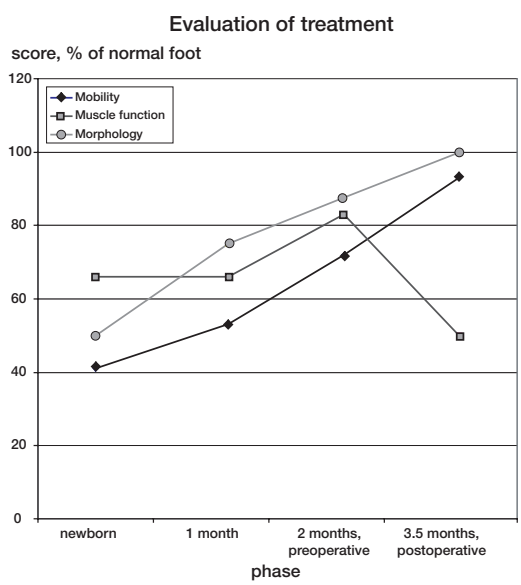
For each subgroup a summary score can be made. This score can than be converted to a percentage score where 100% means within normal variation and 0% means serious functional problems: Sub score / maximal possible score x 100

Missing data is substituted by the average of item scores from the time points before and after the missing data. Items not applicable are distracted from the maximal possible scoring.

Maximal possible scoring:

CAP**Mobility I**: 20 points.
CAP**Mobility II**: 8 points
CAP**Muscle function**: 8 points.
CAP**Morphology**: 16 points.
CAP**Motion I**: 16 points.
CAP**Motion II**: 8 points.

With use of a computer program a profile can visualize the clubfoot different subgroups development (See Graph).



Example: Clinical examination and motion quality assessment (CAP)

Name: Peter N — Date of birth: 011023
Date of assessment: — Assessment number: 5 years
Side: O Left (red mark) — ⊘ Right (blue mark)

Rating	0	1	2	3	4
Passive mobility I					
1. Dorsiflexion	< -10°	-10°– < 0°	0° – < +10°	+10° – +20°	> +20°
2. Plantar flexion	0°– < 10°	10° – < 20°	20° – < 30°	30° – 40°	> 40°
3. Varus/valgus	> 20° varus	20° – > 10° varus	10° – > 0° varus	0° – neutral	> 0° valgus
4. Derotation	> 20° inver.	20° – > 10° inver.	10° – > 0° inver.	0° – 10° evers.	>10° evers.
5. Adduction/abduction	> 20° add.	20° – > 10° add.	10° – > 0° add.	0° – neutral	> 0° abd.
Passive mobility II					
6. Flex. digit. longus	+ reduced		reduced		normal
7. Flex. hallucis longus	+ reduced		reduced		normal
Muscle function					
8. M. peroneus	absent/poor		reduced		normal
9. M. ext. digit. longus	absent/poor		reduced		normal
Morphology					
10. Tibial rotation	+ inward		inward		normal
11. Calcaneus position	> 10° varus		10° – > 0° varus		neutral/valgus
12. Forefoot position	> 20° add.		20° – 10° add.		< 10° add.
13. Foot arch	+ cavus		cavus		normal
Motion quality I					
20. Running 2 year	cannot	+ deviant	deviant	slightly deviant	normal
14. Walking 2 year	cannot	+ deviant	deviant	slightly deviant	normal
15. Toe walking 3 year	cannot	+ deviant	deviant	slightly deviant	normal
16 Heel walking 3 year	cannot	+ deviant	deviant	slightly deviant	normal
Motion quality II					
21. One-leg stance 4 yr	cannot	+ deviant	deviant	slightly deviant	normal
22. One-leg hop 4 year	cannot	+ deviant	deviant	slightly deviant	normal

Standard questions

Pain with activities: Never _ Sometimes _ Regular _ Always _
Stiffness: Never _ Sometimes _ Regular _ Always _
Activity level of the child: Low _ Normal _ High _
Shoe problems: None _ Regular _ Always _ Orthopaedics shoes _
Leisure-time activities:
Does your child experience specific problems in daily life activities such as in sports, cycling, playing and keeping up with peers:

