Dynamic ultrasound investigation of clubfeet in children, 0-4 years of age, with normal controls

Arne Johansson

Department of Orthopaedics

Institute of Clinical Sciences

Sahlgrenska Academy, University of Gothenburg

UNIVERSITY OF GOTHENBURG

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arne.g.johansson.@vgregion.se; arne.0500451435@telia.com

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“A picture is worth a thousand words”

Ancient Chinese proverb

To the children participating in the study and their parents

To my family

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Department of Orthopaedics, Institute of Clinical Sciences
Sahlgrenska Academy, University of Gothenburg
Gothenburg, Sweden

ABSTRACT

Clubfoot is one of the most common congenital deformities, 1-6/1,000 births in Europe and up to 5-6/1,000 in Polynesia. The Ponseti method is now widely regarded as the method of choice for treatment. The treatment starts soon after birth using manipulations and castings until the deformities are corrected, usually 6-10 weeks. To prevent a recurrence, the treatment continues using orthoses to the age of four years. During this period of life, large parts of the foot skeleton are cartilaginous and not visible on radiographs. Using ultrasonography (US), the non-ossified parts of the skeleton can be visualised. This thesis consists or four studies with the overall aim of developing reliable measurement variables for the evaluation of ultrasonographic images of feet during the first four years of life.

Study I was a longitudinal cohort study running over one year. One hundred and eight normal feet in 54 healthy children were examined soon after birth, at four or seven months and at 12 months of age. The aim was to establish reproducible standardised projection planes and reliable measurement variables for assessing the ankle joint, the talo-navicular joint and the calcaneo-cuboid joint. Three scanning planes were used, medial and lateral coronal and dorsal sagittal. Pearson's correlation coefficient for the measurements was r=0.65-0.94 ($p \leq 0.01$) for intra-observer and r=0.53-0.93 ($p \leq 0.01$) for inter-observer. The non-ossified parts of the skeleton were depicted as black with white dots, while the joint cartilage appeared black.

Studies II and III were cross-sectional cohort studies. The control group comprised 105 healthy children and the clubfoot group comprised 46 children with 71 clubfeet. The age of the children was newborn to four years. They were divided into 10 age groups (newborn, 3, 6, 12, 18, 24, 30, 36, 42 and 48 months) and were investigated once.

In **Study II,** the aim was to establish reliable variables, independent of the age-related size or the ossified nuclei, for the assessment of the ankle joint using a posterior sagittal projection. The inter-investigator agreement for the scans was 0.71 to 0.89, Intra-class Correlation Coefficient (ICC). The intra-observer agreement (ICC) was ≥ 0.9 for controls and ≥ 0.8 for clubfeet. The inter-observer agreement for controls was ≥ 0.68 and ≥ 0.84 for clubfeet for all variables.

In **Study III,** the aim was to improve the evaluation of the deformities and mobility in the talo-navicular and calcaneo-cuboid joints by adding new measurement variables; a total of 20 variables were measured. The intra-observer agreement (ICC) ranged from 0.71 to 0.99 for controls and 0.58 to 0.99 for clubfeet. The inter-observer agreement (ICC) ranged from 0.58 to 0.99 for controls and 0.45 to 0.96 for clubfeet. The correlations were higher on the medial side than on the lateral side. The mean ROM in the talo-navicular joint was 59° in the controls and 41° in the clubfeet. The corresponding values for the calcaneo-cuboid joint were 17° and 8.5°.

Study IV was a longitudinal, cohort, observational study from birth to the age of four years. Twenty children with 30 clubfeet and 29 controls were included. The four scanning planes described in Studies I-III were used. The children's feet were investigated by US at the same ages as in Studies II and III and the images were evaluated using the same variables. Clinical data were retrieved from the medical records to assess the correlation between US findings and clinical data and the course of treatment. The medial malleolus-navicular (MM-N) distance and the talo-navicular (T-N) angle showed the highest correlation (r = -0.7 resp. +0.7) with the number of casts needed to correct the deformities. Even after the initial correction phase, some differences between clubfeet and controls in the US findings remained to the age of four years.

Overall conclusion: Ultrasound investigations of normal feet and clubfeet can be conducted with good reliability from birth to the age of four years. US can be a valuable complement to the clinical evaluation of clubfeet.

Keywords: clubfoot, ultrasonography, Ponseti treatment, congenital deformities, range of movement, repeatability, longitudinal study

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SAMMANFATTNING PÅ SVENSKA

Klumpfot är en av de vanligaste medfödda missbildningarna, 1-6/1000 födda i Europa och upp till 5-6/1000 i Polynesien. Ponseti-metoden är nu allmänt accepterad som den bästa metoden för behandling. Behandlingen påbörjas i nyföddhetsperioden med redressioner och gipsningar tills felställningarna är korrigerade, vanligen 6-10 veckor. För att förhindra recidiv fortsätter behandlingen med ortoser till fyra års ålder. Under denna period i livet är stora delar av fotskelettet brosk och syns inte på konventionell röntgen. Med hjälp av ultraljud kan de icke ossifierade delarna av skelettet avbildas. Den här avhandlingen består av fyra studier med det övergripande syftet att utveckla tillförlitliga mätvariabler för utvärdering av ultraljudsbilder av fötter under de första fyra levnadsåren.

Studie I var en longitudinell kohortstudie som löpte över ett år. Etthundraåtta normala fötter hos 54 friska barn undersöktes kort efter födseln, vid fyra eller sju månaders ålder och vid 12 månaders ålder. Syftet var att utveckla reproducerbara standardiserade bildplan och tillförlitliga mätvariabler för att bedöma fotleden, talo-navikularleden och calcaneo-cuboideumleden. Tre bildplan användes, medialt och lateralt coronalt och dorsalt sagittalt. Pearsons korrelationskoefficient för mätningarna var $r=0{,}65–0{,}94$ ($p \leq 0{,}01$) för intra-observer och $r=0.53-0{,}93$ ($p \leq 0{,}01$) för interobserver korrelation. De icke-ossifierade delarna av skelettet avbildades svart med vita prickar, medan ledbrosket återgavs svart.

Studierna II och III var tvärsnitts-kohortstudier. Kontrollgruppen omfattade 105 friska barn och klumpfotsgruppen omfattade 46 barn med 71 klumpfötter. Barnens ålder var från nyfödda till fyra år. De delades in i 10 åldersgrupper (nyfödda, 3, 6, 12, 18, 24, 30, 43 och 48 månader) och undersöktes en gång.

I **Studie II** var syftet att utveckla tillförlitliga variabler, oberoende av den åldersrelaterade storleken av benkärnorna, för undersökning av fotleden med en bakre sagittal projektion. Överensstämmelsen för skanningarna mellan två undersökare var 0,71 till 0,89 Intra-class Correlation Coefficient (ICC). För mätningarna var intraobserver-korrelationen (ICC) $\geq 0{,}9$ för kontrollerna och $\geq 0{,}8$ för klumpfötterna. Interobserver-korrelationen för kontrollerna var $\geq 0{,}68$ och $\geq 0{,}84$ för klumpfötterna för alla variablerna.

I **Studie III** var avsikten att förbättra bedömningen av deformiteter och rörlighet i talo-navikular- och calcaneo-cuboideum-lederna genom att lägga till nya mätvariabler; totalt 20 variabler mättes. Intraobserver-korrelationen (ICC) var 0,71–0,99 för kontrollerna och 0,58–0,99 för klumpfötterna. Interobserver-överensstämmelsen (ICC) var 0,58–0,99 för kontrollerna och 0,45–0,96 för klumpfötterna. Korrelationen var högre på medialsidan än på lateralsidan. Medelvärdet för ROM i talo-navikularleden var 59° för kontrollerna och 41° för klumpfötterna. Motsvarande värden för calcaneo-cuboideumleden var 17 och 8,5°.

Studie IV var en longitudinell kohort-observationsstudie från födelsen till fyra års ålder. Tjugo barn med 30 klumpfötter och 29 kontroller inkluderades. De bildplan som beskrivits i studie I-III användes. Barnens fötter undersöktes med ultraljud vid samma åldrar som i studie II och III och bilderna bedömdes med samma variabler. Kliniska data hämtades från patientjournalerna för att utvärdera korrelationen mellan ultraljudsfynden och kliniska data och behandlingsförloppet. Avståndet från mediala malleolen till navikulare (MM-N) och den talo-navikulära (T-N) vinkeln visade högst korrelation (r = -0,7 resp. +0,7) med antalet gipsningar som behövdes för att korrigera felställningarna. Även efter den initiala korrektionsfasen kvarstod en del skillnader i ultraljudsfynden mellan kontroller och klumpfötter till fyra års ålder.

Övergripande slutsats: Ultraljudsundersökning av normala fötter och klumpfötter kan utföras med god repeterbarhet och interobserver-korrelation från födseln till fyra års ålder. Ultraljud kan vara ett värdefullt komplement till klinisk bedömning av klumpfötter.

LIST OF PAPERS

This thesis is based on the following studies, referred to in the text by their Roman numerals.

I. Aurell, Y., A. Johansson, G. Hansson, H. Wallander and K. Jonsson, *Ultrasound anatomy in the normal neonatal and infant foot: an anatomic introduction to ultrasound assessment of foot deformities.* Eur Radiol, 2002. **12**(9): p. 2306-2312.

II. Johansson, A., Y. Aurell, and B. Romanus, *Assessment of the ankle joint in clubfeet and normal feet to the age of four years by ultrasonography*. J Child Orthop, 2018. **12**(3): p. 262-272.

III. Johansson, A., Y. Aurell, and B. Romanus, *Range of motion in the talo-navicular and the calcaneo-cuboid joints evaluated by ultrasound during clubfoot treatment with normal references up to the age of four years.* J Child Orthop, 2018. **12**(5): p. 526-538.

