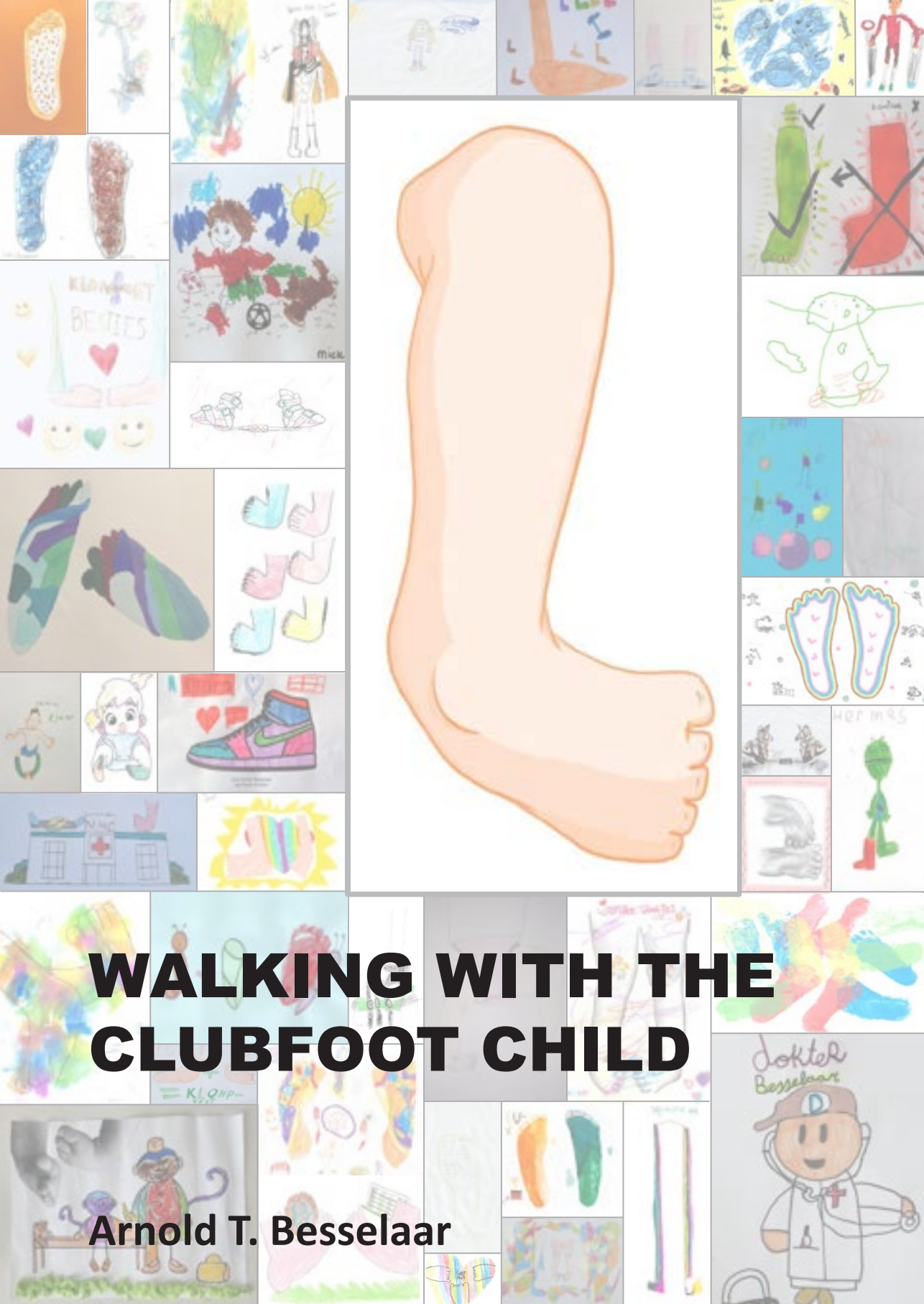

WALKING WITH THE CLUBFOOT CHILD

Arnold T. Besselaar

Walking with the clubfoot child

Evolution of clubfoot care in the Netherlands

Arnold Timotheus Besselaar

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CHAPTER 1

General introduction

The idiopathic clubfoot

The idiopathic clubfoot

The idiopathic clubfoot (talipes equinovarus) is a complex, 3-dimensional congenital deformity with structural changes of the foot and the lower leg. The clubfoot is characterized by a combination of midfoot cavus, midfoot adductus, hindfoot varus and equinus (Figure 1.1 and 1.2).[1,2] One (unilateral) or both (bilateral) feet can be affected. In the majority of patients, the deformity is already visible on ultrasound in the prenatal period.[3] In the Netherlands an ultrasound is performed around the 20th gestational week.

The exact pathogenesis leading to an isolated idiopathic clubfoot deformity remains unclear. Genetic aspects play a role, but also environmental factors have been associated with the occurrence of clubfeet.[4-6] It is important to distinguish idiopathic clubfeet from positional clubfeet, due to limited space in utero, without typical clubfoot appearances and self-limiting.[7] Furthermore, the group of syndromic clubfeet represents a different entity than the idiopathic clubfeet. In syndromic clubfeet the foot deformity is associated with syndromes like arthrogryposis or myelomeningocele.[8,9] Due to the structural changes that occur in an idiopathic clubfoot, the deformity is not reducible acutely and active treatment is indicated.

Figure 1.1 - Typical aspect of untreated idiopathic clubfoot.

Figure 1.2 - Clinically, the clubfoot appears as a foot directed in a fixed cavus, adductus, varus and equinus. (Drawing Saskia Wijnands).

Historic background of the clubfoot

Already in ancient times clubfeet as a foot deformity were noted. Drawings of Hephaestus, the Greek God of fire, technical knowledge and metallurgy, portrayed a man with a clubfoot deformity (Figure 1.3). Around 400 B.C., Hippocrates carried out research on clubfeet and proposed a treatment strategy with repeated manipulations followed by bandages that showed impressive similarities with the treatment nowadays.[10]

Figure 1.3 - Anonymous. Etruscan vase (520 B.C.) stored at the Kunsthistorisches Museum of Vienna. The person sitting at the back of the horse shows a clubfoot deformity.[10]

Many decades ago, researchers reported pathological anatomic changes in clubfoot. The first description dates from Antonio Scarpa (1752-1832), a professor of anatomy and theoretical surgery in Modena, Italy (Peltier 1994). More recently authors described the clubfoot in anatomical and patho-anatomical manners where altered relationships between midtarsal deformities in the bony shapes were noticed. Most of the time these descriptions were based on surgical observations. Ponseti and Ippolito added a histological dissection study of fetal healthy and fetal clubfoot (Figure 1. 4) showing these deformities already existed fetally.[11] Beside bony deformities also differences in muscles, tendons and vessels were described.[6,12,13]

Figure 1.4 - Illustration of one of the dissections made by Ponseti and Ippolito, showing the deformities of a clubfoot of a 3-year-old infant (Ponseti 1996).[2]

Treatment of the clubfoot deformity

The last medical, Dutch thesis giving an overview of clubfeet treatment was written in 1986 by Thomas Hoogland.[14] The regular treatment those days advised to first treat with plaster, changed every two weeks and after four to five times put on a cast for a month. In case of persisting equinus the foot was indicated for an extended surgical posteromedial release, intra-articular surgery. This treatment protocol changed since then radically.[2]

Nowadays the Ponseti method is accepted as gold standard for the treatment of idiopathic clubfeet. Although the pathology and biomechanical aspects of the clubfoot already had previously been described in detail, Ponseti in 1996 added

important aspects to that knowledge which formed the base to change the treatment worldwide. He emphasized the importance of gentle manipulation and casting as a fundamental step in clubfoot treatment and discouraged extensive operative correction which until then was standard care.[2,3,5,15,16]

Ponseti treatment

The Ponseti treatment consist of a series of gentle manipulations of the foot followed by casting, an Achilles tenotomy and a bracing phase. The manipulative technique is well described in detail and easy to learn.[2] Knowledge about the anatomy and kinematics of the foot helps to understand the technique and thus prevents mistakes. To explain the Ponseti method the nomenclature of tarsal movements is necessary to mention. Ponseti followed the definitions of von Langelaan, who defined the movements of the midfoot and hindfoot by kinematical analysis (Figure 1.5).[16]

Figure 1.5 - Van Langelaan's definitions of movements for tarsal bones. Extension, flexion, adduction, abduction and inversion, eversion are shown. The terms supination and pronation are not mentioned in the figure. Supination is a combined movement of adduction, flexion and inversion. Pronation is a combined movement of abduction, extension and eversion (drawn by Saskia Wijnands, after von Langelaan 1983).[17]

When the manipulation starts, all components of the clubfoot are treated except for the equinus. The sequence of correction of the foot is well described (Figure 1.6).[2] First the cavus is corrected by manipulation and held by a well molded

above knee plaster cast (Figure 1.7). Next the foot is, while fixation of the talar head is obtained, abducted, gradually decreasing the supination till neutral. During this maneuver, the heel will correct from varus into valgus. The plaster is changed weekly, and the correction is extended every visit till the talar head is covered and the entire fore- and midfoot is placed laterally in relation to the talus, after approximately 5 casts. In this position enough correction should be reached to indicate an Achilles tendon tenotomy to correct the remaining equinus component of the clubfoot deformity. After this percutaneous surgery performed under local anesthetics, again a plaster cast is applied and removed three weeks later.

Figure 1.6 - Sequence of correction in Ponseti clubfoot treatment from stage 1 to 4.

The first three months after the casting phase full-time, followed by wearing the brace at night and nap times till their 4th or 5th anniversary.[18,19]

When this final cast is removed, a foot abduction brace is applied (Figure 1.8). This brace must be worn full time for the first three months after the casting phase full-time, followed by wearing the brace at night and nap times till their

4th or 5th anniversary.[18,19] During clinical follow up young clubfoot patients should be evaluated to monitor the correction of the clubfoot.

Figure 1.7 - Applying a plaster cast after gentle manipulation as part of the Ponseti treatment.

Figure 1.8 - Foot abduction brace.

Outcome of treatment

The desirable treatment outcome for a clubfoot is a foot with at least a plantigrade position without pain, fitting normal shoes. All activities of daily life should be possible, so not only an anatomical but also a functional satisfying result is important.[2]

At the end every treatment result must be judged in the patients' perspective and in case of children the parents' perspective. Functioning in children can be captured using the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY). The ICF contains three main aspects which affect a child's functioning: body function and structures, activities and participation Body function and structures are defined as the physiological and psychological function of body systems, activity is defined as the execution of a task or action by an individual and participation is defined as involvement in a life situation (WHO-FIC CC, International Classification of Functioning, Disability and Health, Children & Youth version, BSL 2018). To illustrate this clinically, first the structures of the foot need to be corrected to serve as a base to stand on. Standing correctly can induce walking correctly, which can consecutively lead to physical activities. These activities can finally lead to participation in daily life.

Recurrence

Unfortunately, poor results of the Ponseti method are still faced. Mistakes in the Ponseti treatment are already known since the start of the treatment and are possibly one of the clarifications for poor results.[20] But also differences in initial clubfoot severity and amount of initial correction seem to play a role in the outcome of Ponseti treatment.[21] Other factors as non-adherence of the bracing regime are mentioned.[22] Not completely corrected clubfeet with remaining varus, adductus, equinus or cavus components, are often referred to as residual clubfeet. In addition, relapse is a remaining challenge in clubfoot care. Relapse deformities are defined as feet that initially were corrected but in time show again an appearance with components like the original clubfoot deformity.[23]

Treatment strategies for recurrent clubfoot vary from repeated serial casting or Achilles tenotomy to extensive soft tissue release or corrective osteotomies.[24] Ideally, treating recurrent deformities should be as less invasive as possible with as little as burden for the patient.[25]

Aims and outlines of this thesis

Since the end of the 20th century, in Europe and subsequently also in the Netherlands, clubfoot treatment changed drastically. Different questions arose during this transition phase of the surgical treatment towards the Ponseti method. Answers were necessary to give clarity and support to health professionals and, even more important, to parents and their children with clubfoot. This thesis will address the changes in clubfoot care in the Netherlands. The studies in this thesis raised out of clinical questions and unsolved issues according to treatment transition in clubfoot care. Different perspectives of the studies molded the core of the research projects: organization of care, treatment outcome and their measurements and relapse issues.

PART I: Organization of care

Chapter 2 describes the process in which the Dutch guideline on clubfoot treatment was created and established. The need for guidance was felt by care professionals and parents because treatment protocols differed extensively between geographical locations in the Netherlands. Initiated by the parents' association and supported by the Dutch Orthopaedic Society, a guideline committee was formed with scientific help by the Knowledge Institute of Medical specialists to ensure the highest scientific level possible. Overall, this chapter shows the importance of collaboration between different stake holders with a focus on parent participation. Furthermore, an important tool how one can thoroughly and sustainable change patient care is described.

One of the logical implications of improving care should be centralization of care as shown in complex surgery where higher volumes in treatments per institute or medical professional lead to better patient outcome.[26] To translate this for clubfoot treatment we ought to know the incidence of the pathology in the Netherlands. **Chapter 3** describes the process of detecting the incidence of clubfeet in the Netherlands. Clubfoot centers were visited in person and patient records scrutinized for confirmation of the diagnosis clubfoot. Two external registers were used to verify our findings.

PART II: Outcome

In **Chapter 4 and 5** the influence of the Ponseti treatment on clubfoot children was assessed. As an important part of the clubfoot treatment, children must start

with a brace. The first three months after the casting phase full-time, followed by wearing the brace at night and nap times till their 4th anniversary.[18] Nevertheless, the brace period can have seriously impact on the child and its parents.[27] Therefore, we studied **in Chapter 4** the quality of life during the brace period within the Ponseti treatment protocol. We used a validated questionnaire to examine health related quality of life and compared results of children with clubfoot with healthy controls.[28]

In **Chapter 5** we investigated the functional outcome of the Ponseti treatment. This chapter describes the parents' perceived motor abilities and participation levels of their children. To judge the perceived motor abilities, we used a validated instrument for assessment of motor competency[29] and for the participation, a validated instrument to collect information on life habits assessing participation in children and adolescents was used.[30]

PART III: Relapse

In clubfoot treatment one of the most important challenges relate to recurrent clubfeet. In **Chapter 6** we propose definitions of relapsed and residual deformities. Moreover, we identified and classified these late deformities and treatments applied to solve the relapsed/residual deformities in our own patient population. We also described moments when relapses occur.

Often recurrence of a clubfoot can be treated by the Ponseti method itself, including transferring the anterior tibial tendon. Unfortunately, a patient group remains in which, after all Ponseti methods are used, limitations and complaints are still present. In **Chapter 7** a salvage procedure Anterior Distal Tibial Epiphysiodesis (ADTE) to minimize the ankle impairment is studied. A foot in equinus is an important impairment during gait.[24] In this study we measured the sagittal angular influence on the tibial plafond, measured the shape of the talus as indicator for a so called 'flat top talus' and studied in a selected group the one year follow up after removal of the used tension band plate.

PART IV: General discussion

At the end of this thesis, in **Chapter 8**, the findings of the separate studies are discussed in the light of current literature and future perspectives. Furthermore, the impact of this thesis for clubfoot care in the Netherlands is described in **Chapter 9**.

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CHAPTER 2

Guideline on the diagnosis and treatment of primary idiopathic clubfoot

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*Acta Orthopaedica 2017;88(3):305-309**

*adapted version: Question 4, remarkably, was omitted from the published article. The proof included this question but the printed article did not. For completeness, however, it is included in this thesis and commented on in the general discussion. Furthermore, methods and parts of the results published as supplementary materials are incorporated in the main text.

Abstract

A delegation of 6 pediatric orthopedic surgeons from the Dutch Orthopaedic Association (NOV) and 2 members of the board of the Dutch Parents' Association for children with clubfoot created the guideline "The diagnosis and treatment of primary idiopathic clubfeet" between April 2011 and February 2014. The development of the guideline was supported by a professional methodologist from the Dutch Knowledge Institute of Medical Specialists.

This evidence-based guideline process was new and unique, in the sense that the process was initiated by a parents' association. This is the first official guideline in pediatric orthopedics in the Netherlands, and to our knowledge it is also the first evidence- based guideline on clubfoot worldwide.

The guideline was developed in accordance with the criteria of the international AGREE instrument (AGREE II: Appraisal of Guidelines for Research and Evaluation II). The scientific literature was searched and systematically analyzed. In the second phase, conclusions and recommendations in the literature were formulated according to the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) method. Recommendations were developed considering the balance of benefits and harms, the type and quality of evidence, the values and preferences of the people involved, and the costs.

The guideline is a solid foundation for standardization of clubfoot treatment in the Netherlands, with a clear recommendation of the Ponseti method as the optimal method of primary clubfoot treatment. We believe that the format used in the current guideline sets a unique example for guideline development in pediatric orthopedics that may be used worldwide. Our format ensured optimal collaboration between medical specialists and parents and resulted in an important change in clubfoot care in the Netherlands, to the benefit of medical professionals as well as parents and patients.

In this way, it is possible to improve professional collaboration between medical specialists and parents, resulting in an important change in clubfoot care in the Netherlands that will benefit medical professionals, parents, and patients. The guideline was published online and is freely available from the Dutch Guideline Database (www.richtlijnendatabase.nl).

Introduction

Clubfoot (talipes equinovarus) is a well-known deformity in children. The disorder consists of 4 entities: cavus, equinus, varus, and adductus, not spontaneously correcting with growth. There are no specific data on the incidence of clubfoot in the Netherlands, but this incidence can be estimated based on data obtained in nearby countries with similar population characteristics. Based on the incidence of clubfoot in Sweden of 1.4 per 1,000 newborns,[1] it is estimated that 200–300 children with 1 or 2 clubfeet are born in the Netherlands every year.

Starting around 1990, the popularity of the non-surgical treatment of clubfoot with the Ponseti method increased worldwide. Today, the Ponseti treatment of clubfeet is considered to be the first choice of primary treatment in most parts of the world, but some feet are still first treated surgically. For patients and their parents, and also for medical professionals, the choice between these treatment modalities was not an obvious one. For the non-surgically based treatment, unwanted variation resulted in different outcomes in children who were treated, leading to an urgent need for scientifically based guidance.

The Dutch Parents' Association for children with clubfoot organized a forum discussion in 2011, which resulted in the development of the Dutch guideline on primary treatment of clubfoot supported by the Dutch Orthopaedic Association (NOV). The guideline was developed in collaboration with the Dutch Knowledge Institute of Medical Specialists and was co-developed by the Dutch Parents' Clubfoot Association. The initiation by and participation of a parents' association represents a new concept for guideline development in the Netherlands.

The Dutch clubfoot guideline on primary treatment of clubfoot has recently been approved by the general assembly of the NOV. The NOV already has a long history of multidisciplinary reviews leading towards evidence-based guidelines. Also, in the Nordic Orthopaedic Federation the importance of guideline development to improve care is emphasized. These guidelines serve as a standard in specific treatments used by professionals to rule out unwanted variance and to provide guidance for professionals and patients. Guidelines cover the optimal applied care that an orthopedic surgeon should provide. Deviations from a guideline are allowed—and are sometimes even required—to ensure optimal care of an individual patient, but only if they are justified and

properly documented and agreed upon together with the patient (or legal representative). In a research setting, deviations from guidelines are also allowed, but only after proper authorization by an ethics committee. Guidelines from the NOV are always developed together with professional guideline developers to guarantee an optimal, methodologically sound, process. A properly developed evidence-based guideline makes it easier for caregivers to treat according to a well-balanced standard.

After authorization by the relevant medical societies and patient organizations, guidelines are published in the Dutch Guideline Database (Richtlijnen database; richtlijnendatabase.nl/en/). In this database, guidelines are organized in separate modules with each module addressing a specific clinical question. Instead of revising complete guidelines every 5 years, the guideline database allows for regular updates at the level of individual modules according to need. New modules can easily be added, and modular revisions ensure that guidelines in the guideline database remain up to date. This avoids rigidity in guidance and stimulates new scientific initiatives. As part of the guideline development process, knowledge gaps are identified, and research questions formulated. These research questions are published on the guideline database together with the guideline and are used by professional societies to provide guidance for future clinical research in the Netherlands.