Specification motion quality

_ Intoeing
_ Lateral loading
_ No IC
_ Deviant knee motion
_ Limp
_ Decreased propulsion power
_ Co-ordination problems

+ = Pronounced / very, inver. = inversion, evers. =eversion, add. = adduction, abd. = abduction., flex. digit. longus = length of m. flexor digitorum longus, flex. hallucis longus = length of m. flexor hallucis longus

Clinical investigation

CAP**Mobility**

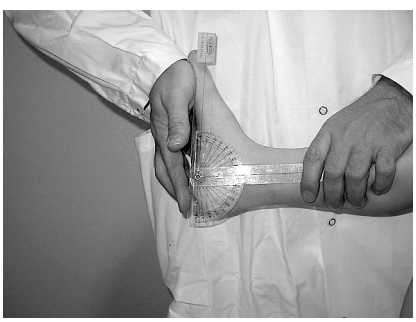
The maximal reducibility is measured. The child is expected to be relaxed and a mild pressure is used.

1. Talar joint (equinus component): dorsiflexion

Method: The child is sitting or supine with the knee extended. Fixation of the tibia with one hand while the other is placed plantar just in front of the heel. The foot is moved from resting position into the direction of dorsiflexion. Horizontal arm of the goniometer along the medial tibia, centre just under the medial malleolus. The vertical arm follows metatarsus I.

Flexion of the knee normally increases dorsiflexion. If not this indicates contracture of the hindfoot capsule.

Problems: Observe where the movement takes place. Movement in the midfoot is easily measured as talar movement.



+ = from plantigrad into dorsiflexion.
– = from plantigrad into plantarflexion.

4 points: > +20°
3 points: +10° – +20°
2 points: 0°– < +10°
1 point: -10° – < 0°
0 points: < -10°

2. Talar joint: plantarflexion

Plantarflexion plays an important role in terminal stance in walking, running and jumping motion. Together with the dorsiflexion movement it provides information about the total ROM available for movement.

Method: Sitting or supine position. Knee flexed. Tibia fixated with one hand while the other holds around the foot and moves the foot into maximal plantarflexion from its resting position. Position of the goniometer same as with measurement of dorsiflexion.

Problems: Observe where the movement takes place. Increased forefoot plantarflexion can compensate decreased motion in the talar joint.

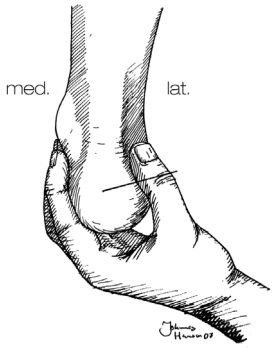
4 points: > 40°
3 points: 30° – 40°
2 points: 20° – < 30°
1 point: 10° – < 20°
0 points: 0° – < 10°

3. Subtalar joint (varus component)

Method: Prone position (baby held with stomach against the parents chest, older child sitting on its knees on a chair with feet hanging over the side). Tibia/fibula is fixated with one hand and the other grips the calcaneus between the thumb and index finger. The foot is hanging freely. Movement in valgus direction and estimation of range of movement is made. A goniometer is used to help with the estimation. The horizontal arm parallel to the transverse of the tibia or floor and the vertical arm following the contour of the heel.

Problems: In new-borns this assessment is difficult as the calcaneus lies more proximal and the posterior part is deviated laterally. Proper assess-

ment cannot be made until the calcaneus has come down in its normal position.



4 points: > 0° valgus
3 points: 0° – neutral
2 points: 10° – > 0° varus
1 point: 20° – > 10° varus
0 points: > 20° varus

4. Derotation around the talus (supination/ inversion/ adduction component)

This is a combined movement of the forefoot, midtarsal and subtalar joints.

