Evaluation of treatment and follow-up methods in children with clubfoot

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LUND
UNIVERSITY

DOCTORAL DISSERTATION

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Title and subtitle: ***Evaluation of treatment and follow-up methods in children with clubfoot***

Abstract

Background: Clubfoot affects on average 1 per 1000 children. Nonsurgical methods are the gold standard for initial treatment. However, relapse i.e., recurrence of the deformity and gait deviations are common even after successful initial treatment. The general aim of this thesis was to expand the knowledge of clubfoot management by evaluating the treatment and follow-up methods and identifying prognostic factors for relapse.

Methods/Results: *Study 1* included 20 consecutive children with clubfoot treated with Ponseti casting and custom-made dynamic orthoses (KAFO/AFO). Data for relapse, compliance reports, and three-dimensional gait analysis (3DGA) at the age of 7 years were used to evaluate the KAFO/AFO treatment. No relapse during the orthosis period was observed, but relapse in need of surgery was observed in 20% of the children after the orthosis period. Good compliance was recorded. 3DGA showed no equinus or calcaneus gait and normal tibia and hip rotation. Increased plantarflexion at initial contact and at the end of swing was observed in 16% of the feet,and increased internal foot progression in 53%. Ankle power and moment decreased.

Study 2 included the 20 consecutive children from Study 1. The overall gait scores were calculated using the Gait Profile Score (GPS) and Gait Variable Score (GVS), which were derived from 3DGA. The Clubfoot Assessment Protocol (CAP) was used to assess the clinical status. Correlations were found between foot-related variables for the GVS and clinical assessments for the CAP, and between clinically observed gait deviations for the CAP item walking and GPS.

Study 3 included 72 consecutive children with clubfoot treated with Ponseti casting or the Copenhagen stretching method followed by the use of KAFO/AFO. Foot length (FL) was measured every 6 months from age 2 to 7 years. Motion quality according to the CAP and relapse were assessed at the age of 7 years. The contralateral feet from children with unilateral clubfoot were used as the reference feet. The development of FL was analyzed in relation to the initial treatment, initial FL, motion quality, and relapse. Clubfeet were smaller than the reference feet at all ages but had a similar growth rate up to age 7 years. Small FL at the first measurement was associated with more relapses and poorer motion quality.

Study 4 included 19 children with clubfoot aged 2.5–7 years treated with the Ponseti method. The reliability of the foot drawing method (FDM), which measures FL and foot rotation, was evaluated. The feet were measured twice on 1 occasion by 2 raters. Systematic differences and limits of agreement (LoA) were used to assess reliability. No statistically or clinically significant systematic differences were observed. The LoA was <6 mm for FL and <18° for foot rotation. The intra- and interrater differences were less pronounced in feet with lower degrees of rotation.

Conclusion: KAFO/AFO can be a useful alternative for clubfoot treatment. Overall 3D gait deviation scores reflect clinically observed gait deviations in children with clubfoot. The FDM is reliable for monitoring foot growth and foot rotation during clubfoot follow-up, though the method is more reliable in stiffer feet. Children with small FL at the age of 2.5 years are prone to a higher risk of relapse and poor motion quality.

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To all children born with clubfoot

Το thesis αυτό γράφτηκε μετά από θυσία, και σκέψη απερίγραπτη κόρης απ΄ τη Σητεία.
Είχε καθήκον και όφειλε εις την ζωή να πράττει σαν γνήσια απόγονος του γέρο Ιπποκράτη.
Το έργο της προσφέρεται ώθηση για να δώσει, στην επιστήμη την ιερή που' χει ανάγκη τόση.

Mantinada (Cretan traditional style humorous rhyme)
By Roussos Bourmpakis

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List of papers

The present thesis is based on the following original papers.

1. Manousaki E, Czuba T, Hägglund G, Mattsson L, Andriesse H. *Evaluation of gait, relapse and compliance in clubfoot treatment with custom-made orthoses.* Gait and Posture. 2016 Oct; 50:8–13, *DOI: 10.1016/j.gaitpost.2016.08.005*
2. Manousaki E, Esbjörnsson AC, Mattsson L, Andriesse H. *Correlations between the Gait Profile Score and standard clinical outcome measures in children with idiopathic clubfoot.* Gait and Posture. 2019 Jun; 71:50–55, *DOI: 10.1016/j.gaitpost.2019.04.009*
3. Manousaki E, Esbjörnsson AC, Hägglund G, Andriesse H. *Development of foot length in children with congenital clubfoot up to 7 years of age: a prospective follow-up study.* BMC Musculoskeletal Disorders. 2021 May 27;22(1):487, *DOI: 10.1186/s12891-021-04323-4*
4. Manousaki E, Andriesse H, Hägglund G, Ström A, Esbjörnsson AC. *The foot drawing method: Reliability of measuring foot length and outward rotation in children with clubfoot*. (Submitted)

Abstract

Background

Clubfoot affects on average 1 per 1000 children. Nonsurgical methods are the gold standard for initial treatment. However, relapse i.e., recurrence of the deformity and gait deviations are common even after successful initial treatment. The general aim of this thesis was to expand the knowledge of clubfoot management by evaluating the treatment and follow-up methods and identifying prognostic factors for relapse.

Methods/Results

Study 1 included 20 consecutive children with clubfoot treated with Ponseti casting and custom-made dynamic orthoses (Knee-Ankle-Foot Orthosis (KAFO)/Ankle - Foot Orthosis (AFO)). Data for relapse, compliance reports, and three-dimensional gait analysis (3DGA) at the age of 7 years were used to evaluate the KAFO/AFO treatment. No relapse during the orthosis period was observed, but relapse in need of surgery was observed in 20% of the children after the orthosis period. Good compliance was recorded. 3DGA showed no equinus or calcaneus gait and normal tibia and hip rotation. Increased plantarflexion at initial contact and at the end of swing was observed in 16% of the feet and increased internal foot progression in 53%. Ankle power and moment decreased.

Study 2 included the 20 consecutive children from Study 1. The overall gait scores were calculated using the Gait Profile Score (GPS) and Gait Variable Score (GVS), which were derived from 3DGA. The Clubfoot Assessment Protocol (CAP) was used to assess the clinical status. Correlations were found between foot-related variables for the GVS and clinical assessments for the CAP, and between clinically observed gait deviations for the CAP item walking and GPS.

Study 3 included 72 consecutive children with clubfoot treated with Ponseti casting or the Copenhagen stretching method followed by the use of KAFO/AFO. Foot length (FL) was measured every 6 months from age 2 to 7 years. Motion quality according to the CAP and relapse were assessed at the age of 7 years. The contralateral feet from children with unilateral clubfoot were used as the reference feet. The development of FL was analyzed in relation to the initial treatment, initial FL, motion quality, and relapse. Clubfeet were smaller than the reference feet at all ages but had a similar growth rate up to age 7 years. Small FL at the first measurement was associated with more relapses and poorer motion quality.

Study 4 included 19 children with clubfoot aged 2.5–7 years treated with the Ponseti method. The reliability of the foot drawing method (FDM), which measures FL and foot rotation, was evaluated. The feet were measured twice on 1 occasion by 2 raters. Systematic differences and limits of agreement (LoA) were used to assess reliability. No statistically or clinically significant systematic differences were observed. The LoA was <6 mm for FL and <18° for foot rotation. The intra- and interrater differences were less pronounced in feet with lower degrees of rotation.

Conclusion

KAFO/AFO can be a useful alternative for clubfoot treatment. Overall 3D gait deviation scores reflect clinically observed gait deviations in children with clubfoot. The FDM is reliable for monitoring foot growth and foot rotation during clubfoot follow-up, though the method is more reliable in stiffer feet. Children with small FL at the age of 2.5 years are prone to a higher risk of relapse and poor motion quality.

Abbreviations

KAFO = knee–ankle–foot orthosis

AFO = ankle–foot orthosis

3D = three-dimensional

3DGA = three-dimensional gait analysis

GPS = Gait Profile Score

GVS = Gait Variable Score

CAP = Clubfoot Assessment Protocol

FL = foot length

FDM = foot drawing method

FR = foot rotation

FTR = foot and tibia outward rotation

LoA = limits of agreement

PMR = posteromedial release

ICFSG = International Clubfoot study group

PF = plantar flexion

DF = dorsal flexion

CAP_{MQI} = CAP domain Motion Quality I

FLG% = foot length growth percentage

uniFLD% = foot length difference percentage

D = drawings

AT = Achilles tenotomy

Thesis at a glance

Study	Aim	Methods	Results	Conclusions
1	To evaluate treatment with custom-made dynamic Knee Ankle Foot Orthosis (KAFO) and Ankle Foot Orthosis (AFO) in children with clubfoot.	Twenty consecutively born children were included. Three-dimensional gait analysis (3DGA) at the age of 7 years and relapse and compliance reports were analyzed.	No relapse during the orthosis period was observed. Relapse in need of surgery was observed in 20% of the children. Good compliance was recorded. 3DGA showed no equinus or calcaneus gait and normal tibia and hip rotation. Increased plantarflexion at the initial contact and at the end of swing was observed in 16% of the feet and increased internal foot progression in 53%. Ankle power and moment were decreased.	KAFO/AFO can be considered useful and effective alternatives for clubfoot treatment. Relapse frequency and level of gait deviations were similar to those in children treated with foot abduction orthosis reported in literature.
2	To identify the relationships between measures of overall gait deviations, such as Gait Profile Score (GPS) and Gait Variable Score (GVS), and clinical assessments of children with clubfoot.	Twenty children were evaluated at 7 years of age. GPS and GVS were calculated from 3DGA data. The Clubfoot Assessment Protocol (CAP) was used to assess the clinical status.	Correlations were found between foot-related variables for the GVS and clinical assessments according to the CAP, and between clinically observed gait deviations according to the CAP item walking and GPS.	GPS and GVS reflect gait deviations as observed clinically in children with clubfoot. Hence, GPS with GVS may be useful for clubfoot follow-up.
3	To describe the development of foot length (FL) in children treated for clubfoot from 2 to 7 years of age and to analyze foot growth in relation to relapse and motion quality.	Seventy-two children were included. FL was measured every 6 months. Motion quality and relapses were assessed at the age of 7 years. The contralateral feet from children with unilateral clubfoot were used as reference feet.	Clubfeet were smaller than the reference feet at all ages but had a similar growth rate up to age 7 years. Unilateral clubfoot with a larger difference in size relative to the contralateral foot at the first measurement relapsed more frequently, and this correlated with poorer motion quality.	Clubfeet had the same growth rate as the reference feet. FL at the age of 2–2.5 years may be a prognostic tool for identifying relapse and poor motion quality at the age of 7 years.
4	To evaluate the intra- and interrater reliability of the foot drawing method (FDM), which measures FL, foot rotation (FR), and combined foot and tibia rotation (FTR) in children with clubfoot	Nineteen children with clubfoot, age 2.5–7 years being treated with the Ponseti method were included. Feet were measured twice on 1 occasion by 2 raters. Systematic differences and limits of agreement (LoA) were calculated.	No systematic differences were found. The LoA for FL was <6 mm and that for FR and FTR was <18°. The intra- and interrater differences were smaller in feet with lower degrees of rotation.	The FDM is useful for clinical practice and research. However, the results of the foot and foot–tibia rotation analyses imply that caution is needed when interpreting changes in feet with high degrees of rotation.