This guideline covers the primary diagnosis and treatment of idiopathic clubfoot in children presenting with the deformity in the first 6 months after birth. This cohort has been studied because it has not been biased by previous treatments. The current guideline does not cover the treatment of clubfeet after delay or in children with residual deformities. At a later stage the guideline might be extended to these patient groups. To our knowledge, this is the first evidence-based guideline on clubfoot.

Methods

Guideline committee

This guideline was developed and sponsored by the NOV, using governmental funding from the Quality Foundation of the Dutch Association of Medical Specialists in the Netherlands. The early preparative phase started in April 2011, and the guideline was officially authorized by the Dutch Orthopaedic Association

in February 2014. The guideline committee consisted uniquely of members of the parents' association, pediatric orthopedic surgeons selected by the NOV, and a methodologist (KB) of the Knowledge Institute of Medical Specialists (KiMS). The methodologist was included to ensure a proper design and systematic evidence-based development of the guideline using the GRADE methodology, to meet all the criteria of the AGREE instrument. Decisions within the guideline group were made by consensus. At the start of guideline development, all guideline committee members completed forms regarding conflicts of interests and this information was published together with the guideline.

Target group and aims

This guideline was developed for Dutch providers of healthcare for children with clubfoot, in particular (pediatric) orthopedic surgeons, but also pediatricians, gynecologists, obstetricians, and general practitioners. The main purpose of the guideline is to provide the best possible care for children with idiopathic clubfoot. This is achieved by informing healthcare providers about optimal treatment decisions, thereby reducing unwarranted variation in the delivery of care. The guideline is also meant to facilitate the development of uniform information for patients, parents, and caregivers.

Methodology and workflow

The guideline was developed in agreement with the criteria set by the advisory committee on guideline development of the Association of Medical Specialists in the Netherlands (Adviescommissie richtlijnen, 2011), which are based on the AGREE II instrument.[2] The guideline was developed using an evidence-based approach endorsing the GRADE methodology and meets all criteria of AGREE II. Grading of Recommendations Assessment, Development and Evaluation (GRADE) is a systematic approach for synthesizing evidence and grading of recommendations offering transparency at each stage of the guideline development.[3,4] The guideline development process involves a number of phases: a preparative phase, a development phase, a commentary phase, and an authorization phase. After authorization, the guideline has to be disseminated and implemented. Furthermore, uptake and use must be evaluated. Finally, the guideline must be kept up-to-date. It has to be revised every 5 years. Because the guideline consists of different modalities, stored in the Dutch Guideline Database, important changes can be incorporated earlier. Each phase involves

a number of practical steps.[4] As a first step in the early preparative phase, a broad forum discussion was held and all relevant stakeholders were consulted to define and prioritize key issues, which were extensively discussed in the guideline committee. The selected, high-priority, issues were translated into carefully formulated clinical questions. These questions defined patient problems, intervention, comparison, and outcomes. Furthermore, the patient outcomes relevant to decision-making were prioritized and minimal clinically important differences were defined. In the development phase, the literature was systematically searched using the databases MEDLINE (Ovid), Embase, and the Cochrane Database of Systematic Reviews. Selection of the relevant literature was based on predefined inclusion and exclusion criteria and was carried out by one of the orthopedic surgeons (AB) in collaboration with the methodologist (KB). For each of the clinical questions, the evidence was summarized by the guideline methodologist using the GRADE approach. A systematic review was performed for each of the relevant outcomes and the quality of evidence was assessed in 1 of 4 grades (high, moderate, low, very low) by analyzing limitations in study design or execution (risk of bias), inconsistency of results, indirectness of evidence, imprecision, and publication bias. The evidence synthesis was complemented by a guideline committee member (AB) considering any additional arguments relevant to the clinical question, including patient values, preferences, and resource use (costs, organization of care issues). Evidence synthesis, complementary arguments, and concept recommendations were extensively discussed in the guideline committee. Then, final recommendations were formulated. The final recommendations are based on the balance between desirable and undesirable outcomes, the quality of the body of evidence across all relevant out-comes, values and preferences, and resource use. The strength of a recommendation reflects the extent to which the guideline panel was confident that desirable effects of the intervention would outweigh undesirable effects or vice versa, across the range of patients for whom the recommendation is intended. The strength of a recommendation is determined by weighing all relevant arguments together. This includes the weight of the body of evidence from the systematic literature analysis, and also the weight of all complementary arguments formulated, the so called considerations. When using the GRADE approach, guideline panels must use judgement in integrating these arguments to make a strong or weak recommendation. Thus, although a low quality of the body of evidence from the systematic literature analysis will generally result in a weak recommendation, it does not a priori exclude a strong recommendation, and weak recommendations

may also result from high-quality evidence.[4] After reaching consensus in the guideline committee, the concept guideline was subjected to peer review by all the relevant stakeholders: the commentary phase. Amendments were made and agreed upon by the guideline committee, and the final text was presented to the Dutch Parents' Clubfoot Association for approval and to the NOV for formal authorization. In this authorization phase, additional amendments were made to the guideline text based on the outcome of a general assembly of the NOV. The guideline was finally approved by the Dutch Parents' Clubfoot Association and officially authorized by the NOV.

Results

Results of literature review and analysis:

The following questions were formulated by the guideline committee:

1. *What is the optimal treatment for clubfoot?*
2. *What is the importance of brace compliance and other patient-related factors in the successful treatment of clubfoot?*
3. *What is the optimal method to be used for the diagnosis and classification of a clubfoot?*
4. *Is screening for developmental dysplasia of the hip (DDH) in idiopathic clubfoot useful?*
5. *With respect to organization of care, what are the preconditions for optimal treatment of patients with clubfoot?*

Below, the main (sub)questions are elaborated.

Clinical Question 1: What is the optimal treatment for clubfoot?

This clinical question consisted of several subquestions. The first subquestion should determine the preferable treatment, surgically (postero-medial release (PMR)) or non-surgically. The second subquestion judged the non-surgical treatments of clubfoot used in the Netherlands. Based on the available evidence, is the Kite or the Ponseti method preferred? After first answering subquestions 1 and 2, some subquestions were added. In the Ponseti method, are accelerated treatment protocols useful and is the use of different plaster materials effective?

Also, in the brace treatment there are differences within the Ponseti method, so the question of which brace needs to be used was addressed.

Our systematic literature analysis comparing surgical treatment (PMR) and non-surgical treatment (Ponseti) showed that the results of the Ponseti treatment were at least as effective as treatment with a PMR; level of evidence according to GRADE: LOW.[5-9] Mobility and also ankle and foot position in childhood are reported to be better after Ponseti treatment than after surgical treatment by PMR; level of evidence according to GRADE: LOW.[5,9-14] Serious clubfoot recurrence is reported less frequently after Ponseti treatment than after surgical treatment by PMR; level of evidence according to GRADE: LOW.[6-9] A serious recurrence was defined as a recurrence with the need for intra-articular surgery (intracapsular or bony). Quality of life in adulthood was reported to be higher after Ponseti treatment than after surgical treatment by PMR; level of evidence according to GRADE: VERY LOW.[10-14] Cost-effectiveness studies considering advantages between 1 of the 2 treatments are scarcely available. One study reported 2 times lower costs for Ponseti treatment than for PMR in New Zealand. Also, in the USA, the costs related to the Ponseti method were lower than when an extensive surgical treatment was used.[7] For any surgical treatment, one should consider local and systemic complications. Also, the severity and frequency at which these complications (risks) occur are factors that are important in deciding a favorable treatment. If a surgical treatment does not result in better treatment (effects vs. benefits) according to nonoperative treatment for the same deformity, the nonoperative modality is preferable. If one cannot prove significant advantages of a surgical procedure, the least invasive treatment should be recommended. If literature searches conclude that there is low evidence according to the GRADE classifications, these factors play an important role and can—although the literature-based evidence is low—lead to a strong recommendation. Considering this aspect in relation to the above-mentioned literature, the guideline group came to a clear recommendation of the Ponseti method in preference to surgical strategies such as PMR. The systematic literature analysis also provided evidence in favor of using the Ponseti treatment in the primary correction of idiopathic clubfoot instead of using a treatment according to the Kite method; level of evidence according to GRADE: LOW.[15-18] In the Netherlands, besides the Ponseti method, Kite's method is also used as a non-surgical treatment of clubfoot. The literature analysis indicated that the Ponseti method was more effective than Kite's method; level of evidence according to GRADE: LOW.[18] Comparison of the

standard Ponseti method with the accelerated Ponseti method showed similar effectiveness in primary correction of idiopathic clubfeet; level of evidence according to GRADE: MODERATE.[19] Because of greater and more widespread experience with the standard Ponseti method, using weekly serial manipulations, the guideline committee advised use of this method. The accelerated Ponseti method may be considered if poor compliance during the plaster phase is to be expected and if the hospital outpatient scheduling permits it. In primary correction of idiopathic clubfoot, the available evidence indicates that the standard use of plaster of Paris in the Ponseti treatment is more effective than using synthetic plasters; level of evidence according to GRADE: LOW.[20] Moreover, long-term results are decisive and are not yet fully known for Ponseti treatment using materials other than plaster of Paris. Therefore, the guideline committee advised that the standard plaster of Paris should be used for the Ponseti treatment. Ponseti treatment using a Dennis-Brown type of foot abduction brace resulted in greater effectiveness in preventing recurrences than Ponseti treatment followed by ankle/foot orthotics according to the literature analysis; level of evidence according to GRADE: VERY LOW.[21] The guideline committee recommended the use of a Dennis-Brown type of foot abduction orthotic with a bar, as used in the standard Ponseti method. Further scientific research will be needed to support future brace modifications.

Clinical Question 2: What is the importance of brace compliance and other patient-related factors in the successful treatment of clubfoot?

Firstly, the significance of the severity of clubfoot in relation to treatment success using the Ponseti method was addressed. Secondly, compliance regarding wearing of an abduction brace in relation to the success of Ponseti treatment was an important issue to investigate. Thirdly, the importance of other patient-related factors in relation to the treatment success in Ponseti treatment was investigated. The literature search for these questions was similar to the search used in the treatment section. Due to heterogeneity between the studies and a lack of quantitative data required, a meta-analysis was not possible. In the literature on wearing a foot abduction brace, different definitions of compliance are used. The duration of wearing the brace differs in different treatment protocols, and as a result non-compliance is defined differently in the studies that are included. Another important finding is that in all studies, patient compliance was reported by the parents and may have been overestimated. Even so, taking these limitations into account, a statistically significantly reduced risk of recurrence was found in compliant patients in all but one of the studies

included. Only in the study by Halanski et al. was no statistically relevant relationship found, but the statistical power of this study was limited because of the small size of the patient sample.[7] Non-compliant patients regarding foot abduction brace wear are strongly positively associated with the risk of recurrence, although no strong scientific evidence is available; level of evidence according to GRADE: MODERATE.[7,22-29] A relatively large distance to the treatment center might possibly have a negative influence on treatment success. Treatment in a local urban patient group resulted in significantly greater improvements in Pirani score and a lower risk of recurrence; level of evidence according to GRADE: MODERATE.[27] The other statistically significantly positive associations are a married marital status of the parents, private insurance status, and high educational level and income level; level of evidence according to GRADE: MODERATE.[27] It is important to mention the use of univariate data analysis; without correction for potential confounders, the relationship between the different factors remains unclear. Some of these factors could indirectly affect the treatment results, from their effects on compliance regarding wearing of the foot abduction brace. Considering the low statistical power (small study sample size), the univariate analyses without adjustment for confounding variables, the testing of a wide range of factors (multiple testing), and the large heterogeneity between the studies, the guideline committee concluded that these findings cannot be used to predict the success rate of Ponseti treatment. The severity of clubfoot (determined at the start of the treatment) is a likely prognostic factor regarding the risk of recurrence.[7,22,23,25-30] The guideline committee therefore recommends that parents should be warned that in cases of severe clubfoot, the duration of the treatment and the number of plaster casts could be higher than usual. Caregivers are advised to register factors such as severity, mobility, and brace compliance. Each of these factors can influence the treatment outcome. By frequent contact between parents and caregivers this possible influence can be monitored and if necessary, positively influenced.

Clinical Question 3: What is the optimal method to be used for the diagnosis and classification of a clubfoot?

Because there is international consensus on defining the deformities in clubfoot, a systematic literature analysis was not required. Instead, authoritative standard works in orthopedics were used.[31,32] A clubfoot consists of 4 typical entities: equines (mid- and hindfoot), varus (hindfoot), cavus (midfoot) and adduction (forefoot). Often there are typical folds in the sole of the foot, an altered heel formation, and an altered formation of calf and peroneal muscles. These typical,

clinically addressable entities can reliably lead to the diagnosis of clubfoot. The additional value of radiological examination as a diagnostic tool for clubfoot is limited.[24] However, radiological examination can be of additional help when there is doubt about progress in treatment. Radiographs can be used to follow treatment progress.[33] The guideline committee supports the opinion that radiological examination is not primarily indicated in diagnosing clubfoot. No uniform, generally accepted classification system for quantification of the severity of the clubfoot deformation is used either in the Netherlands or worldwide. A classification system would ideally provide a solid prognosis for treatment outcomes and also a method to measure treatment outcomes. Ideally, the scoring system for clubfoot would be reliable and reproducible and would include separate information on the different parts of the foot and its position in 3 dimensions and provide information on stiffness or flexibility. Last but not least, an ideal scoring system would have strong predictive value regarding treatment outcomes. The severity of a clubfoot can be identified using various classification systems. The value of these classification systems was systematically analyzed according to patient-related prognostic factors in treatment outcomes. Whether or not the severity of the clubfoot is prognostic of relapse has not been reported. The classification systems according to Pirani [34] and Diméglio[35] are most frequently reported. No significant differences in reliability or reproducibility have been described,[36] and there are currently no clear reasons to prefer one system to the other. The guideline committee recommends the simultaneous use of both of these classification systems, in order to facilitate comparison of data in the future. Scoring of the foot has to be performed at the start of the treatment as well as during the follow-up, to evaluate correction and notice negative alterations early on.

Clinical Question 4: Is screening for developmental dysplasia of the hip (DDH) in idiopathic clubfoot useful?

The systematic search revealed 5 studies in which the incidence of DDH was determined. All but 1[37] compared the incidence of DDH in clubfoot with already published data in a healthy population without clubfoot. Important again here is the definition of the disease: in one study, DDH was defined as an acetabular deficiency that must be treated; in other studies, every sonographic or radiological abnormality is mentioned as a DDH. The studies selected from our search mostly found an increased incidence of DDH in the clubfoot population, although the cases detected with DDH often do not need any treatment; level of evidence according to GRADE: LOW to MODERATE.[37-41] New study results on

this topic are expected in the near future. This new literature can be incorporated in a revision of the guideline. This is one of the great advantages of a dynamic, modular guideline format. New insights or new literature can easily be added.

Clinical Question 5: Regarding organization of care, what are the preconditions for optimal treatment of patients with clubfoot?

Currently in the Netherlands, every orthopedic surgeon is allowed to treat children with clubfoot. Of the 800 orthopedic surgeons, around 75 are members of the Dutch Pediatric Orthopedic Association, but not necessarily every one of them is treating clubfoot on a regular basis. An unknown number of non-members also treat children with clubfoot. With an estimated incidence resulting in 200–300 clubfoot newborns every year, the annual number of clubfoot patients per orthopedic surgeon will be low. For this clinical question the optimal process of patient referral, including dividing and allocating responsibilities according to the caregiver, has been addressed. The guideline committee formulated recommendations on how integrated care should be optimized and guaranteed. To answer this question no systematic research was conducted. An important issue was the question of how treatment should be organized nationally and the way in which centralization and specialization should be established. In addition, directly related issues concerning the qualifications and training of orthopedic surgeons are also mentioned. Finally, information and communication with patients, including the reimbursement of prenatal counseling, has been addressed. The guideline group discussed the optimal process in referring patients with clubfoot. If, based on sonographic investigation, clubfoot pathology is suspected during pregnancy, the parents should be referred to a specialized member of a clubfoot treating team for prenatal counseling. Dutch insurance will carry the costs, as consultation is part of the mother's insurance in the period of the pregnancy. During this consultation, the protocol according to diagnostics and treatment can be discussed and the logistic process can be explained. Prognosis can also be discussed, although this is mainly covered by the severity of the pathology to be addressed after the child is born. If the clubfoot is diagnosed just after delivery, the gynecologist, midwife, or general practitioner must contact the local orthopedic surgeon who can transfer the child to a specialized center. According to the responsibilities within the care process, the guideline group stated that the orthopedic surgeon should be the leading member of the treating team. He or she should always diagnose the pathology and initiate the correct treatment. Adequately trained members of the clubfoot team should perform serial

manipulation and plastering themselves. Members of the team should also explain the features of the foot abduction brace and take care that there is a good fit. The treating team should consist of at least 2 trained orthopedic surgeons, 2 trained plaster physicians, and—if necessary—a technician. The optimization of the clubfoot care and aftercare should be coordinated by the orthopedic surgeon. Frequent contact with the patient and the parents appears to be essential. In the primary phase of the treatment until the start of brace wearing, the patient should be checked frequently, mostly on a weekly basis. After at least 2 weeks of starting with the brace, the child should be checked clinically, continuing with check-ups between 3 and 6 months. This is especially important due to the higher risk of residual deformities in non-compliers (See Clinical Question 2). The guideline committee does not involve physiotherapists in the primary treatment initiated before the age of 6 months, because there is no evidence that it would have positive effects. A walking child with locomotive deficits can of course be treated by a physiotherapist, although thorough research on the effectiveness of this therapy would help formulation of recommendations on this subject. Because of the population density in the Netherlands and the high number of hospitals in a relatively small area, patients never have to travel far to reach care although, of course, the traffic increases the travel time. If care is centered because of a low number of patients, the implication is a longer travel time. Based on a questionnaire filled out by their members, the Dutch Clubfoot Parents' Association stated that if the care should be of a higher quality, there would be no objection at all to such an investment of time (Questionnaire NVK 2012). Training of caregivers should be guaranteed by nationally or internationally certified courses, orthopedic surgeons should attend (certified, GAIA) refresher courses, and be members of the Dutch Pediatric Orthopedic Society. Because of the higher number of patients treated in a clubfoot center, the infrastructure can be kept at a high standard. The NOV should audit the centers on an annual basis and publish the results. A specialized center should have a website with up to date information, and should also refer to information that is available on international websites.

Guideline recommendations according to the following clinical questions:

Clinical Question 1: What is the optimal treatment for clubfoot?

- Treat primary clubfoot with the standard Ponseti method.
- Do not use plaster casting according to the Kite method.

- Do not use synthetic plaster casts but Plaster of Paris for the Ponseti method.
- Do not use foot orthotics in follow-up treatment but use a foot abduction brace.
- If possible, the Achilles tendon tenotomy is to be carried out under local anesthetics.
- Only carry out surgery for primary treatment for idiopathic clubfoot as described in the standard Ponseti method.

Clinical Question 2: What is the importance of brace compliance and other patient-related factors in the successful treatment of clubfoot?

- Inform the parents of children with a severe clubfoot because of an increased risk of a higher number of plaster changes and recurrences.
- Motivate the parents strongly so that they succeed with high compliance in the after-treatment with a Dennis Browne- type foot abduction brace up to the age of 4, in order to minimize the chance of recurrences.
- Register and document all factors that could possibly be related to the outcome of the Ponseti treatment on a regular basis for evaluation of the long-term follow-up.

Clinical Question 3: What is the optimal method to be used for the diagnosis and classification of a clubfoot?

- Use physical examination to establish a clubfoot diagnosis.
- Do not use standard radiological examination; radiological examination should only be used when there are doubts about the diagnosis, or when there is a lack of progression of the foot correction, or if there is a recurrence.
- Use both the Dimeglio and the Pirani score as classification systems in order to obtain sufficient long-term data for both classification systems.

*Clinical question 4: Is screening for Developmental Dysplasia of the Hip (DDH) in idiopathic clubfeet useful?**

- Screen children with idiopathic clubfoot, for dysplastic hip development (DDH) by ultrasound or X-ray, regardless of findings on physical examination of the hips.

Clinical Question 5: With respect to organization of care, what are the preconditions for optimal treatment of patients with clubfoot?