IV. Johansson, A., Y. Aurell, and B. Romanus, *A prospective longitudinal study, with dynamic ultrasound and clinical examinations, during the first 4 years of life of children with clubfoot treated according to Ponseti, and of feet in a control group. The medical reports of all the children with clubfoot were reviewed up to the age of 8 years.* Submitted

CONTENTS

ABBREVIATIONS

C-C	Calcaneo-cuboid (distance/angle)
CR	Conventional radiology
ICC	Intra-class correlation coefficient
MM-N	Medial malleolus–navicular distance
MM-T-N	Medial malleolus-talar head-navicular distance
MRI	Magnetic resonance imaging
ROM	Range of motion
Skin-talus	Skin-talus distance
Skin-tib. E	Skin-tibial epiphysis distance
Tib.phys.- C	Tibial physis-calcaneus distance
Tib.phys.-TCJ	Tibial physis-talo-calcaneal joint distance
STT	Soft tissue thickness
T-N angle	Talo-navicular angle
T-Tang-N	The perpendicular distance from the medial talar tangent to the medial border of the navicular
OR	Odds ratio
US	Ultrasonography

DEFINITIONS

Clubfoot	A congenital foot deformity; the foot is in the equinus, varus and adducted position. Synonyms: talipes equinovarus and pes equino-varus adductus (PEVA)
Atypical clubfoot	At birth, these feet have more severe deformities, especially the cavus, resulting in a deep plantar crease and an extension of the big toe. They are short and stubby with severe equinus deformity and require a modified casting technique.
Complex clubfeet	These feet look like "typical clubfeet" at birth, but they develop into the atypical shape due to slipping casts. They should be treated as atypical clubfeet.
Positional clubfoot	Synonym: postural clubfoot. These feet are supple, have mild deformities and can easily be put into the normal position. They usually correct without treatment.

1 INTRODUCTION

1.1 PREVALENCE

Clubfoot is one of the most common congenital deformities, with a prevalence of 1-2/1,000 live births in Europe and North America and up to 5-6/1,000 in Polynesia [1-3]. On some islands in Polynesia, where marriages between cousins are frequent, the prevalence is higher. If one child in the family has a clubfoot, the likelihood that future children will also have clubfeet is 1/35, indicating some genetic predisposition.

1.2 AETIOLOGY

The aetiology is considered multifactorial. The most common risk factors are family history (OR = 7.8), medication with selective serotonin reuptake inhibitors (SSRI) during pregnancy (OR = 4.26-1.64), early amniocentesis (11th-12th gestational weeks) [4], maternal/paternal smoking, maternal obesity and gestational diabetes. Several factors are associated with a moderate increase in the odds of clubfoot [5].

1.3 BIOLOGY AND ANATOMY

Clubfoot is a developmental deformity occurring during the second trimester of pregnancy when a foot that hitherto is developing normally turns into a clubfoot. Clubfoot is rarely detected by prenatal ultrasound before the 16th week of gestation. In clubfeet, the tibialis posterior, the gastrosoleus, the tibialis anterior and the long toe flexors are smaller and shorter than in normal feet, pulling the foot into equinus and the calcaneus and navicular into adduction and supination. The ligaments of the posterior and medial aspect of the ankle and tarsal joints are thick and tight, keeping the foot in equinus and the navicular and calcaneus in adduction and inversion [6]. Collagen synthesis is excessive in ligaments, tendons and muscles [7, 8]. The navicular is medially dislocated in relation to the talar head, the calcaneus is in the varus position and the fore foot is pronated in relation to the hind foot, resulting in cavus deformity. As a result of the altered position of the tarsal bones, the shape and the position of the ossified nucleus are altered. This will be aggravated during growth, if untreated [6, 9, 10].

1.4 DIAGNOSTICS

In congenital clubfoot, the deformities are obvious, and the diagnosis is made at a clinical examination shortly after birth. This should not be confused with positional clubfoot, where the foot is supple and can easily be corrected. In Sweden today, most clubfeet are detected by a prenatal ultrasound.

1.5 CLASSIFICATION

There is a large variation in the severity of clubfoot deformity. In order to choose the type of treatment and to enable comparisons between different treatment protocols, several classification systems have been proposed. Well-known classification protocols are: Catterall [11], Harrold and Walker [12], Diméglio [13] and Pirani [14].

1.5.1 DIMÉGLIO CLASSIFICATION

In the Diméglio classification [13], four essential parameters are each scored 1-4: equinus in the sagittal plane, varus deviation in the frontal plane, rotation of the calcaneo-forefoot block around the talus and adduction of the forefoot in relation to the hind-foot. If present, 1 point is added for each of the following elements: marked posterior crease, marked midtarsal crease, cavus and muscular imbalance. The maximum score is therefore 20 points. The feet are then classified into 4 grades: Grade I = benign (soft-soft) feet 0 – < 5 points, Grade II = moderate (soft-stiff) feet 6 – < 10 points, Grade III = severe (stiff-soft) feet 10 – < 15 points and Grade IV = very severe (stiff-stiff) feet 15-20 points. The Diméglio score was used in many clinics in Sweden from its publication in 1995 to 2015.

In this study, the Diméglio classification was used, as it was the routine at the participating departments when the study was performed

1.5.2 PIRANI CLASSIFICATION

In the Pirani classification [14], six clinical signs are scored: curved lateral border, medial crease, talar head coverage, posterior crease, rigid equinus and empty heel. Each sign is scored: 0 = normal, 0.5 = moderately abnormal or 1 = severely abnormal. The maximum total score is therefore 6. The Pirani score is used in the Swedish Paediatric Orthopaedic Quality (SPOQ) register. Since the register started in 2015, the Pirani score has become the most used scoring protocol in Sweden.

1.6 TREATMENT: A HISTORICAL REVIEW

The deformity is obvious at birth and has been known for thousands of years. It has been documented in literature, art and archaeological findings. Many treatment methods have been used through the millennia [15].

1.6.1 ANCIENT TREATMENT

Clubfoot was documented by ancient civilisations from different continents. It has been found on 5,000-year-old wall paintings in Egyptian tombs and it is mentioned in the Indian prayer book Yajur-Veda from the 10th century B.C. The treatment was described in India in 1,000 B.C. [15, 16]. The oldest known written description of clubfoot and its treatment was given by Hippocrates (approx. 460-377 B.C.), who believed that the deformity was a medial luxation in the ankle joint caused by mechanical pressure. Knowledge of the foot anatomy was incomplete at that time; for example, the talus and the calcaneus were regarded as one bone. Despite this, Hippocrates reported that the majority of the cases could be treated successfully. He started the treatment soon after birth with gentle serial manipulations and, between the manipulations, the improved position of the foot was maintained by bandages. When correction was achieved, special shoes were worn to maintain the correction and prevent relapses [17]. Hippocrates' methods are remarkably similar to current non-surgical treatment. From the Middle Ages, very little information is available.

In 1658, Arcaeus described his stretching technique and two mechanical devices for maintaining the correction, one of them similar to Scarpa's shoe. In the mid-18th century, Cheselden treated clubfeet by repeated stretching and tape to maintain the correction. Scarpa published *Memoar on Congenital Club-foot of Children* in 1803 [18]. He considered the talus to be normal in shape and position and that the deformity was due to an inward dislocation of the forefoot upon the head of the talus. He used forceful manipulation using a mechanical device later known as Scarpa's shoe. The method was not successful in other hands and not widely accepted.

In 1806, Timothy Sheldrake published his essay *Distortion of the Legs and Feet of Children* [19]. Like Hippocrates, he used bandages and he claimed that, if the treatment started before the age of two months, most of the feet could be corrected in two to three months but should not be left free until the child was able to walk. If the child was older than two months at the start of treatment, the correction took longer.

1.6.2 SURGICAL TREATMENT

In 1823, Delpech performed percutaneous tenotomy of the Achilles tendon in two patients with acquired clubfeet and both developed sepsis. The high incidence of postoperative infection was a problem. However, in 1831, Stromeyer percutaneously divided the Achilles tendon in several patients without infection. His opinion was that the deformity was caused by insufficiency in the development of the medial malleolus. Little learned how to perform the tenotomy from Stromeyer and introduced it in England. He believed that the deformity was due to abnormal muscles during intra-uterine development and that the foot deformity was associated with outward rotation of the thigh. In 1834, Rogers and, in 1835, Dickson started to use tenotomy of the Achilles tendon in the USA.

Adams performed dissections on stillborn infants with clubfeet and published the results in 1866. He made microscopic examinations of the muscles but found no structural changes. He examined the bones and found that the only one that exhibited any marked change was the talus, which tilted medially. Adams believed that the changes in the talus were due to the altered position of the talus and calcaneus and that, anatomically, clubfoot is a dislocation of the talo-calcaneo-navicular joint (peritalar subluxation) and the muscles are the deforming forces. He emphasised that the talus can only adopt its normal shape and position after the reduction of the dislocation of the calcaneus and the navicular. To achieve anatomic reduction, he therefore recommended early surgery. Ryöppy performed surgical corrections in the neonatal period in 1983 [20]. This was later abandoned. The feet are very small, the technique is demanding, and the risk of complications is very high.

More radical operations of clubfoot could be performed following two medical developments: the introduction of general anaesthesia by the dentist William Morton in 1846 and the introduction of antiseptic principles of surgery by Lister in 1867 [21].

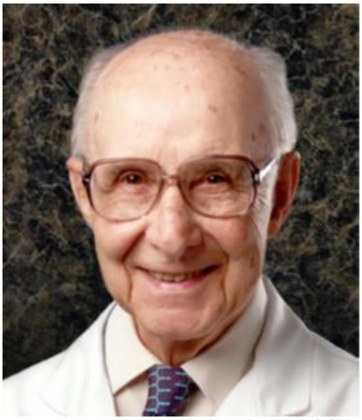
In 1891, Phelps performed a tenotomy of the Achilles tendon combined with the medial release of soft tissues: the elongation of the tibialis posterior, division of the medial ligament of the ankle joint, the plantar fascia, the abductor hallucis longus and all the short flexors and he finally performed an osteotomy of the neck of the talus and a wedge resection of the calcaneus. Similar radical procedures were performed by other orthopaedic surgeons, Duval, Ogston and Lane. Several variants of medial releases were introduced during the late 19th and early 20th century. In 1971, Turco introduced internal fixation by Kirschner wires combined with plaster after posteromedial releases to prevent relapses [22]. The transfer of the anterior tibial tendon was described by Dunn in 1922. During the same period, many surgical procedures on the skeleton were used: talectomy, the removal of the ossified nucleus of all the tarsal bones and the wedge resection of the tarsal bones. Extensive surgery was followed by deep scarring and joint stiffness and many complications were reported [23].

1.6.3 RETURN TO NON-SURGICAL TREATMENT

In 1838, M. Guerin started to use plaster of Paris in clubfoot treatment. Sir Robert Jones gave up surgical treatment in favour of manipulation and casting [24]. Michael Hoke (1874-1944), the first medical director of the Scottish Rite Hospital in Decatur, Georgia, advocated manipulation and maintaining the correction with plaster casts. Kite succeeded Hoke as medical director and continued to practise what he had learned from Hoke. Kite corrected each component of the deformity separately. He recommended correction by abducting the foot "at the midtarsal joint" with counter-pressure at the calcaneo-cuboid joint [25]. However by applying counter-pressure at the calcaneo-cuboid joint, the abduction of the calcaneus is blocked and it took a long time to correct the heel varus [15].