Introduction

Congenital clubfoot, also referred to as congenital talipes equinovarus, is a complex, three-dimensional (3D) foot deformity that is present at birth. It is characterized in the sagittal plane by hindfoot equinus, in the frontal plane by hindfoot varus, and in the transverse plane forefoot adductus [1]. Midfoot cavus is also commonly present [1-3].

If clubfoot remains untreated, the child starts to walk on the lateral edge or, in severe cases, on the dorsal side of the foot [4, 5], which will lead to activity limitations, pain, and stiffness. However, successful treatment of clubfoot leads to a functional and pain-free foot [2, 6-9].

In the past few decades, nonsurgical treatment has become the gold standard for treatment of clubfoot, and the Ponseti method is the current method of choice [2, 6, 10-14]. Nevertheless, relapse, i.e., recurrence of the deformity and gait deviations, is common even after successful initial treatment [6, 7, 14-17]. This thesis focused on evaluating treatment results, follow-up methods, and identifying prognostic factors for relapse.

Epidemiology

The average prevalence of clubfoot is 1 per 1000 live births but has been reported to vary between 0.6 and 7 per 1000 live births [18-22]. The lowest reported birth prevalence is among Chinese people and the highest among Hawaiian and Maori people [18, 20, 22]. In Sweden, the birth prevalence is 1.35 per 1000 live births [23]. Both feet are involved in 50% of cases, and boys are affected at least twice as often as girls [21-24]. Clubfoot is most often idiopathic (isolated) but can be syndromic or associated with other congenital malformations such as arthrogryposis, spina bifida, tethered cord syndrome, and amniotic band syndrome [19, 23].

This thesis included only children with idiopathic clubfoot.

Pathoanatomy

Clubfoot can be detected by ultrasound in utero from the 12th week of gestation [25, 26]. The severity of the deformity varies, and similar structural defects in the foot and lower extremity can lead to the characteristic abnormal position of the foot and ankle joints of clubfoot [1, 3, 27-29].

The deformity results from altered alignment, orientation, and shape of bones and joints of the lower extremity [1, 2]. The components of the deformity include hindfoot equinus and varus, forefoot adductus, and midfoot cavus [1, 2]. Medial and posterior creases are also commonly present and are more distinct in severe cases of clubfoot [3]. The main factors that influence these components include medial subluxation of the navicular bone, medial deviation of the head and neck of the talus, and equinus of the talus and calcaneus (Figure 1) [1, 2]. The anterior part of the calcaneus deviates medially and downwards, and the posterior part deviates laterally and upwards. The cuboid bone and calcaneocuboid joint are also displaced medially, and the fifth metatarsal is in the correct position in relation to the cuboid bone, but the first metatarsal bone is plantarflexed, which causes the cavus deformity [1, 2].

The malalignment of the joints is caused by the contraction of soft tissues. It is believed that connective tissue structures lose their spatial orientation and become contracted, which leads to ligament contractions [27, 30]. Severe clubfoot is accompanied by extensive soft-tissue abnormalities such as atrophy, shortening, abnormal activity of the leg muscles, and neuromuscular imbalance [31-33]. Moreover, alterations in pedal circulation, such as the absence of the dorsalis pedis and hypoplastic tibialis anterior arteries, have been reported [34]. A high proportion of adipose tissue in the muscles has been observed, and this is more pronounced in severe cases; triceps surae and tibialis posterior are mainly affected [31, 35]. Clubfeet are also smaller in size than normal because of bone hypoplasia caused by intrinsic primary growth disorder [36].

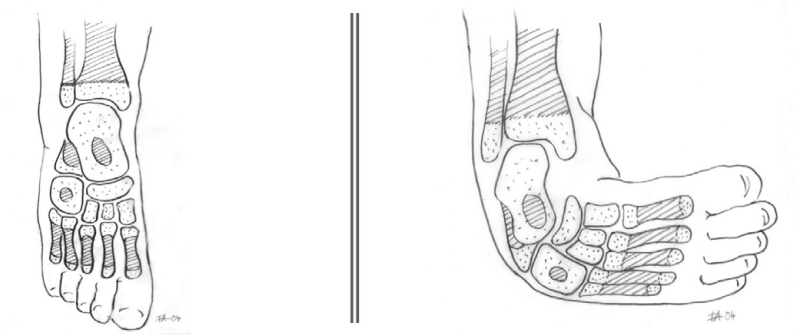

Figure 1. Normal foot (left), clubfoot (right). By permission of Finn Aurell

Etiology

The etiology of clubfoot is believed to be multifactorial. The genetic, environmental, and medical factors associated with idiopathic clubfoot have been described [19, 21, 37-42]. Family and twin studies have shown a clear genetic association [43, 44]. One-quarter of all cases are familial [37, 40], and a sibling of a child with clubfoot has a 2% to 4% chance of having clubfoot [43]. Boys are twice as often affected as girls even in ethnic groups with different prevalence [18-21, 23, 45, 46]. The sex discrepancy in clubfoot enforces the hypothesis of genetic involvement. However, no specific gene has been identified as the clubfoot gene [39, 47-52].

The relationship between environmental and medical factors, and the risk of clubfoot have also been investigated [42, 53-60]. For example, antidepressants taken during pregnancy, amniocentesis, parental smoking, maternal obesity, and gestational diabetes have been reported to increase the odds of clubfoot in some studies [42, 53-60].

Treatment

The goals of clubfoot treatment are to normalize mobility and alignment of the joints and bones affected by the clubfoot pathology and, consequently, to achieve appropriate lower limb function to allow participation in everyday activities without pain. The treatment is long lasting and requires dedication from the affected child, family, and caregivers.

History

The first descriptions of the clubfoot deformity come from archeological studies. In antiquity, deformities were believed to be divine punishment and there was no interest in treating them [61].

The first reports of clubfoot treatment came from Hippocrates in 400 BC [61-63]. In the 'Hippocratic Corpus' clubfoot was described as a treatable congenital deformity, probably caused by intrauterine factors. It was recommended to start the treatment as soon as possible because the deformity becomes more severe with time. Gentle manipulations and strong bandages were proposed for maintaining the correction. This knowledge was ignored later, and clubfeet were again left untreated and associated with evil until the Renaissance[61, 62, 64], when nonsurgical techniques, such as manipulation, straps, plasters, custom-made shoes, and special machines were used [61, 62, 64]. Surgery was not performed because of the risk of infection. After the introduction of antiseptic surgical methods in the late 1800s, the

treatment of clubfoot became progressively more surgical [61, 62, 65]. Extended surgery led to stiffness and pain and, consequently, the treatment shifted again during the 1980s to nonsurgical methods, when treatments such as the Ponseti, French, and Copenhagen methods were introduced [65].

Contemporary treatment

Several methods are currently used to correct the deformity, and these are usually divided in 2 phases: initial correction and maintenance phase. The correction phase starts soon after birth and is mainly nonsurgical. The Ponseti technique is the most widely used initial treatment, although the French and modified Copenhagen methods are also used occasionally [7, 10, 12, 66-70]. Surgical treatment is rarely performed, except in cases in which conservative methods have failed to achieve full correction [71]. Studies have shown that the Ponseti casting technique is the method that requires less extensive surgery at the end of the initial correction [70, 72-75]. After successful initial correction, orthoses are usually recommended to preserve the results because of the high risk that the deformity will reappear [76-80]. The children included in this thesis have been treated either with a modified Copenhagen (modified maintenance phase), Ponseti, or a modified Ponseti method (modified maintenance phase).

Copenhagen method

The initial correction lasts about 3 months and includes daily manipulation followed by the use of a plexidur orthosis. At the end of this period, soft tissue surgery, which most often involves posteromedial release (PMR) and Achilles lengthening, is performed to correct any persisting components of the deformity [79, 81, 82]. After the initial correction phase, the Copenhagen method recommends the use of a hinged custom-made dynamic orthosis (dynamic knee–ankle–foot orthosis or KAFO) until the age of 3 years [79].

Ponseti method

The initial correction phase includes weekly serial manipulation of the foot to correct the clubfoot components followed by a specific casting technique (Ponseti casting technique) [2]. The procedure usually lasts 5–10 weeks. After 5–8 casts, some degree of the equinus deformity remains in 80–85% of cases [2, 10, 13, 83]. To correct the remaining equinus, percutaneous Achilles tenotomy is performed under local anesthesia, and a cast is worn for 3 weeks [2]. After the initial correction phase, the Ponseti method recommends the use of a foot abduction orthosis (FAO) until the age of 4–5 years [77, 78, 80].

Relapse

Relapse i.e., recurrence of 1 or more of the components of the deformity, is a common problem in fully corrected feet [6, 14, 16, 84-88]. Studies have shown that the risk of relapse is strongly related to poor orthosis compliance [16, 71, 89-92]. Different types of orthoses have been used with the aim of improving comfort and have produced diverse outcomes [93-98]. However, even with high compliance, some feet relapse. Other factors associated with relapse include the initial severity, poor evertor muscle activity, difficulty in achieving the initial correction, and educational level of the parents [89, 90, 92, 99-101].

The management of relapse is initially nonsurgical, as in the initial correction, and involves serial casting and/or orthosis treatment. Surgery is performed only when conservative treatment is not sufficient; tibialis anterior transfer is the most frequent procedure performed for relapse [14, 102-111]. Early detection of relapse is crucial because it can reduce the need for extensive surgery [84].

Relapse often occurs before the age of 5 years but can occur later [2, 6, 14, 16, 84, 86, 103, 112], and follow-up even after the age of 5 years is required [14, 84-86, 103, 112, 113]. During treatment and follow-up, various instruments and measurements are used to evaluate the treatment results, assess the need for secondary treatment, and as prognostic factors [89, 92, 99-101, 114]. However, there is a lack of agreement about which measurements are the most appropriate [115].

Classification, follow-up, and outcome measures

The most widely used instruments were developed primarily for classification purposes, such as the Pirani and Dimeglio scoring systems, or as an outcome score, such as the International Clubfoot Study Group's (ICFSG) score [116-121]. A relatively new instrument, the PBS (Pirani Böhm Sinclair) score, was introduced as an alternative to the Pirani score for use in ambulatory children [122]. One validated instrument is the disease-specific questionnaire of Roye, which is a patient-reported outcome measure [123, 124]. The Clubfoot Assessment Protocol (CAP), which was developed for long-term follow-up and evaluates multiple aspects of a child's clinical status, is also a well-established validated follow-up instrument [125-127]. During the past decade, 3D gait analysis (3DGA) was introduced to evaluate clubfoot treatment [15].

In the current thesis, the Dimeglio score, 3DGA, and CAP were used, and a new follow-up instrument, the foot drawing method (FDM), was evaluated.

Dimeglio classification

The Dimeglio classification system is an instrument with high intra- and interrater reliability [117, 128, 129]. It is used widely to assess the initial severity of the clubfoot deformity and the progress of treatment [128, 130-132]. The Dimeglio contains 8 items/parameters [117]. The score ranges from 0 to 20, where 20 represents the most severe deformity. The items included are equinus, varus, supination, forefoot adduction, posterior crease, medial crease, cavus, and deviant muscle function (Table 1).

Table 1. A summary of the Dimeglio classification system [117] (Table reprinted with permission [133])

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.