- Refer pregnant mothers, when there is suspicion of clubfoot pathology in their unborn children, to a specialized member of a clubfoot treatment team.
- Refer newborns with a clubfoot without previous suspicion—preferably on the first working day—to a local orthopedic surgeon who can take care of referral to a clubfoot center, preferably within 48 hours but not more than one week after delivery.
- The orthopedic surgeon of the clubfoot center should be responsible for the correct diagnosis, initiation of the correct treatment, and the logistic pathway.
- The treatment itself (plaster treatment, tenotomy, brace fitting, and follow-up) must be carried out by adequately trained and qualified members of the specialized team.
- During the primary treatment, the child should be seen on a weekly basis. After the primary process until wearing of the brace starts, there must be frequent contact with the child and parent between 3 and 6 months.
- In the first year, routine physiotherapy is not advisable.
- Clubfoot treatment and the aftercare should be performed in appointed centers with a specialized clubfoot treatment team. Also, extended operations on clubfeet should be performed in specialized centers.
- The treating team should consist of at least 2 trained orthopedic surgeons, 2 trained plaster physicians, and, if necessary, a technician.
- The Dutch Orthopedic Society yearly evaluate the centers on an annual basis according to the recommendations in the guideline and publishes the results.
- A clubfoot center must use a website with adequate and up to date information.

Prenatal counseling should be provided.

Discussion

To our knowledge, this is the first evidence-based guideline by a national orthopedic association to be initiated by a parents' association. This initiative resulted in a scientifically based guideline with optimal support in the medical and parent/ patient communities. The guideline is aimed at providing evidence-based advice both to clinicians and to parents, in order to minimize unwanted variation in treatment of clubfoot and to improve therapeutic compliance.

Because of the frequent lack of high-level evidence in pediatric orthopedics in general, and in clubfoot management in particular, it was essential to use the GRADE method. This is a systematic and transparent approach to collection and grading of available evidence and to weighing the evidence together with complementary arguments, so-called considerations, relevant to the clinical question including patient values and preferences, and resource use (costs, organization of care issues).[3,4] Guideline panels must use judgement in integrating these factors to make a strong or weak recommendation. In the GRADE approach, the strength of a recommendation reflects the extent to which the guideline panel was confident that the desirable effects of the intervention would outweigh the undesirable effects, across the range of patients for whom the recommendation is intended. Guidelines that are produced as a result of a thorough methodological process and used in the correct manner can direct patient care in a positive way. Unwanted variance can be avoided and both caregivers and patients can use a guideline as a basis for communication. A medical professional can always deviate from the recommendations in the guideline but must justify this decision with valid arguments. As a part of a guideline one should identify lack of knowledge, leading towards new research, which can lead to revisions of the guidelines in the future. In this way, guidelines only have advantages and should lead to positive changes in care. The Dutch Guideline Database is formatted in a modular fashion. This means that if there are new insights, it is easier to alter a particular module, so the guideline is more of a dynamic tool. The most important clinical question addressed in the current guideline concerns the primary treatment of clubfoot, whether to use extensive surgery or a non-surgical method. A systematic analysis of the current scientific literature showed similar benefits from surgical treatment (posteromedial release) and non-surgical treatment (Ponseti). Due to the lack of high-quality comparative studies, the overall quality of the evidence was low. Despite this, the guideline panel decided on a strong recommendation of the standard Ponseti method for the treatment of primary idiopathic clubfoot, because of the higher intrinsic risk of complications in surgical treatment than with the non-surgical (Ponseti) treatment. Because of the enormous popularity and favorable results with the Ponseti treatment in idiopathic clubfoot, parents and ethical committees will not support randomized clinical trials comparing surgical release and non-surgical (Ponseti) treatment. Thus, higher levels of evidence cannot be expected in future. New studies should instead focus on optimizing the Ponseti method by determining the optimal length of brace treatment, improving compliance with brace treatment, developing diagnostic tools to identify clubfeet

that are at high risk of treatment failure, and as a consequence developing optimal treatment for these clubfeet. The possibility of using hybrid methods between different non-surgical methods and physiotherapy could also be investigated. Optimal diagnostic and classification tools should also be designed. There is no evidence-based reason to only use the Pirani or the Dimeglio score, so the guideline group decided to use both. In this way, we can probably solve this knowledge hiatus in the future. In a recent study to evaluate both classification systems, simultaneous use of both systems was recommended, as they are different and complement each other.[42]

This guideline was produced for idiopathic clubfoot children starting treatment within 6 months of delivery. Because of this narrow definition, the literature survey could be focused but could be a base for further guideline development in regarding clubfoot patients with residual deformities, relapses, and syndromic clubfoot. The importance of having a guideline for these specific entities, which are not covered by the present document, must be emphasized and this should be addressed in the near future.

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CHAPTER 3

Incidence of congenital idiopathic clubfoot in the Netherlands

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Abstract

The incidence of clubfoot patients is an important factor for centralization of care. Medical records of 21 accredited clubfoot centers were selected using the diagnosis treatment codes and checked to confirm diagnosis. All idiopathic clubfoot cases born during 2013–2014 were analyzed with respect to sex, affected foot, regional distribution, and seasonal variation. Among the 346 522 live births, 377 idiopathic clubfoot cases were registered. The incidence of the congenital idiopathic clubfoot in the Netherlands during 2013 and 2014 was 1.09 per 1000 live births, indicating that every year, ~ 200 children with one or two clubfeet are born in the Netherlands. On the basis of this finding, we can start to refine clubfoot care.

Introduction

Care for patients with idiopathic clubfoot has recently experienced some major changes in the Netherlands, especially with the implementation of the Dutch Guideline on clubfoot care.[1] Recommendations of the guideline include the initiation of dedicated clubfoot treatment centers to centralize the clubfoot care and thereby ensuring a high standard of care. Centralization of care is an important subject in medicine nowadays. An important factor influencing the number of clubfoot treatment centers is the incidence of new clubfoot patients, which, before this study, was not known in the Netherlands.

Worldwide, the incidence of the congenital idiopathic clubfoot is estimated to be ~1 per 1000 live births.[2-4] Previous studies have reported distinct differences in the birth prevalence among various racial, ethnic groups, and geographic regions[5], varying from a high prevalence of 6.8 cases per 1000 live births among Polynesian populations to a low incidence of 0.39 cases per 1000 live births among the Chinese populations.[2] For White populations, an incidence of 1.12 per 1000 live births is reported, whereas among the Hispanic population, 0.76 per 1000 live births is reported.[2,3,6] Irrespective of the ethnicity, the clubfoot consistently shows a male-to-female ratio of 2 : 1. Half of all clubfoot patients are bilaterally affected and in case of unilateral deformity, the right foot is slightly more frequently affected than the left foot.[6,7] Furthermore, several authors have suggested a seasonal variation in the incidence of the congenital idiopathic clubfoot[8-11], whereas several others did not find any seasonal variation.[7,12,13] An explanation for these conflicting findings remains unclear, although there is the speculation of an unidentified environmental influence during the gestation period.[7,8,10,13]

The Netherlands, with a population of 17 million and about 175 000 births each year, has no central register of congenital, orthopedic, deformities. As a result, there are no specific data on the incidence of clubfoot in the Netherlands available. On the basis of the incidence of clubfoot in Sweden, a nearby country with similar population characteristics, of 1.4 per 1000 newborn[4], it is estimated that yearly, about 250 children with one or two clubfeet are born in the Netherlands. To verify this hypothesis, we carried out a cross-sectional nationwide multicenter study. Sex differences, bilateral or unilateral appearance, and seasonal and geographic distribution were studied as secondary endpoints. Knowledge of the real incidence of clubfoot and any seasonal or geographic

distribution will aid informed policy decisions and addresses the issue of centralization of clubfoot care.

Materials and methods

Data collection

In collaboration with the Dutch Pediatric Association (WKO), the 21 accredited clubfoot treatment centers (2014) in the Netherlands were visited for data collection (Figure 3.1). At each of these 21 departments, we recruited a contact person to be responsible and approachable for the data collection. Medical records were selected using the diagnosis treatment codes (DBC) 2040 (clubfoot), 2097 (congenital foot anomaly – orthopedics), and 5108 (deformity – pediatrics). These DBC codes are specific codes in the Dutch healthcare system with information on the diagnosis and treatment of a disease.

Figure 3.1 - The Netherlands is divided into four regions and the 21 clubfoot treatment centers (2014).

By visiting all clubfoot centers, the obtained records were personally scrutinized by one of the authors (M.K.) to confirm the diagnosis of congenital idiopathic clubfoot and minimize the sampling bias. All children diagnosed with a congenital idiopathic unilateral or bilateral clubfoot and date of birth between 1 January 2013 and 31 December 2014 were included in the study. Exclusion

criteria were neurologic clubfoot (e.g. myelodysplasia), syndromic clubfoot (e.g. arthrogryposis multiplex congenita, Down's syndrome), and other congenital foot deformities.

Furthermore, details on sex, month of birth, place of residency, and affected foot were extracted from the medical record. After data collection, duplicate individual cases were identified and excluded on the basis of coded personal data.

This study was approved by the ethical committee (reference niet-WMO 2015-47).

To verify the accuracy of our data, we contacted central registries in the Netherlands, which might have information relevant to the incidence of clubfoot available. We contacted the European Surveillance of Congenital Anomalies in the northern Netherlands (EUROCAT- NNL), which registers congenital anomalies in the three northern provinces of the Netherlands to investigate risk factors. Perined is a registration organization consisting of obstetricians, gynecologists, pediatricians, and general practitioners in the Netherlands; they register a core set of perinatal variables mainly for quality purposes. Furthermore, we collaborated with the Dutch Clubfoot Parents' Association (NVK) and the Dutch Pediatric Association (WKO) for this project (Figure 3.2).

Statistical analyses

The incidence was defined as the number of new clubfoot patients during 2013–2014 in the Netherlands per 1000 live births, calculated as the ratio between the number of children born with congenital idiopathic clubfoot during 2013–2014 and the number of live births during the same period. To obtain the number of live births in the Netherlands, we used the official reports from Statistics Netherlands (Central Bureau of Statistics). This Dutch governmental institution gathers statistical information about the Netherlands.

For the incidence on the basis of the total number of cases and separated for year and sex, the 95% confidence intervals were calculated following the Poisson distribution. To assess whether there was a geographical distribution of clubfoot deformities, we looked at the places of residency of the newborn patients grouped into four regions of the Netherlands (Figure 3.1). Furthermore, we assessed seasonal variation by analyzing the monthly distribution of clubfoot deformities over the year (monthly cumulative incidence). The monthly totals

were combined into four quarters: December to February (winter), March to May (spring), June to August (summer), and September to November (fall). Differences between groups were analyzed using the χ^2-test. Statistical analyses were carried out using SPSS statistical software (IBM Corp. Released 2013, IBM SPSS Statistics for Windows, Version 21.0; IBM Corp., Armonk, New York, USA). For all statistical analyses, *P* values of less than 0.05 were considered significant.

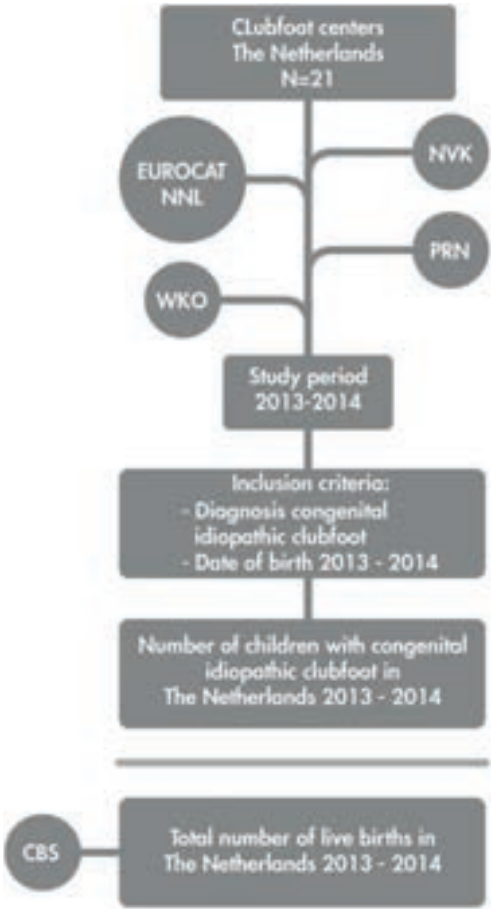

Figure 3.2 - Flowchart data collection. CBS, Central Bureau of Statistics; EUROCAT, European Surveillance of Congenital Anomalies in the Northern Netherlands; NVK, 'Nederlandse Vereniging Klompvoetjes – Dutch Clubfoot Parents' Association; PRN, Netherlands Prenatal Registry; WKO, Dutch Pediatric Association.

Results

Between January 2013 and December 2014, 393 cases of children born with a congenital idiopathic clubfoot deformity were identified in the 21 clubfoot treatment centers. We excluded 16 duplicate individual cases between the clubfoot treatment centers, leading to a total of 377 unique congenital idiopathic clubfoot patients. Among the 346 522 newborns recorded in the Netherlands between January 2013 and December 2014, the incidence of congenital idiopathic clubfoot was 1.09 per 1000 live births. There was no significant difference between the incidence in the 2 years studied, respectively, 1.16 and 1.02 per thousand (*P*>0.05) (Table 3.1).

Table 3.1 - Number of children with congenital idiopathic clubfoot in relation to total number of live births in the Netherlands between 2013 and 2014.

Year	No. of children		Incidence per thousand
	Idiopathic clubfoot	Live births	[95% CI]
2013	198	171 341	1.16 [1.0-1.3]
2014	179	175 181	1.02 [0.9-1.2]
Total	377	346 522	1.09 [1.0-1.2]
Boys	248	177 467	1.40 [1.2-1.6]
Girls	129	169 055	0.76 [0.6-0.9]

As commonly found, the clubfoot deformity occurred more frequently in boys (66%) compared with girls (34%). We found bilateral clubfoot in 188 (50%) cases and unilateral clubfoot in 189 (50%) cases; in case of unilateral clubfoot, the deformity equally affected the right side (50%) and the left side (50%). We also determined whether the distribution of patients with right side- affected or left side-affected unilateral clubfoot or bilateral clubfoot was similar for boys and girls. No sex-related side difference could be detected (*P*>0.05) and there was no significant difference between sex and the proportion of unilateral or bilateral clubfoot cases (*P*>0.05) (Table 3.2).

Table 3.2 - Distribution according to sex, side, bilateral occurrence and affected side of the clubfoot.

	Boys (n=248)	**Girls** (n=129)
Bilateral	131 (53%)	57 (44%)
Right side	56 (23%)	38 (30%)
Left side	61 (25%)	34 (26%)

Sex difference of side; P=0.2; Sex difference of bilateral/unilateral clubfoot; P=0.1; Sex difference of right versus left side; P=0.6.

There was a large variation in the number of treated patients between the 21 clubfoot centers, range 1–48 a year. The eight academic hospitals in the Netherlands treated 55% of all clubfoot patients. The five largest centers, four academic hospitals and one large peripheral hospital, treated 52% of all newborn patients.

After dividing the Netherlands into four regions, we found a statistically significant heterogeneity in the geographical distribution of clubfoot deformities. Confidence intervals showed overlap, indicating that no difference was found between regions (Table 3.3). The seasonal variation in children with clubfoot in relation to the total number of children born per season was also not significantly different (P>0.05).

Table 3.3 - Incidence of idiopathic clubfoot in 4 regions of the Netherland.

Province	No. of children		Incidence per thousand [95% CI]
	Idiopathic Clubfoot	Live births	
North Netherlands	20	31 789	0.6 [0.4-1.0]
East Netherlands	92	73 386	1.3 [1.0-1.5]
South Netherlands	65	65 653	1.0 [0.8-1.3]
West Netherlands	200	175 694	1.1 [1.0-1.3]

Discussion

The search for the incidence of congenital idiopathic clubfoot in the Netherlands was inspired by the Dutch guideline on the primary treatment of idiopathic clubfoot patients. In this guideline, centralization of clubfoot care was advocated to improve results and eliminate unwanted variance. Between January 2013 and December 2014, we registered 377 unique cases of children with congenital idiopathic clubfoot in the 21 clubfoot centers of the Netherlands, resulting in an incidence of 1.09 per thousand live births. No statistical difference was found between the two years studied. In line with the previous literature, we found a higher incidence in boys compared with girls. Furthermore, the distribution of bilateral, unilateral cases, and affected side was in line with the available literature.[6,7] No regional or seasonal difference in incidence was found. This latter finding is in line with studies from Lochmiller *et al.*[7] and Carney and Coburn.[12]

In surgical oncology, it has been shown that in the treatment of complex disorders high volume leads to better treatment outcome.[14-17] The relationship between oncology and clubfoot according to centralization, however, may be debated. Although the initial treatment of an idiopathic clubfoot is relatively simple, ~5–10% of all clubfeet can be classified as complex.[18] Complex clubfeet are extremely stiff feet that contain extreme cavus, a very short first row, and deep, sometimes transverse plantar, creases. These feet require a modified treatment to prevent the need for an adjusted Ponseti protocol.[18] Early detection of this special clubfoot type is important as they demand an even more precise casting technique and require more attention during treatment to achieve a reasonable correction. Furthermore, in up to 48% of the idiopathic clubfoot relapse deformities occur.[19] Initial clubfoot treatment in a dedicated Ponseti clinic compared with treatment in a general clinic decreases the relapse rate.[20] Identification of problematic cases can be difficult because of the large variety in which they can occur.[21] In literature, the best way to treat these relapses is not quite clear.[22,23] Overall, early recognition of complex and relapse clubfoot is important to prevent the need for extensive surgery and a favorable outcome.[18,21,24] Relapse deformities consist not only of clubfoot treated by Ponseti. An important and difficult group of relapsed feet includes former extensively operated feet with disappointing results and problems; these difficult feet also can only be treated adequately by experienced teams. In selected centers, higher number of patients can also be treated, experience in identifying and treating clubfoot will increase, and unwanted variance can be eliminated. In this way, one can expect optimal treatment results.[20]

There are some limitations to our research. Although the Dutch Orthopaedic Association determined the clubfoot centers in 2014, it is still possible to treat clubfoot outside these centers. Therefore, we attempted to verify the accuracy of our data. Perined registered less newborn clubfoot cases as we did. This under-registration might be related to difficulties in recognizing a clubfoot by healthcare professionals who encounter clubfoot sporadically because of the relatively low incidence in their setting and the wide variety of care givers. At a later stage, these clubfoot deformities will be recognized in specialized centers but are then missed in the perinatal database Perined. EUROCAT-NNL, however, had registered eight cases more than we did in the same region. Unfortunately, EUROCAT-NNL is only avail- able for the Northern part of the Netherlands. Our search started off with a broad identification on the basis of several relevant treatment codes, after which all medical records were checked to verify the

diagnosis of congenital idiopathic clubfoot and duplicates were removed. We are certain that all our 377 cases are single idiopathic clubfoot patients born in 2013 or 2014 in the Netherlands. Unfortunately, in studies on the incidence of disorders it is inevitable that a number of cases are missing.[4] In this study, the obtained incidence of clubfoot might be a slight under-registration but is definitely not an over-registration of the true number of clubfoot patients in the Netherlands. Furthermore, in our collaborations for this project with the Dutch Pediatric Association and Dutch Clubfoot Parents' Association, no orthopedic surgeons treating clubfoot patient outside a clubfoot center or patients treated in another than club- foot center came forward. Overall, because of a lack of a nationwide prospective registration of congenital (orthopedic) disorders, we believe that our method of data gathering was the best possible method.

According to the available literature, the incidence of congenital idiopathic clubfoot differs between ethnic groups. On the basis of data available through the governmental institution Statistics Netherlands, ~22% of the Dutch population has an immigration background. Unfortunately, the ethnicity of the identified clubfeet patients is unknown. As a result, it was not possible to determine incidences of different ethnical groups in the Netherlands.

Although determining the number of dedicated clubfoot centers is necessary in the Netherlands to ensure the best possible care for our clubfoot patients, aspects such as the minimum number of patients treated yearly by an orthopedic surgeon but also geographic distribution and availability of trained staff should be considered. The current study contributes to this discussion by determining the number of newborn clubfoot patients who yearly will require treatment. On the basis of the current study, an incidence of congenital idiopathic clubfoot of 1.09 per thousand live births was found. This indicates that every year, ~200 children with one or two clubfeet are born in the Netherlands.

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CHAPTER 4

Quality of life of clubfoot patients during the brace period of the Ponseti method

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The Foot 2022;52:101895

Abstract

Purpose
Quantifying the quality of life in clubfoot patients during bracing following the Ponseti method compared with healthy controls.