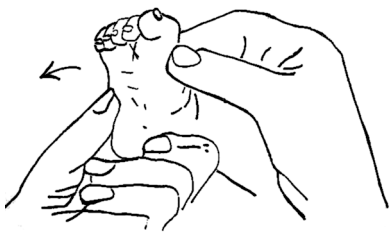
Method: Supine or sitting position. Stabilize the tibia and fibula to prevent rotation on knee level. Identify the tuberositas tibia and palpate talus. A derotation (eversion/abduction) movement is made of the calcaneo-forefoot kinematic chain including external rotation of the foot distal to the talus. One arm of the goniometer is placed parallel to the line through the second toe. The other arm is projected between the malleoli.

4 points: neutral, mobile in eversion (> 10°)
3 points: neutral (0° – 10°)
2 points: slight inversion position (10° – > 0°)
1 point: inversion position (20° – > 10°)
0 points: pronounced inversion position (> 20°)

5. Midtarsal/ metatarsal joint (adduction component)

Method: Supine or sitting. Fixation of the calcaneus (thumb on cuboideum) with one hand and abduction force on middle of metatarsus I with the other hand.

Problems: Observe that increased plantarflexion(cavus) of the forefoot often decreases this movement and dorsiflexion (planus) often increases this abduction mobility(combined movement with the midtarsal and subtalar joint). The forefoot must then first be aligned with the hindfoot.

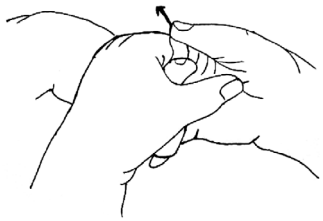


4 points: neutral, mobile in abduction (> 0°)
3 points: neutral
2 points: slight adduction (10° – 0°)
1 point: adduction (20° – > 10°)
0 points: pronounced adduction, crease (> 20°)

6. and 7. Muscle length of toe flexors

The mobility of the m. flexor hallucis longus and m. flexor digitorum longus are assessed. Contracture of these muscles influence the possibility of the foot to roll over normally especially in the end of stance phase.

Method: Supine or sitting. Fixation just proximal of the metatarsophalangeal joints. The foot is held as plantigrad as possible. Have in mind that the ankle position influences the length of the flexors. Thumb or finger underneath the toes (phalanx II–V together and phalanx I by its self) and an extension movement is made.



4 points: normal range of motion (50° – 90°)
2 points: decreased range of motion (20°– 50°)
0 points: significantly decreased range of motion (< 20°)

CAP**Muscle function**
(eversion-dorsalflexion power)
Specific muscle activity

8. Activity of m. peroneus

This muscle is of importance for controlling eversion movement and stabilising in stance.

In small children this is estimated by stimulation on the outside of the fibula and downwards, around the back of the lateral malleoli towards the fifth toe.

Observation in playing situation of spontaneous movement. Foot reaction on balance disturbance is also a possibility.

Older children that can cooperate are asked to move the foot into eversion and hold against resistance.

The grading scale 0–5 is followed in an attempt to estimate muscle function.

4 points: normal (4–5), can hold against resistance
2 points: reduced (3–4), problems holding against resistance
0 points: absent (0–2) or poor (2–3), cannot hold against resistance

9. Activity of m. extensor digitorum longus

This muscle is of importance in pre-swing and loading response phase together with m. tibialis anterior and m. extensor hallucis. It is active in dorsiflexion but because of its insertion it has slightly more eversion influence than m. tibialis anterior.

Observation of activity of this muscle group against resistance. With babies and small children this is done during voluntary movement. Pressure is applied against the dorsal surface of the toes in the direction of flexion while stimulated to movement. Older children that are able to cooperate can be asked to hold against resistance.