Gait analysis

3DGA is used to provide detailed information about normal and pathological gait. 3DGA measures joint angles (kinematics) in 3 planes, joint moments, and powers (kinetics) and spatiotemporal parameters [134]. 3DGA is used widely for research purposes to evaluate the effects of treatment on gait [135-137]. 3DGA is seldom used as a standard follow-up method in everyday practice given both the high cost and the time required for assessment and interpretation. Several gait deviations, such as increased internal foot rotation and decreased ankle dorsiflexion (DF) along with decreased ankle power and moment, have been reported in children treated for clubfoot [7, 15, 96, 138-143]. Recent studies using 3DGA have focused on the gait characteristics of children with relapsed clubfoot and on identifying gait indicators for clubfoot relapse [143-146].

The output from 3DGA is often presented as specific parameters, such as knee extension in stance or hip flexion in swing. To complement these specific parameters with measures indicating a person's overall level of gait deviations, measures have been introduced to summarize gait deviations as a single score [135, 137, 147, 148]. In this thesis, the Gait Deviation Index (GDI) and the Gait Profile Score (GPS) along with its subscale the Gait Variable Score (GVS) were used. The

GDI and GPS/GVS are highly correlated, and both express the same concept [137]. These overall gait indexes are used to evaluate gait deviation in children with various diseases, including clubfoot [136].

Clubfoot Assessment Protocol

The CAP is a multilevel, disease-specific, observer-administered test for long-term follow-up [125-127]. It assesses various aspects of a child's clinical status in terms of body structure, function, and activity level according to the ICFSG [121]. Both feet, including the healthy foot in unilateral cases, are assessed separately. The CAP (version 1.2) contains 19 items separated into the 4 domains (Figure 2) of mobility, muscle function, morphology, and motion quality. The scoring for each item is from 0 (severe reduction/no capacity) to 4 (within normal). The CAP clinimetric properties have been shown to have moderate to good reliability, responsiveness, and validity in previous studies [125-127].

Clinical examination and motion quality assessment (CAP version 1.2)

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°var	20°- > 10°var	10°- > 0°var	0°- neutral	>0°vlg
4. Derotation	>20°inv	20°- > 10°inv	10°- > 0°inv	0°- 10°evr	>10°evr
5. Add/abd, ff	>20°add	20°- > 10°add	10°- > 0°add	0°- neutral	>0°abd
II.					
6. Flx.dig.long.	+ reduced		reduced		normal
7. Flx.dig.hall.	+ reduced		reduced		normal
Muscle function					
8. M. peroneus	absent/poor		reduced		normal
9.M. ext.dig.long	absent/poor		reduced		normal
Morphology					
10. Tib.rotation	+ inward		inward		normal
11. Calcaneus pos.	> 10° varus		10°- >0°varus		neutral/ vlg
12. Forefoot pos.	> 20°add		20°- 10°add		<10° add
13. Foot arch	+ cavus/planus		cavus/planus		normal
Motion quality					
I					
14. Running 2y	cannot	+deviant	deviant	± deviant	normal
15. Walking 2y	cannot	+deviant	deviant	±.deviant	normal
16. Toe walking 3y	cannot	+deviant	deviant	± deviant	normal
17. Heel walking 3y	cannot	+deviant	deviant	± deviant	normal
II					
18. 1- leg stand 4y	cannot	+deviant	deviant	± deviant	normal
19. Hop 1 leg 4y	cannot	+deviant	deviant	± 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:
Never □ Sometimes □ Regular □ Always □
Specify problem(s):

Specification motion quality

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

+ = pronounced / very, ± = slightly, var= varus, vlg= valgus, inver= inversion, evr= eversion, add= adduction, abd=abduction, inw= inward rotation, outw= outward rotation, flx= flexor, dig= digitorum, long= longus, hall= hallucis, ext= extensor, tib= tibial, calc= calcaneus, pos= position, y = years

Figure 2. The Clubfoot Assessment Protocol (reprinted with permission of Hanneke Andriesse)

Clubfoot clinic and research in the Department of Orthopedics in Lund

In the Department of Orthopedics in Lund, children with clubfoot have been prospectively followed up in a standardized way since 1995. The following treatment methods were used: modified Copenhagen method until the year 2000, modified Ponseti method (Ponseti casting followed by dynamic KAFO/AFO) until 2010, and the Ponseti method since 2011.

In 2000, a modified Ponseti method was introduced because, at the time of the change, no studies had evaluated its efficacy. The dynamic KAFO/AFO treatment was already in use in the department and had a history of good compliance and results. Hence, it was decided to continue using the dynamic orthoses after Ponseti casting before applying the whole Ponseti concept.

After 2011, following international studies and recommendations, the whole Ponseti concept was applied. This gradual change in treatment provided a unique opportunity to study separately the effects of each phase. Figure 3 shows the timeline of clubfoot treatment in the Department of Orthopedics in Lund.

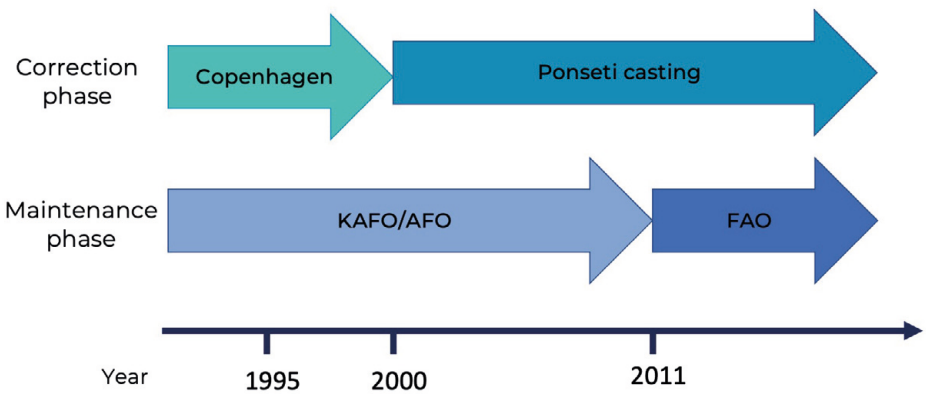

Figure 3. Timeline of clubfoot treatment at the Department of Orthopedics in Lund

This thesis is an extension of a previous thesis in this department: "Follow-up of children with congenital clubfoot. Development of a new evaluation instrument" [133]. The previous thesis developed and validated the CAP follow-up instrument and compared the correction phase between the Ponseti casting technique and the Copenhagen stretching and manipulation treatment. The Ponseti casting technique was found to be superior in terms of outcomes. This thesis evaluated further clubfoot treatment and follow-up methods, and was the introduction of the follow-up FDM instrument.

Aims

The general aim of this thesis was to expand the knowledge of clubfoot management.

Specific aims

1. To evaluate clubfoot treatment using custom-made dynamic orthoses with focus on gait characteristics, relapse rate, and compliance (Study 1).
2. To evaluate the relationship between overall gait deviations, as measured with the Gait Profile Score and Gait Variable Score and clinical status in children with clubfoot (Study 2).
3. To describe the development of foot length in children with clubfoot and to evaluate the relationships between foot length, relapse, and motion quality (Study 3).
4. To evaluate the intra- and interrater reliability of the Foot Drawing Method (Study 4).

Methods

Participants

A total sample of 91 children born with idiopathic clubfoot participated in the studies included in this thesis. All children were followed up prospectively according to a standardized protocol [133]. Figure 4 illustrates the year the children in each study were born in relation to the treatment received.

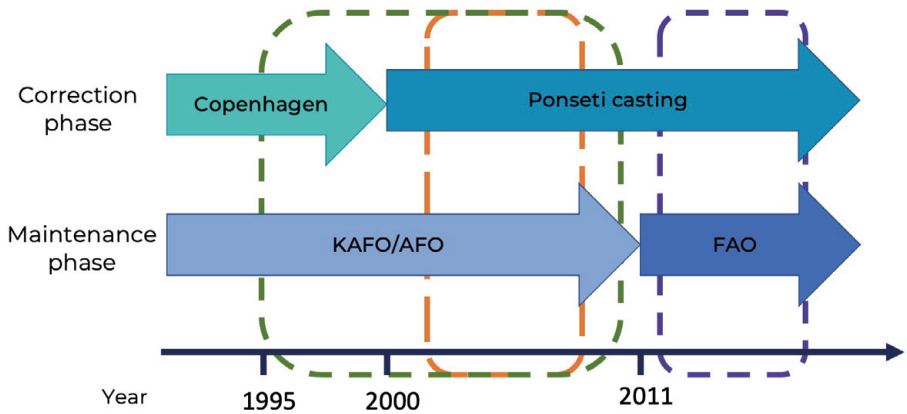

Figure 4. Illustration of treatment received in relation to the year of birth. Treatments included in Studies 1 and 2 are marked in **orange**, Study 3 in **green**, and Study 4 in **purple**.

Studies 1 and 2

Twenty-two consecutive children from our catchment area born with idiopathic clubfoot between 2001 and 2005 were invited for gait analysis at the age of 7 years. Two children did not respond to the invitation, yielding a cohort of 20 children (3 girls, 10 with unilateral clubfoot).

In the first study, all feet with clubfoot were included (30 feet). In the second study, only 1 foot was included from the children with bilateral clubfoot, giving a cohort of 20 feet. The right foot was chosen for every second patient based on their inclusion number in the study.

Study 3

Seventy-eight consecutive children born with idiopathic clubfoot within our catchment area between 1995 and 2007 were invited to participate in this prospective longitudinal study. The standardized follow-up protocol did not change over the study period. Two children declined to participate, and 4 children were excluded because they had ≤4 complete follow-up assessments. In total, 72 children (55 boys and 17 girls, 43 unilateral and 29 bilateral clubfoot) were included. Thirty children were treated according to the modified Copenhagen method and 42 children according to the modified Ponseti method. In children with bilateral involvement, only 1 foot was selected. The right or left foot was chosen based on inclusion numbers to include an equal number of left and right feet.

Study 4

Twenty children with clubfoot born between 2013 and 2018 and in different stages of treatment were invited and accepted to participate. All children were treated according to the Ponseti method. One canceled the appointment because of illness, leaving a sample of 19 children (15 boys, 12 with unilateral clubfoot) between 2.5 and 7 years of age.

Study designs

The designs of the studies are shown in Table 2.

Table 2. Designs of the studies included in this thesis

Study	Aim	Study design	Sample size
1	Evaluation of the effectiveness of custom-made dynamic orthoses	Cohort study	20 children/30 clubfeet
2	Relationships between measures of overall gait deviations and clinical assessments	Cross-sectional study	20 children/20 clubfeet
3	FL development and relationships with motion quality and relapses.	Longitudinal cohort study	72 children/72 clubfeet
4	Intra- and interrater reliability of the FDM	Reliability study	19 children/38 feet (26 clubfeet)

Treatment

The children in this thesis were treated with the modified Copenhagen method, modified Ponseti method, or Ponseti method. Figure 4 shows a chronological overview of the 4 studies and the different treatment methods applied.

Modified Copenhagen method (Study 3)

Treatment started within 2 weeks after birth and included daily manipulations followed by using a plexidur orthosis to maintain the maximal obtained correction [81]. After 2 months, surgery was performed with the child under general anesthesia when needed. This was followed by 4–5 weeks of plaster. The criteria for surgery are shown in Table 3.

Table 3. Criteria for surgery

Criteria	Type of surgery
Isolated equinus <5° dorsiflexion	Percutaneous Achilles tendon lengthening
Varus adductus <15° mobile into valgus abduction	Tibialis posterior lengthening and capsulotomy of the talonavicular joint and, occasionally, tibiotalar capsulotomy
Remaining toe-flexion contracture	Lengthening of the flexor hallucis longus and/or flexor digitorum tendon

When all the components of the deformity were fully corrected, treatment with dynamic orthoses was introduced (Figure 5). The dynamic KAFO was used for 18 h/day during the first 2 months and then gradually reduced to 12 h/day from the age of 8 months (10 h at night and 2 h during the day).