Methods
Data collected during the brace period of the Ponseti method and of a reference sample was retrospectively analyzed to investigate health-related quality of life scale (TAPQOL) in clubfoot patients compared with healthy controls. The TAPQOL instrument consists of 12 subscales comprising the 4 domains of health-related quality of life namely physical, social, emotional and cognitive functioning.

Results
Responses of 80 parents of clubfoot patients and 238 parents of healthy controls were analyzed. On average both study groups scored high on the 4 domains of the TAPQOL instrument. The clubfoot group scored significantly (P<0.0125) lower on the subscales motor functioning, sleep, lung and skin problems during bracing. No difference was observed between the study groups in the year the bracing had ended.

Conclusion
Dutch clubfoot patients show an overall good health related quality of life. However, during the brace phase of the Ponseti treatment they score lower in subscales in the physical functioning domain. These results can be used in the counselling of parent and might alleviate some concerns that parents have about the bracing period.

Level of evidence
Level III, Case control study.

Introduction

Approximately 175–200 children with clubfeet are born in the Netherlands every year.[1] The initial treatment of choice is the Ponseti method.[2] The last phase of this treatment consists of wearing a foot abduction brace until the age of four. The brace period is an important as well as intense part of the Ponseti method. The Dutch Guideline for clubfoot care states that the brace has to be worn full time for three months, continued at night-time hours until the age of four.[2,3] A lack of brace compliance is often reported as the main risk factor for a relapsed clubfoot with a 17 times higher rate of relapse in non-compliant cases reported in some series.[3]

Insufficient bracing can have different causes. Physicians may prescribe the brace for a too short period.[4] Residual components of the clubfoot or problems due to the material or manufacture of the brace itself might impede proper brace compliance. Three main reasons for parental noncompliance with regard to the bracing are parents' perception of achieved correction and therefore assuming the brace is no longer necessary, parents' lack of education and parents' belief that the brace is uncomfortable.[4] Other factors include an emotional response by the patients and difficulties in usage of the brace.[4] These findings are in line with our experiences during counseling of the parents of clubfoot patients in our tertiary clubfoot center. Parents often display concerns about the motor development of the children and a possible effect of the brace on the quality of life, which may lead to early discontinuation of brace wearing. A mild, self-limiting delay in the gross motor function was already mentioned in earlier studies but seemed to be no reason for premature ending of brace treatment.[5,6]

The aim of this study is to investigate the health-related quality of life (HRQoL) of clubfoot patients' during the brace period of the Ponseti method. The results may improve counselling, aid brace compliance and therefore, optimize the Ponseti treatment.

Methods

Responses on the TNO-AZL Preschool Children's Health Related Quality of Life (TAPQOL) obtained during regular consultation within the brace period and first consultation after the brace period were compared with scores of healthy

children in a similar age range.[7] During consultation clubfoot patients are examined extensively by an orthopedic surgeon specialized in clubfoot treatment. The Clubfoot Assessment Protocol is administrated to provide an overall profile of the clubfoot child's functional status within the domains mobility, muscle function, morphology and motion quality [8,9]. An indication of brace compliance is obtained by asking the parents how much the brace is worn and by inspecting the brace itself for traces of use. Based on this information the orthopedic surgeon gives a classification for brace compliance [good, fair or poor]. This study was approved by the local ethics committees (Niet-WMO 2014-69/METC number 2014-67).

TAPQOL

The TAPQOL was developed to measure parents' perception of HRQoL in children aged between one and five years.[7] The questionnaire is designed primarily for research purposes focusing mainly on data aggregated at a group level. The TAPQOL can be used to evaluate the impact of disease by comparing a clinical sample with a reference sample obtained in a general Dutch population.[7,10] Following the original design, the TAPQOL was administrated via e-mail and filled in by one of the parents[7] after consultation by the orthopedic surgeon. The TAPQOL is a validated multidimensional questionnaire with 43 items covering 12 subscales (Table 4.1).[7] The subscales motor function, social function and communication are only applicable for children older than 1.5 years.[7] The subscales comprise four domains of HRQoL, namely physical, social, emotional and cognitive functioning (Table 4.1). For each subscale a score is calculated and transformed into a 0–100 scale. A higher score indicates a better HRQoL.[7]

Participants

Clubfoot patients

For the current analyses, uni- and bilateral idiopathic clubfoot patients who were initially treated with the Ponseti method were included. Children were at least one year of age, within the brace period of the treatment or in their first year of regular follow up. Patients where treatment commenced within the first six months after birth, and with parents who consented to participate and were reachable via e-mail were eligible for inclusion. Patients included in the analyses only used the standard foot abduction brace with a connecting bar between the

two shoes. Patients who wore the ADM-brace (Abduction Dorsiflexion Mechanism brace, C-Pro Direct, Great Britain) without such a specific connecting bar were excluded from the current study. Furthermore, syndromic and positional clubfoot patients, adopted children and patients of whom parents did not have sufficient knowledge of the Dutch language were excluded from the current analyses. Data was collected between December 2014 and October 2019.

Table 4.1 - Domains and subscales of the TAPQOL. Adapted from Fekkes et al., 2000.

DOMAIN	**Physical functioning**	**Social functioning**	**Emotional functioning**	**Cognitive functioning**
SUBSCALES	Stomach problems Skin problems Lung problems Sleeping Appetite Motor functioning	Problem behavior Social functioning	Positive mood Anxiety Liveliness	Communication

Reference sample

The reference data of the TAPQOL is available through the Netherlands Organization for Applied Scientific Research (TNO) and can be online downloaded via https://www.tno.nl/media/5004/vragenlijsten_01032012.pdf. This dataset was collected in a general population of children who visited a child health care center in the Netherlands as part of their periodical checkup.[7] For the current analyses children of whom the parents reported a chronic illness were excluded. Furthermore, age was restricted between 12 and 60 months.

Analyses

Differences between clubfoot patients and the healthy reference group on the TAPQOL were compared by means of independent sample *t*-test. In case of non-normal distributed data, a the Mann Whitney *U* test was used. If multiple questionnaires from different phases of the brace period were available, only the first questionnaire was included for the primary analyses.

Furthermore, in order to explore the influence of brace wearing during the Ponseti method on the quality of life, differences on the TAPQOL between clubfeet patients and the reference group were compared within separate age categories. For the latter analysis, we also included a group of patients within

the first year after the end of the brace period (aged between 4 and 5 years). In addition, we compared a group of clubfoot patient within the final year of the brace period (aged between 3 and 4 years) with a group of patients within the first year after the end of the brace period (aged between 4 and 5 years).

Analyses were performed with Statistical Package for the Social Sciences Version 25.0 (SPSS Inc., Chicago, IL, USA). A two-tailed *P* value <0.05 was considered to be statistically significant. A Bonferroni corrected p value of <0.0125 was considered to be statistically significant when comparing the subscales of the TAPQOL.

Results

Participants

Clubfoot patients

Within the study period, 119 clubfoot patients with parents who consented to participate in the data collection visited our clinic within the brace period or the first year of regular follow up of the Ponseti treatment. Of these patients, 21 patients were excluded from the analyses because of non-idiopathic clubfeet (n=12), usage of the ADM-brace prior to the assessment (n=6), adoption (n=1) or insufficient knowledge of the Dutch language by the parents (n=2). Parents of 80 of the potential 98 patients responded (82%) to invitations to complete the TAPQOL. In 8 cases, only data acquired during the first year after the brace period was available for analyses. In 34 cases multiple TAPQOL questionnaires were acquired during the study period. Only the first questionnaire was used for the primary analysis. The additional questionnaires were included in the analyses comparing the different age groups.

Reference sample

Furthermore, data of 238 healthy controls from the reference sample provided by TNO[7] could be included in the current study (Table 4.2). Eleven of these healthy controls were due to their age (4–5 years) only included in the sub-analyses comparing patients and controls in the year after the bracing had ended.

Table 4.2 - Demographic characteristics of participants included in the primary analysis.

		Clubfoot patients (n=66)	Reference group (n=227)
Age	(median [range] months)	29 [14-47]	29 [12-48]
Gender	(% male)	74.2%	54.2%
Affected side	(% bilateral)	40.9%	-
	(% unilateral left)	28.8%	-
	(% unilateral right)	30.3%	-
Relapse	(% yes)*	13.6%	-

*8 patients received a re-APT prior to assessment, 1 patient was scheduled for a re-APT one month after assessment.

Quality of life within brace period

On average both healthy controls and clubfoot patients scored high on the 4 domains of the TAPQOL. Lowest HRQoL was seen on the sub-scales for problem behavior and sleep; with the highest scores on positive mood and liveliness (Figure 4.1). Clubfoot patients scored significantly lower on the subscales for motor functioning, sleeping, lung problems and skin problems (P<0.0125). The difference between group means on the four domains were respectively 4.4, 11.6, 4.3 and 4.5 points.

Figure 4.1 - Overall mean scores on the 12 subscales of the TAPQOL. *Represents significant difference between groups (<0.0125).

There were no differences between patients with uni- or bilateral clubfoot or those treated following relapse in any of the TAPQOL subscales. Responses provided by the parents of clubfoot patients on the individual items of the TAPQOL subscales for motor functioning, sleeping, lung problems and skin problems are presented in Figure 4.2.

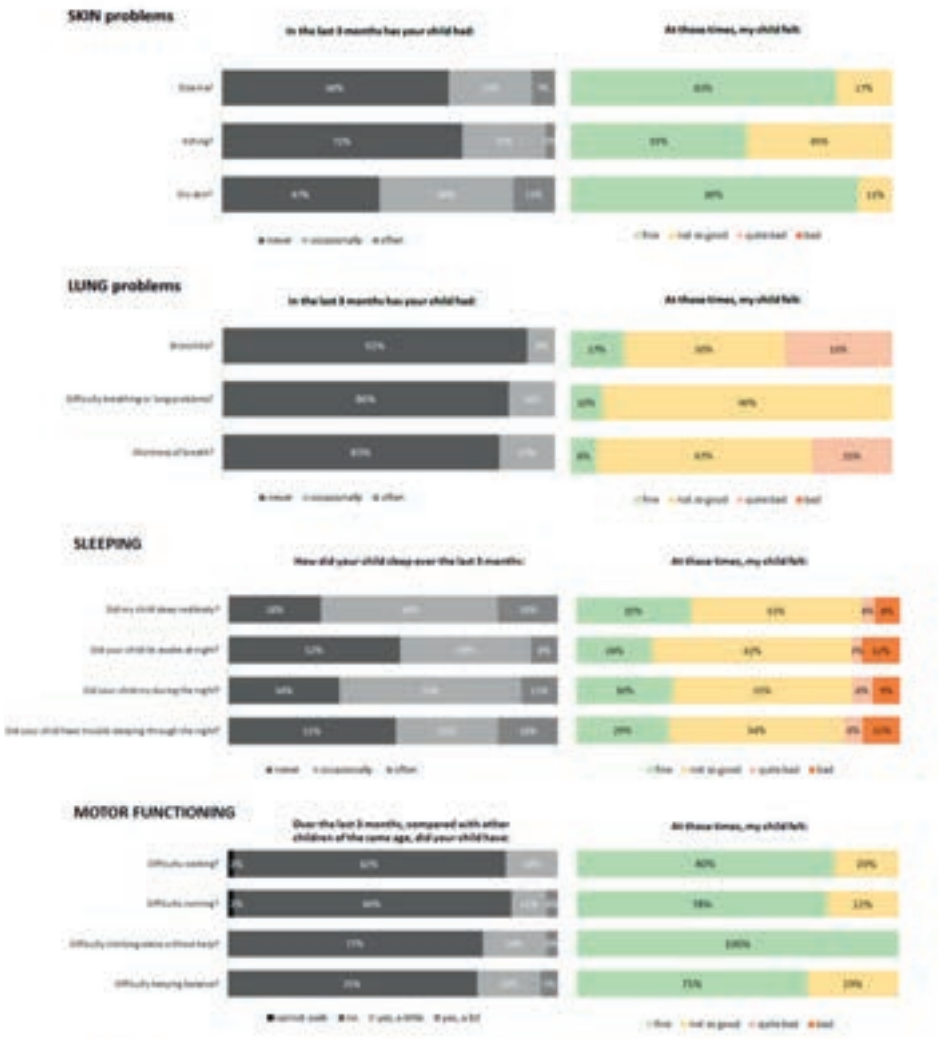

Figure 4.2 - Responses of the parents of clubfoot patients on the items of the TAPQOL subscales with significant differences. In case parents report problems on an item, also the degree to which a child shows negative emotions to such problems are addressed in the TAPQOL.

Within the subscale skin, problems with dry skin were reported the most often. In case of problems, patients showed no or only mild negative emotions to these problems. This in contrary to the subscale lung problems. Where only a small portion of the parents reported problems, the majority of these patients however showed a stronger negative emotion. Often parents reported problems with sleeping, with moderate to strong negative emotions of the clubfoot patients to these sleeping problems. Consistently approximately 20% of the parents of clubfoot patients reported problems on the four items comprising the subscale motor functioning (Figure 4.2).

Quality of life for different age groups

When comparing the clubfoot patients with the healthy controls within different age categories, the subscales sleeping and motor functioning showed significant differences. For the other TAPQOL subscales no difference between clubfoot patients and healthy controls were found in any of the age categories. During the first year of bracing, clubfoot patients (aged between 1 and 2 years) scored lower on the subscale sleep (Table 4.3). While clubfoot patients scored lower on the subscale motor function during the later phases of the bracing period (2–3 and 3–4 years of age). During the first year after the brace period ended (patients 4–5 years of age) no difference between the clubfoot patients and healthy controls were found (Table 4.3).

Table 4.3 - Mean scores of the TAPQOL subscales with significant differences presented per age group. *Represents significant difference between groups (*P*<0.0125).

Group (n)		***Skin problems***	***Lung problems***	***Sleeping***	***Motor functioning***
		Mean (SD)	*Mean (SD)*	*Mean (SD)*	*Mean (SD)*
Total group	***1-4 year***ˆ	*P=0.009**	*P=0.008**	*P=0.004**	*P<0.001**
	Clubfoot (72)	*89 (13)*	*93 (14)*	*71 (27)*	*94 (9)*
	Healthy (227)	*93 (10)*	*98 (8)*	*83 (17)*	*98 (4)*
Brace period	***1-2 year***ˆ	*P=0.21*	*P=0.09*	*P=0.002**	*P=0.08*
	Clubfoot (27)	*89 (13)*	*93 (14)*	*60 (31)*	*93 (9)*
	Healthy (80)	*94 (8)*	*97 (9)*	*80 (17)*	*96 (6)*
	2-3 year	*P=0.07*	*P=0.05*	*P=0.05*	*P<0.001**
	Clubfoot (39)	*88 (15)*	*92 (16)*	*75 (23)*	*93 (13)*
	Healthy (81)	*93 (10)*	*97 (8)*	*84 (18)*	*99 (4)*
	3-4 year	*P=0.02*	*P=0.15*	*P=0.28*	*P<0.001**
	Clubfoot (35)	*87 (12)*	*95 (13)*	*78 (24)*	*94 (9)*
	Healthy (66)	*92 (11)*	*98 (6)*	*85 (15)*	*99 (3)*
Follow–up	***4-5 year***	*P=0.38*	*P=0.69*	*P=0.83*	*P=0.13*
	Clubfoot (23)	*86 (13)*	*91 (17)*	*82 (20)*	*95 (7)*
	Healthy (11)	*87 (15)*	*95 (12)*	*85 (15)*	*99 (2)*

ˆTAPQOL sub scale motor functioning only contains items for children aged ≥1,5 years. As a result these results are based on resp. 57 and 13 clubfoot patients and 227 and 48 healthy controls.

Comparing clubfoot patients within the final year of the brace period (aged between 3 and 4 years) with patients within the first year after the end of the brace period (aged between 4 and 5 years) showed no significant differences (Figure 4.3).

Figure 4.3 - Scores on the 12 subscales of the TAPQOL for patients within the final year of bracing (3–4 year old) and patients during the first year after bracing ended (4–5 year old).

Discussion

Evaluation of our dataset of Dutch clubfoot patients showed an overall good HRQoL during the brace period of Ponseti treatment. However, compared to healthy controls, clubfoot patients showed a statistically significant decrease in scores on the TAPQOL related to sleeping, motor functioning, lung and skin problems. Despite there was no significant difference between clubfoot patients within the final year of the brace period and patients within the first year after the end of the brace period, HRQoL on all subscales seems to reach normal levels in clubfoot children after the brace period has finished.

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Problems with interrupted sleep and challenges related to sleeping behavior are frequently reported by parents of healthy children under the age of 5 years.[11] In our cohort the youngest clubfoot children (aged between 12 and 24 months) showed more problems related to sleeping compared to age matched healthy controls. This particular subscale of the TAPQOL measures sleeping problems related to crying at night and sleeping through the night.[7] A key component of the foot abduction brace is a bar that connects both feet of the child.[12,13] Children are limited in their movements whilst in the brace and sleeping on their side is difficult. Berger and colleagues listed sleep disturbances as the main reason for non-compliance with the foot abduction brace.[14] Parents often report difficulties with sleeping at the start of the brace phase during consultation. In clubfoot support groups it is often mentioned that it is important to incorporate bracing into the bedtime routine to minimize these problems. In doing so the brace becomes a habitual element of sleeping.

In the overall comparison between the control group and the clubfoot patients, patients scored lower on the lung subscale. This difference was not seen when comparing the different age groups but only in the overall group. It is difficult to understand why clubfoot patients would score lower on this subscale. There are no indications both in literature as from our clinical experience that clubfoot patients would experience more often lung problems compared to other children nor that they would respond differently in case of lung problems. It should however be noted that all children with chronic illnesses (e.g. asthma, epilepsy, rheumatism, heart conditions) were excluded from the control group in the current analysis. If these children were included in the analyses, clubfoot patients did not score different on the HRQoL related to lung problems. Respiratory tract infections are very common in the infant population.[15]

Clubfoot patients scored statistically significantly lower in the skin domain of the TAPQOL than healthy controls. This domain refers to general skin problems such as eczema and dry skin.[11] From clinical practice, it is known that several local skin problems, such as mycosis, eczema and blistering, can occur during the brace wearing phase of the Ponseti method. Besides the material from which the shoe of the brace is made, a suboptimal position of the foot in the brace can cause skin problems and negatively affect the corrective mechanism of the brace. Blisters should be a sign for the treating physician to check the position of the foot in the brace and be aware of possible under correction.[12,13] It should be

notated that the differences in HRQoL on the subscale skin were only visible when comparing the overall groups.

Idiopathic clubfoot patients show minimal delay in gross motor function compared to healthy controls. Most milestones have a delay of 1.5-months, while independent ambulation is delayed by approximately two months.[6,16] In the current cohort a difference on the TAPQOL subscale for motor function between clubfoot children and healthy controls was found. This difference was seen in the overall analyses as well as in the analyses comparing children between the age of 2-3 and 3-4 years old. It seems to normalize in the year after the bracing had ended. Compared to the healthy controls in the similar age range relatively less 4-5 year old clubfoot patients scored lower on the subscale motor function than 3-4 year old patients. This subscale of the TAPQOL addresses if children have problems with walking, running, stair climbing and balance.[7] Many parents express their concern whether the Ponseti method will negatively affect the motor development of their child, which is most notable by the achievement of independent walking.[16] The brace positions the affected feet of the clubfoot patients in a subtle overcorrection of 70^0 of abduction in order to prevent relapse of the clubfoot.[17] As a result, in the beginning, some children show out-toeing when start walking. For some parents, this is a reason to consider ending bracing. It is therefore extremely important for the treating physician to explain that it is a harmless, temporary side effect of bracing and to stress the importance of the brace in preventing relapse.[5] Muscle imbalance[18] and soft tissue abnormalities[19] are known problems in clubfoot patients and likely to contribute to delayed motor development. Our results also seem to suggest that after the bracing period, clubfoot patients reach normal levels on the TAPQOL subscale motor functioning and other subscales. Other studies showed development of gross motor skills at the age of 5 and athletic abilities to be comparable with age matched peers at elementary school.[20,21] Some clubfoot patients however, experience gait impairments and limited motor function in later life.[22]

In order to further investigate the effect of the brace on HRQoL of clubfoot patient a longitudinal study design monitoring the HRQoL of the same clubfoot patients during and after the brace period would be interesting. In addition, including patients at the early stage of the brace period, i.e. in the first year of life, would aid the understanding of clubfoot patients' responses to the brace. It is however challenging to measure HRQoL in very young children.[7] Furthermore,

HRQoL might differ on the subscales of the TAPQOL as children grow older and develop physically, mentally and verbally. A comparison with a healthy control group in a similar age range is therefore in the authors' opinion necessary.