In his work *Precis de Manual Operatoire,* first published in 1872, Farabeuf described how, as the foot goes into varus, the calcaneus adducts and inverts under the talus, while the cuboid and navicular adduct and invert in front of the calcaneus and the talar head respectively. He also explained that, in a clubfoot, the ossification centre in the talus responds to the abnormal pressure caused by the displaced navicular. He also observed that bony deformities in infants with clubfeet were reversible and that the recurrence rate was high due to soft-tissue contractures. In 1961, Huston published his PhD thesis entitled *A functional and anatomical study of the tarsus*. He showed that the tarsal joints do not move as a single hinge but rotate around moving axes and that motion of the joints occurs simultaneously and is interdependent. If the motion in one joint is blocked, the others are also functionally blocked. Ponseti added to this knowledge, by dissections in the anatomical department on normal feet of children and adults and on clubfeet in stillborn foetuses. In a histological study of the abundant collagen in the medial ligaments from virgin clubfeet and stillborn foetuses, he found that the abundant collagen could be easily stretched [7, 8, 26]. Based on these findings, Ponseti developed his principles of treatment and presented them in his monography in 1996 [1]. This is now widely accepted as the method of choice for clubfoot treatment in many countries [1, 27]. Ponseti's work has been continued by his successor, Jose Morquende, in Iowa City and the Ponseti International Organisation.

1.7 THE PONSETI METHOD



Ignacio V. Ponseti 1914-2009
Photo reproduced with permission from Stuard Weinstein.

The fundamental principles of the Ponseti method are as follows [1, 28, 29].

- All components of the deformity have to be corrected simultaneously, apart from the equinus, which should finally be addressed when the rest is corrected [1, 30].
- The cavus is caused by pronation of the forefoot in relation to the hind foot and is corrected by supinating the forefoot until the arch looks normal. Counter-pressure is applied at the lateral aspect of the head of the talus (which is always prominent directly under the skin), while the foot in front of the talus is abducted in supination. As a result, the navicular moves laterally in front of the talus, the calcaneus rotates laterally under the talus and the adductus and the varus deformities are corrected. The equinus improves as the calcaneus dorsiflexes when it abducts under the talus. However, in the majority of clubfeet, it is not enough when the adductus, varus and cavus are corrected and most clubfeet therefore require a percutaneous tenotomy of the Achilles tendon before finishing the cast treatment [1, 30].
- To maintain the correction, a foot-abduction orthosis (FAO) is used after plaster removal, because the deforming forces caused by the hypercollagenosis at the medial side are still there. [31] The FAO is used 23 hours/day during the first three months and then at night until the age of four years. The hypercollagenosis decreases at three to four years of age and FAO treatment can therefore usually be terminated at the age of four years [1, 30].

1.8 IMAGING

1.8.1 CONVENTIONAL RADIOLOGY (CR)

Imaging methods have been used since 1896 when Barwell introduced radiology in the investigation of clubfoot. One problem associated with the radiological imaging of feet during the first years of life (when the treatment of clubfeet takes place) is the fact that large portions of the tarsal bones have not yet ossified and cannot therefore be visualised by radiography. The navicular, whose position is crucial in clubfoot treatment, starts to ossify at three to four years of age. Several attempts have been made to standardise the measurements on conventional radiographs to obtain normal reference values [32]. It is projection sensitive, especially regarding angles, and the correct positioning of the foot can be difficult. Another shortcoming of conventional radiography is the assumption that the images of the ossification centres on the radiograph represent the true position of the whole cartilaginous anlage. Autopsy studies and MRI studies have revealed that ossification in the talus, calcaneus and navicular bones does not begin in the centre of the cartilaginous anlage. As a result, the long axes of the ossification centre and the long axes of the anlage do not coincide [6, 33-39]. Ossification begins and proceeds eccentrically within the cartilaginous anlage, which explains why only parts of the changes are seen on plain radiographs during growth [39]. Furthermore, the ossification centres in clubfeet are positioned differently and the longitudinal axes are oriented differently compared with normal feet [7, 9, 38].

1.8.2 ARTHROGRAPHY

Conventional arthrography is able to provide a detailed depiction of the talo-crural and talo-navicular joints, but it has the disadvantage of being invasive and requiring sedation [40, 41].

1.8.3 COMPUTED TOMOGRAPHY (CT)

The advantages of CT are that three-dimensional reconstructions can be achieved, visualising the deformity of the bones and dislocations in the joints [42-44]. The disadvantages are the amount of radiation, the limited visualisation of the non-ossified part of the tarsal bones, motion artefacts, the need for sedation and the costs.

1.8.4 MAGNETIC RESONANCE IMAGING (MRI)

The advantages of MRI are the capacity for imaging the ossified nuclei, as well as the cartilaginous anlage and the soft-tissue structures. Three-dimensional reconstructions illustrating the displacement and the deformities of the bones can be made [10, 35, 36, 45]. The axes of the bones and cartilaginous anlage can be calculated [36]. The disadvantages are the need for sedation and the cost.

1.8.5 ULTRASONOGRAPHY (US)

The first report on the US imaging of clubfeet was presented in 1989 by Dahlström, describing the shape of the talus in a clubfoot [46]. From studies of neonatal hip joints, it was known that, with ultrasonography, articular cartilage appears black because of its homogeneous structure, while the non-ossified cartilaginous anlage is black, with white dots representing blood vessels [47]. In 1994, Bensahel et al. presented an ultrasound protocol with five projection planes: a dorsal sagittal visualising the Achilles tendon, a medial vertical and a lateral vertical plane with the probe over the malleoli, a longitudinal dorsal projection revealing the alignment in the talo-navicular joint in the sagittal plane and a transverse plane at the level of the navicular bone [48, 49]. In a study from 1995, Tolat et al. used a longitudinal dorsal projection and a perpendicular transverse scanning plane [50]. In normal feet, longitudinal dorsal scanning visualises the relationship between the tibia, talus and navicular, but, in clubfeet, the navicular is dislocated medially out of this plane but is visualised in the transverse plane. In 1996, Chami et al. presented a US study of the feet in 50 normal children [51]. They used three posterior sagittal scans (with the foot in the neutral, plantar-flexed and dorsiflexed positions) and one anterior sagittal and, in addition, one medial projection including the medial malleolus, the talus and the navicular. They measured the distance between the medial malleolus and the medial part of the navicular, but no values were presented. The posterior and the medial scans are similar to the posterior and medial projections in this study. Important knowledge of the ultrasonography of infant feet was presented by Aurell [52-54].

2 AIM

2.1 GENERAL AIMS

To develop measurement variables with good repeatability for the assessment of ultrasound examinations of feet in children for the age range from newborn to the age of four years.

To monitor the correction of the malalignments in clubfeet throughout the treatment period by ultrasonography.

To use dynamic ultrasonography to explore the range of motion and movement patterns in the joints of the clubfoot, with the emphasis on the ankle joint and talo-navicular joint and compare this with the corresponding parameters in normal feet.

2.2 SPECIFIC AIMS

1. To establish a protocol for US assessment of the ankle joint and the talo-navicular and the calcaneo-cuboid joints during the first year of life, including reproducible standardised projections and reliable variables for measurements (Paper I).
2. To develop reliable measurement variables for the posterior scans, which are independent of the age-related size of the ossified nuclei, applicable from birth to the age of four years. To establish normal values for this age span. To compare measurements in clubfeet with these age-matched normal values (Paper II).
3. To extend the reliability evaluation from one year to the age of four years for the variables introduced in Paper I. To improve the assessment of the talo-navicular and the calcaneo-cuboid joints by adding new reliable variables (Paper III).
4. To follow the progress of treatment of individual clubfeet by ultrasound from the neonatal period to the age of four years and compare this with the development of normal feet in a control group. To correlate the ultrasound findings with clinical variables and follow the course of treatment at group and individual level. To explore the development of the feet after the end of orthosis treatment, usually at the age of four years, to the age of eight years (Paper IV).

3 SUBJECTS AND METHODS

3.1 STUDY DESIGN

Study I was a longitudinal cohort study running for one year.

Studies II and III were cross-sectional cohort studies.

Study IV was a longitudinal cohort study running for four years.

3.2 SUBJECTS

3.2.1 STUDY I

From the maternity unit at the County Hospital of Halmstad, 54 healthy newborn children (24 boys and 30 girls) were recruited. All these children were investigated in the neonatal period, 28 of them were examined a second time at the age of four months and 23 at the age of seven months. All but three of the children were examined at 12 months of age.

3.2.2 STUDEIES II AND III

One hundred and five controls (45 boys and 60 girls) aged from newborn to four years were recruited from the local child care centre, the Billingen health centre, in Skövde, and the maternity unit at Skaraborg Hospital, Skövde. Forty-six children (33 boys and 13 girls) with 71 clubfeet (25 bilateral and 21 unilateral) were recruited from the Department of Orthopaedics, Sahlgrenska University Hospital/Östra, Gothenburg. The inclusion criteria were that the children were undergoing treatment for idiopathic clubfoot in 2007 when the study started and were aged from newborn to four years. The clubfeet were all treated according to the Ponseti method and were in different stages of treatment. Since some of the variables used are age dependent, all the children were divided into ten age groups (newborn, 3, 6, 12, 18, 24, 30, 36, 42 and 48 months of age). For the control group, the recruitment continued until there were at least ten children in each group. In the clubfoot cohort, the number of children in the age groups varied and there were no newborns. The aim was to perform the ultrasound investigation within one month of the planned age. However, this was not always possible in the clubfoot group, due to illness, the parents' work and other reasons and, as a result, the age group limit was set at ± 2.6 months in the age-dependent statistical calculations (Table 1).

Table 1. Number of feet included per age group.

Age months	0	3	6	12	18	24	30	36	42	48	Total
Controls	20	26	20	20	22	22	20	20	20	20	210
Clubfeet	0	2	12	3	8	8	12	5	6	13	69
Normal*	0	2	2	1	2	2	4	1	4	3	21

Comments: In the age-correlated statistical calculations, the limit for the age groups was set at ± 2.6 months. As a result, a nine-month-old boy with bilateral clubfeet was outside this age span and is not included in this table. *Normal feet in unilateral cases.

3.2.3 STUDY IV

Twenty-two consecutive children were treated for congenital clubfoot at the Department of Orthopaedics, Skaraborg Hospital, Skövde, Sweden, from 2006 to July 2011. Of these, 20 (14 boys and 6 girls) who met the inclusion criteria were included. The control group, 29 healthy children (18 boys and 11 girls), were recruited from the maternity ward, Skaraborg Hospital, Skövde, Sweden, and the maternity ward, Sahlgrenska University Hospital/Östra, Gothenburg, Sweden. The ultrasound investigations that were analysed were made at the same ages as in Studies II and III.