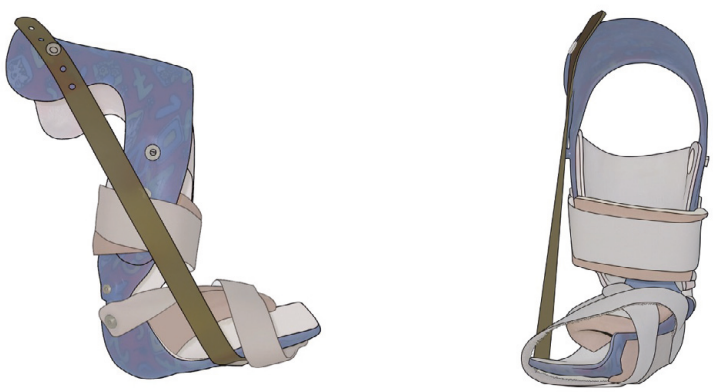

Figure 5. Side and front views of the custom-made dynamic knee–ankle–foot orthosis (KAFO). By permission of Sverrir Kiernan

The original Copenhagen method recommends KAFO until the age of 3 years [81]. Because of compliance problems, which start mainly after the age of 2 years, a dynamic AFO (Figure 6) was developed and used in the modified Copenhagen method. This solution was chosen as a better option than discontinuing the orthotic treatment.

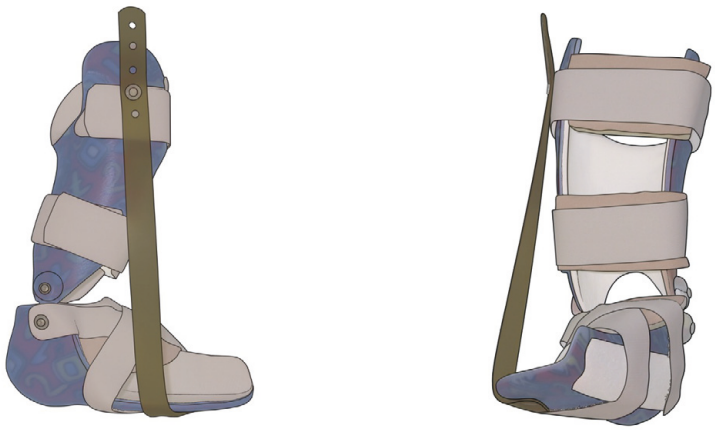

Figure 6. Side and front views of the custom-made dynamic ankle–foot orthosis (AFO). By permission of Sverrir Kiernan

The custom-made dynamic AFO was used for a minimum of 10 h every night from age 2 to 4 years.

Both the KAFO and AFO are designed to allow mobility of the foot, which allows the child to move legs and feet separately, and enable the child to walk if necessary. The dynamic hinged KAFO and AFO are made from individual casts that position the child's foot in maximal outward rotation, as in during Ponseti casting, and provide additional correction of the components of the deformity. The elastic adjustable strap attached to the orthosis stretches the foot into DF when the foot is at rest but does not prevent active plantar flexion. The pad positioned over the lateral part of talus forces the foot toward the medial part of the brace to control forefoot abduction.

Modified Ponseti method (Studies 1–3)

Treatment started within 2 weeks after birth. The initial treatment included weekly serial manipulations of the foot followed by casting according to the Ponseti method [2]. The procedure lasted 5–10 weeks. Surgery was performed at the end of the correction phase with the child under general anesthesia using the same criteria

described above (Table 3). When the feet were fully corrected, dynamic KAFO and AFO were used as described above for the modified Copenhagen method.

Ponseti method (Study 4)

The Ponseti casting technique was used as described above. The remaining equinus was corrected with a percutaneous Achilles tenotomy with the child under local anesthesia when passive DF was <10°. After the initial correction, FAOs were used (Figure 7).

Figure 7. The foot abduction orthosis (FAO) currently used in the Department of Orthopedics in Lund. By permission of Sverrir Kiernan

FAOs are made from 2 shoes connected by a bar that keeps the affected foot in 60–70° of abduction and 10° of dorsiflexion. For unilateral cases, the unaffected foot was positioned in 30–40° abduction. For the first 3 months, the FAO was used for 23 h/day and then gradually decreased to about 10 h/day until the age of 4 years, or longer if needed.

Classification and outcome measures

An overview of the outcome measures and data sources used in each study are shown in Table 4.

Table 4. Overview of the outcome measures and data sources

Outcome measure	Data source	Study 1	Study 2	Study 3	Study 4
Dimeglio classification	Medical records	x	-	x	-
Gait parameters	3DGA	x	-	-	-
GDI, GPS/GVS	3DGA	x	x	-	-
Relapse	Medical records	x	-	x	-
Orthosis compliance	Medical records	x	-	-	-
CAP scores	Physical examination according to CAP	-	x	x	-
Foot measurements	FDM	-	-	x	x

3DGA: three-dimensional gait analysis, GDI: Gait Deviation Index, GPS: Gait Profile Score, GVS: Gait Variable Score, CAP: Clubfoot Assessment Protocol, FDM: foot drawing method

Dimeglio classification (Studies 1 and 3)

All clubfeet were assessed according the Dimeglio classification by the same specialized physiotherapist at the child's first visit to our department. The score was calculated according the Dimeglio manual (Table 1) [117]: 0–4 points (where 0 is best) for the variables of equinus, varus, talus, supination, and forefoot adduction and 1 point if posterior crease, medial crease, cavus, and muscle imbalance was present. The scores (0–20) were registered in the child's medical journal.

Three-dimensional gait analysis (Studies 1 and 2)

3DGA was performed in the gait laboratory in Lund using the Vicon plug-in gait lower-body marker set (Vicon, Oxford, UK). Standard procedures were applied to help make the children feel comfortable and to collect valid data. The procedure was first explained, the general examination was performed by 2 physiotherapists who specialize in gait analysis, and the markers were placed on the child. The child then walked barefoot on a 10-m walkway at a self-selected speed.

Marker data were collected with 6 MX40 cameras capturing at 100 fps. Force data were collected from an AMTI force plate embedded in the middle of the walkway (Advanced Mechanical Technology, Inc., Watertown, MA). Kinematic and kinetic modeling was performed using the Vicon plug-in gait model [149]. Three representative gait cycles from each affected foot were selected. The selected

trials were midsession and of similar speed. The reference data were from 16 typically developing children aged 6.1–12.0 years (median 8.5).

Gait parameters (Study 1)

The kinematic gait parameters analyzed in the sagittal plane were plantar flexion (PF) at initial contact and push-off (toe-off ±2%), and maximum DF during stance and at the end of swing (last 10% of the gait cycle). In the transverse plane, the average hip rotation, shank rotation, and foot progression angle during stance were analyzed. The stance phase was identified individually for each cycle. The maximum peak of ankle moment and ankle power was analyzed from the kinetic parameters. Gait deviations were assessed according to the criteria in Table 5.

Table 5. Criteria for kinematic gait deviations

	Description
Foot drop	Decreased DF/increased PF at the end of swing and increased PF at initial contact (>1 SD from the control group's average values)
Calcaneus	Decreased DF at the end of swing and increased max DF in stance (>1 SD from the control group's average values).
Equinus	Max DF <0° during stance
In-toeing	Internal foot progression during stance (>0°)

DF: dorsiflexion, PF: plantarflexion, SD: standard deviation. Definitions from Karol et al 2009 **[150]**

Figure 8 shows an illustration of the normal gait cycle from the initial heel contact to the next heel contact with the same leg.

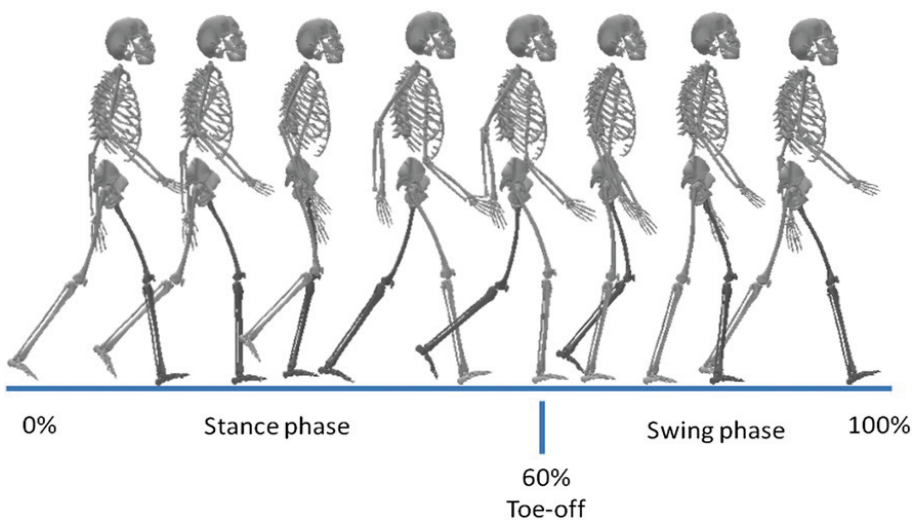

Figure 8. Normal gait cycle defined from the initial heel contact to the next heel contact with the same leg. Reprinted with permission [151]

Overall gait scores (Studies 1 and 2)

The GDI, GPS, and GVS were calculated. The scores are overall gait scores based on kinematics that describe gait deviations throughout the entire gait cycle [147, 148].

GDI contains 9 parameters: rotation angles for the pelvis and the hip (in 3 planes), knee and ankle angles (in the sagittal plane), and foot progression angle (in the transverse plane). GDI measures the scaled distance between patient's data and reference data (for typically developing children). A GDI score >100 indicates no gait pathology and each 10 points indicates 1 standard deviation (SD) away from the control material [148]. In other words, a GDI of 90 implies that the gait pattern deviates by 1 SD from the control, and a GDI of 80 means that the deviation is 2 SD away.

The GPS and GVS are scores reported in degrees that represent the root mean square differences between the patient's data and reference data (for typically developing children). The GPS can be decomposed into a subscale comprising 9 kinematic components called the GVS, which helps to highlight the gait deviations that contribute to the deviant GPS. The 9 components of the GVS are the same as those on which the GDI is based. GPS/GVS values higher than the reference values indicate a deviant gait [147].

The GDI was calculated separately for each affected leg as described by Schwartz and Rozumalski [148]. The GPS was calculated separately for each affected leg ($GPS_{affected\ side}$) and as an overall score ($GPS_{overall}$) for each child. The 9 GVSs were calculated for each affected side. The averages of GDI, $GPS_{overall}$, $GPS_{affected\ side}$, and GVSs over 3 gait cycles for each child were used in the analyses. The GDI, GPS, and GVS were calculated using the spreadsheet provided by the original authors and our reference data for 16 typically developing children.

Relapse (Studies 1 and 3)

Relapse was defined according to the criteria of reappearance of ≥1 of the components of the deformity. These criteria were DF <0° with extended knees, subtalar joint mobility in valgus <0°, foot outward rotation/abduction in relation to the tibia <5°, forefoot adduction >10°, and/or in-toeing gait >10°. Relapse was treated by reintroduction of orthosis, serial casting, surgery, or a combination of these treatments.