Potential limitations of the study must be taken into account when interpreting the results. The TAPQOL instrument was not developed specifically for clubfoot children but as a general tool to measure HRQoL.[7] The different domains are however also relevant for clubfoot children but the TAPQOL lacks the specificity to identify problems to specific aspects of the clubfoot. For example, it is unknown if the problems reflected in the subscale skin problems were localized around the foot. Moreover, one could wonder if the relatively small differences found between the control and clubfoot group are of clinical relevance. The minimal clinical important difference (MCID) for the TAPQOL is only available for children with Cerebral Palsy on the TAPQOL.[23] This study described that caregivers already perceive a relevant change in HRQoL of their children at a relatively low treatment efficacy. Although this MCID was established in children with cerebral palsy, this MCID gives some guidance of the relevance of the differences between clubfoot patients and the healthy reference sample in the current study. The mean differences on the subscales lung, skin, sleep and motor functioning that showed to statistically differ between clubfoot patients and controls all exceeded the MCID established for the domain physical function that covers these subscales.[7,23] Furthermore, the subgroup used to determine differences between patients and the reference group in the age range when patient do not wear the brace anymore was relatively small. As a consequence, these results should be interpreted with caution. Considering the high percentage of patient with good brace compliance (93%) in this cohort, it remains the question if our results apply for all clubfoot patients. In clubfoot literature a wide range of brace compliance and methods to assess this compliance have been reported.[24,25] Overall, Dutch clubfoot patients have high scores concerning their health-related quality of life. However, compared to healthy controls, clubfoot patients showed problems related to sleep, lung, skin and motor functioning during the brace phase of the Ponseti method. These problems seem to resolve in the year bracing had ended. The results may be used to counsel parents. Physicians will hopefully be able to alleviate some concerns that parents have about the bracing period. In combination with the physicians' emphasis on the important positive effect of the brace on preventing relapses[3], this may lead to a better understanding by the parents of clubfoot patients on the role of the brace which subsequently may improve compliance.

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CHAPTER 5

Participation and motor abilities in children aged 5 to 9 years with idiopathic clubfeet after treatment with the Ponseti method

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Abstract

Background
Functioning in children consists of different aspects, including their ability to execute activities and participate in life situations. Several studies on children with clubfeet showed limited motor abilities and walking capacity compared with healthy control children, while other studies showed comparable athletic abilities and gross motor development. Although participation in activities of daily life plays an important role in the development of children, this has not yet been investigated in children with clubfeet. The study aims to determine the level of parents' perceived motor ability and participation in Ponseti-treated children with clubfeet compared with age-matched healthy controls.

Methods
Parents of children aged 5 to 9 years with and without idiopathic Ponseti-treated clubfeet were asked to complete an online questionnaire about their child's motor abilities and participation level using the Dutch version of the Assessment of Life Habits for Children (LIFE-H) version 3 to assess participation and the Dutch Movement Assessment Battery for Children-2 Checklist (MABC-2 Checklist) to assess motor abilities. Statistical analysis focused on differences between groups and the relationship between motor abilities and level of participation.

Results
Questionnaires of 86 children with clubfeet (mean age 7.1, 73% boys) and 62 controls (age 6.7, 53% boys) were analyzed. Despite a large variation, results showed no significant differences between groups on the total scores of the LIFE-H and the MABC-2 Checklist. Children with clubfeet, however, scored lower on Mobility and better on the categories Communication and Responsibility of the LIFE-H. Furthermore, children with clubfeet showed lower scores on the MABC-2 Checklist subscale "movement in a static and/or predictable environment." High levels of the parents' perceived participation correlate with good results, as perceived by the parents, in motor ability.

Conclusions
Although differences on some aspects of motor ability and participation existed, children with clubfeet in general showed high levels of parents' perceived motor ability and participation. High levels of participation correlated with good results in motor ability. Level of Evidence: Level II.

Introduction

Idiopathic clubfoot or talipes equinovarus is a well-known deformity of the lower extremity in children. The disorder is characterized by a 3-dimensional foot deformity with 4 components: cavus, equinus, varus, and adductus.[1,2] If left untreated, a clubfoot will lead to deformity, functional disability, and pain. The Ponseti method is nowadays the recommended primary treatment in clubfeet.[2]

Despite the large body of literature available on good initial outcomes of the Ponseti method,[3] far less is known about the functional long-term outcomes of this treatment. Thorough analyses of the gait pattern by means of 3-dimensional gait analysis showed several impaired gait characteristics in children with clubfeet treated with the Ponseti method compared with healthy age-matched controls, such as significantly lower ankle plantarflexor moment and ankle power.[4-6]

More recent studies focused on motor abilities of children with clubfeet in daily life. Some studies showed no differences in gross motor development as well as athletic performance in elementary school sports between children with clubfeet and controls.[7,8] However, other research showed gross motor deficits, increased risk of lower motor ability, as well as asymmetries in heel walking, 1 leg stand and 1 leg hop, and lower walking capacity in children with clubfeet.[9-12]

According to the International Classification of Functioning, Disability and Health for Children and Youth (ICF-CY),[13] there is a relationship between function, motor abilities, and participation in children. This suggests that the above described physical impairments of children with clubfeet might lead to limitations in activities and therefore impede a child's participation in life.[13,14]

Therefore, the aim of this study is to determine the level of perceived participation, as indicated by the parents, in Ponseti-treated children with clubfeet compared with age-matched healthy controls and to investigate if this level of participation is associated with motor ability.

Methods

Data within this explorative study was prospectively gathered between September 2020 and July 2021 through a digital survey. Parents of children with clubfeet and healthy controls aged 5 to 9 years old were asked to fill out an online questionnaire about their child's motor abilities and participation level. The Medical Ethical Committee Máxima Medisch Centrum declared that this study was not subjected to the Medical Research Involving Human Subjects Acts (WMO) (N20.057). All parents had given informed consent.

Participants

Children with clubfeet were recruited through 11 dedicated clubfoot treatment centers located throughout the Netherlands. All children with clubfeet aged between 5 and 9 years old were screened by the attending physician based on their medical files, after which parents of potential participants were approached by letter. In addition, a call was placed on the Facebook page of the Dutch Parents' Association for children with clubfoot (Stichting Klompvoet Nederland). Children with clubfeet were included based on unilateral or bilateral idiopathic clubfeet, which were primarily treated with the Ponseti method. They were excluded if treatment started 6 or more months after birth. Parents of controls were recruited among elementary schools and through the personal network of the research team. In addition, parents of children with clubfeet were asked if there were siblings without a clubfoot that could be included. Controls were excluded when they had health issues that may affect physical function and participation (e.g., fractures). Inclusion criteria for both groups were boys and girls between the ages of 5 and 9 years old, and exclusion criteria were obesity, underlying syndromes, neurological diseases, and insufficient Dutch language skills (parents). Obesity was set as an exclusion criterion to be able to compare children with clubfeet and healthy controls without additional "movement challenges," such as movement impairments caused by excess weight.

Questionnaires

Besides questions regarding demographics and clinical background of their child, parents filled in the Dutch version of the Assessment of Life Habits for Children (LIFE-H) questionnaire version 3, short version,[15] and the Dutch translation of the Movement Assessment Battery for Children-2 Checklist (MABC-2 Checklist).[16] These questionnaires have been developed and validated

to screen children either for social participation (LIFE-H) or for movement difficulties (MABC-2 Checklist) regardless of the type of underlying impairment.

Participation

The short version of the LIFE-H consists of 64 items, covering 12 life habits categories. In the current study, the categories Community Life and Employment were excluded, as the items in these categories are not relevant for the 5 to 9 age group. Table 5.1 shows the categories, number of items, and a short description of each category of the LIFE-H used in this study. The individual item scores range from 0 (total impairment) to 9 (optimal participation), developed by the combination of 2 concepts of the scale: degree of difficulty and required assistance. A transformation of scores on a 0 to 10 scale is used to give similar weighting to each category of life habits. The lower the score, the poorer the child's participation. In addition, the parent's perceived level of satisfaction about the child's execution for each item is measured using a 5-point-scale ranging from 1 (very unsatisfied) to 5 (very satisfied).[15]

Table 5.1 - Description of used categories of the LIFE-H questionnaire.

Category	Nr of items	Description
Nutrition	4	choosing, preparing and eating food
Fitness	4	sleeping, participation in movement activities for physical fitness or relaxation
Personal care	8	personal hygiene, use of the bathroom, putting on clothes
Communication	8	reading, writing, communicating with adults and young people, and the use of a phone
Housing	6	helping with chores in and around the house, moving in and around the house
Mobility	4	moving outside, cycling
Responsibility	7	money, respect for others and taking responsibility for themselves
Interpersonal relationships	6	keeping relationships with parents and family members, maintaining friendships
Education	6	participating in school en extracurricular activities
Recreation	8	participating in recreational activities (e.g. sports, music, arts)

Based on Noreau et al. 2007[15]

Motor abilities

The MABC-2 Checklist contains motor and non- motor parts. The motor part contains 30 items divided into 2 sections. Section A measures movement in a static and/or predictable environment, where the child is in control of how it moves without pressure of time. Section B measures movement in a dynamic

and/or unpredictable environment, where the child has to react to a moving object (e.g., a ball) and has to move itself within a moving environment (eg, running with other children). For every item on the checklist, parents had to rate the motor competence of their child on a 4-point scale (0=very well; 3=not close). All individual items of the checklist can be found in Figures 5.2A and 5.2B. The total motor score (TMS) is the sum of the 30-item scores. The higher the TMS, the poorer the child's performance. In addition, the TMS is transformed into percentiles, where children with scores at or above the 95th percentile are highly likely to have a motor impairment in daily life. Children with scores between the 85th and 95th percentile were at risk, and children with scores up to the 85th percentile had no detectable motor impairment.[16] The nonmotor part, Section C, concentrates on nonmotor factors that might affect the child's movement skills.[17]

Data-analysis

Differences in participation and motor abilities between children with clubfeet and healthy controls were assessed using the nonparametric Mann-Whitney U test for independent-samples. In addition, differences between children with unilateral and bilateral clubfeet, and between 5 to 6 versus 7 to 9 years old in the clubfoot group were analyzed. Effect size for nonparametric data using r=z/N was calculated for significant differences, with z being the sum of the ranks, and N being the number of cases.[18] The effect size was interpreted as 0.5 for a large effect, 0.3 for a medium effect, and 0.1 for a small effect.[19] A $\gamma 2$ test was used to explore associations between the groups and the transformed TMS percentiles. Correlations between accomplishment and satisfaction of participation, and motor abilities and participation were examined using Spearman rank-order correlation. Correlation coefficients of >70 were considered as a strong correlation, 0.40 to 0.69 as moderate, and <0.40 as weak.[20] Statistical analyses were performed with IBM SPSS Statistics 25. The level of significance was set at $P<0.05$.

Results

A total of 260 questionnaires were sent out to parents who were interested in participating. Considering inclusion/exclusion criteria and incomplete questionnaires, a total of 148 participants were included for further analysis (Figure 5.1). In the majority of these cases the entire set of questionnaires was

filled in completely (LIFE-H and MABC-2 Checklist, n=118). In addition, in some cases only the MABC-2 Checklist (n=17) or the LIFE-H (n=13) questionnaire could be used for data-analysis.

Table 5.2 shows the demographics of the study population. Significantly more boys were present in the clubfoot group versus the control group (P = 0.012). However, participation and motor abilities in the entire population did not differ between boys and girls. Additional surgical treatment reported by parents within the clubfoot group (34%) were posteromedial release, tibialis anterior transfer, anterior distal tibial epiphysiodesis, and lengthening of the Achilles tendon.

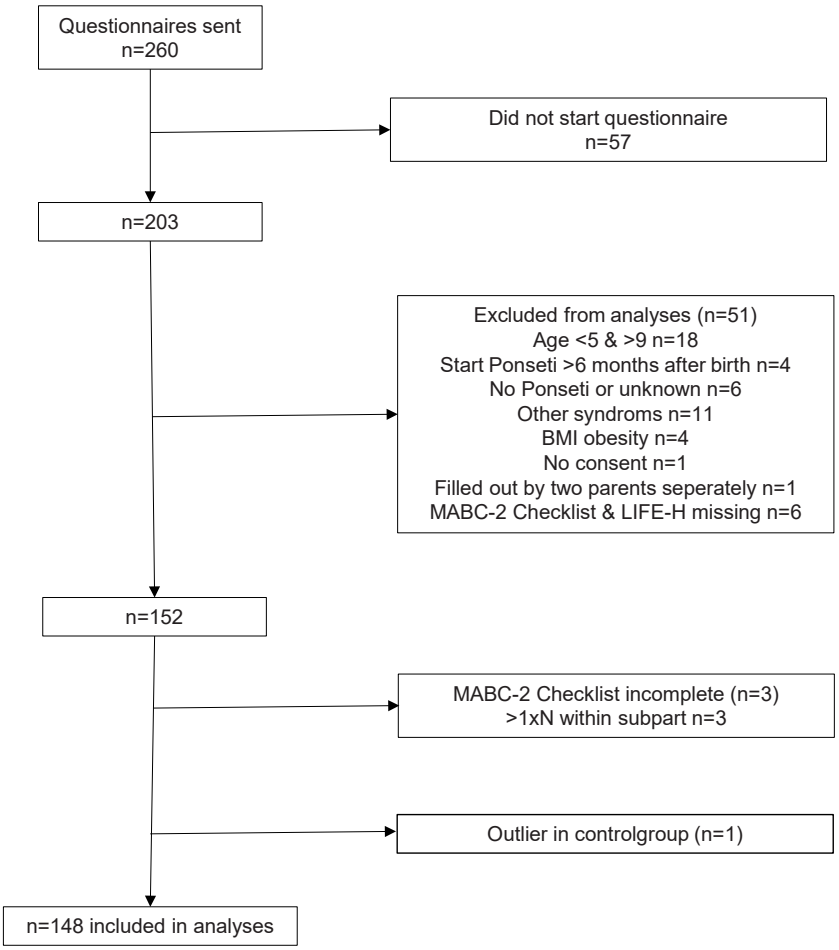

Figure 5.1 - Flowchart inclusion participants. BMI indicates body mass index; LIFE-H, Assessment of Life Habits for Children; MABC- 2 Checklist, Movement Assessment Battery for Children-2 Checklist.

Participation

In general, children with clubfeet and controls scored high on accomplishment and satisfaction scales of the LIFE-H for the total and separate categories (Table 5.3). Significant differences (all $P<0.05$, with small effect sizes all r<0.3) between groups on accomplishment were found in communication, mobility and responsibility categories. Children with clubfeet scored significantly lower on mobility, but significantly better on communication and responsibility. Within the communication category, questions were asked about reading, writing, communicating with adults and peers, and the use of a phone. The responsibility category consisted of questions regarding money, respect for others and taking responsibility for themselves. In regard to satisfaction with execution, the children with clubfeet scored significantly lower on Education ($P<0.05$, with a small effect r=0.179). No significant differences were found between children with unilateral and bilateral clubfeet concerning parents' perceived accomplishment and satisfaction. When examining the 3 age groups, the 5 to 6-year-old children with clubfeet scored significantly worse than 7 to 9-year old on personal care for accomplishment as well as satisfaction ($P<0.05$, with small effect sizes both r<0.3).

Table 5.2 - Demographics of the study population. Significantly more boys were present in the clubfoot group versus the control group ($P=0.012$). However, participation and motor abilities in the entire population did not differ between boys and girls. Additional surgical treatment reported by parents within the clubfoot group (34%) were posteromedial release, tibialis anterior transfer, anterior distal tibial epiphysiodesis, and lengthening of the Achilles tendon.

Total	**Controls n=62**		**Children with clubfeet n=86**	
Gender [n] (boys/girls)	**33/29**		**63/23**	
Age [mean (SD)] (y)	6.7	(1.2)	7.1	(1.4)
BMI [mean (SD)]*	15.4	(1.8)	15.5	(1.9)
Unilateral [n (%)]			44 (51%)	
Unilateral left [n (%)]			*27 (61%)*	
Number of casts				
Less than 3 (n)			1**	
3 to 5 (n)			*9*	
5 to 7 (n)			*32*	
More than 7 (n)			*39*	
Unknown (n)			*5*	
Achilles tendon tenotomy [n (%)]			75 (86%)	
Additional surgical treatment [n (%)]			29 (34%)	
Within Ponseti protocol			*14*	
*Outside Ponseti protocol****			*15*	
Age at surgery [mean (SD)] (y)			*5.4 (2.4)*	

Significant difference $P<0.05$ presented in bold. * Results for BMI could not be calculated for 4 participants (2 controls and 2 children with clubfeet) due to missing data concerning the height or weight of the child; ** Reported by parents, but patient did receive Achilles tendon tenotomy and brace period; *** Ponseti protocol defined as manipulation, casting, Achilles tenotomy and bracing.

A moderate, significant, positive correlation between parents' perceived accomplishment and satisfaction was found within the entire population (rs=0.480, *P*<0.0001), as well as in the clubfoot group (rs=0.506, *P*<0.0001) and control group (rs=0.479, *P*<0.0001).

Table 5.3 - LIFE-H scores for parents' perceived participation in terms of accomplishment and satisfaction in the control and clubfoot group.

	Accomplishment (0-10)					**Satisfaction (0-5)**				
	Controls (n=53)		**Children with clubfeet (n=78)**		***P***	**Controls (n=53)**		**Children with clubfeet (n=78)**		***P***
	Median	**(IQR)**	**Median**	**(IQR)**		**Median**	**(IQR)**	**Median**	**(IQR)**	
Nutrition	8.9	(7.8-10)	9.3	(7.8-10)	.119	4.5	(4.0-5.0)	4.4	(3.8-5.0)	.536
Physical fitness and social well-being	10.0	(9.2-10)	10.0	(8.9-10)	.694	4.5	(4.3-5.0)	4.8	(4.3-5.0)	.599
Personal care	8.7	(7.8-10)	8.9	(7.8-10)	.877	4.6	(4.3-4.9)	4.5	(4.0-5.0)	.388
Communication	**8.4**	**(7.7-9.9)**	**9.5**	**(8.3-10)**	**.007**	4.5	(4.1-4.9)	4.6	(4.0-5.0)	.296
Housing	10.0	(8.5-10)	10.0	(8.5-10)	.980	4.7	(4.4-5.0)	4.8	(4.3-5.0)	.824
Mobility	**10.0**	**(9.4-10)**	**10.0**	**(8.3-10)**	**.022**	4.8	(4.5-5.0)	4.8	(4.2-5.0)	.186
Responsibility*	**7.4**	**(6.4-9)**	**8.8**	**(6.5-10)**	**.021**	4.3	(4-4.5.0)	4.3	(4.0-4.9)	.130
Interpersonal relationships**	10.0	(9.1-10)	10.0	(9.3-10)	.372	4.8	(4.3-5.0)	4.8	(4.2-5.0)	.781
Education	10.0	(8.9-10)	9.6	(8.4-10)	.256	**4.8**	**(4.3-5.0)**	**4.6**	**(4.0-5.0)**	**.041**
Recreation	10.0	(8.6-10)	9.6	(8.7-10)	.168	4.8	(4.4-5.0)	4.8	(4.0-5.0)	.427
Total	*8.9*	*(8.4-9.6)*	*9.2*	*(8.3-9.8)*	*.249*	*4.6*	*(4.3-4.8)*	*4.5*	*(4.1-4.8)*	*.312*

* n=76 in clubfoot group; ** n=77 in clubfoot group; IQR=interquartile range. Significant difference P<.05 presented in bold

Motor abilities

A significant difference (*P*<0.0001, with a moderate effect r=0.312) in motor abilities was found between groups in Section A of the MABC-2 Checklist, where the children with clubfeet scored lower (Table 5.4). On individual items of Section A, children with clubfeet appeared to have more difficulty with maintaining balance whilst standing to pull on articles of clothing, jumping whilst keeping two feet together on take-off and landing, and hopping on either foot (Figure 5.2A). Section B (Figure 6.2B), the TMS and Section C showed no significant differences between groups. No significant differences were found between children with unilateral and bilateral clubfeet. When examining the 2 age groups within the clubfoot group, a significant difference was found

concerning part B—movement in a dynamic and/or unpredictable environment (*P*<0.05, with a small effect r<0.3) and the TMS (*P*<0.05, with a small effect r<0.3). Scores were worse in the younger group.