4 points: normal (4–5), can hold against resistance
2 points: reduced (3–4), problems holding against resistance
0 points: absent (0–2) or poor (2–3), cannot hold against resistance

CAP**Morphology**

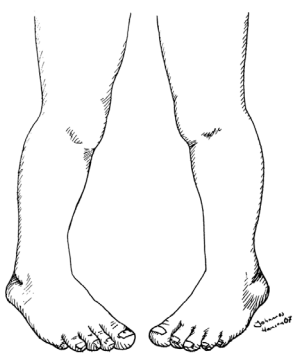
11. Tibia rotation

An increase or decrease in the tibia torque can influence foot progression with either outward toeing or inward toeing.

Method: Sitting or lying prone. Knee extended. The relation between the bimalleolar line and the medial/lateral axis of the proximal articular surface of the tibia is assessed. This normally lies around 20° outwards. With a decrease the foot tends to turn inwards.

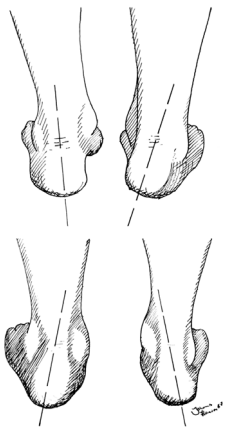
Problems: Difficult to get exact axis. Often in clubfoot the lateral malleoli lies more posterior than normal (caused in most cases by not fixating the talus properly against lateral rotation during the correction phase). This can work confusing when the bimalleolar line is taken as a reference.

With in-toeing also hip movement should be assessed to determine how much is caused by hip anteversion.



4 points: normal
2 points: slightly increased inward rotation
0 points: significant increased inward rotation

12. Calcaneus position



4 points: neutral or valgus (note if excessive valgus)
2 points: slight varus (≤ 10°)
0 points: significant varus (> 10°) or significant valgus

13. Forefoot position

4 points: neutral to slight adduction less than 10°
2 points: moderate adduction (10° – 20°)
0 points: significant adduction (> 20°

14. Foot arch

Estimate (if possible measure) the angle made by a line through the first metatarsal shaft and the plantar surface. On weight-bearing a foot perceived to be normal has an angle between these two landmarks of 25° in a range of 20° to 40°. Feet with this angle greater than 40° have excessive forefoot equinus and can be termed cavus deformity.

With the little child who is not able to stand yet the foot is passively dorsiflexed with the whole hand under the foot.

4 points: normal, nothing specific.
2 points: slight to moderate pes cavus or planus.
0 points: excessive pes cavus or planus.

CAP**Motion quality: Quality of basic motor performance**

Movement quality focuses on the child's ability to perform an activity in an age appropriate manner, which includes sufficient and efficient power generation, joint kinematics and body balance.

Assessing movement quality in children is sometimes difficult. Experience of motion analysis, knowledge of child neuromotor development and experience in working with children influences reliability and validity while testing. The child's ability to cooperate, as well as the level of concentration and task comprehension required, combine to make this task even more difficult.

The intention of this category of the CAP is to try to provide structure as well as to standardise the observation of various activities which after literature study, clinical experience and colleques discussion were thought to be valid for a profile of the child's functional ability.

Rating: Each activity is rated between 0–4 points (5 level scales). For each rating a description of criteria are provided. The amount of points is related to the impact it is expected to have on the child's activity-participation level in accordance with the classifications levels used in the ICF.

The assessment is to be done in a surrounding where the child feels at ease. The younger child will need to be stimulated to perform the movements we wish to observe. Playing situation can provide us with rich information. Even observation of older children performing non-specific tasks, such as playing a ball, provides us with information.

If problems with performance are thought to be caused by other factors other than decreased mobility and periphery muscle function such as hypo/hypertoni, hyperactivity, concentration problems, clumsiness and/or obesity this should be noted separately.

It should also be noted that the information gathered from the clinical assessment should be disregarded while assessing the activity. It is very easy to be influenced and prejudiced, altering ones judgement. The aim is to assess the quality on how the activity is preformed and not to analyse it.

Testing environment: Normal clinical situation with a corridor approximately 10 meters in length. Parent standing on one side and examiner on the other side.