Orthosis compliance (Study 1)

The orthosis compliance was assessed by the same specialist physiotherapist at each visit. The criteria used to define compliance were observations of orthosis wear and tear, the parents' confidence and speed when placing the orthosis on the child, and the parents' self-report. Assessment of orthosis compliance was registered in the

medical record as compliant (orthosis used according to schedule), irregular (orthosis used, although probably not according to schedule), and noncompliant (orthosis rarely used).

Clubfoot Assessment Protocol (Studies 1 and 3)

Children in Studies 2 and 3 were evaluated using the CAP by a single experienced assessor who was responsible for the children's treatment and follow-up at the time. The examination took place in a child-friendly environment (spacious, quiet room with toys), and the time for completion was 10–15 min depending on the child's cooperation.

The CAP contains 19 items separated into 4 domains (Figure 2). These domains cover mobility (mobility I, 5 items and mobility II, 2 items), muscle function (2 items), morphology (4 items), and motion quality (motion quality I, 4 items and motion quality II, 2 items). The scoring for each item is from 0 (severe reduction/no capacity) to 4 (within normal). Each score is defined by specific criteria in the CAP manual and is based on the expected effect on function. Cutoff points have been established previously for each item, domain, and total score [152]. Scores below these points are considered to indicate poor clinical outcomes.

Both the left and right sides were assessed regardless of whether there was uni- or bilateral involvement (Figure 2). In Study 2, the assessment was performed after the gait analysis. Scores from all domains and separately for the item walking were used. In Study 3, the latest registered domain motion quality I (CAP_{MQI}) measured at age 6 to 7 years was used in the analysis. The median CAP_{MQI} and previously established cutoff points were used to evaluate motion; scores ≤12 were considered to indicate poor clinical outcomes [152].

Foot drawing method (Studies 3 and 4)

The FDM is an instrument that measures FL and foot outward rotation. It was developed in the Department of Orthopedics in Lund. The FDM was used in Studies 3 and 4. Only the FL measurement was used in Study 3. The entire method was described, and intra- and interrater reliability were evaluated in Study 4.

The FDM standardized procedure is divided into 2 parts as follows.

1. Drawing

The footprint is drawn as described by Hazlewood et al [153]. The child sits on a chair with the ankles, knees, and hips in 90° of flexion. A line is drawn around each foot with the pen kept vertical. The foot is then rotated in maximal external rotation in relation to the tibia, and the medial foot margin is drawn again. During the rotation of the foot, the tibia is held stable with 1 hand, while the other hand is used to rotate the foot at the subtalar joint. Thereafter, the foot is rotated further outward. One hand continues to rotate the foot and the other hand moves from the tibia to the

distal femur to add tibia and fibula rotation at the knee level to the foot rotation. The medial foot margin is drawn once more. Both feet are drawn, even in unilateral cases.

2. Measurements (Figure 9)

FL is the distance between 2 parallel lines, including the footprint, and is measured as follows. Two parallel lines are drawn distally and proximally to the footprint. The proximal line is perpendicular to an imaginary line passing through the middle of the hindfoot. The distal line is parallel to the proximal line, including the footprint. The distance is measured with a ruler.

Foot outward rotation (FR) is the angle between the medial foot margin with the foot in maximal outward rotation and a line drawn vertically to the long side of the paper (Figure 9). The angle is measured with a goniometer.

Foot and tibia outward rotation (FTR) is the angle between the medial foot margin with the foot and tibia in maximal outward rotation and the line drawn vertically to the long side of the paper. The angle is measured with a goniometer.

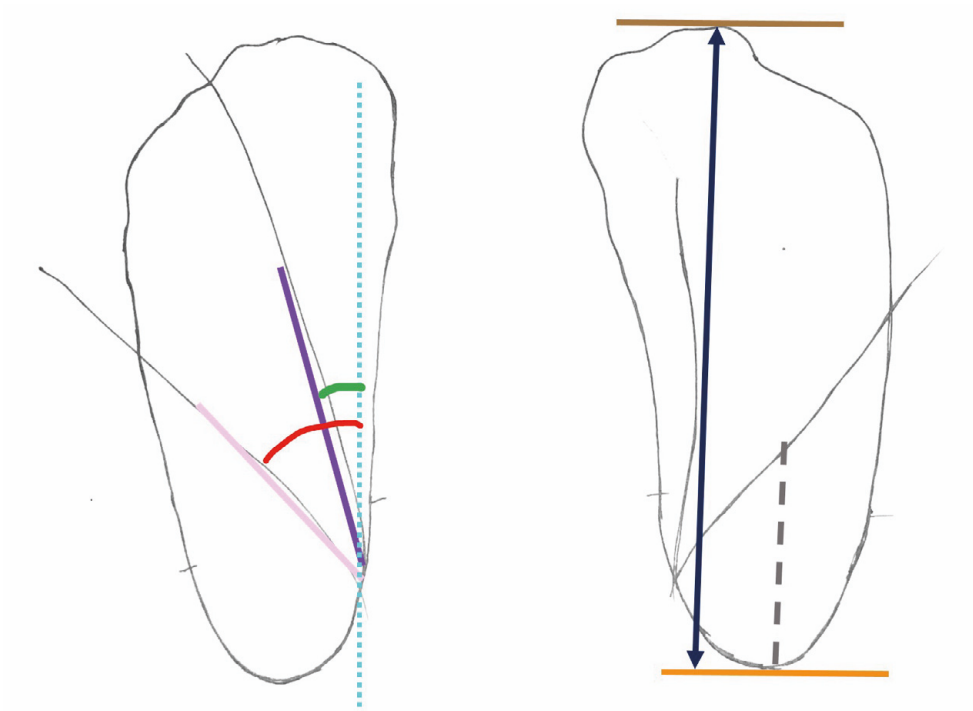

Figure 9. Foot length (**dark blue line**) is defined as the distance between 2 lines. The proximal line (**orange**) is perpendicular to the imaginary line that passes from the middle of the hindfoot (**gray** line). The distal line (**brown**) is parallel to the proximal line and includes the whole footprint. Foot rotation (**green**) and foot and tibia rotation (**red**) are measured with a goniometer between each foot margin (foot rotation margin: **purple**, foot tibia rotation margin: **pink**) and a line (**light blue**) drawn vertically to the long side of the paper.

For Study 3, the same experienced physiotherapist made all the drawings during the children's follow-up visits, and the first author later measured the FL. In Study 4, all drawing and measurements were performed by 2 experienced physiotherapists.

FL calculations (Study 3)

Clubfeet were divided into 4 groups based on their FL at the first measurement using the mean and SD of the normal feet at the same age as a reference. Clubfeet with FL <–2 SD of the FL of normal feet were referred to as xsmall, –2 SD to –1 SD of FL of normal feet as small, –1 SD to the mean FL of normal feet as medium, and clubfeet with FL greater than the mean FL of normal feet as large.

The FL, FL growth percentage (FLG%), and FL difference percentage (uniFLD%) were calculated.

FLG% was calculated by subtracting the previous FL value from the current value. Thereafter, that value was divided by the previous FL value and multiplied by 100 to express the FL growth as a percentage. For example, the FLG% between ages 4 and 4.5 years was calculated as follows: (FL at 4.5 years – FL at 4 years) / FL at 4 years × 100.

UniFLD% was calculated for the children with unilateral clubfoot. The FL value of the clubfoot was subtracted from that of the contralateral unaffected foot. Thereafter, the difference was divided by the contralateral unaffected FL to standardize the difference and multiplied by 100 to express the difference in percentage. For example: (contralateral unaffected foot FL – clubfoot FL) / contralateral unaffected foot FL × 100.

The unaffected foot in children with unilateral clubfoot served as the reference foot in the statistical analysis.

Reliability assessment of the FDM

For Study 4, all drawings and measurements were made by 2 experienced physiotherapists, Rater 1 and Rater 2. The results were used to calculate the systematic differences between raters and to evaluate the intra- and interrater reliability of the method.

First, Rater 1 and Rater 2 made drawings (D) of the children`s feet twice (D1 and D2), independently of each other. The order the raters started altered for every new child. Each of the 2 raters scanned their drawings (D1 and D2) and measured FL, FR, and FTR directly on the copy of their own first drawing (D1). Thereafter, an independent person collected both the original drawings and the copies. After a period of 3–4 weeks, the raters repeated the measurements of FL, FR, and FTR on their anonymized D2 drawing. These were once again collected for further analysis.

The children were divided into 2 age groups (younger and older than 4.5 years of age) to study the effect of age.

Interrater reliability was assessed by comparing measurement from the first D1 of each rater. Intrarater reliability was assessed by comparing the measurements of each rater's D1 and D2.

Statistical methods

Study 1

A nested mixed model was used for the analysis [154]. This method was chosen because it allows one to consider that bilateral cases are related. All available measurements were evaluated (3 cycles for the study group and 2 cycles for the reference group). Each gait parameter was analyzed separately using the independent parameters in the model of group and side. P-values ≤0.05 were considered to be significant. Stata 13 (Stata Statistical Software, release 13; StataCorp LP; College Station, TX, USA) was used for all analyses.

Study 2

Demographic and disease characteristics were described using the mean and standard SD or median and range. The Spearman correlation coefficient was used to identify correlations between clinical assessments and gait analysis because CAP values are ordinal variables. Correlations were interpreted according to Cohen's method (low (0 to ±0.29), moderate (±0.30 to ±0.49), and strong (±0.5 to ±1.0)) [155]. P-values ≤0.05 were considered to be significant. Statistical analyses were performed using IBM SPSS Statistics (version 22; IBM Corp., Armonk, NY, USA).

Study 3

Student's *t* test was used to identify differences in FL and FLG% between clubfeet and reference feet, between clubfeet with and without relapse, and between clubfeet with different initial treatment. In unilateral feet, the paired Mann–Whitney–Wilcoxon test was used to compare the distribution of uniFLD% for the initial measurement between children with and without relapse before the age of 7 years. Spearman correlation was used to identify correlations between uniFLD% for the initial measurement and CAP_{MQI} at the age of 7 years. Fisher's exact test was used to compare differences in relapse frequency and motion quality between children with xsmall clubfeet and those with large clubfeet. The Mann–Whitney *U* test was used to compare distributions of CAP_{MQI} between the xsmall and large clubfeet.

P-values ≤0.05 were considered to be significant. For multiple comparisons, the Bonferroni correction was used [156]. α was set at 0.05, and the P value was adjusted to 0.005. Correlations were interpreted according to Cohen's method [155]. Statistical analyses were performed using IBM SPSS Statistics software (version 25; IBM SPSS, Armonk, NY, USA).

Study 4

The measurements from each rater's D1 were used to assess interrater reliability. The measurements from each rater's D1 and D2 were used to assess intrarater reliability. The mean, 95% confidence interval, and SD of the differences were calculated for all measurements. The *t* test was used to identify systematic differences. Bland–Altman graphs were used to visualize the limits of agreement. The independent samples *t* test was used to compare the foot rotation measurement results from the 2 age groups. All analyses were performed using R version 3.6.3 [157].

Ethical approval

Studies 1–3 were approved by the local ethics committee (Dnr LU 666-3, LU667-3) and Study 4 by the Swedish Ethical Review Authority (Dnr 2020-03008).