3.3 ULTRASONOGRAPHY

3.3.1 INVESTIGATION CIRCUMSTANCES

The child was sitting on a parent’s lap or lying on an examination table during the examination. In Study I, the foot was held in an appropriate position by one of the examiner’s hands, while the other conducted the US probe. In Studies II-IV, the same paediatric orthopaedic surgeon (AJ) held the foot in the desired position, while an experienced radiologist performed the US scans. No sedation was used, but some children were fed or were given a sugar solution during the examination in order to be relaxed.

3.3.2 PROJECTIONS

3.3.3 STUDY I

Three scanning planes were used: medial coronal, lateral coronal and dorsal sagittal (Fig. 1a-c). *The medial scan* was performed with the probe at the medial border of the foot in a slightly oblique plane so that the medial and lateral malleoli, the talus and the navicular were visualised in the same scan (Fig. 1a and Fig. 3). All the feet were examined in a neutral position. To evaluate the mobility of the talo-navicular joint, frozen images in maximum abduction and adduction were saved for 20 feet in each age group. *The lateral scan* was performed with the probe at the lateral border of the foot parallel to the footpad (Fig 1b). In the *dorsal sagittal scan,* the probe was placed on the upper side of the foot with the foot in a neutral position regarding ab- and adduction and plantar flexion to visualise the whole length of the talus (Fig. 1c). If the navicular was plantarly dislocated in relation to the talus, an additional scan was performed during passive elevation of the metatarsals to evaluate the reducibility of the plantar dislocation of the navicular.

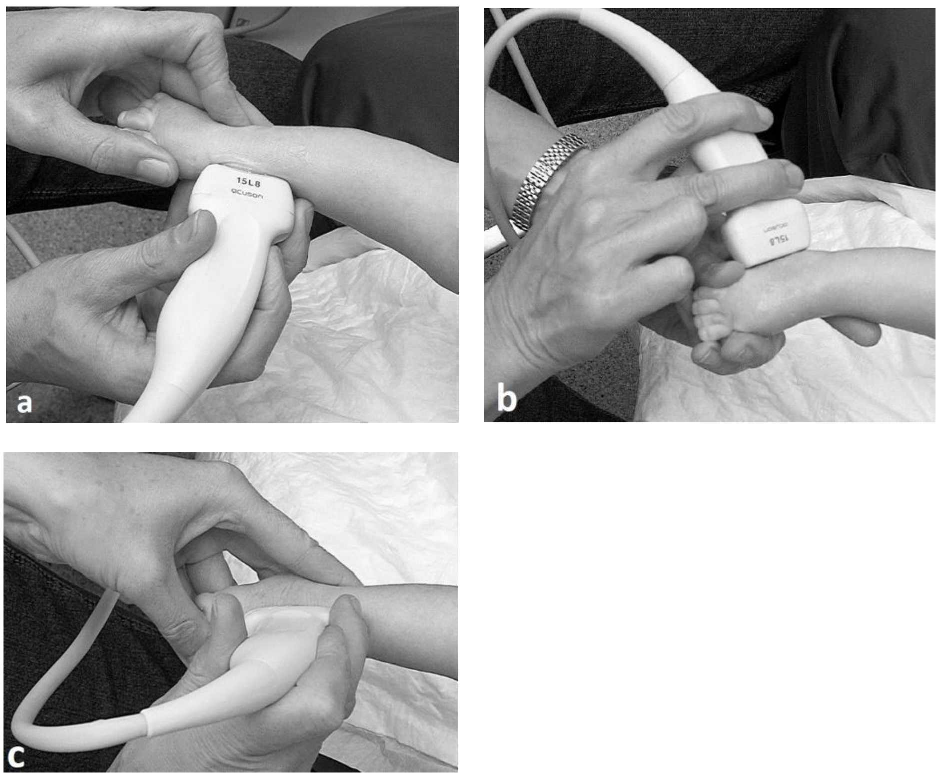


*Figure 1. Transducer positions (**a**) medial coronal (**b**) lateral coronal (**c**) dorsal sagittal.*

3.3.4 STUDY II

A posterior sagittal plane was used to evaluate the mobility of the ankle joint and the degree of equinus. The probe was placed over the heel parallel to the Achilles tendon (Fig. 2). Scans were made with the foot in neutral, plantar flexed and dorsiflexed positions.

Figure 2. Transducer position in the posterior sagittal projection.

3.3.5 STUDY III

The same projections as in Study I were used and scans were performed in the neutral position, with maximum adduction and maximum abduction on all feet in the medial and lateral coronal projections.

3.3.6 STUDY IV

Scans were performed in all the four projections and foot positions used in Studies II and III.

3.4 EVALUATION OF THE ULTRASOUND IMAGES

In Study I, the measurements were performed on hardcopy laser prints of the ultrasound images. In Studies II to IV, the measurements were made digitally using PACS software (Centricity PACS, General Electric Healthcare, and SECTRA PACS, Sweden).

In Studies I and III, measurements were made on the two coronal and the dorsal projections. In Study II, measurements made on the posterior sagittal projection. In Study IV, evaluations were performed on all four projections.

Distance measurements were made from the cartilage border to the cartilage border of the bones in order to obtain measurements independent of the age-related size of the osseous nuclei. Moreover, the angle measurements were performed with the cartilaginous borders as anatomical landmarks for the same reason.

3.4.1 MEASUREMENTS ON THE MEDIAL CORONAL SCANS

On the medial coronal scans, the following measurements were made.

1. The distance between the medial malleolus and the proximal medial corner of the navicular (MM-N distance) (Figs. 3 and 4a).
2. Soft-tissue thickness (STT) (Fig. 3b).
3. Visual assessment of the medial displacement of the navicular, semi-quantitative grading as normal, subluxated or luxated.
4. Medial malleolus-talar head-navicular distance (MM-T-N) was introduced in Paper III to improve the assessment when the navicular turns around the head of the talus in the abducted position (Fig. 4b), because then the MM-N does not measure the real distance the navicular moves.
5. The distance between the medial tangent of the talus and the medial border of the navicular (T-Tang-N) was introduced in Paper III to improve the assessment of the medial-lateral position of the navicular (Fig. 5).
6. The angle between the longitudinal axes of the talus and a line drawn from the centre of the talar head to the most proximal medial border of the navicular (T-N angle) (Fig. 6). Note that the same terminology has been used for another way of measuring the angle between the talus and the navicular in a few previous papers [55, 56].

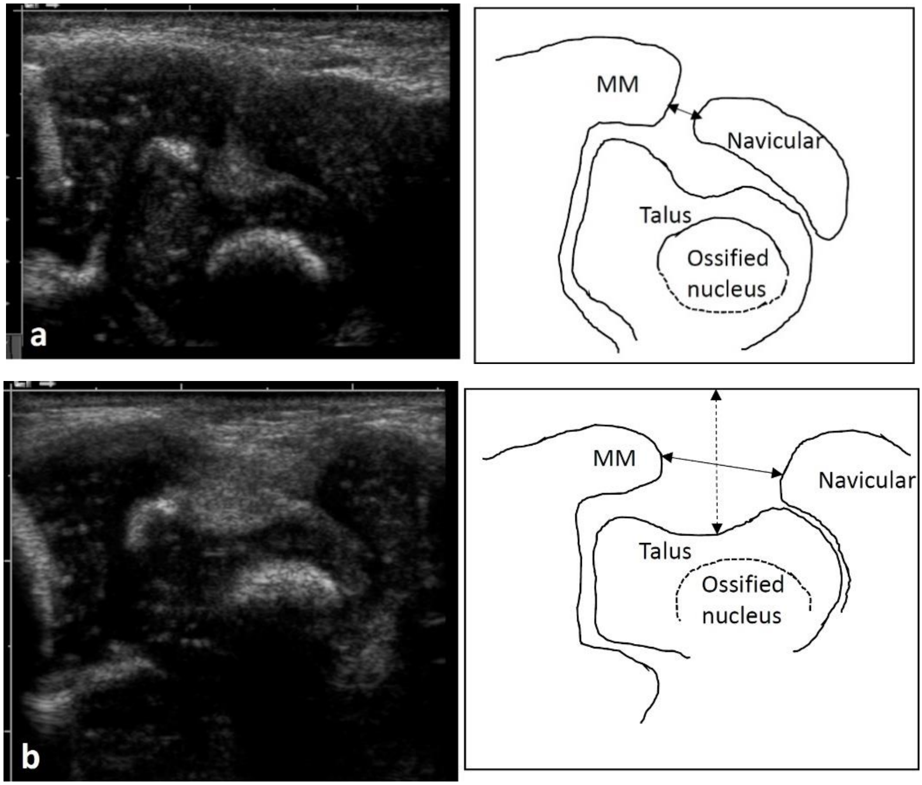


Figure 3. A clubfoot after four castings in a one-month-old baby.
*(**a**) In the neutral position. (**b**) In the abducted position (Ponseti manipulation)*
Unbroken double arrow = medial malleolus-navicular (MM–N) distance
Dashed double arrow = soft-tissue thickness (STT).