Testing procedure: A good way of starting the performance test is to let the child decide if he/she wants to begin with running or walking. Being able to have some control over the situation makes the child feel at ease.

CAP**Motion quality I**

15. Running

Have in mind that some children have a tendency to run more on the forefoot, which can be a normal variation in running.

Observe in the same way as described for walking.

Instruction: "Run as fast as you can to your mother/father and than back again to me. Ready steady go….".

4 points – normal:

Nice flow, no compensation on knee/hip level, good foot position during the whole stride; straight-line.

3 points – slightly deviant:

a. Clear intoeing without lateral loading and /or varus, normal progression over the foot
   or
b. tendency to intoeing and lateral loading.

2 points – deviant:

a. Intoeing and lateral loading, tendency for "rolling" over the lateral foot border creating a feeling that the child is bow-legged
   or
b. IC lays more lateral and anteriorly causing a short step length, increased hyperextension on knee level
   or
c. increased knee flexion in stance, clear decreased push-off power, difficult to increase speed.

1 point – very deviant:

Same as 2 points though together with clearly increased pelvic movement. Energy consuming.

Difficult keeping a straight progression line.

0 points:

Cannot accomplish the task.

16. Walking

Observe the child's ability to walk straight, step length, initial contact of heel (IC), knee/hip movement (hyperextension/flexion), roll over the foot (loading), intoeing (varus-adductus foot, tibia inward rotation, hip-ante version).

Instruction: "Walk straight towards your mother/ father and (once there) back to me" (= the examiner).

4 points – normal:

Smooth pattern, normal initial heel contact (IC), good progression over the ankle, normal knee flexion/extension pattern, hip stabilisation, normal foot progression angle. A very slight in- or outtoeing.

3 points – slightly deviant:

a. Only in toeing or out toeing with normal IC, loading and progression over the ankle/foot
   or
b. a tendency to in toeing together with a tendency for tipping laterally. Normal knee/hip movement.

2 points – deviant:

a. A clear in toeing with lateral loading and/or varus of the heel
   or
b. normal/nearly normal foot progression though compensation on knee/hip level such as genu varum, hyperextension in knee in stance phase; short stride length, no clear initial heel contact
   or
c. increased ankle dorsal flexion and/or knee flexion in stance.

1 point – very deviant:

a. No IC and/ or significant lateral loading hyperextension /instability on knee level; increased hip flexion and increased pelvic rotation
   or
b. highly increased in toeing with clear compensation mechanism on knee/hip level such as increased hip outward rotation; the movement looks energy consuming; difficulties at keeping a straight walking path.

0 points:

Cannot accomplish the task.

17. Toe walking

Observe how much maximal plantar flexion the child is able to achieve. Look at stabilisation around knee and ankle and position/ alignment in relation to upper body. Does the child manage to keep good maximum height or does he/she "start dropping" the heel in stance after 3–5 steps?

Disregard any intoeing!

Procedure: The child is standing on the side of the examiner and walks toward the parents.

Instruction: Visual by showing what tiptoeing means and verbally by saying: *"let's see if you can tiptoe all the way to mummy/daddy and try to make yourself really long"*.

If the child starts dropping the heel try to correct it by urging once again to go high up. Observe if child manages to correct him/herself.

4 points – normal:

Manages without particular problems; good ability to roll over to the toes, lifts up relatively high and keeps balance; retains this position throughout the walking path.

3 points – slightly deviant:

a. Sufficient power to get up on toes and keeping position throughout the walking path, good upper body alignment though decreased plantar flexion
   or
b. appropriate plantar flexion but not enough power to retain position, the heel starts dropping to the ground after a couple of steps.

2 points – deviant:

Decreased plantar flexion and insufficient power to retain position, drops heel to the ground with weight shift, can correct but soon drops again.

1 point – very deviant:

Clearly decreased active plantar flexion together with compensation on knee/hip level (flexion).