When transforming the TMS into percentiles, 86.4% of the control group and 71.1% of children with clubfeet have no detectable perceived motor impairment in daily life (Figure 5.3). Both controls and children with clubfeet were found in the "at risk for" motor impairment zone as well as 'highly likely' to have motor impairment zone. Although the percentages of children with clubfeet are higher in both zones, no statistical association was found between having a clubfoot and motor impairments (*P*=0.068).

Table 5.4 - MABC-2 checklist scores for parents' perceived motor abilities in the control and clubfoot group.

	Controls (n=59)	**Children with clubfeet (n=76)**	
	Median (IQR)	Median (IQR)	*P*
Section A*: movement in a static and/or predictable environment (0-45)	0 (0-2)	1 (0-3.75)	**<.0001**
Section B*: movement in a dynamic and/or unpredictable environment (0-45)	3 (0-6)	2 (0-6)	.567
TMS: total motor score (A+B) (0-90)	3 (0-7)	3 (1-9)	.317
Section C: non-motor factors that might affect movement skills (0-13)	2 (0-4)	2 (0.25-5)	.717

* Figure 5.2 visualizes the items of Section A and B; IQR = interquartile range; Significant difference P<0.05 presented in bold.

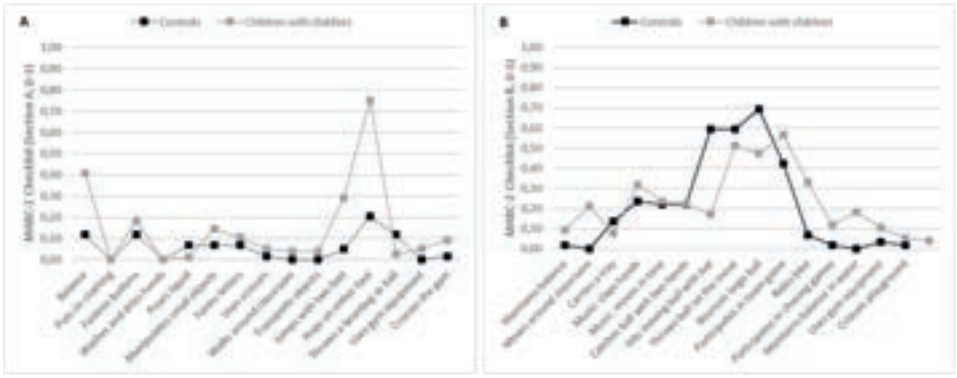

Figure 5.2 - A, Mean MABC-2 Checklist scores (Section A, static/predictable environment) of the control and clubfoot group per item. B, Mean MABC-2 Checklist scores (Section B, dynamic/unpredictable environment) of the control and clubfoot group per item.

Relationship participation and motor abilities

In the complete study population, a weak but significant negative correlation (rs=-0.362, *P*<0.0001) was found between the total scores, where higher scores of parents' perceived participation are related with a lower TMS on motor abilities. The relationship was stronger in the clubfoot group (rs=-0.446, *P*<0.0001).

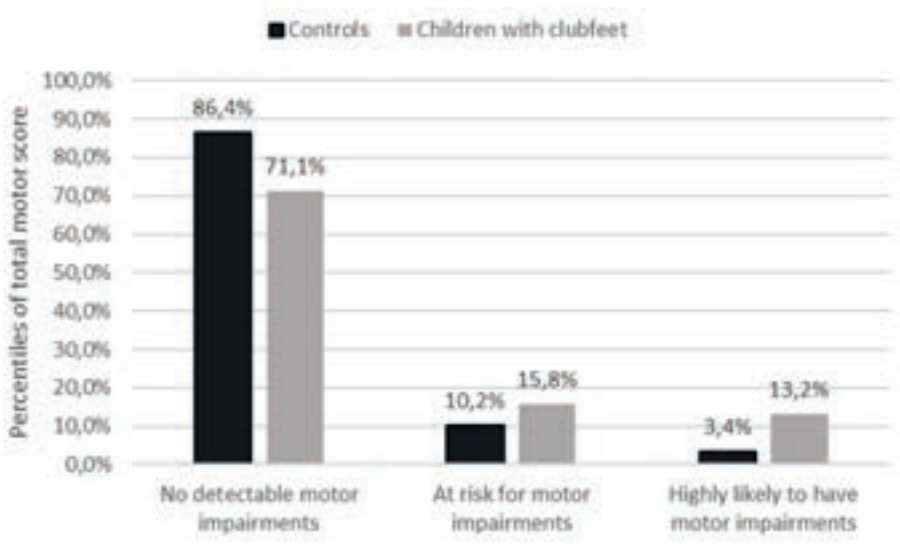

Figure 5.3 - Total motor scores of the MABC-2 Checklist trans- formed into percentiles for the control and clubfoot group. MABC-2 Checklist indicates Movement Assessment Battery for Children-2 Checklist.

Discussion and conclusion

The aim of this study was to compare parental perceived levels of participation and motor abilities in Ponseti-treated children with clubfeet with age-matched healthy controls. Furthermore, the association between participation and motor

abilities was investigated. Our results showed that children with clubfeet have high levels of motor ability and participation which are overall comparable to age-matched controls. However, children with clubfeet scored significantly lower on accomplishment of mobility, but higher on communication and responsibility. Motor ability was found to be significantly lower in children with clubfeet in a static and predictable environment, and a trend was observed that children with clubfeet show a higher risk for deviating motor abilities. Furthermore, a significant correlation was found between participation and motor ability, indicating that better motor abilities are associated with higher levels of participation.

Participation in daily life plays an important role in the development of a child's social relationships and social skills, in long-term mental and physical health.[14,21] It is therefore not surprising that the participation-aspect of the ICF-CY is increasingly considered when assessing the outcome of children with disabilities.[13,14] To our knowledge, there is no previous literature on participation in daily life of children with clubfeet aged 5 to 9 years old. Findings in previous literature on motor abilities in similar patient populations vary. Some studies observed no differences in gross motor development and athletic performance,[7,8] others showed gross motor deficits and increased risk of low motor ability in children with clubfeet.[9,11,12] These studies used quantitative measures scored by health care professionals, instead of parent-reported measures to judge motor abilities. Measures scored by health care professionals provide a momentary snapshot of the child, whereas parents can assess their child's participation and motor abilities throughout daily life. The questionnaires used in our study, the LIFE-H and the MABC-2 Checklist, were specifically designed to measure parents' perceived participation and motor abilities of children.[15,16]

Although our research shows high levels of perceived motor ability and participation in children with clubfeet, this does not mean that all children with clubfeet are doing well. The large variation in scores of the clubfoot group on both the LIFE-H and MABC-2 Checklist suggest that some patients do experience problems. This might be because of a relapse, which is a well-known challenge within clubfoot care. This recurrent deformity of the clubfoot requires further treatment.[22] However, because of poor definitions and diverse duration of follow-up, reported percentages of relapse after initial Ponseti treatment vary tremendously in literature.[22,23] Very little literature comparing relapse and non-

relapse patients is available. Relapse patients show multiple structural impairments which contribute to gait issues at the ankle and knee.[24,25] Recent studies show lower clinical status in relapse patients compared with non-relapse patients,[26] but after relapse treatment, comparable physical activity was found.[27] In the current clubfoot group, 34% of the parents reported additional surgical interventions. However, because of the diversity of terminology used to describe interventions, we decided not to use this information to further analyze the data in respect to possible relapse. Information from patients' medical charts would have ensured more accurate treatment data in comparison to the information the parents have provided. In our opinion, only medical history provided by the treating physician can be reliably used to define relapse, additional treatment and good functional outcome.

A limitation in the current project was the inability to verify clinical background information of the children with clubfeet provided by the parents because of privacy issues. Patients were treated in multiple clubfoot centers and access to the patients' medical charts was because of the General Order Protection of Personal Data not possible. Some descriptive parameters might therefore be incomplete. For instance, 11 parents reported that no Achilles tendon tenotomy was performed, an unusually high percentage (13%) for Ponseti treatment. Another child with clubfeet was reported to have had less than three casts, an unusually low number. However, this patient was selected for participation by the treating physician, and parents did report an Achilles tendon tenotomy and subsequent brace period. In addition, in this study the severity of the clubfoot was not structurally classified according to the Pirani score or Dimeglio score.[28] As a result, no analysis could be performed related to the severity of the clubfoot. Furthermore, the LIFE-H questionnaire may lack sensitivity because of substantial ceiling effects and may require refinement for use in children with disabilities.[29] As an alternative to assess participation in children, the Participation and Environment Measure- Children and Youth (PEM-CY) questionnaire might be considered for future research. It would also be interesting to investigate the child's own perspective on its motor abilities and participation levels. The presence of pain could then also be included because pain can affect participation and motor abilities. Finally, the recruitment of controls amongst siblings of children with clubfeet, might have been cause for some potential bias as those parents may be either more or less critical of certain abilities compared with parents who have never seen a child with a clubfoot. However, the number of siblings in the control group that participated

was limited. Also, to our knowledge, this is the first study we know of that studied parents' perceived participation in children with clubfeet. This research is in accordance with the ICF-CY, which states that functioning in children is affected by body structures and function, activities, and participation.[13] The addition of including the data on parents' perceived satisfaction leads to a better interpretation of the results.[30]

Overall, the results of this study show that children with clubfeet aged 5 to 9 years have high levels of parents' perceived motor ability and participation, which are similar to age-matched controls. High levels of participation correlate with good results on motor ability, but on an individual level each child must be assessed to define possible problems. More research is needed to study the potential association between more severe forms of clubfoot on participation and motor function (eg, high clubfoot scores at diagnosis, relapse clubfoot, etc.). In conclusion, the Ponseti method leads to overall high levels of participation and motor abilities in children aged 5 to 9 years.

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CHAPTER 6

Identification and treatment of residual and relapsed idiopathic clubfoot in 88 children

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Abstract

Background and purpose

The Ponseti treatment is successful in idiopathic clubfoot. However, approximately 11–48% of all clubfeet maintain residual deformities or relapse. Early treatment, which possibly reduces the necessity for additional surgery, requires early identification of these problematic clubfeet. We identify deformities of residual/relapsed clubfeet and the treatments applied to tackle these deformities in a large tertiary clubfoot treatment center.

Patients and methods

Retrospective chart review of patients who visited our clinic between 2012 and 2015 focused on demographics, deformities of the residual/ relapsed clubfoot, and applied treatment. Residual deformities were defined as deformities that were never fully corrected and needed additional treatment. We defined relapse as any deformity of the clubfoot reoccurring, after initial successful treatment, with necessity for additional treatment.

Results

We identified 33 patients with residual and 55 patients with relapsed clubfeet. In both groups decreased dorsal flexion and adduction were the most often registered deformities. Furthermore, often equinus/decreased dorsiflexion, active supination, and varus occurred. In more than half, typical profiles of combined deformities were found. Relapses occurred at all stages of treatment and follow - up; half of the residual or relapsed clubfeet were identified before the end of the bracing period. In half of the patients, additional treatment consisted of the Ponseti treatment, one – quarter also required adaptation of the brace protocol, and one – quarter needed additional surgery. The Ponseti treatment was mainly reapplied if feet presented with relapses or residues until the age of 5.

Interpretation

Practitioners should especially be aware of equinus/decreased dorsiflexion, adduction, and active supination as a sign of a residual or relapsed clubfoot. Due to the heterogeneous profiles of these clubfeet, treatment strategy should be based on a step-by step approach including recasting, bracing, and if necessary surgical intervention.

Introduction

The Ponseti treatment has shown to be a very successful treatment for idiopathic clubfoot.[1,2] Unfortunately, approximately 11–48% of treated clubfeet show problems during follow up.[2-4] Some feet do not fully correct, while others have a tendency to relapse.[3] In the literature, the distinction between these problematic clubfeet is not always clear.[4] In the current study we define residual deformities as deformities that underwent primary treatment but were never fully corrected and need additional treatment.[5] A relapse is defined as any feature of the clubfoot reoccurring after initial successful treatment, which needs additional treatment.[6]

The pathology of residual or relapsed clubfeet is still unknown. It is clear that inappropriate bracing leads to relapses.[2,7] But non-compliance with the bracing protocol does not explain all relapses. Any deformity of the initial clubfoot can be present in a residual or relapsed clubfoot. Furthermore, toe deformities, stiffness, or articular incongruence might be present.[8,9]

The treatment of residual and relapsed clubfeet involves many challenges that were, in the past, frequently tackled by means of extensive surgical interventions.[5] Nowadays, opinions have shifted and the additional treatment takes on a more reserved or nonoperative approach. A step-by-step approach is important and based on the original Ponseti treatment, involves repeated casting, a proper bracing period, and only if necessary surgical intervention.[5,10,11] The Ponseti treatment includes the possibility to expand the foot correction with a tibialis anterior tendon transfer to redress the common problem of adduction and supination in residual and relapsed clubfeet.[5,9] Furthermore, treatment should be specific to the pathoanatomy of the deformity and functional needs of the patients should be taken into account during treatment planning.[5] Applied to residual, relapsed, neglected, and complex clubfoot, the Ponseti treatment has shown positive results with respect to pain, functionality, and cosmesis.[5,10,12,13] Early identification of residues and relapses allows for early treatment and therefore may diminish the necessity for major surgical interventions.[3,10] However, due to the variable manner in which a residual or relapsed clubfoot may occur, they may be difficult to identify at an early stage. Therefore, the aim of this study is to gain insight into the deformities of residual and relapsed clubfeet and the applied treatment at our clubfoot treatment center. Being aware

which deformities occur most frequently and at what stage of the treatment aids in determining the optimal treatment and timing of this.

Patients and methods

We performed a retrospective dossier study of clubfoot patients treated by one orthopedic surgeon specialized in clubfoot pathology (ATB) between 2012 and 2015. Potential study participants were identified by means of the Dutch diagnosis and treatment code (DBC) for clubfoot. Patients were included in the study if they had idiopathic clubfoot and underwent treatment for a residual or relapsed deformity of their clubfoot at our tertiary institute. Patients were excluded if they did not have idiopathic clubfoot but rather a syndromic, neurogenic, or positional clubfoot.

Data on demographics, clubfoot deformities, the primary treatment, and additional treatment were gathered from the electronic patient files. Known clubfoot deformities were recorded: adduction, equinus, varus, and cavus. Since decreased dorsiflexion and active supination are early signs of relapsing clubfeet and as such require treatment to prevent further problems, we documented these as well. Active supination is caused by suboptimal function of the tibialis anterior (TA) muscle. In residual or relapsed deformities, the shape of the foot results in relative medialization of the insertion of the TA. This leads to an over-supination in the early swing phase but also by landing on the lateral border of the foot. This mechanism can be studied during walking but also when sitting on a bench with the lower legs free. If the patient is asked to raise the foot, it can clearly be seen if the foot is dorsiflexed neutrally or with a supination component. Because equinus and decreased dorsiflexion are strictly related, and both mark a deformity in the sagittal plane of the ankle, we decided to combine the equinus and decreased dorsiflexion (EqDD) and treated them as a single entity.

The data on the deformities were gathered retrospectively and were cumulatively gathered per individual. Because of the retrospective character of this study, the moment at which specific deformities occurred could not be determined. Given these facts, the deformities in a single patient formed a profile of the deformities that occurred over time.

Primary treatment was defined as the initial treatment of any kind that was performed with the intention to fully correct the primary clubfoot. Treatment for residual or relapsed clubfoot that had been performed outside our own clinic was also recorded. Additional treatment comprised the treatment applied for a residual or relapsed clubfoot at our center. For the different treatment stages, we registered the date of the initial corrective casts, usage of braces, and any surgical treatment.

We composed 3 treatment groups that differ from each other in extensiveness of the additional treatment necessary to treat the residual or relapsed clubfoot. In the first group (extended Ponseti protocol) additional treatment consisted of additional treatment following the Ponseti protocol. This could entail a second casting phase, renewed Achilles tendon tenotomy (re-ATT), and/or a tibialis anterior tendon transfer (TATT) (Ponseti 2002). In the second group (brace adaptation), the treatment protocol of the Ponseti group was combined with adaptation of the bracing phase. In this group bracing was prolonged according to age or adjusted by the use of another type of brace. In those—often older—children who did not tolerate the standard foot abduction brace with a bar, an abduction dorsiflexion mechanism brace was used as an alternative. This brace consists of an alternative abduction, dorsiflexion mechanism. It is constructed without a bar between both feet and therefore is usable unilaterally. The third group (additive surgery) entailed patients who received additional extra- and/or intra-articular surgical treatment that is not part of the aforementioned extended Ponseti protocol.

Ethics, funding, and potential conflicts of interest

Ethics approval was obtained from the local medical ethical committee (niet-WMO 2016-23). No funding was obtained for this study. No conflicts of interest declared.

Results

Patient selection

Initially, we identified 416 patients by means of the diagnosis/treatment code (DBC) for clubfoot. First, 103 patients were excluded from the database because these consisted of mothers expecting a child with clubfoot who were counselled

before delivery by the orthopedic surgeon or patients who had been labelled incorrectly as clubfoot patients, leaving 313 clubfoot patients. Ultimately, 88 patients with 122 residual or relapsed clubfeet were identified and included in the following analyses (Figure 6.1).

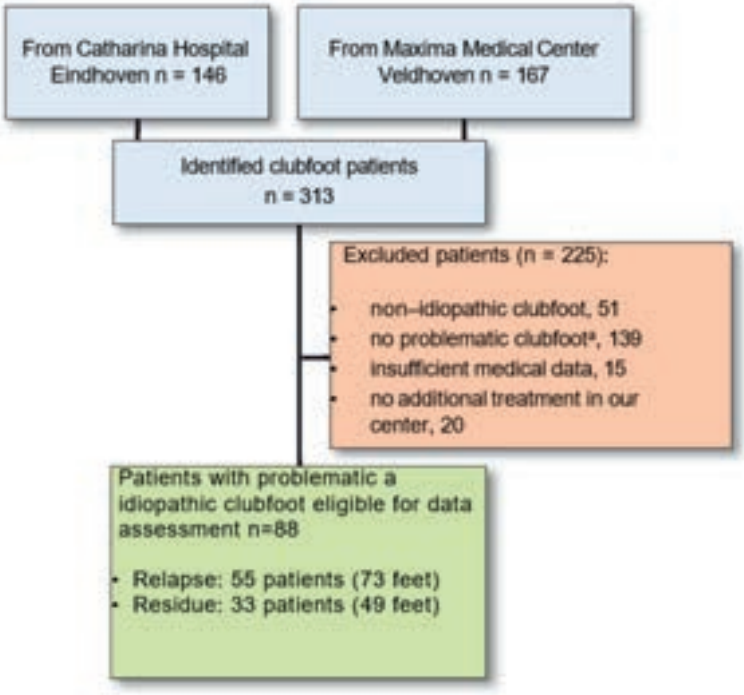

Figure 6.1 - Flowchart of patient selection. [a] Residual and relapsed clubfeet.

Patient characteristics

The residual group comprised 33 patients (49 feet) and 55 patients (73 feet) had relapsed deformities. A substantial part (52/88) of the population was referred to our clinic from other centers. This explains the relatively high percentage of residual and relapsed feet in our population. As expected, the mean age at identification of the relapse was considerably higher than the age at identification of residual deformities (respectively 4.9 (0–15) years and 3.4 (0–16) years). The mean follow-up since identification of the residual and relapsed club- foot was similar in both groups; in the residual group this was 3.8 (0.4–10) years, and in the relapse group 3.7 (0.3–21) years.(Table 6.1)

Table 6.1 - Descriptive data on both groups for patients and feet.