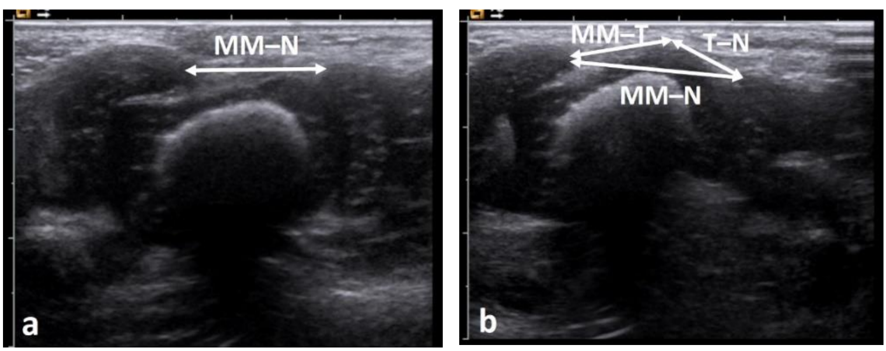


Figure 4. Normal foot in a three-month-old girl.
*(**a**) In the neutral position. MM–N = medial malleolus-navicular distance.*
*(**b**) In the abducted position. MM–T = medial malleolus-medial aspect of the talus distance. T–N = medial aspect of the talus-navicular distance. The sum of MM–T and T–N = MM–T–N.*

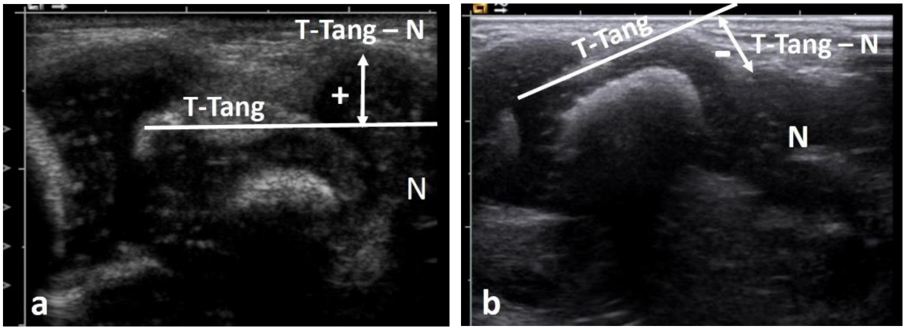


*Figure 5. T-Tang-N distance = the perpendicular distance from the medial tangent of the talus to the medial border of the navicular. When the medial border of the navicular is medial to the tangent, the values are positive (+) and, when it is lateral to the tangent, the values are negative (-). N = navicular bone. (**a**) Clubfoot during early treatment, the navicular is medial to the tangent. (**b**) Normal foot in abduction, the navicular is lateral to the tangent.*

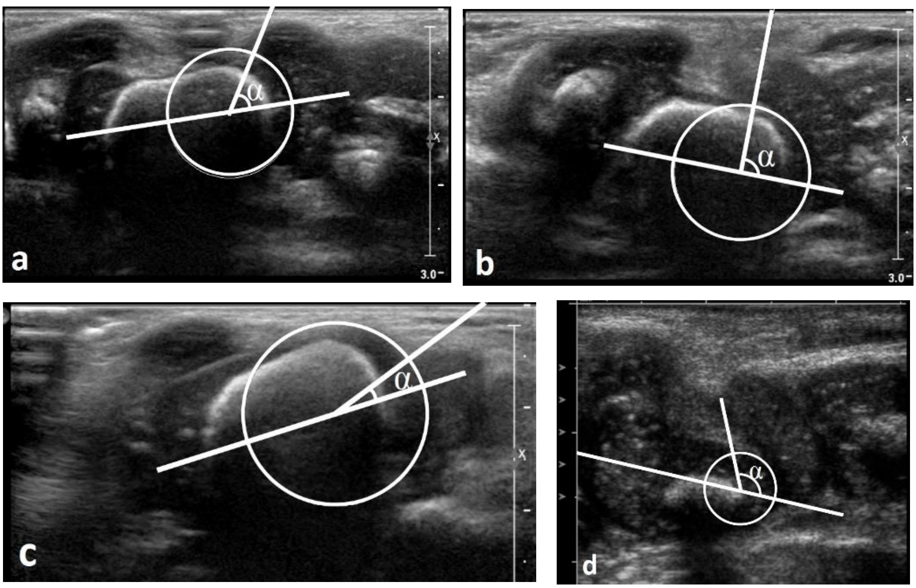


Figure 6. T-N angle (α) = the angle between the longitudinal axis of the talus and a line from the centre of the talar head to the medial border of the navicular bone. The centre of the talar head was determined using the "region of interest (ROI)" tool in the PACS. The size-adjustable circle was laid over the periphery of the talar head and the lines were drawn through the marked midpoint.
*(**a**) Normal foot in a one-year-old boy in the neutral position. (**b**) In the adducted position. (**c**) In the abducted position. (d) In an untreated clubfoot in a five-day-old girl.*

3.4.2 MEASUREMENTS ON THE LATERAL CORONAL SCANS

On these scans, the relationship between the calcaneus and the cuboid bone was evaluated by the following variables.

1. The calcaneo-cuboid distance (C-C distance) (Fig. 7)
2. The calcaneo-cuboid angle (C-C angle) (Fig. 7)

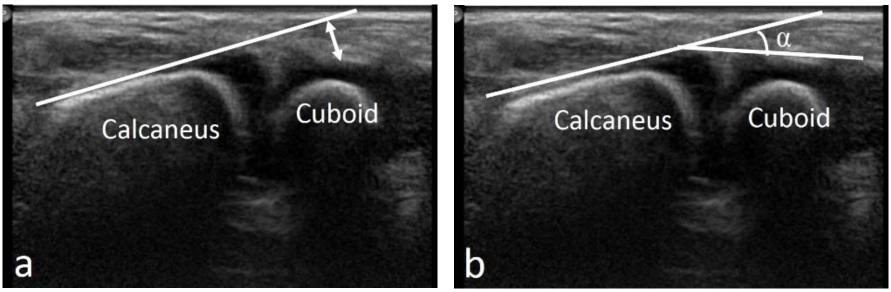


*Figure 7. Lateral coronal projection of a normal foot in the neutral position in an 11-month-old girl. (**a**) C-C distance (double arrow) = the perpendicular distance between the lateral tangent of the calcaneus and the middle of the lateral border of the cuboid. The values were defined as positive (+) when the lateral border of the cuboid was medial to the lateral tangent of the calcaneus and negative (-) if the lateral border of the cuboid was lateral to the tangent of the calcaneus (**b**) C-C angle (α) = the angle between the lateral tangents of the calcaneus and the cuboid. The values were defined as positive (+) when the angle was medially open and negative (-) when the angle was laterally open in relation to the tangent of the calcaneus.*

3.4.3 MEASUREMENTS ON THE DORSAL SAGITTAL SCANS

On the dorsal scan, two variables were assessed.

1. The length of the talus was measured (Fig. 8).
2. The position of the navicular in relation to the talar head was assessed as normal, plantarly or dorsally dislocated.

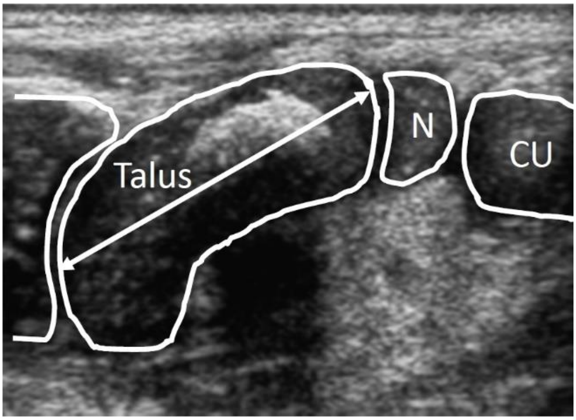


Figure 8. Dorsal sagittal projection. N = navicular bone, CU = cuneiform bone. Double arrow = length of the talus.

3.4.4 MEASUREMENTS ON THE POSTERIOR SAGITTAL SCANS

On the posterior scan, the motion in the ankle joint and the antero-posterior position of the talus in the ankle joint were evaluated.

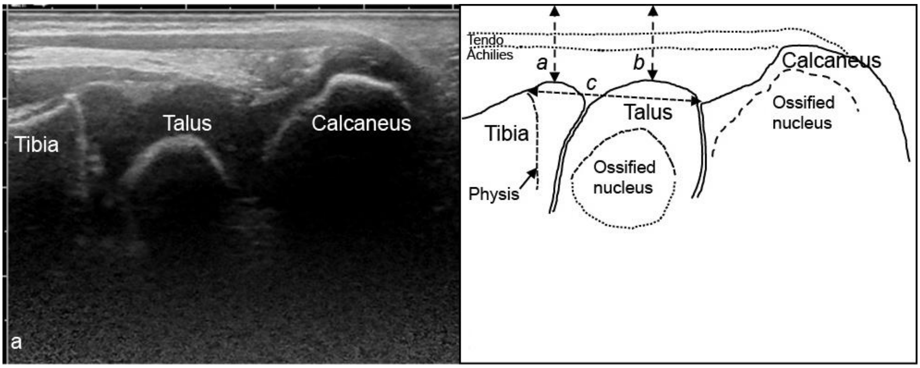


*Figure 9. Posterior ultrasonography scan of the left foot in a neutral position in a 1.5-year-old boy with bilateral clubfeet. (**a**) Skin – tibia distance. (**b**) Skin – talus distance. (**c**) Tibial physis – talo-calcaneal joint distance (Tib.phys.-TCJ).*

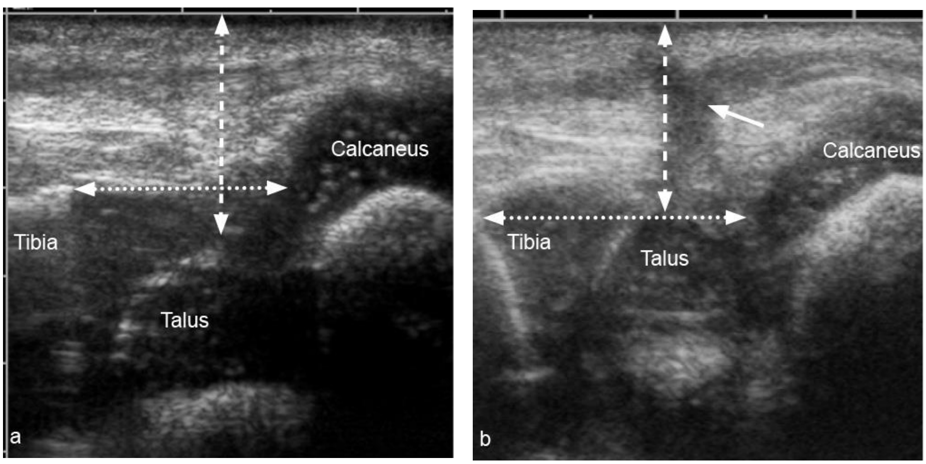


*Figure 10. Posterior sagittal projection of the left foot in a 10-week-old boy with bilateral clubfeet (**a**) before percutaneous tenotomy of the Achilles tendon and (**b**) three weeks after the tenotomy. Note the more dorsal position of the posterior border of the trochlea tali in relation to the tibia and the shorter skin-talus distance (dashed arrow), the increased distance between the tibial physis and calcaneus (dotted arrow) and the black depicted scar after the tenotomy (solid arrow). Before the tenotomy, the posterior border of the talus was not aligned with the posterior surface of the tibial epiphysis, but, after the tenotomy, it was aligned.*

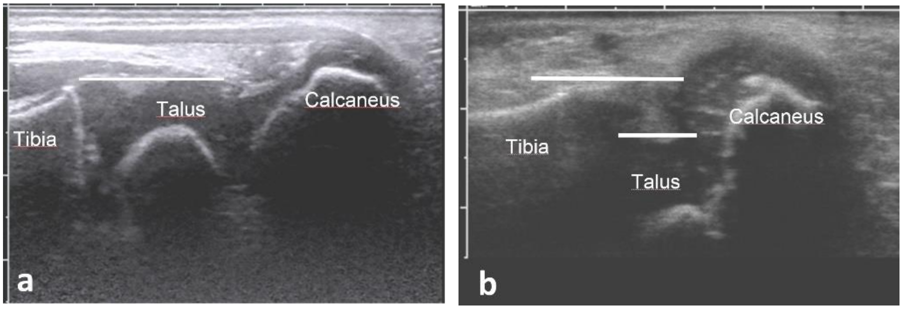


*Figure 11. Posterior alignment (**a**) Aligned, the posterior border of the talus is in line with the tangent of the posterior surface of the tibial epiphysis. (**b**) Not aligned, the posterior border of the talus is anterior to the tangent of the tibial epiphysis.*

3.4.5 EVALUATION OF THE REPEATABILITY OF THE MEASUREMENTS

One of the purposes of this work was to evaluate the repeatability and reliability of the scans and the variables used to assess the images. This was done in the three first studies. The variables tested for reliability were then used in Study IV.