Main results

Sample characteristics (Studies 1–3)

Table 6. Characteristics of participants in Study 3 (The sample characteristics for the 20 children of Studies 1 and 2 are integrated in the Table inside {})

	Total (n)	Modified Copenhagen method (n)	Modified Ponseti method (n)
Total (n)	72	30	42 {20}
Sex			
Male	55	21	34 {17}
Female	17	9	8 {3}
Laterality			
Unilateral	44	18	26 {10}
Bilateral	28	12	16 {10}
Dimeglio score			
Median (range)[1]	10 (7–14)	10 (7–14)	11 (8–13) {11 (9–13)}
Surgery before applying KAFO			
PMR	22	17	5 {2}
AT	27	1	26 {14}
No surgery	23	12	11 {4}
Relapse treatment			
Total	23	9	14 {9}
Serial casting/prolonged orthosis	10	4	6 {6}
Minor surgery + serial casting/prolonged orthosis	6	2	4 {3}
Minor surgery	6	4	2 {0}
Major surgery	1	0	1 {0}

Equivalent numbers included in Studies 1 and 2 are presented inside {}
n, number of children; AT, Achilles tenotomy; KAFO, knee–ankle–foot orthosis; PMR, posteromedial release
No statistically significant differences were found between any of the groups for any parameter.
[1]Dimeglio missing values, 19 (9 from the Copenhagen group and 10 from the Ponseti group {2 from Studies 1 and 2}).

Study 1

The gait analysis at age 7 years showed that no children walked with calcaneus or equinus gait. Hip and shank rotation, PF at push-off, and DF at the end of swing were within the reference values. The deviations observed were greater internal foot progression in 16/30 feet (53%), foot drop in 5/30 feet (16%), and statistically significant greater PF at initial contact and less DF at midstance between the children in the study group and the reference group. Peak ankle moment and peak ankle power were statistically significant lower in the children with clubfoot compared with the reference group. The mean GDI of the study group was 89.4 (SD ± 10), with 6 feet <80.

No relapse occurred during the orthosis treatment, which was continued to 4 years of age. The relapses that occurred after the age of 4 years are shown in Table 6.

Eighteen of the 20 children used their KAFO and AFO with good compliance according to the observations. Two children (3 feet) had irregular compliance.

Study 2

The median CAP total scores and median scores of all CAP domains and the item walking were all over the cutoff points previously established [127], except for the domain of motion quality II. The median CAP score for each affected side was 63 (max, 76; range, 52–76, cutoff point, 57). The median score for the CAP item walking was 3 (max, 4; range, 2–4; cutoff point, 2).

Higher $GPS_{overall}$ score and GPS on the affected side were observed in children with clubfoot than for reference data. The most deviating GVSs were for ankle DF/PF and foot progression.

As expected, the $GPS_{overall}$ score showed low to moderate correlations with most of the different aspects of the disease-specific follow-up CAP scores but statistically significant strong negative correlation with the CAP morphology score. The CAP item walking showed a statistically significant strong negative correlation with $GPS_{affected\ side}$ and with the GVS of ankle DF and foot progression. Low or non-statistically significant correlations were found for the remaining GVSs.

Study 3

Study 3 confirmed that clubfeet in general are smaller than normal feet. Fifty percent of the clubfeet had FL <−1 SD of that of normal feet, and 18% had FL greater than or equal to the mean FL of normal feet (Table 7).

Table 7. Size distribution in relation to CAP_{MQI} at the last registered measurement and relapses

Size	Total (n)	Poor CAP_{MQI} (n)	P-value1	CAP_{MQI} median (IQR)	P-value2	Relapse (n)	P-value1
xsmall	11	2 (18%)		13 (12–13)		5 (45%)	
small	25	7 (28%)		13 (10.5–15)		7 (28%)	
medium	23	7 (30%)	0.576	12 (11–14)	0.134	7 (30%)	0.675
large	13	1 (8%)		15 (12–16)		4 (30%)	
Total	72	17 (24%)		13 (12–15)		23 (32%)	

CAP, Clubfoot Assessment Protocol; CAPMQI, CAP domain motion quality I; IQR, interquartile range; n, number of measurements; Poor CAPMQI, under the cutoff of 12; xsmall, extra small. P-value1 between xsmall and large after Fisher's exact test. P-value2 between xsmall and large after Mann–Whitney *U* test. Significance was set at P ≤0.05.

Clubfeet in this study grew at the same speed as healthy feet. The FL and FL growth did not differ statistically significant between clubfeet treated with different initial correction methods nor between clubfeet that relapsed and those that did not (Figure 10).

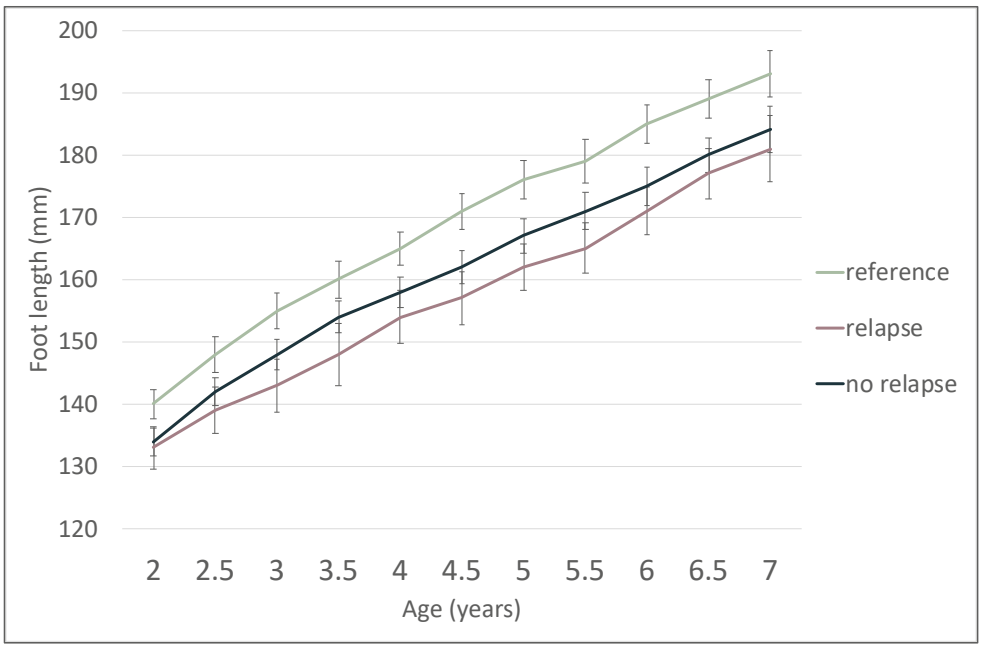

Figure 10. Foot length development in reference feet (**green**), relapsed clubfeet (**pink**), and non-relapsed clubfeet (**dark blue**). The gray lines show the confidence interval.

Before the age of 7 years (median 5 years), 23 of the 72 children (32%) included in the study were treated for relapse (Table 6). Twelve of these 23 children had minor surgery and 1 child had major surgery. In total, 13 of the 72 children (20%) had relapse that required surgery. During the time of relapse, there was a decrease in FL growth between clubfeet that relapsed compared with non-relapsed clubfeet. However, after the Bonferroni correction was applied, this decrease was not statistically significant.

More relapses were observed in children with xsmall clubfeet than in those with larger clubfeet, but the difference was not statistically significant (Table 7). Unilateral clubfeet with a larger difference in FL from the contralateral foot at baseline had statistically significant more relapses and worse motion quality at the age of 7 years.

Study 4

No statistically or clinically significant systematic differences were observed.

The LoA for FL were 4.5 mm to 5.9 mm between raters, -5.9 mm; 4.8 mm and -5 mm; 5.1 mm within raters.

The LoA for FR were -12° to 10.6° between raters, -8.4°; 6.6° and -14°; 14.1° within raters.

The LoA for FTR were -17.8° to 14.3° between raters, -12°; 12.2° and -12.7°; 13.6° within raters.

The intra- and interrater differences were less pronounced in feet with smaller degrees of rotation. This indicated that the method was more reliable in stiffer feet. The average FR and FTR was larger for younger than for older children.

Discussion

This thesis evaluated treatment and follow-up methods in children with idiopathic clubfoot. The results from these studies provide new insights into and extend the current knowledge of clubfoot management.

Are KAFO/AFO good options during the maintenance phase?

Before the introduction of the Ponseti method, orthosis treatment was not used consistently in many clinics. Not using orthosis after the initial treatment has a negative impact on the maintenance of the correction [2, 16]. In feet treated surgically, the use of orthosis is even more important, as scar tissue can become contracted when not stretched regularly.

The 3DGA results from children with clubfoot initially treated with Ponseti casting and dynamic KAFO/AFO (Study 1) showed gait deviations similar to those in studies of children treated with the strict Ponseti protocol (Ponseti casting and FAO) [15, 138, 150, 158]. In this study, the mean GDI was 89.4 (SD ± 10), which is similar to the value of 90.6 (SD ± 6.8) for children treated with the strict Ponseti protocol as reported by Duffy et al. [158].

The peak ankle moment and power were lower than those in children with typical gait patterns, as has been reported in other studies of children treated with the strict Ponseti protocol [15, 138, 150]. Similarly, the deviation observed in ankle DF in the sagittal plane in our study was similar to values reported in other studies [15, 150]. In the transverse plane, internal foot progression was greater than in the reference group. However, in contrast to other studies, no statistically significant differences were found in hip or shank rotation between the study and the reference group [15, 150]. Most other studies report greater external hip and shank rotation in children with clubfoot than in their reference group [15, 138, 150].

The observed greater internal foot progression angle and normal hip and shank rotations in Study 1 may be explained by the design of the orthoses. The dynamic orthoses (KAFO/AFO) used do not engage knee and hip rotation at the same extent as the FAO. The greater external rotation at the hip and shank level observed in other studies using FAO suggests that the correction of the deformity observed was the sum of changes at multiple levels and not at the foot level only.

The low relapse rate observed in the children treated with modified Ponseti in Study 1 was also observed at the 7-year follow-up of the children included in Study 3. The children included in Study 3 had different initial treatments (Copenhagen stretching vs Ponseti casting technique) but the same follow-up protocols and same

orthotic treatments (KAFO and AFO) during the maintenance phase. Twenty-three of the 72 children (32%) were treated for relapse, and 13 (20%) of these were treated surgically. This relapse rate was lower than that in other studies with a similar follow-up period [14, 85]. The relapse rate after treatment with the Ponseti method ranges from 3.7% to 67.3% and correlates highly with the duration of the follow-up and the definition of relapse used, as some studies have reported only relapses that needed surgery [88, 101, 103]. Although the aim of Study 3 was not to evaluate treatment, the relapse rate did not differ statistically significant between children treated with the modified Copenhagen method and those treated with the modified Ponseti method.

Some studies have reported worse results with the use of KAFO and AFO models for clubfoot treatment compared with FAO [93, 95, 97, 98]. However, the biomechanical designs of these orthoses differ from the hinged dynamic orthoses used in Studies 1–3. The orthoses used in the above-mentioned studies included models with a rigid neutral ankle joint and/or less foot outward rotation.

The abovementioned findings from Studies 1 and 3 suggest that dynamic KAFO/AFO can be used during maintenance treatment without compromising the outcome.

How can compliance be improved?

The custom-made dynamic orthoses (KAFO/AFO) have some advantages over the traditional FAO. These advantages should be considered when treating children who are noncompliant with FAO. KAFO/AFO allow the child to move the legs independently, which makes walking possible. In cases of unilateral clubfoot, the orthoses are applied only to the affected foot, whereas FAO engages both feet.