	Residual		***Relapse***
Patients, n	33		*55*
Age at identification in months			
mean (range)	41	(3–187)	*59 (3–182)*
Follow up since identification in months			
mean (range)	45	(5–120)	*45 (3–250)*
Male/female ratio (n)	4.5/1.0	(27/6)	*2.0/1.0 (36/19)*
Unilateral:bilateral ratio (n)	1.1/1.0	(17/16)	*1.1/1.0 (29/26)*
Feet, n	49		*73*
Pirani at start of primary treatment			
median (IQR) [a]	5.8	(4.8–6.0)	*5.5 (4.5–6.0)*
Ponseti treatment used in primary treatment, n			
yes	26		*49*
no	19		*16*
missing	4		*8*
Number of casts used in Ponseti			
median (IQR) [b]	8.0	(6.5–15.0)	*5.0 (4.0–6.0)*
Deformity, n			
equinus	20		*12*
decreased dorsiflexion	22		*35*
adduction	20		*34*
active supination	15		*21*
cavus	2		*3*
varus	13		*13*
Group of additional treatment, n			
Ponseti protocol	32		*27*
brace adaptation	8		*23*
additional surgery	*9*		*23*

[a] Data only available of 22/10 feet; [b] Data only available of 37/17 feet.

The male-to-female ratio in the residual group was 4.5:1 and in the relapse group 2:1. Unilateral and bilateral clubfoot occurred as frequently in the relapse group, as it did in the residual group (1.1:1.0). It should be noted that not all patients with bilateral clubfeet in the relapse group had a bilateral relapse. In the relapse group 8 patients with a bilateral clubfoot had a unilateral relapse.

Often Pirani scores and number of casts are used to give an idea of the primary severity of the clubfoot. Unfortunately, data on these two particular variables, especially of referred patients, were not complete (Table 6.1). Based on the available data, clubfeet in the residual group had a Pirani score at the start of the primary treatment of 5.8 with an interquartile range (IQR) of 4.8–6.0. For the relapse group the median Pirani score was 5.5 with an IQR of 4.5–6.0.

In the residual group the number of primarily treated patients according to the Ponseti method was 26 of 49 patients and a median of 8.0 casts were used (IQR=6.5–15) (Table 6.1). In the relapse group 49 of the 73 patients were treated with the Ponseti method as primary treatment and a median number of 5.0 casts was used to achieve initial correction (IQR=4.0–6.0).

Deformities (Table 6.2)

Table 6.1 shows the occurrence of the different deformities. Portraying these deformities as single entities does shed some light on the deformities that appear in residual and relapsed clubfeet, but it does not grasp the full complexity of the clubfeet. In 26 of the residual clubfeet and 33 of the relapses, a profile with multiple deformities evolved over time. In total, 13 profiles with multiple deformities were identified in addition to 4 deformities occurring as a single entity (Table 6.2).

EqDD occurred in 30 of the residual clubfeet and 42 of the relapsed clubfeet. In the majority of the profiles, EqDD played a role. Furthermore, EqDD is the most prevalent single deformity that occurs in residual and relapsed clubfoot. Adduction as a single entity, however, occurs in a rather large proportion in the relapse group as well. Adduction was present only once as a single entity in the residual group. Profiles in which active supination and adduction played a role were abundant as well (28 in the residue group and 41 in the relapse group). Varus was always combined with other deformities (Table 6.2).

As a means of examining the relation between profiles of deformities and the age at which the residual and relapsed clubfeet were initially identified, we plotted these variables against each other (Figure 6.2). As stated in the methods section, it should be noted that not all feet had sufficient data available to determine the age at identification (20/122 missing).

Table 6.2 - Proportions of profiles of deformities in the relapse and residue group. Values are number of feet.

Profile	Residual	*Relapse*
Single deformity	17	*31*
EqDD [a]	14	*22*
active supination	1	*2*
adduction	1	*7*
cavus	1	*0*
EqDD involved	16	*20*
EqDD + active supination	2	*2*
EqDD + adduction	5	*4*
EqDD + varus	0	*1*
EqDD + active supination + varus	1	*0*
EqDD + active supination + adduction	2	*7*
EqDD + adduction + cavus	0	*1*
EqDD + varus + adduction	4	*3*
EqDD + active supination + varus + adduction	2	*2*
Other combinations	10	*13*
active supination + adduction	3	*4*
active supination + varus	4	*3*
active supination + varus + adduction	0	*1*
varus + adduction	2	*3*
adduction + cavus	*1*	*2*

[a] EqDD = equinus/decreased dorsiflexion

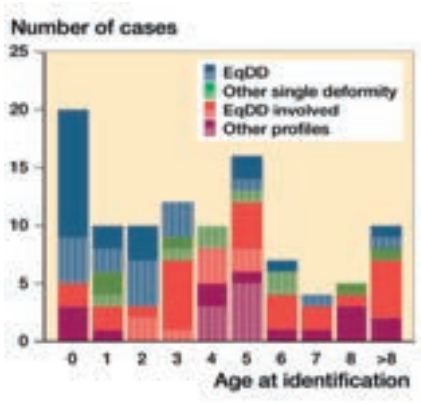

Figure 6.2 - Age at identification of the residual and relapsed clubfoot compared to the group of profiles of deformities in the clubfoot. Blue bars show the feet with solitary equinus and/or decreased dorsiflexion and green bars are those with other solitary deformities. Red bars indicate feet with combined profiles that contain EqDD and purple bars show feet with other combined profiles. Plain tones show patients that were referred to our clinic. (N=number of cases).

An important proportion of the solitary EqDD deformity (27 of 36) occurs in the first year of life (Figure 6.3). When patients become older the number of deformities occurring as a single entity diminishes. In our cohort 50% of the residual and relapsed clubfeet were identified before the end of the bracing period (before the age of 4). Furthermore, a peak is seen at the age of 5, 1 year after the end of the bracing period. After this peak, the amount of new residual and relapsed clubfeet decreases even further. Additionally, the proportion of clubfeet where single deformities play a role seems to decrease with age and these are seldom seen at the age of 6 and older. Combined deformities are detected more often as residual or relapsed deformities at a higher age. Figure 6.3 also distinguishes our own (striped) from referred patients (plain color). In the first two years of life patients are often referred with clubfeet that display EqDD. Referred patients often demonstrate a more complex profile as they are diagnosed later in life.

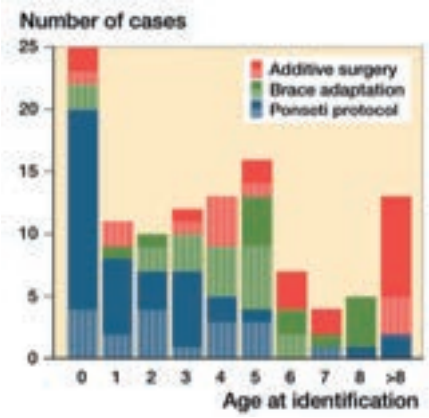

Figure 6.3 - Age at identification of residual and relapsed clubfoot compared with their treatment group. Blue bars represent patients that had satisfying results with the Ponseti protocol. For patients displayed by green bars the Ponseti protocol was not sufficient and adaptation to a bracing protocol was needed to get good results. The red group contains those patients in which the previous 2 treatment options did not suffice and additive surgery was needed. Plain toned bars contain patients that were referred to our clinic. (N=number of cases).

Treatment groups

Three treatment groups were composed (see Methods section). The extended Ponseti protocol was sufficient in 32 out of 49 of the residue group and 27 out of the 73 clubfeet in the relapse group. An extra brace adaptation was sufficient in 8 of the residual clubfeet and in 9 of these feet additional surgery was needed (Table 6.1). In the relapse group 23 of the feet were treated with brace adaptation and 23 of the patients needed additional surgical interventions that were not a part of the Ponseti protocol.

The Ponseti protocol was mainly used when feet presented with residues or relapses until the age of 5 (Figure 6.2). Brace adaptations increase up to the age of 6, but by the age of 9 brace adaptation was not used in any of the cases. The additional surgery group was not influenced by age.

Surgical interventions

In the 3 previously depicted treatment groups, ATT and TATT were considered part of the extended Ponseti protocol, whereas any other surgery was not part of the extended Ponseti protocol and was classified as additional surgery.

Surgical treatment mostly consisted of extra articular procedures, of which re-ATT was applied 67 times out of 135 surgeries. Until the age of 4, re-ATT was almost exclusively the surgical treatment of choice. After that, TATT was used more often as well as other extra articular procedures (Figure 6.4).

The additional surgeries which are not part of the Ponseti protocol (see Figure 6.4, in green and red) were mostly pre-served for older children in whom other treatment did not prove to be effective. We subdivided these additional surgeries into extra-articular treatments (green) and intra-articular treatments (red). The partial epiphysiodesis (PED) of the anterior segment of the distal end of the tibia was the most predominantly used extra-articular treatment that was not part of the Ponseti protocol. Intra-articular surgeries (see Figure 6.4, in red) were used in older children as well and consisted of posteromedial releases, closing wedge osteotomies, a triple arthrodesis, a fascia plantaris release, and the use of external fixators (in medial column lengthening and metatarsal osteotomies).

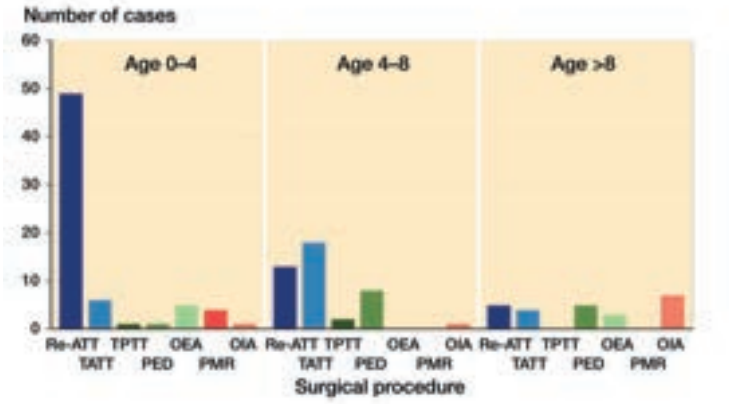

Figure 6.4 - Age of patients at the moment of surgical intervention. Blue color marks the surgical treatments that are part of the Ponseti protocol. Green portrays extra-articular (EA) treatments that are not part of the Ponseti protocol. The red bars show the intra-articular (IA) treatments. Re-ATT: renewed Achilles tendon tenotomy, TATT: tibialis anterior tendon transfer, TPTT: tibialis posterior tendon transfer, PED: partial epiphysiodesis of the ventral distal tibia, OEA: other extra-articular surgery, PMR: posteromedial release, OIA: other intra-articular surgery.

Discussion

We describe a cohort of residual and relapsed clubfeet treated at a tertiary clubfoot treatment center. By identifying the profiles of deformities occurring in these problematic clubfeet and describing the treatment performed we depict the strength of the Ponseti treatment used in these patients but also show the necessity for additional surgical interventions in some patients.

Our population showed high resemblance to the normal clubfoot population in terms of affected foot and sex.[14] The majority of the included patients had already initially been treated with the Ponseti method, nowadays the preferred initial treatment for idiopathic clubfeet in the Netherlands.[15] The residual group showed a high number of casts used (median 8 casts), while the initial Pirani score was not higher. This suggests that these clubfeet already showed difficulties during the initial correction. Zhao et al.[16] also showed that difficulty in correcting the initial deformity was predictive for a relapse. Furthermore, Ponseti et al.[17] noted a specific group of complex clubfeet that are difficult to treat and require a modified Ponseti treatment. All known clubfoot deformities could be present in residual and relapsed clubfeet. As was pointed out by Ponseti in 2002,[3] equinus/decreased dorsiflexion is the most frequent reoccurring

deformity, as we found as well. We also found many different profiles of deformities, though deformities in children younger than 4 years old often occurred solitarily.

The majority of relapses occur in the first 2 to 3 years of life and rarely after the age of 5.[10,17] However, we also saw an increase in cases at the end of the bracing period. This difference might be related to ours being a tertiary clubfoot treatment center. As a consequence, patients may be older at presentation than they would be in other centers. However, the peak of residuals and relapses around the age of 5 might also point toward the important role of the foot adduction brace in preventing relapses. It should, however, be noted that difficulties during bracing often are a sign of a residual or relapsed foot causing a fitting problem as a result of decreased dorsiflexion.[10] In our study, however, in children over the age of 5 relapses often occurred in more complex deformity profiles.

Deformities, age, and treatment are all associated with each other, as age and deformities determine the suitable treatment. Treatment using the Ponseti protocol is particularly prevalent in the first 4 years of life, while brace adaptations and additional surgery are commoner in older children; we found the same in our cohort. This is in line with the step-by-step approach described by Radler and Mindler and Jowett et al.[5,11] Early identification seems to be essential in preventing the need for additional surgical interventions, which have been associated with less positive outcomes in pain, functionality, and cosmesis.[5] In our retrospective cohort no objective scores on pain, functionality, and cosmesis were available.

Of course, the retrospective nature of our study also imposes several other limitations. The most important of these are incomplete data and difficulty distinguishing between residual and relapsed clubfeet, especially in referred cases. We defined a relapse in line with the Iowa group. Radler and Mindler[5] suggest that the differentiation has minimal effect on further treatment. Perhaps a classification of residual and relapsed clubfeet based on severity might be more useful. However, it is difficult to compare the severity of the different profiles of deformities. Bhaskar and Patni[18] identified 5 relapse patterns based on the involvement of dorsiflexion, adduction, or supination and whether the deformity was either dynamic or fixed. As we found many profiles of deformities, regularly also including cavus and varus, we felt that the Bhaskar classification

was not capable of embracing the complexity of both residual and relapsed clubfeet. It should be noted that as we are a tertiary center, we generally treat the more severe cases and therefore the profiles of deformities might be more complex than in a standard clubfoot population.

In summary, our study showed that relapses occur at all stages of treatment and follow-up. All deformities of the initial clubfeet can (re)occur in residual and relapsed clubfeet and often a combination of deformities is seen. Practitioners should especially be aware of EqDD, adduction, and active supination as signs of a relapse. Due to the heterogeneous nature of residual and relapsed clubfeet, the treatment strategy should be based on a step-by-step approach including recasting, bracing, and if necessary surgical intervention. In the majority of our cases, especially if identified in an early stage, treatment according to Ponseti was sufficient to treat the residual and relapsed clubfeet. Identifying residues or relapses at an early stage could prevent the need for additional surgery.

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CHAPTER 7

Anterior distal tibia hemi-epiphysiodesis in relapsed Ponseti treated clubfoot patients: beware of the rebound phenomenon!

Rebound in a positive way
Torino 2025, FISU World University Games. Skier Tobias Besselaar

A.T. Besselaar, I. van Tilburg, M.C. van der Steen

Abstract

Purpose

Anterior hemi-epiphysiodesis of the distal tibia (AHDT) with an 8-plate has been proposed as treatment for clubfoot patients with recurrent equinus deformity. However, to date, clinical results have been inconsistent and particularly focused on correction during treatment. In the current project, we specifically added evaluation of outcomes at least one year after 8-plate removal.

Methods

We included Ponseti treated idiopathic clubfoot patients aged between 4 and 12 years who underwent AHDT between 2015 and 2022. Patients were assessed preoperatively at indication for 8-plate removal and if available at least one year after 8-plate removal. A senior orthopaedic surgeon specialized in clubfoot treatment determined passive ankle dorsiflexion. Furthermore, on standardized lateral radiographs, the anterior distal tibial angle (ADTA) was measured.

Results

The median treatment duration of 46 feet (32 patients, 38% female) was 22.1 months (95%-CI: 19.3-24.9) months. Both ankle dorsiflexion and ADTA improved significantly during AHDT. Of 35 feet at least 1 year follow up after removal of hardware was available. The ADTA showed significant increase ($p<0.001$) after 8-plate removal, by modelling to preoperative values. The measured ankle dorsiflexion one year after 8-plate removal showed no change with the pre-treatment situation. One patient undergoing bilateral AHDT experienced a wound infection and screw migration.

Conclusions

AHDT using an 8-plate, is an effective method to achieve slope correction. However, after 8-plate removal, rebound of ADTA occurs. Physicians, parents, and patients should be informed about this mechanism which might result in different timing of the treatment and possibly the need for renewed AHDT.

Introduction

Treatment of idiopathic clubfoot can be complicated by a relapse tendency.[1] The Ponseti method is nowadays the preferred treatment for clubfoot with good to excellent results.[1-3] However, percentages of relapses after Ponseti treatment in literature vary significantly between 1.9 and 67%.[4,5] These relapse deformities include hindfoot varus, midfoot adduction, cavus and impaired dorsiflexion.[6,7] The most common relapse deformity is impaired ankle dorsiflexion, possibly responsible for the pain and functional limitation relapse patients encounter.[6,7] Patients experiencing a relapse show deviations in their gait pattern and score lower on both patient reported and clinical outcome measures.[8-10] The decreased ankle dorsiflexion may be the result of a flat top talus, joint stiffness, soft tissue imbalance or posterior tightness.[11,12]

Previously reported relapse treatment strategies to influence impaired dorsiflexion vary from repeated serial casting or Achilles tenotomy to extensive soft tissue release or corrective osteotomies.[11,13] These intra-articular surgical interventions are however associated with an increased risk of perioperative complications and prolonged recovery.[13] Another, less applied technique uses guided growth by means of a temporary anterior hemi-epiphysiodesis of the distal tibia (AHDT) with a small 8-plate. Due to this minimally invasive surgical procedure the growth plate is partially inhibited through a tension band principle, resulting in a planned axis deviation.[14] As a consequence a slight recurvatum of the distal tibia occurs, theoretically leading to a foot position in more dorsiflexion.[14-16]

Limited literature is available on the results of AHDT with an 8-plate as relapse treatment for clubfeet patients with recurrent equinus deformity.[15-18] Often these findings were based on small populations of initially surgically treated clubfoot patients. While some studies found both clinical and radiological benefits after hemi-epiphysiodesis in carefully selected groups of clubfoot patients,[15-17] the effects of epiphysiodesis have not been unequivocally reported in literature. In one study of 31 clubfeet treated with epiphysiodesis, no clinically significant improvement was mentioned.[18] No or only few minor related complications were reported.[15-18] However, follow-up over a longer period is lacking. As a result, possible decline of recurvatum of the distal tibia after implant removal has scarcely been considered.

The aim of this study was to establish whether AHDT with 8-plate is a safe and effective treatment for sagittal residual deformity in clubfeet after Ponseti treatment. To this end we radiologically and clinically evaluated patients preoperatively, at indication for 8-plate removal and if available at least one year after 8-plate removal.

Materials and methods

This retrospective analysis of a single-centre cohort included data from children who visited the outpatient clinic of the tertiary clubfoot centre at Máxima MC with a relapsed clubfoot and who were treated with AHDT between 2015 and 2022. Approval of the local medical ethical committee was obtained [reference MMC: 2014-67], and legal guardians gave their permission to collect and analyse this data.

Study population

Patients with uni- or bilateral idiopathic clubfoot were included if they were initially treated with the Ponseti method and experienced a relapse for which AHDT was indicated. A hemi-epiphysiodesis was indicated if dorsiflexion -passively tested- was less than 0 degrees. In addition the so-called 'end feeling' had to be rigid as an indication that the Achilles tendon did not interfere with raising the foot. Furthermore, flattened talus seen on X-ray, added to the indication for relapse treatment by means of AHDT. To be included in the study, removal of the 8-plate needed to be between January 2015 and December 2022, with an age not older than 12 years due to the limits of the cohort. Anterior distal tibial angle (ADTA) needed to be measurable on the pre-operative X-ray of the ankle, intra-operatively or within 3 months after implantation before influence of guided growth was seen. Syndromic, neurogenic, or positional clubfoot were excluded. In children with bilateral clubfoot, each foot was considered separately.

Treatment

The procedure of implantation is performed in a supine position under general anaesthesia completed with a popliteal nerve block by a standard anterior approach. One 8-plate (Pro-Motion Medical®, Netherlands) is placed in the anterior centre of the distal tibia on the intact periosteum as advised.[14] Under

image intensification the centre of the tibia in the frontal plane is marked with a smooth Kirschner (K-)wire. Subsequently the leg is rotated to perform a lateral image to place the K-wire in the epiphyseal part of the distal tibia. First the distal screw and plate were mounted, then the proximal screw is placed. The wound is closed in layers. Post-operative regime includes immediate functional and full weight-bearing instructions, no plaster is used. Physiotherapy is prescribed if desirable.