Study I

To evaluate the inter-observer repeatability, all the investigations were blindly interpreted by two independent investigators (YA and AJ). To estimate the intra-observer repeatability, all the images were interpreted a second time by one of the investigators (YA).

Studies II and III

All the images of the feet of the children with a clubfoot and the controls were measured by AJ and these measurements were used for the statistical evaluations. To evaluate the intra-observer repeatability, 60 of the feet in the control group (age: 3, 6, 12 and 48 months) and 36 clubfeet (age: 6, 18, 24 and 48 months) were measured twice by AJ. For the inter-observer evaluation, all 71 clubfeet and 66 feet from the control group (age: 6, 12 and 48 months) were also measured by YA.
To evaluate the repeatability of the new posterior scanning plane introduced in Study II, the posterior scans of 14 feet (10 clubfeet and 4 normal feet) in seven children were examined twice on the same day independently by two experienced US examiners (YA and S-BH).

3.5 CLINICAL INFORMATION

In Studies II-III, on the visit at which the US examination was performed, the feet were assessed and the presence of any adductus, varus, supination or equinus was registered as yes or no. The plantar flexion and dorsiflexion were measured with a handheld goniometer from the lateral side, with one goniometer arm parallel to the fibula and the other parallel to the sole of the foot.

In Study IV, the US was performed on the same day as the clinical follow-ups to the age of four years. Plantar flexion, dorsiflexion and any presence of adductus, abductus, supination or cavus were registered. The Diméglio score was retrieved from the medical record on the first visit to the clinic. Clinical data were retrieved from the medical record: clinical assessment of the feet, progression of the treatment, number of castings, type and use of orthoses and tenotomy of the Achilles tendon, as well as other complementary surgical treatment.

3.6 ETHICAL APPROVAL

Study I
The study was approved by the Ethics Committee at the University of Lund, Sweden (Dnr. LU: 288-96).

Studies II and III
These studies were approved by the Regional Ethical Review Board in Gothenburg, Sweden (Dnr. 031-06 and T397-07).

Study IV
The study was approved by the Swedish Ethics Authority (EPM Dnr. 2019-04403) and the Regional Ethical Review Board (EPN), Gothenburg (Dnr. T397-07 ad. 031-06).

3.7 STATISTICS

Study I

Student's t-test was used when the data were approximately normally distributed; otherwise the Mann-Whitney U-test was used. Pearson's correlation was used for intra- and inter-observer agreement.

Studies II and III

The calculations and diagrams were made using IBM SPSS Statistics 22 (IBM Corp., Armonk, New York). The Intraclass correlation coefficient (ICC) was used for the calculation of inter-examiner reliability in Study II and for the inter-observer and intra-observer reliability in Studies II and III. The Mann-Witney U-test was used for the statistical comparisons. In the comparative statistical calculations, the age group limits were set at ± 2.6 months and, as a result, one nine-month-old child (two feet) and three children (six feet) who had passed the age of 48 + 2.6 months were excluded from the comparative calculations. Sensitivity analyses were performed with and without the three patients (six clubfeet) who had passed the age of 48 + 2.6 months, but this did not change the results of the sensitivity tests.

Study IV

Descriptive statistics for continuous variables were presented as the mean with 95% CI (confidence intervals). Frequencies and percentages were used for categorical variables. The Mann-Whitney U-test was used for comparisons of continuous variables dealing with non-normally distributed data or small groups. Spearman's correlation was used to explore bivariate correlations between different continuous variables. All the calculations were performed using IBM SPSS Statistics 25.

4 RESULTS

4.1 STUDY I

4.1.1 RELIABILITY

The reliability was calculated for each age group and was r=0.65-0.94 for intra-observer reliability and r=0.53-0.93 for inter-observer reliability, $p \leq 0.01$ (Pearson's correlation coefficient).

4.1.2 MEASUREMENTS

As the ossified nuclei are highly echogenic they are depicted as very bright with acoustic shadowing deep to the calcified surface. The non-ossified cartilage appeared as black, with white dots representing blood vessels, while the articular cartilage was anechoic and depicted as black in the image. The medial projection visualised the ankle mortis, talus and the navicular. The MM-N distance and the STT increased with age and the difference between the age groups was significant ($p < 0.001$). The C-C distance increased to a statistically significant degree to the age of seven months (($p \leq 0.009$), but, between the ages of seven and 12 months, the difference was not significant.

4.2 STUDY II

4.2.1 RELIABILITY

The intra-observer agreement, intra-class correlation coefficient (ICC) for all the variables was ≥ 0.90 for the controls and ≥ 0.86 for the clubfeet. The inter-observer ICC was ≥ 0.8 for controls, except for the Tib. phys.-C distance in plantar flexion (0.68), and ≥ 0.84 for all the variables in the clubfeet. The inter-examiner ICC was 0.71 to 0.89.

4.2.2 MEASUREMENTS

The Tib. phys.-TCJ distance in the neutral and dorsiflexed position was statistically significantly shorter in the clubfeet than in the controls in some age groups and the same tendency was seen in the other age groups. There was no significant correlation between the clinically measured dorsiflexion and the Tib. phys.-C distance. After the correction phase of the clubfeet (from the age of six months), the clinical range of motion in the ankle joint calculated per age group was 14° to 34° less in clubfeet.

4.3 STUDY III

4.3.1 RELIABILITY

The intra-observer agreement (ICC) was calculated for a total of 20 variables in controls and 18 variables in clubfeet, including different foot positions.
The intra-observer ICC was > 0.8 in 18/20 variables (exceptions: T-N angle in neutral and adducted) in controls and in 16/18 variables (exceptions: T-N angle adducted and C-C distance neutral position) in clubfeet.

The inter-observer ICC was calculated for 20 variables in the controls and 19 variables in the clubfeet. The inter-observer ICC was ≥ 0.6 for all variables, except the C-C distance in controls, T-N angle adducted, C-C angle neutral and C-C angle adducted in clubfeet (for details, see Table 2).

			Intra-observer						Inter-observer					
			Controls			Clubfeet			Controls			Clubfeet		
Projection	Variable	Position of foot	n	ICC	95 % CI	n	ICC	95 % CI	n	ICC	95 % CI	n	ICC	95 % CI
Medial	STT	Neutral	57	0.95	0.92-0.97	34	0.82	0.68-0.91	64	0.85	0.77-0.91	65	0.96	0.94-0.98
Medial	MMN	Neutral	57	0.98	0.97-0.99	35	0.96	0.92-0.98	64	0.97	0.95-0.98	66	0.94	0.91-0.96
Medial	MMN	Adducted	54	0.97	0.95-0.98	29	0.96	0.91-0.98	51	0.88	0.79-0.93	61	0.87	0.80-0.92
Medial	MMN	Abducted	53	0.99	0.98-1.00	35	0.99	0.98-0.99	57	0.98	0.97-0.99	64	0.95	0.92-0.97
Medial	MM-T-N	Abducted	47	0.97	0.95-0.99	4	≠		55	0.95	0.92-0.97	17	0.85	0.63-0.94
Medial	T-Tang-N	Neutral	57	0.92	0.87-0.96	33	0.95	0.91-0.98	64	0.71	0.56-0.81	69	0.88	0.81-0.92
Medial	T-Tang-N	Adducted	55	0.95	0.91-0.97	29	0.88	0.76-0.94	58	0.68	0.51-0.79	64	0.67	0.51-0.79
Medial	T-Tang-N	Abducted	52	0.87	0.79-0.92	35	0.94	0.89-0.97	59	0.75	0.62-0.84	67	0.94	0.91-0.96
Medial	T-N angle	Neutral	55	0.75	0.61-0.85	31	0.91	0.83-0.96	64	0.86	0.77-0.91	69	0.84	0.76-0.90
Medial	T-N angle	Adducted	47	0.71	0.54-0.83	22	0.58	0.22-0.80	50	0.75	0.60-0.85	62	0.59	0.40-0.73
Medial	T-N angle	Abducted	51	0.86	0.77-0.92	34	0.93	0.87-0.96	58	0.79	0.66-0.87	66	0.90	0.84-0.94
Medial	Talar length	Neutral	39	0.99	0.98-1.00	17	0.99	0.98-1.00	45	0.97	0.95-0.98	35	0.87	0.77-0.93
Medial	Talar length	Abducted	29	0.99	0.98-1.00	11	0.99	0.97-1.00	37	0.99	0.98-0.99	25	0.76	0.53-0.89
Dorsal	Talar length	Neutral	32	0.99	0.98-0.99	5	≠		33	0.85	0.72-0.93	8	≠	
Lateral	C–C dist.	Neutral	55	0.89	0.83-0.94	34	0.75	0.56-0.87	64	0.86	0.78-0.91	64	0.76	0.63-0.85
Lateral	C–C dist.	Adducted	48	0.92	0.86-0.95	28	0.88	0.77-0.94	56	0.90	0.84-0.94	56	0.73	0.58-0.83
Lateral	C–C dist.	Abducted	52	0.89	0.82-0.94	30	0.89	0.77-0.94	62	0.58	0.39-0.73	55	0.87	0.79-0.92
Lateral	C-C angle	Neutral	55	0.94	0.90-0.96	32	0.93	0.86-0.96	64	0.69	0.53-0.80	63	0.59	0.40-0.73
Lateral	C-C angle	Adducted	49	0.88	0.80-0.93	28	0.93	0.85-0.97	57	0.80	0.68-0.88	55	0.45	0.22-0.64
Lateral	C-C angle	Abducted	52	0.94	0.90-0.96	26	0.85	0.70-0.93	61	0.60	0.42-0.74	55	0.70	0.53-0.81

Table 2. Intra- and inter-observer reliability measured using the Intraclass correlation coefficient (ICC).
n = number of valid measurements of the variable. Measurements may be missing because the variable is not applicable to all feet, the child did not co-operate in obtaining a complete investigation or the image quality was insufficient. STT = soft-tissue thickness, MM-N = medial malleolus-navicular distance, MM-T-N = medial malleolus-talar head-navicular distance, T-Tang-N = distance from the medial tangent of the talus to the medial corner of the navicular bone, T-N angle = talo-navicular angle, C-C dist. = calcaneo-cuboid distance, C-C angle = calcaneo-cuboid angle

4.3.2 MEASUREMENTS

MEDIAL PROJECTION

The mean STT was thicker in clubfeet than in controls in all age groups ($p < 0.05$).