0 points:

Cannot accomplish the task.

18. Heel walking

Observe where the child has its center of gravity (COG). Is it perpendicular to the ankle or behind? Is there hyperextension in the knee and or increased hip flexion? How is the position of the foot? Does the foot drop laterally or is there a good balance between invertors/evertors.

The child is standing on the parent's side of the walkway and walks towards the examiner.

Instruction: Visual by showing the child how to walk on heels and saying: *"let's see if you can walk all the way on your heels to me"*.

Make clear to the child that it is important that he/she lifts his/her feet properly from the floor (show again if necessary).

4 points – normal:

Manages without great effort, body position relaxed and the centre of gravity lies over the ankle or slightly shifted backwards. Good ankle dorsal flexion.

3 points – slightly deviant:

a. Some effort is needed to keep balance; decreased dorsal extension, slight compensation such as shifting the COG behind the ankle; tendency to hyperextend on knee level and flex on hip level

   or

b. appropriate forefoot lift though a tendency to drop the lateral border of the foot and / or varus of the heel.

2 points – deviant:

a. COG clearly behind the ankle joint, clear hyperextension in knee and hip flexion but still good control over the foot

   or

b. nearly normal position of COG over the ankle but cannot lift the lateral border sufficient enough from the floor.

1 point – very deviant:

There is both highly increased hyperextension on knee level and hip flexion together with varus and drop of lateral border; compensation mechanism is evident; the child has great difficulties keeping balance.

0 points:

Cannot accomplish the task

CAPMotion quality II

In the following two parameters both quality and quantity are assessed. Have in mind the child's age when assessing performance quality. A child of four does not have the same motor maturity as a child of five and balance maturity may even vary between children of the same age.

The ability for task understanding and to concentrate is an important factor for good performance. Therefore a note should be made about the child's ability for these factors if deviating from normal.

Definition: Balance = keeping the centre of gravity within the stance limb surface. Instability is avoided by a shift of the body vector toward the standing limb and strong contraction of the hip abductors to support the pelvis.

19. One-leg stance

Besides determining the quality of performance the number of seconds the child manages to stand on one leg is counted. The best result out of three trials is taken.

Before testing balance on one leg the child should be allowed to have a short rest.

Instruction: Visual by showing and verbally by saying: "*let see how long time you can manage to stand on one leg. I will be counting one, two, three….*".

If the child has problem focusing try to make it look straightforward and concentrate on an object slightly under eye level.

4 points – normal:

The child easily finds its balancing point; good alignment of upper body over the standing leg; the child looks confident in its position; no significant postural sway.

3 points – slightly deviant:

Some effort is needed to find the balancing point; can keep good alignment with upper body most

of the time though (slight postural sway) needs to adjust slightly now and then.

2 points – deviant:
The child needs more time to find the balancing point, finds it but soon starts working with upper body and arms to keep balance; upper body centre of gravity falls outside the foot.

1 point – very deviant:
The child needs help to find the balance position; no alignment of upper body; needs to work a lot with arms and legs to keep standing on one leg.

0 points:
Cannot accomplish the task. None of the criteria above.

20. One-leg hop

Besides assessing quality also the numbers of hops are noted for each leg.

Instruction: Visual by showing the child how to hop on one leg and verbal by asking the child to hop as far as possible: *"How far can you hop!"*

4 points – normal:
Stable body position and alignment; the child seems secure in its task; finds its balance quickly and hops away with good stride and power; keeps a straight path.

3 points – slightly deviant:
Some effort to find balancing point; needs some more help from his arms to get going and for maintaining balance.

2 points – deviant:
Problem with keeping balance; difficulty in keeping a straight path; insufficient propulsion power and/or either short hop stride or unregulated hop stride.

1 point – very deviant:
Great difficulties in getting started, finding balance and propulsion upwards/ forwards, manages a couple of hops but falls over.

0 points:
Cannot accomplish the task. Cannot even get started.

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