However, reports of compliance can be challenging because they rely mainly on subjective observations and reports by the patient or parents. Some studies have proposed the use of sensors to assess compliance objectively [159-162]. The sensors allow close monitoring of compliance and, consequently, provide more active support to families with compliance issues, and the use of sensors may give extra motivation to the family to use the orthosis because the use is registered [162]. This extra motivation may improve compliance. However, use of sensors during a study that assesses compliance may not reflect real-life compliance because the awareness of monitoring may change the behavior and monitoring during treatment outside a study can be considered ethically questionable.

An important issue that should be considered during orthosis treatment is the increased presence of neurodevelopmental problems in children with clubfoot [17, 152, 163, 164]. Neurodevelopmental issues have been reported in one-third of children with idiopathic clubfoot and can affect orthosis compliance negatively [164]. Thus, the presence of these problems should be considered when treating children with poor compliance with the orthotic treatment. The ideal standardized clinical examination assesses neurodevelopmental symptoms and addresses orthosis

issues in an open and trusting relationship, and this may be sufficient for detecting major orthosis compliance problems.

How and when should clubfoot follow-up be managed?

It is agreed that children with clubfoot should be followed up regularly and past the age of 5 years [14, 84-86, 103, 112, 113]. However, there is a lack of agreement about which outcomes should be measured and reported in clubfoot follow-up and research [115]. All children with clubfoot in our studies were examined using the CAP and FDM. After the age of 2 years, the children were examined at least every 6 months and more often if we observed problems with orthosis compliance or when a relapse occurred. Previous studies have shown good reliability and validity of the CAP as a follow-up instrument [125-127].

The low frequency of major relapse surgery observed in Studies 1–3 compared with studies with the same follow-up period supports the idea that regular follow-up with CAP and FDM is sensitive enough to detect relapses early, before major surgery is needed. In the studies of this thesis, the relapse rate was low regardless of the correction treatment (Copenhagen stretching method or Ponseti casting technique). This may have resulted from the combination of the regular and standardized follow-up, patient-centered care, and parent support we provided to the children and their families, and possibly the use of the custom-made dynamic orthoses.

Are overall 3D-gait measures useful in clubfoot follow-up?

Overall measures of gait deviation such as the GDI and GPS with GVS have been used to describe various pathologies [135, 137] but are not used widely to describe gait quality in children with clubfoot.

Study 2 aimed to evaluate whether and how the GPS and GVS correlate with visually assessed gait deviations and aspects of clinical status assessed with the CAP. The GPS and GVS showed low to moderate correlations with most of the CAP aspects. Most of the low correlations were expected because the CAP assesses multiple clinical aspects of clubfoot pathology and the GPS and GVS are indexes based exclusively on kinematic gait parameters. Nevertheless, the CAP item walking had a statistically significant moderate to strong correlation with overall gait deviations and the gait parameters most affected in children with clubfoot (i.e., foot-related GVSs).

After the publication of Study 2, a new specific gait index, the Foot Profile Score [165], was introduced. This index was derived from the Oxford Foot Model [166], which was shown to be useful in detecting gait deviations associated with relapse [144]. The Foot Profile Score may correlate more strongly with clinical status and may be more useful during clubfoot follow-up, and this relationship should be investigated further. However, the overall scores do not add information about the direction or timing of the deviations and should be used mainly as a complement during clubfoot follow-up.

In view of the cost effectiveness and user friendliness, the use of a standardized observational gait analysis, such as the CAP item walking, should be the first choice in clinical practice. 3DGA may be useful for difficult cases and research.

Is FL a worthwhile variable to assess during follow-up of clubfoot?

In Study 4, the reliability of a new follow-up instrument that measures FL, FR, and FTR was introduced and assessed in children with clubfoot. The findings indicate that the FDM is applicable in both clinical practice and research.

Using FDM, the development of foot growth was followed in 72 children. The risk for relapse was non statistically significant higher in children with clubfoot 2 SD smaller than normal feet. It is possible that the small number of participants in each foot size group influenced the results. However, in children with unilateral clubfoot, those with a larger difference in FL compared with normal feet at the age of 2.5 years had statistically significant more relapses and poorer motion quality at the age of 7 years. These findings suggest that the initial foot size may be a risk factor for relapse.

Differences in FL and growth were also observed between children with and without relapse at the age of 3–5 years, when most of the relapses occurred. These findings are consistent with earlier clinical observations that clubfoot growth occasionally slows during relapse and normalizes after an appropriate intervention. Even though the differences were not statistically significant, this observation needs further investigation.

Statistical considerations

Bilateral clubfeet are related because feet from the same person are related, and this relationship must be considered in the statistical analysis [167]. Study 1 addressed the problem of bilaterality by using mixed models [154]. However, for Study 2, the mixed-models approach was difficult to apply in the correlation analysis, and to avoid bias, only 1 foot was included for each bilateral case. The same approach was used for Study 3. For Study 4, a sensitivity analysis was used before including all feet. Thus, the statistical tests were performed twice, first by including both feet and then using only 1 foot. Because the results were similar, the results for both feet have been presented here.

Another statistical issue was the multiple comparisons performed for Study 3 [156]. When multiple statistical tests are performed, the chance of statistically significant results increases for some tests, and the null hypothesis may be rejected incorrectly (i.e., type 1 error). This problem was handled by applying the Bonferroni correction. However, this correction may have led to a type 2 error.

Finally, systematic differences and Bland–Altman plots were used to evaluate the FDM, as a newly developed method. This approach was chosen instead of the

intraclass correlation coefficient because the presence of systematic differences and the visual interpretation of the differences were more interesting to investigate.

Methodological considerations

The children in the first 3 studies were recruited consecutively, and all were examined and followed up by the same senior physiotherapist in close collaboration with the responsible orthopedic surgeon. The fact that the same physiotherapist was responsible for the assessments increases the reliability and strengthens the study. On the other hand, the good results for relapse rate may have reflected the capacity of the physiotherapist to detect relapse and not the efficacy of the instruments used.

Experienced raters also participated in Study 4, which could have affected the reliability of the method [126, 128]. The reliability of inexperienced raters has not yet been determined.

A small number of participants were included in Studies 1 and 2 because of the population of the catchment area. However, the age range and their SD were narrow compared with other outcome studies, which yielded low variability in the sample. Less variability increases the power of small-sample studies [168]. The reference group was not age matched with the study group, but studies have shown that at around the age of 7 years, age no longer has an important impact on gait parameters except for temporospatial parameters [169, 170].

Study 1 compared children with clubfoot with a reference group of children without gait pathology. A randomized study that included the FAO would have been ideal. In general, a better research design would have included children treated with FAO during the maintenance phase in the first 3 studies. However, the small population of the catchment area and the prevalence of clubfoot pathology makes such a study design difficult to apply.

The contralateral feet were used as reference feet in Study 3. In gait studies, the contralateral unaffected foot cannot be considered normal in children with unilateral clubfoot [140, 171]. Nevertheless, in Study 3, the size of the unaffected feet was used as reference and did not differ from that reported in the literature of typically developing feet [172].

In Study 4, a clinical approach was used to evaluate reliability. Both feet were included in the analysis, even the unaffected foot in children with unilateral clubfoot. In addition, the procedures of drawing around the foot and measuring FL, FR, and FTR were not separated when assessing the reliability. These approaches were chosen because, in the clinical setting, both feet are drawn and the measurements are performed directly.

General conclusions

- Custom-made dynamic orthoses (KAFO/AFO) can be useful and effective alternatives for clubfoot treatment.
- Overall 3D-gait deviation scores reflect clinically observed gait deviations in children with clubfoot.
- The FDM is reliable for monitoring foot growth and foot rotation during clubfoot follow-up.
- Children with small foot size at the age of 2.5 years are prone to a higher risk of relapse and poor motion quality.

Future perspectives

After the introduction of the Ponseti casting and the good results achieved from this method, the focus of clubfoot treatment has been preservation of the initial correction. The main trends in clubfoot research are developing follow-up methods sensitive to early detect relapse, identifying factors predisposing to relapse, and studying the possible role of genetics [38-40, 42, 47, 49, 84, 87-90, 100, 101, 107, 108, 144].

Follow-up recommendations

The optimal clubfoot follow-up should include monitoring the factors associated with relapse. The family's cooperation is essential for the success of the treatment. Patient-centered care, proper information, and easily accessible support improve the results of the treatment.

Orthosis compliance should be assessed regularly through open and trustful communication and without disapproval in cases of irregular compliance. The option of alternative orthoses that would not compromise the results should be given to children who are noncompliant with the FAO. The results from the 3 first studies of this thesis imply that the dynamic KAFO/AFO is an adequate choice.

The initial severity, evertor muscle activity, and foot size at 2.5 years of age should be thoroughly assessed and registered because these have been shown to be associated with relapse [89, 173]. Children presenting with ≥1 of these factors should be monitored carefully, and prolonged orthotic treatment should be considered.

Use of the CAP is recommended during clubfoot follow-up because this instrument is standardized, sensitive over time, and covers the child's overall physical function. The FDM is a valuable complement to the CAP and can be used to monitor FL, FR, and FTR. Using the FDM during clubfoot follow-up may contribute to both the early detection of relapse and identification of predisposing factors.

Future research

No randomized studies investigated the effects of different orthosis models before the Ponseti method was introduced. Most studies have evaluated the initial treatment [70, 72-75] or both the correction and maintenance phases together but not separately [7, 67, 138, 158, 174-176]. More studies that evaluate the orthotic treatment are needed [93-95, 97, 176-178]. The presence of compliance issues with the orthosis and the high risk of relapse in cases of noncompliance indicate the need for further evaluation of the maintenance phase.

Study 3 found that irregularity in foot growth can occur before and during relapse, and that small clubfeet have a greater tendency to relapse. Future research is needed to establish a procedure for identifying small clubfeet at risk of relapse and whether measuring the FL regularly can predict relapse.

Recently, an international group identified 31 key outcomes that may be valuable to report in research and clinical practice [179]. These outcomes include information about static and dynamic measures, the definition of relapse, and patient-reported outcome measures. However, more studies are needed to establish the usefulness of these key outcomes.

Another new trend in clubfoot research is treatment of the pathological tissues responsible for the clubfoot deformity [180]. The development of pharmacological treatment for clubfoot that reduces tissue stiffness and contraction may improve the outcomes. Few studies have focused on the treatment of fibrotic tissue in clubfoot [181, 182], although research on antifibrotic therapies for other diseases has been reported [183, 184]. Recent studies have shown promising results in vitro, and further studies are needed to explore this new field of clubfoot research [185, 186].

Summary in Swedish

Pes Equino Varus Adduktus (PEVA), också kallat klumpfot, är en medfödd fotdeformitet som drabbar drygt 1 av 1000 födda barn per år. Båda fötterna är påverkade i hälften av fallen och pojkar är oftare drabbade. Obehandlat ger PEVA en allvarlig felställning där barnet går på utsidan av foten. Målet med behandlingen är att barnet kan gå och springa på fötter som är väl korrigerade och smärtfria. Trots behandling kan uppemot hälften av barnen drabbas av återfall eller ha avvikande gångmönster, samt, vid ensidig klumpfot, olika stora fötter och vader samt benlängdsskillnad.