The MM-N distance was significantly shorter within some age groups and the same tendency was seen in the others in all foot positions except at the three-month adducted position.

The MM-T-N distance in the neutral position was applicable (the medial border of the navicular was lateral to the medial tangent of the talus, thereby the MM-T-N > MM-N) in 70/210 feet in the controls and 5/70 in the clubfeet. The mean difference between the MM-T-N and the MM-N distance without age stratification, in the neutral position, was 2.62% (0 to 7.91) in controls and 3.07% (0.45 to 8.76) in clubfeet.

The MM-T-N distance was applicable in abduction in 164/210 feet in controls and 15/71 in clubfeet. The difference between the MM-T-N and the MM-N in the abducted position was a mean of 3.87% (0 to11.33) in controls and 2.78% (0 to 7.74) in clubfeet.

The T-Tang-N distance in the abducted position had positive values in 4/198 feet in controls and in 48/70 clubfeet. This means that the medial corner of the navicular moved laterally in relation to the talar tangent in only 31.5% (22/70) of the clubfeet in abduction.

The T-N angle in the abducted position was statistically significantly larger in clubfeet in all age groups except the 36-month group.

The mobility (ROM) in the talo-navicular joint evaluated without age stratification as the mean difference between the T-N angle in the adducted and abducted positions was 59.2° in controls (SD 11.8°) and 41.1° in clubfeet (SD 19.7°). The mean difference between the controls and the clubfoot cohort was therefore 18.1°.

LATERAL PROJECTION

The mean difference between the C-C angle in ad- and abduction for the whole control group was 17.0° (SD 11.7°) and, in the clubfoot group, it was 8.5° (SD 9.0°), making the difference between controls and clubfeet 8.5°.

The C-C angle in controls was smallest in the younger age groups and increased with increasing age. In the clubfoot cohort, the development was the opposite, as the angle decreased with age. In newborns, the angle was larger in clubfeet than in controls, but from the age of two years it was the opposite.

DORSAL PROJECTION

The length of the talus was measured on both the dorsal and medial projections to evaluate whether it matters which projection the measurements are made on. The measurements on the medial projection tended to be shorter than on the dorsal projection, but the mean percentage difference was small, -1.8% (SD 7.9%) in the neutral position, -4.4% (SD 7.65%) in the adducted position and -2.6% (SD 6.6%) in the abducted position. The length of the talus tended to be shorter in the clubfeet compared with controls.

CORRELATION WITH CLINICAL ASSESSMENT

In eight clubfeet, the navicular was assessed as subluxated and in these feet the MM-N was shorter ($p < 0.01$), while the T-Tang-N distance and the T-N angle were significantly larger ($p < 0.01$) than in the clubfeet where the navicular was not displaced. Of the eight clubfeet with a subluxated navicular, only one had documented clinical adductus, while data were missing for three.

Correlation between clinical and ultrasonographic evaluation:
Eight clubfeet had a clinical remaining adductus. Compared with the other 47 clubfeet with complete data available (16 missing), the MM-N distance was shorter in the neutral ($p < 0.05$), adducted ($p < 0.05$) and abducted ($p < 0.01$) positions. The T-Tang-N distance was longer ($p < 0.05$) in the adducted position. The T-N angle was larger ($p < 0.05$) in the neutral and abducted positions. The C-C angle was larger ($p < 0.05$) in the adducted position.

4.4 STUDY IV

4.4.1 MEASUREMENTS

MEDIAL PROJECTION

The STT was thicker in clubfeet than in controls and contralateral normal feet during the first four years of life. In clubfeet, it decreased during the first six months (correction phase) and it then increased with growth to four years of age. In the controls and the normal contralateral feet, it increased from birth, but the increase slowed down from 1-1.5 years, while in clubfeet the increase continued and was significantly greater than in controls from 1.5-4 years ($p < 0.001$).

The MM-N distance was shorter in clubfeet than in controls and contralateral normal feet in all foot positions from newborn to four years of age ($p < 0.05$-0.001). The ratio between the length of the talus and the MM-N in the neutral position was smaller in clubfeet. The difference decreased after the initial cast treatment, but there was a difference to the age of four years.

The T-Tang-N distance was longer in clubfeet compared with controls and contralateral normal feet in all age groups from newborn to four years of age, in the neutral position ($p < 0.001$ and in abduction ($p < 0.01$).

The T-N angle was larger in clubfeet than in controls ($p < 0.01$) and contralateral normal feet in unilateral cases ($p < 0.05$) in the neutral and abducted positions from newborn to four years of age.

The ROM in the talo-navicular joint (the T-N angle difference between the adducted and abducted positions) was about 20° less in clubfeet than in controls to the age of four years ($p < 0.001$).

LATERAL PROJECTION

The measured values for the C-C distance as well as the difference between the adducted and abducted positions were small (mean < 4 mm in all age groups in clubfeet as well as controls).

DORSAL PROJECTION

The talus was shorter in clubfeet compared with controls. The growth of the talus, expressed as the mean increase in the talar length in mm from birth to the age of four years, was less in clubfeet ($p < 0.003$). Expressed as a percentage of the talar length at birth, the increase was larger in clubfeet than in controls and contralateral normal feet; from the age of one year the difference was statistically significant ($p < 0.05$).

At the first US investigation of the newborn children with clubfeet, the navicular had a plantar dislocation in 16/30 and a normal position in 11 (data missing for 3). At the age of one year, they had all normalised, apart from two atypical clubfeet, which needed another six months to normalise.

POSTERIOR PROJECTION

In newborns, the overall correlation between Tib.phys.-C (clubfeet)/Tib.phys.-TCJ (controls and contralateral normal feet) compared with clinically measured foot dorsiflexion was $r = 0.7$ ($p < 0.001$). At the age of two years, the correlation between the Tib.phys.-TCJ distance and clinical foot dorsiflexion was $r = 0.4$ ($p = 0.001$) and at age four years it was $r = 0.3$ ($p = 0.009$).

4.4.2 CORRELATION BETWEEN US AND DIMÉGLIO SCORE

The T-Tang-N distance was longer ($p = 0.004$) in clubfeet with a Diméglio adductus variable score of 3 compared with those with a score of 2 (there was only one foot with a score of 1 and no with a score of 4). The length of the talus had a negative correlation to the total Diméglio score, $r = -0.599$.

4.5 CORRELATION BETWEEN US AND THE COURSE OF TREATMENT

The correlation between measurements on the medial projection in the abducted position (Ponseti manoeuvre) before treatment and the number of casts needed to correct the deformities (with two atypical outliers excluded) was negative for MM-N, $r = -0.7$ ($p < 0.01$), positive for the T-Tang.-N distance, $r = 0.4$ ($p < 0.01$), and the T-N angle, $r = 0.7$ ($p < 0.01$). With the two outliers included, the Spearman's rho for MM-N was -0.66, T-Tang-N 0.496 and for the T-N angle 0.576 ($p < 0.01$).

Between the MM-N distance at the age of 1.5 and four years, there was still a negative correlation with the number of casts they had needed as newborns to correct the deformities.

Tib.phys.-C data before treatment were available for 19 feet. The 10/19 who subsequently required a tenotomy of the Achilles tendon had a shorter Tib.phys.-C distance than the others ($p < 0.001$ Mann-Whitney test).

4.5.1 COURSE OF TREATMENT

All 30 clubfeet were treated using the Ponseti method. During the casting treatment, 21 feet had a tenotomy of the Achilles tendon and, of these, three (in two children) had a second tenotomy. They were all treated with orthoses, after the casting period, 29 received a foot-abduction orthosis (FAO) and one a knee-ankle-foot orthosis (KAFO). In six, the type of orthosis was subsequently changed, most often because of sleeping problems.

In addition to the tenotomies of the Achilles tendon performed during the casting treatment, complementary surgery was required in 12 feet, five of which had atypical signs. Three percutaneous and five fractional percutaneous Achilles tendon lengthenings were performed. Two atypical clubfeet required posterior release, including the lengthening of the posterior tibial tendon. The transfer of the anterior tibial tendon was performed on ten feet and one had a tenodesis of the anterior tibial tendon.

5 DISCUSSION

5.1 STUDY I

By placing the proximal end of the probe over the medial malleolus and the distal part over the navicular and tilting the probe slightly towards the plantar side, a reproducible image plane is achieved, visualising the ankle mortis, the talus and the navicular in the same image, similar to an anteroposterior radiograph. The lateral projection is defined by the centre of the calcaneus and cuboid in a plane parallel to the planta pedis. The dorsal sagittal plane is where the talus, the navicular and one of the cuneiform bones are optimally depicted. To visualise the posterior border of the trochlea tali, the scan is performed with the foot in plantar flexion. These three scanning planes were reproducible. The medial and dorsal projections visualise the position of the navicular, which is essential in clubfoot treatment. These planes are therefore particularly useful for following the course of treatment of clubfeet, as the navicular does not ossify and become visible on plain radiographs until the age of three to four years.

5.2 STUDIES II-IV

5.2.1 MEASUREMENTS ON THE MEDIAL PROJECTION

The three continual variables, MM-N, T-Tang-N distance and T-N angle, are used to evaluate the degree of dislocation in the talo-navicular joint and there is an interdependent covariation between these variables, as well as for the variable “navicular position” (classified as normal, subluxated or luxated). With increasing dislocation of the navicular, the MM-N distance decreases, while the T-Tang-N distance and the T-N angle increase.

In addition to the navicular dislocation, the MM-N distance is influenced by the length of the talus, which is in turn influenced by the severity of the clubfoot and the size of the child. When the navicular moves mainly in the distal direction away from the medial malleolus, the MM-N measures the increased distance sufficiently well, but it is not as sensitive to motion in the medial-lateral direction. To overcome this problem, the MM-T-N distance was introduced, but the percentile difference compared with the MM-N was not so large, the mean values for controls and clubfeet in the neutral and abducted positions were all between 2.5 and 4% (range 0 to 11.33) and the variable was mainly applicable in normal feet in abduction and only in 20% of the clubfeet in abduction. The clinical value of this variable is therefore limited.