Behandlingen av PEVA är primärt icke kirurgisk. Den består av en korrigerings- och en underhållsfas. Ponsetimetoden är den internationellt rådande behandlingen. Denna innebär att fötterna under korrigeringsfasen gipsas enligt en specialteknik varje vecka i sex veckor, efterföljt av ett kirurgiskt ingrepp, där hälsenan förlängs och foten gipsas ytterligare 3 veckor. Efter korrigeringsfasen inleds underhållsfasen där barnen använder en skena, Fot Abduktion Ortos (FAO). För att undvika återfall bör denna skena användas fram tills minst 4–5 års ålder.

I Lund före år 2000 behandlades barn med PEVA enligt Köpenhamnsmetoden. Under korrigeringsfasen behandlades fötterna med en speciell manipulerings- och töjningsteknik dagligen under ca 2 månader. Däremellan användes en plexidurskena för att bibehålla fotens korrigerade läge. Ofta behövdes slutligen ett kirurgiskt ingrepp. Därefter började underhållsfasen där barnen upp till ca 2 års ålder användes en dynamisk Knä-Ankel-Fot-Ortos (KAFO) och från 2–4 års ålder användes en dynamisk Ankel-Fot-Ortos (AFO).

Under perioden 2001–2010 infördes korrigeringsfasen enligt Ponseti, medan underhållsfasen med KAFO/AFO förblev samma. Från 2011 infördes Ponsetiortosen, FAO, som standard. Detta stegvisa införande av en ny metod gjordes för att kunna utvärdera långtidseffekten, dels av gipsbehandlingen, dels av skenbehandlingen. Även om barn med PEVA behandlats med olika metoder så har uppföljningen har skett på samma strukturerade och standardiserade sätt över åren.

Syftet med denna avhandling var att öka kunskapen om PEVA med fokus på utvärdering av behandling- och uppföljningsmetoder, och att identifiera faktorer som påverkar det långsiktiga behandlingsresultatet.

I Studie 1 undersöktes 20 sjuåriga barn med PEVA med tredimensionell gånganalys (3DGA). Barnen korrigerades enligt Ponseti men använde sedan dynamiska KAFO/AFO skenor. Syftet med studien var att utvärdera följsamhet av användning av skena, återfallsfrekvens och gångmönstret hos barn behandlade med

KAFO/AFO. Inget återfall observerades under behandlingen med skena men 20 % av barnen utvecklade senare återfall som krävde operation. 3DGA visade liknande gångavvikelser som vid andra studier som har utvärderat barn behandlade med FAO.

I Studie 2 inkluderades samma 20 barn och 3DGA data från Studie 1. Barnets kliniska status undersöktes enlig ett specifikt instrument som kallas Clubfoot Assessment Protocol (CAP). Syftet var att utvärdera användbarhet av övergripande gångindex som Gait Profile Score (GPS) och Gait Variable Score (GVS) hos barn med PEVA. Resultatet visade att GPS/GVS återspeglade de kliniska observerade gång-avvikelserna (CAP).

I Studie 3 inkluderades 72 barn med PEVA. Fotlängd (FL) mättes var sjätte månad från 2 till 7 års ålder. Syftet var att analysera fotlängdsutvecklingen hos barn med PEVA och utvärdera relationen mellan initiala behandlingen, FL vid 2 år, rörelsekvalitet och återfall. Fötter med PEVA var mindre än fötter utan PEVA i alla åldrar men växte i samma takt. Liten fotstorlek vid 2,5 årsålder var associerat med fler återfall och sämre rörelsekvalitet upp till 7 års ålder.

I Studie 4, inkluderades 19 barn med PEVA. Syftet var att utvärdera intra- och interreliabiliteten vid mätning av FL och fotrotationer med en ny metod som kallas "The Foot Drawing Method" (FDM.) Fötterna mättes 2 gånger av 2 oberoende bedömare. Det framkom inga systematiska skillnader inom eller mellan bedömarna. Skillnaderna var mindre uttalade i vid fötter som hade mindre utåtrotation

Dynamiska KAFO/AFO är användbara alternativ hos barn med PEVA under underhållsfasen. GPS/GVS reflekterar de kliniskt observerade gångavvikelserna hos barn med PEVA. FDM är en reliabel metod för att följa fotlängd och fotrotationer hos barn med PEVA, men metoden är mer säker vid stela fötter. Barn med liten fotstorlek vid 2,5 års ålder har en högre risk för återfall och sämre rörelsekvalitet.

Summary in Greek

Η ραιβοϊπποποδία (Pes EquinoVarus Adductus, PEVA), είναι μία εκ γενετής παραμόρφωση του άκρου ποδιού, που πλήττει 1 στα 1000 νεογέννητα παιδιά το χρόνο στη Σουηδία. Χαρακτηρίζεται από στροφή του πέλματος προς τα μέσα. Η πάθηση εμφανίζεται συχνότερα στα αγόρια και είναι αμφοτερόπλευρη στις μισές περιπτώσεις. Χωρίς θεραπεία, τα παιδιά με ραιβοϊπποποδία περπατάνε με την εξωτερική και τη ραχιαία επιφάνεια του ποδιού, αλλά με σωστή αντιμετώπιση έχουν φυσιολογική κινητικότητα. Παρόλη τη θεραπεία, τα μισά περίπου παιδιά υποτροπιάζουν ή παρουσιάζουν ανωμαλίες στο περπάτημα καθώς και ανισότητα στο μέγεθος του πέλματος και της γάμπας, κυρίως στην μονόπλευρη ραιβοϊπποποδία.

Η θεραπεία είναι κυρίως συντηρητική με καλό τελικό αποτέλεσμα και αποτελείται από 2 φάσεις, τη διόρθωση και την διατήρηση. Η μέθοδος που χρησιμοποιείται περισσότερο διεθνώς λέγεται Ponseti, η οποία συνδυάζει την εφαρμογή μηρο-κνημο-ποδικών γύψων και διαδερμικής τενοντομής του Αχιλλείου τένοντα κατά την φάση της διόρθωσης, ενώ κατά την φάση της διατήρησης μπαίνει το πόδι σε νυχτερινούς νάρθηκες-υποδήματα (Fot Abduktion Ortos (FAO). Για να αποφευχθούν οι υποτροπές και να μειωθεί η συχνότητα των χειρουργικών επεμβάσεων συνιστάται η χρήση νάρθηκα μέχρι να γίνει το παιδί 4–5 χρονών τουλάχιστον.

Έλλειψη συμμόρφωσης των γονέων στη σωστή εφαρμογή του νάρθηκα (FAO) μετά το πέρας της θεραπείας με διορθωτικούς γύψους είναι ο κύριος λόγος αποτυχίας της μεθόδου Ponseti.

Ο στόχος της παρούσας διδακτορικής διατριβής είναι να εξετάσει την πάθηση της ραιβοϊπποποδίας με έμφαση στην αξιολόγηση και την παρακολούθηση των παιδιών μετά τη θεραπεία. Επίσης, στοχεύει στην επισήμανση παραγόντων που επηρεάζουν το αποτέλεσμα της θεραπείας σε βάθος χρόνου.

Στην πρώτη μελέτη, 20 παιδιά με ραιβοϊπποποδία εξετάστηκαν με τρισδιάστατη ανάλυση βάδισης (3DGA). Η θεραπευτική αντιμετώπιση των παιδιών έγινε σύμφωνα με τη μέθοδο του Ponseti κατά τη διάρκεια της διόρθωσης και με εναλλακτικούς νάρθηκες κατά την φάση της διατήρησης. Μέχρι να συμπληρώσουν τα 2 χρόνια χρησιμοποιήσαν τα παιδιά μηροκνημοποδικό νάρθηκα (Knee-Ankle-Foot Orthosis, KAFO) και μεταξύ 2 και 4χρονών, κνημοποδικό νάρθηκα (AFO). Ο στόχος της μελέτης ήταν να αξιολογήσει τη συμμόρφωση με τη χρήση του νάρθηκα, τη συχνότητα των υποτροπών και τον τρόπου βαδίσματος. Μετά τη διακοπή της χρήσης του νάρθηκα, το 20% των παιδιών υποτροπίασαν και χρειάστηκαν

εγχείρηση. Η συμμόρφωση με τη χρήση του νάρθηκα ήταν υψηλή. Η 3DGA έδειξε παρόμοιες αποκλίσεις βαδίσματος όπως και σε παρόμοιες μελέτες σε παιδιά που αντιμετωπίστηκαν με FAO.

Στη δεύτερη μελέτη συμμετείχαν τα 20 παιδιά και τα 3DGA αποτελέσματα από την πρώτη μελέτη. Η κλινική εικόνα εξετάστηκε σύμφωνα με το πρωτόκολλο αξιολόγησης της ραιβοϊπποποδίας. Ο στόχος ήταν να εξεταστεί ο συνολικός δείκτης βαδίσματος (Gait Profile Score (GPS)) και ο δείκτης μεταβλητών βαδίσματος Gait Variable Score (GVS)) ως προς την ακρίβεια αξιολόγησης της κλινικής εικόνας. Τα αποτελέσματα έδειξαν ότι οι GPS/GVS ήταν ενδεικτικοί των αποκλίσεων στο περπάτημα στα παιδιά με ραιβοϊπποποδία.

Στην τρίτη μελέτη συμπεριλήφθηκαν 72 παιδιά με ραιβοϊπποποδία. Το μήκος του πέλματός τους μετρήθηκε ανά έξι μήνες από τότε που ήταν 2 μέχρι που έγιναν 7 χρονών. Ο στόχος ήταν να μελετηθεί η ανάπτυξη του πέλματος και να αξιολογηθεί η σχέση μεταξύ του αρχικού του μήκους, της ποιότητας της κινητικότητας και των υποτροπών. Τα πέλματα με ραιβοϊπποποδία ήταν μικρότερα από τα φυσιολογικά αλλά αναπτύσσονταν με παρόμοιο ρυθμό. Αν όμως το πέλμα ήταν μικρότερο του κανονικού στα δυόμιση χρόνια, ο κίνδυνος για υποτροπές και χειρότερη κινητικότητα πριν τα επτά χρόνια ήταν αυξημένος.

Στην τέταρτη μελέτη συμπεριλήφθηκαν 19 παιδιά με ραιβοϊπποποδία. Ο στόχος ήταν να αξιολογηθεί η αξιοπιστία των μετρήσεων του μήκους και της περιστροφής του πέλματος με μια νέα μέθοδο που ονομάστηκε “σχεδιασμός πέλματος” (Foot drawing method” (FDM)). Τα πέλματα μετρήθηκαν δυο φορές από δυο ανεξάρτητους παρατηρητές. Δεν υπήρχαν στατιστικά ή κλινικά σημαντικές συστηματικές διαφορές μεταξύ των μετρήσεων γεγονός που ενισχύει την άποψη ότι η νέα μέθοδος είναι αξιόπιστη.

Οι νάρθηκες KAFO/AFO είναι μια χρήσιμη εναλλακτική κατά τη διάρκεια της φάσης της διατήρησης για την αντιμετώπιση παιδιών με ραιβοϊπποποδία. Οι συνολικοί δείκτες βαδίσματος GPS/GVS είναι ενδεικτικοί των αποκλίσεων στο περπάτημα και το FDM είναι μια αξιόπιστη μέθοδος για τη μέτρηση του μήκους και της περιστροφής πέλματος. Η πρόγνωση των παιδιών με ραιβοϊπποποδία που έχουν μικρά πέλματα στην ηλικία των δυόμιση χρονών είναι χειρότερη όσον αφορά την κινητικότητα και τον κίνδυνο υποτροπών.

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