When the navicular is completely luxated, the value for T-Tang-N is equal to the thickness of the navicular (just below 5 mm in newborns). When the navicular turns medially around the caput tali, the maximum value is reached when the navicular is subluxated and it does not increase further with increasing dislocation, making this variable most sensitive to moderate dislocations of the navicular.

5.2.2 MEASUREMENTS ON THE LATERAL PROJECTION

C-C distance

The C-C distance is affected by the angle between the calcaneus and the cuboid and by whether there is a medial dislocation of the cuboid in relation to the calcaneus. The consequences of persistent calcaneo-cuboid malalignment have been discussed [57]. A more detailed analysis of these two components would be interesting in order to reveal a spurious correction taking place distally to the cuboid. Images including the calcaneus, cuboid and the fifth metatarsal, making it possible to evaluate the alignment of the whole lateral border of the foot, would enable this. Technical ultrasound features like the extended field of view (panorama imaging) or a longer ultrasound probe would be helpful.

C-C angle

The C-C angle in controls increased slightly with age, i.e. the lateral border of the cuboid becomes more medially angled in relation to the lateral border of the calcaneus. In untreated clubfeet, the C-C angle was larger than in controls and decreased when the adductus deformity was corrected, but it tended to continue to increase even during the orthosis treatment during the maintenance phase (Study III). Further studies are needed to investigate whether this is significant and whether it can be an effect of the foot abduction orthosis. One explanation could be that the outward rotation of the calcaneus under the talus is restricted and a compensatory outward angulation of the cuboid occurs.

5.2.3 RANGE OF MOTION (ROM)

In Study III, the mean range of motion in the talo-navicular joint, expressed as the mean difference between the T-N angle in the ad- and abducted positions, was 59.2° (SD 11.8°) for the controls and 41.4° (19.7°) for the clubfoot cohort. In the calcaneo-cuboid joint, the mean ROM expressed as the difference between the C-C angle in ad- and abduction was 17.7° (SD 11.7°) for the control group and 8.5° (SD 9.0°) for the clubfoot cohort. The motion in the talo-navicular and the calcaneo-cuboid joints occurs simultaneously and is interdependent, but there is a considerable difference in range of motion in the two joints. This is possibly due to the simultaneous rotation of the calcaneus underneath the talus.

5.2.4 MEASUREMENTS ON THE DORSAL PROJECTION

If possible, the length of the talus should be measured on the dorsal sagittal projection, because the posterior border of the trochlea tali is most accurately depicted in this projection. If measured on the medial projection, the thin posterior edge of the trochlea tali may be outside the image plane, resulting in measurements that are too short. On the other hand, the mean percentile difference between measurements on the dorsal and medial projection was not so great, 1.8% (SD 7.9%) shorter in the neutral position and 2.6% (SD 6.6%) shorter in the abducted position measured on the medial projection.

5.2.5 MEASUREMENTS ON THE POSTERIOR PROJECTION

We chose to measure the shortest distance from the posterior border of the tibial physis to the calcaneus and the talo-calcaneal joint respectively. Some authors have measured orthogonally to the tibial physis, but this is more difficult and time consuming. Furthermore, in older children, it is not always possible to see the whole physis, while the posterior border of the physis is easy to identify. We evaluated both variants of measurement, but the difference was minimal, usually < 1 mm, and we therefore chose the easiest method. Distally, the measurements were made to the cartilaginous surfaces of the bones, not to the ossified nuclei, the diameter of which increases with age.

5.2.6 INVESTIGATION OF ATYPICAL CLUBFEET

In atypical clubfeet, it may be difficult to obtain a medial scan of good quality, due to the thick folded soft tissues and the deep medial crease. The cavus deformity with plantar dislocation of the navicular makes it difficult or impossible to visualise the medial malleolus, the central part of the talus and the navicular in the same plane. On the dorsal sagittal scan, the extreme medial displacement of the navicular causes problems. When the scan is centred and parallel to the long axis of the talus, the navicular may be out of the plane or only the border of the navicular is visible. These problems can be overcome by tilting the probe about 45° laterally using an oblique plane to scan the navicular through the cartilaginous distal part of the talus, revealing the severe dislocation of the navicular. This scanning plane is a useful complement when it is difficult to obtain good medial and dorsal scans in young children as long as the caput tali has not ossified.

5.2.7 PERSISTENT DEFORMITIES AND SPURIOUS CORRECTION

On the medial projection, there was a persistent difference between clubfeet and normal feet for the variables of MM-N distance, T-Tang-N distance and the T-N angle in all age groups (even after the initial correction phase). This means that the navicular was more medially positioned in clubfeet than in normal feet, i.e. the dislocation of the navicular had not completely normalised. In the majority of the clubfeet, there was no corresponding clinical adductus deformity after the correction phase (only 8/71 in Study III had persistent adductus). The remaining dislocation of the navicular therefore appears to be compensated for by a spurious correction in more distal joints. This is in accordance with other reports [58]. In 1981, Ponseti et al. published a study comparing the radiographs of clubfeet and normal feet in 32 patients, aged 14 to 32 years, with unilateral clubfeet treated according to the Ponseti method [1, 59]. They found a medial displacement of the navicular in the majority of the clubfeet and a lateral angulation of the cuneiforms in more than half of them.

In corrected and well-functioning clubfeet, there are often some radiological anomalies [60, 61]. One very important question relates to the degree to which the remaining deformities and spurious correction are of clinical relevance to the function of the foot. Most follow-ups are short term and children do not have the same demands or the same problems with the feet as adolescents and adults [1, 60].

Which persistent deformities have a negative impact on the function of the foot and which are radiological cosmetics? To answer this question, long-term follow-ups from the first years of life into adulthood and several years into working life or to retirement are needed. The patient’s demands on his/her feet are affected by many factors, such as type of occupation, sports activity, BMI and so on, and this should be considered in the assessment.

5.2.8 CORRELATIONS BETWEEN INITIAL US AND THE COURSE OF TREATMENT

The correlation between the MM-N distance and T-N angle and the number of casts needed to correct the deformities was high (-0.7 and +0.7 respectively), while the correlation for T-Tang-N distance was low (0.4) (Study IV). One reasonable explanation for the low correlation for T-Tang-N distance is the fact that the maximum value for this variable is equal to the thickness of the navicular and does not increase beyond this value, as further dislocation means that the navicular tilts medially around the talar head.

5.3 STRENGTHS AND LIMITATIONS

5.3.1 GENERAL STRENGTHS AND LIMITATIONS

In order to generalise the results of the present study, a larger group of children with clubfeet should be investigated. However, the size of the study cohorts is comparable to those in many previous studies in this field [23, 60, 62, 63].

There is a lack of longitudinal studies of clubfoot treatment. This study, combining four years of morphological and eight years of clinical follow-up, will contribute to our understanding of treatment progress.

Even though the Ponseti method is used all over Sweden, there may be differences in the implementation of the treatment. Nevertheless, the two hospitals involved in the actual study have been following the same treatment guidelines for many years, which is a strength.

The schedule for the US investigations was based on the chronological age of the children. No adjustment was made for any premature birth, birth weight or height, which may have influenced the results. This was compensated for in some way by measuring the length of the feet in connection with the US investigations.

If changes in the treatment are made between two US investigations, for example, adjustment or exchange of orthosis, it is difficult to evaluate how many of the changes in the US findings are due to the change in treatment or unexpected incidents in the meantime. A US examination co-ordinated with the change of treatment is desirable.

5.3.2 STUDIES II AND III

In these cross-sectional studies, children with clubfeet who were under treatment when the study started were included. So, no untreated feet were included in the clubfoot cohort. At the start of the studies, the plan was to conduct the US investigations on the same day as the check-ups at the clinic. This proved to be difficult to co-ordinate for the older children and the routine was therefore changed so that they came for an extra visit for the US. For a period, the oldest children had to be given priority so that they did not become too old to participate in the study. This was one of the reasons the number of patients was unevenly distributed between the age groups, resulting in too few clubfeet in some age groups to permit cross-sectional calculations of all variables.

5.3.3 STUDY IV

There were only ten children with unilateral clubfoot and sometimes the image quality did not permit the measurement of all variables. As a result, statistical comparisons between the normal feet in unilateral cases and the controls and clubfeet were not always possible.

6 CONCLUSIONS

6.1 GENERAL CONCLUSION

Studies I-III have concentrated on the development of US assessment protocols and evaluating new measurement variables concerning repeatability and reproducibility. Study IV; with the experience acquired from Studies I-III as the background, it was important to follow the same children longitudinally up to the termination of the orthosis treatment, usually at the age of four years.

6.2 STUDY II

Ultrasound images can be achieved with good inter-examiner reliability. An evaluation of the ultrasound images can be made with good intra- and inter-observer agreement, but the correlation to clinical measurements was not as good. The information from single measurements on frozen US images of the dorsal aspect of the ankle joint is limited, but dynamic US provides a good visualisation of the movement in the ankle joint, which can provide clinically useful information and act as a valuable tool in education.

6.3 STUDY III

Using ultrasonography, the morphology and mobility in the talo-navicular and calcaneo-cuboid joints can be evaluated with fair to very good reliability during the first four years of life. The length of the talus is best measured on the dorsal projection. The ROM in the talo-navicular and the calcaneo-cuboid joints was lower in clubfeet than in normal feet. Ultrasonography can be a valuable complement to a clinical evaluation of the anatomy in clubfeet.

6.4 STUDY IV

Ultrasonography is a useful tool for evaluating the progress of correcting the deformities and the growth in clubfeet. Significant differences were found between clubfeet, controls and contralateral normal feet. Ultrasonography provides information that can be a useful complement to the clinical evaluation in deciding the need for further treatment.

7 FUTURE PERSPECTIVES

7.1 CLINICAL APPLICATION

Ultrasonographic scans and measurements can be made with a high level of reliability. The possibility to get dynamic investigations in real time might be the greatest clinical benefit of ultrasonography [64]. A basic knowledge of ultrasound physics and imaging is a prerequisite, as are familiarity with the scanning planes and interpretation of the images.

The most optimal clinical set-up would be to have close access to ultrasonographic equipment and to make a check whenever there are doubts about the degree of misalignment or what happens during the manipulation process.

7.2 LONG-TERM FOLLOW-UP

New investigation techniques like ultrasonography generate new knowledge, which have to be put into a wider clinical context to evaluate what is clinically useful and what is not. Long-term follow-ups into adulthood and further are needed to understand the clinical significance of persistent anomalies.

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