Two weeks after AHDT post-operative wound assessment took place. Guided growth was monitored by means of clinical follow up with X-rays at 3 months and then every 6 months. The indication for removal was based on clinical and radiological grounds. If the children clinically showed a walking pattern with proper heel landing and/ or a radiological posterior coverage with the tibial plafond at the talus or an ADTA change of 15-20 degrees was seen, material was removed.[16,17] Removal of the 8-plate was performed by a similar surgical procedure as implantation.

Radiographic measurements

The effect of AHDT was based on the difference in ADTA measured at the X-ray showing the starting point of treatment and the X-ray made at indication for removal of the 8-plate. In addition, radiographically measured ADTA at least 12 months after implant removal was used to investigate whether the effect of epiphysiodesis was maintained. ADTA was measured on standard weight-bearing lateral X-rays of the ankle (Figure 7.1). The X-ray was defined as measurable if there was an overlap of the medial and lateral dome of the talar trochlea and the distal fibula was superimposed by the tibia.[19,20]

All radiographic measurements were performed by the same researcher (IT). To assess intrarater reliability, a random sample of 19 X-rays were re-evaluated by the researcher. The blinded re-evaluation was executed at least three weeks after initial assessment of the X-ray.

Figure 7.1 - Standard weight-bearing lateral X-rays of the ankle with 8-plate in situ.

Anterior Distal Tibial Angle

The ADTA is the angle between the shaft of the tibia and the tibial plafond.[15,16,21] The ADTA was determined at the intersection of the tibial shaft axis with a straight line through the posterior and anterior border of the tibial plafond, as illustrated in Figure 7.2A. The midpoint of the tibial shaft and consequently the axis of the tibia were determined using two supporting lines. A normal value in non-clubfeet for ADTA is 80 ± 2 degrees.[21,22]

Alpha Angle

The alpha angle is measured as an indication of flattening of the talus. This angle reflects the opening of the angle of the talar dome and is measured in the sagittal plane by means of a Mose ring spanning between the anterior and posterior trochlea (as illustrated in Figure 7.2B).[23] A normal alpha angle in non-clubfeet is 117.9 ± 15.0 degrees.[23] Decrease of the alpha angle indicates more talar flattening, as illustrated in Figure 7.2C/D.

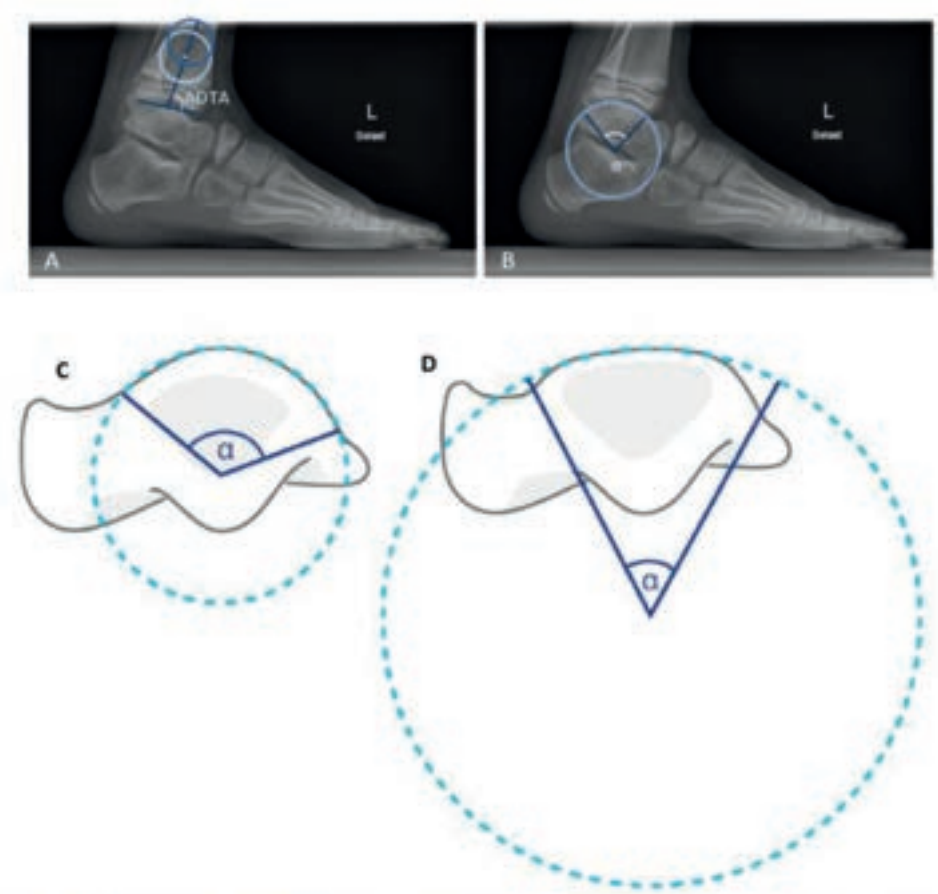

Figure 7.2 - Description of radiographic measurements. 2A: lateral weight-bearing radiograph anterior distal tibial angle (ADTA). 2B: lateral weight-bearing radiograph of the alpha angle. 2C-D: illustration of decrease in alpha angle with talar flattening. 2C: 'normal' shaped talar dome with alpha angle. 2D: flattop talar dome with decreased alpha angle.

Dorsiflexion Measurement

Dorsiflexion with an accuracy of 5 degrees was assessed during outpatient clinic visits by an experienced paediatric orthopaedic surgeon with a specialized in clubfoot pathology (AB), using a goniometer. The measurement was taken from the medial side of the foot with the sole of the foot and the tibia shaft as reference.

Statistical analyses

The interrater reliability of the radiologically measurements was analyzed with an interclass correlation coefficient (ICC). An ICC of <0 was classified as poor, 0<ICC<.2 as slight, .21<ICC<.4 as fair, .41<ICC<.6 as moderate, .61<ICC<.8 as substantial, .81<ICC as almost perfect.[24]

To determine the effect of AHDT by means of the 8-plate implantation, the preoperative and postoperative ADTA and dorsiflexion measurements were compared using the paired t-test and Wilcoxon Signed Rank test respectively. Relations between improvement in ADTA and dorsiflexion measurements were explored by means of Spearman's rho correlation. In the subgroup of patients with an additional evaluation after at least 1 year after 8-plate removal, differences between the 3 time points were assessed by means of a Friedman test. Wilcoxon Signed Rank tests with Bonferroni correction were used as post hoc analyses. The effect of timing on changes in ADTA were explored by means of a visual representation.
Statistical analysis was conducted using "IBM SPPS Statistics" software, version [22]. A p-value <0.05 was considered statistically significant.

Results

Patients

Out of 49 patients under 12 years of age, who underwent AHDT within our tertiary centre for clubfoot deformities, 32 children (46 relapsed clubfeet) were included in this project, as illustrated in the flow-chart in Figure 7.3. Of these patients, 20 (63%) patients had unilateral clubfoot, 38% were female and the median age was 7.5 years at the time of 8-plate implantation. For only two children, it was not possible to determine if they underwent Achilles tendon tenotomy as part of the primary Ponseti treatment, as they were initially treated elsewhere. The remaining 30 children (94%) did receive primarily an Achilles tendon tenotomy. Measured alpha angles indicated that all patients had flattening of the talus. Notably, 21 of the 32 patients (66%) received additional treatment before AHDT was indicated (Table 7.1). Mean treatment duration was 22.1 months (95%-CI: 19.3-24.9). In one patient, there were two minor complications namely superficial wound infection and screw migration. The latter could be accepted without the necessity for treatment adjustments, the infection

was successfully treated with oral antibiotics. There were no neurovascular complications.

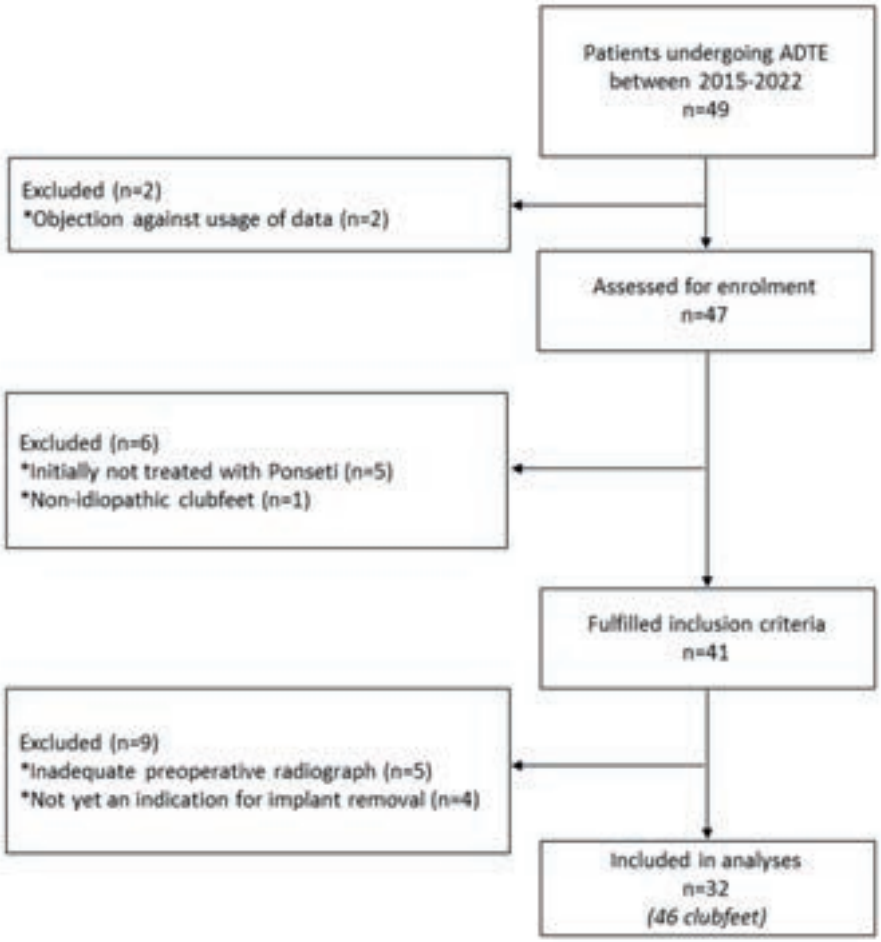

Figure 7.3 - Flow-chart of the study-population.

Table 7.1 – Patient characteristics.

Study no.	Sex	Side	Age at time of Implantation (years old)	Alpha angle (°) On pre-operative radiograph	Additional treatment before 8-plate implantation
1	Male	Right	7	58.4	TATT
2	Female	Bilateral	8	42.4 / 32.0	Bilateral TATT
3	Female	Right	7	49.3	None
4	Male	Left	10	60.2	None
5	Male	Bilateral	6	42.8 / 56.1	None
6	Female	Left	9	28.0	Additional bracing
7	Male	Right	8	59.5	TATT
8	Male	Bilateral	7	82.6 / 73.8	None
9	Male	Bilateral	9	84.5 / 79.4	None
10	Male	Left	8	25.6	TATT
11	Female	Bilateral	8	109.3 / 91.1	Bilateral TATT + PMR
12	Male	Right	11	45.3	None
13a*	Female	Left	5	33.3	PMR
13b*	Female	Left	10	66.6	8-plate (see case 13a)
14	Female	Left	5	15.0	Re-do AT
15	Male	Right	6	16.0	Additional casting
16	Female	Bilateral	7	36.3 / 41.9	Additional casting + bracing
17	Male	Bilateral	7	27.1 / 33.6	Bilateral TATT + re-do AT
18	Male	Bilateral	8	49.7 / 30.0	Bilateral additional casting + TATT
19	Female	Bilateral	8	95.0 / 81.4	None
20	Female	Left	10	43.9	TATT
21	Male	Right	7	53.6	Additional bracing + re-do AT
22	Male	Bilateral	6	37.5 / 55.0	Bilateral TATT
23	Female	Right	7	88.0	Additional bracing
24	Female	Left	11	-	None
25	Male	Right	11	49.1	TATT + twice re-do AT
26	Female	Right	8	80.8	Additional casting + additional bracing
27	Male	Left	6	83.2	None
28	Male	Bilateral	8	75.9 / 70.2	None
29	Male	Right	6	60.1	Additional bracing
30	Male	Bilateral	7	82.8 / 72.0	None
31a°	Female	Left	7	72.9	None
31b°	Female	Right	7	53.0	None
32	Male	Right	9	-	TATT

* Represents 1 patient that had a re-do 8-plate implantation in the same foot. ° Represents 1 patient that has bilateral clubfoot with same 8-plate implantation date, however with a different date of implant removal. PMR = Posteromedial Release; AT = Achilles tenotomy; TATT = tibialis anterior tendon transfer; - = Missing

Additional interventions

Of the 32 included children, 15 children received additional interventions during 8-plate implantation (Table 7.2). These interventions included (re-do) Achilles tendon tenotomy, tibialis anterior tendon transfer and derotation osteotomy. During 8-plate removal, an additional 13 patients underwent addition treatment. In the majority of these cases, a TATT was performed (Table 7.2).

Table 7.2 - Additional interventions during follow-up period.

			Additional interventions during 8-plate implantation		Additional interventions during 8-plate removal	
Study no.	**Clubfoot**	**Time (months) 8-plate in situ**	**Side**	**Additional intervention**	**Side**	**Additional intervention**
1	Right	29.6	None		Right	ATT
2	Bilateral	27.7	None		None	
3	Right	15.8	None		Right	TATT
4	Left	22.2	None		Left	TATT
5	Bilateral	20.1	None		Bilateral	TATT
6	Left	12.8	Left	Derotation osteotomy	Right	Epiphysiodesis foot
7	Right	23.6	None		Right	TATT
8	Bilateral	23.6	Bilateral	TATT	None	
9	Bilateral	29.1	Bilateral	TATT	None	
10	Left	26.6	None		None	
11	Bilateral	24.9	Bilateral	TATT	Bilateral	Epiphysiodesis distal fibula
12	Right	23.6	None		None	
13a*	Left	23.0	Left	TATT	None	
13b*	Left	24.0	Left	ATT	None	
14	Left	17.2	None		Left	TATT
15	Right	18.2	Right	TATT + ATT	None	
16	Bilateral	6.8	Bilateral	TATT	None	
17	Bilateral	21.7	None		None	
18	Bilateral	32.4	None		None	
19	Bilateral	14.0	Left Right	ATT + TAT ATT	Right	Osteotomy Mt1-5
20	Left	29.4	None		None	
21	Right	34.7	Right	ATT	None	
22	Bilateral	26.0	Bilateral	Derotation osteotomy distal tibia	Bilateral	Removal derotation osteotomy plate

Table 7.2 – (continued)

			Additional interventions during 8-plate implantation		Additional interventions during 8-plate removal	
Study no.	**Clubfoot**	**Time (months) 8-plate in situ**	Side	Additional intervention	Side	Additional intervention
23	Right	34.0	Right	TATT	None	
24	Left	11.4	None		None	
25	Right	32.3	None		None	
26	Right	13.1	None		None	
27	Left	13.8	None		None	
28	Bilateral	22.5	Bilateral	ATT	Right	TATT
29	Right	11.7	None		Right	TATT
30	Bilateral	18.7	None		None	
31a°	Left	9.4	None			TATT
31b°	Right	18.3	None		None	
32	Right	40.5	Right	TATT	None	

* Represents 1 patient that had a re-do 8-plate implantation in the same foot. ° Represents 1 patient that has bilateral clubfoot with same 8-plate implantation date, however with a different date of removal. PMR = Posteromedial Release; AT = Achilles tenotomy; TATT = tibialis anterior tendon transfer.

Intrarater reliability of radiographic measurements

The intrarater reliability for the measurement of the ADTA was almost perfect, with an ICC of 0.930.

Effect of hemi-epiphysiodesis measurements

Dorsiflexion and ADTA for each clubfoot at the different time points is presented in Table 7.3. The median ankle dorsiflexion significantly increased during 8-plate treatment ($p<0.001$) from range -20-5° (median 0°) to range 0-10° (median 0°). Dorsiflexion of 19 patients improved with at least 5 degrees. ADTA significantly improved from 84° (95%-CI: 82-85° to 72° (95%-CI: 70-74°) during implantation ($p<0.001$). No significant correlations were found between amount of improvement in ADTA and dorsiflexion.

Table 7.3 - Results per clubfoot.

Study no.	Side	Dorsiflexion			ADTA		
		Preoperative measurement	Indication for 8-plate removal	At least one year follow-up	Preoperative measurement	Indication for 8-plate removal	At least one year follow-up
1	R	0	0	-	76	72	87
2	L	0	10	-10	91	79	88
2	R	5	0	0	94	76	88
3	R	-5	0	0	81	73	-
4	L	0	5	15	83	70	78
5	L	0	0	0	84	76	-
5	R	0	0	10	83	72	-
6	L	0	0	-	86	63	-
7	R	5	0	-5	89	67	92
8	L	0	0	-	86	63	-
8	R	0	5	-	86	71	-
9	L	-	-	10	84	69	79
9	R	-	-	10	86	72	82
10	L	-20	5	0	84	71	78
11	L	0	-	10	80	72	81
11	R	0	-	0	81	64	67
12	R	-10	5	0	83	71	83
13a*	L	-5	0	-	79	63	81
13b*	L	-20	-5	-	71	67	-
14	L	0	0	-	87	73	-
15	R	0	0	-	88	72	-
16	L	0	10	-	85	81	-
16	R	0	10	-	86	86	-
17	L	0	0	-10	80	67	82
17	R	0	0	-10	77	-	87
18	L	0	0	0	85	70	82
18	R	0	0	-10	88	68	82
19	L	0	10	0	84	73	79
19	R	0	-	10	84	80	67
20	L	-	5	10	72	60	83
20	R	-10	5	0	85	62	83
21	L	5	0	0	77	69	82
21	R	5	0	0	81	67	85
22	R	-10	5	0	82	64	76
23	L	5	-	-	77	75	-
24	R	-	-	-	88	69	-
25	R	0	10	-	80	73	-
26	L	0	0	10	80	-	97
27	L	0	10	10	89	71	-
27	R	-10	10	0	91	97	-
28	R	0		0	75	70	83
29	L	0	10	-	84	72	84
30	R	0	10	-	86	73	83

Table 7.3 – (continued)

Study no.	Side	Dorsiflexion			ADTA		
		Preoperative measurement	Indication for 8-plate removal	At least one year follow-up	Preoperative measurement	Indication for 8-plate removal	At least one year follow-up
31a°	L	-5	0	0	84	74	80
31b°	R	0	-	10	84	83	-
32	R	-	0	-	94	73	88

* Represents 1 patient that had a re-do 8-plate implantation in the same foot. ° Represents 1 patient that has bilateral clubfoot with same 8-plate implantation date, however with a different date of removal. PMR = Posteromedial Release; AT = Achilles tenotomy; TATT = tibialis anterior tendon transfer; - = Missing.

In the subgroup of patients of whom also at least one year after 8-plate removal clinical and radiological assessment was available no overall effect on dorsiflexion was found. ADTA significantly changed between the three intervals ($p<0.001$). ADTA significantly decreased during 8-plate treatment ($p<0.001$), but it significantly increased after at least one year of 8-plate removal compared to the situation at indication for 8-plate removal ($p<0.001$) (Table 7.4).

Table 7.4 - Functional and radiographic outcomes in 3 measurement intervals, presented as median [range] or mean [95%-Confidence Interval].

	Preoperative measurement	Indication for 8-plate removal	At least one year follow-up	Significance
Dorsiflexion n=22 feet	0° [range: -20-5°]	0° [range: 0-10°]	0° [range: -10-15°]	*P*=.076
ADTA n=27 feet	83° [95%-CI: 81-85°]	70° [95%-CI: 68-72°]	82° [95%-CI: 79-84°]	*P*<0.001*

* post hoc analyses indicated a significant decrease in the ADTA from preoperative measurement to the indication for 8-plate removal and a significant increase in ADTA from indication for 8-plate removal and follow up.

Timing of hemi-epiphysiodesis

The age at which the AHDT was performed in this cohort varied between 5 and 12 years. No clear relation between change in ADTA during implantation or amount of rebound after 8-plate removal with age was found (Figure 7.4A). Changes in ADTA showed large variation between patients within an age group.

Treatment duration, i.e. number of months the 8-plate was in situ before removal varied. Again, variation of the accomplished change in ADTA was large, with no clear relation with treatment duration (Figure 7.4B).

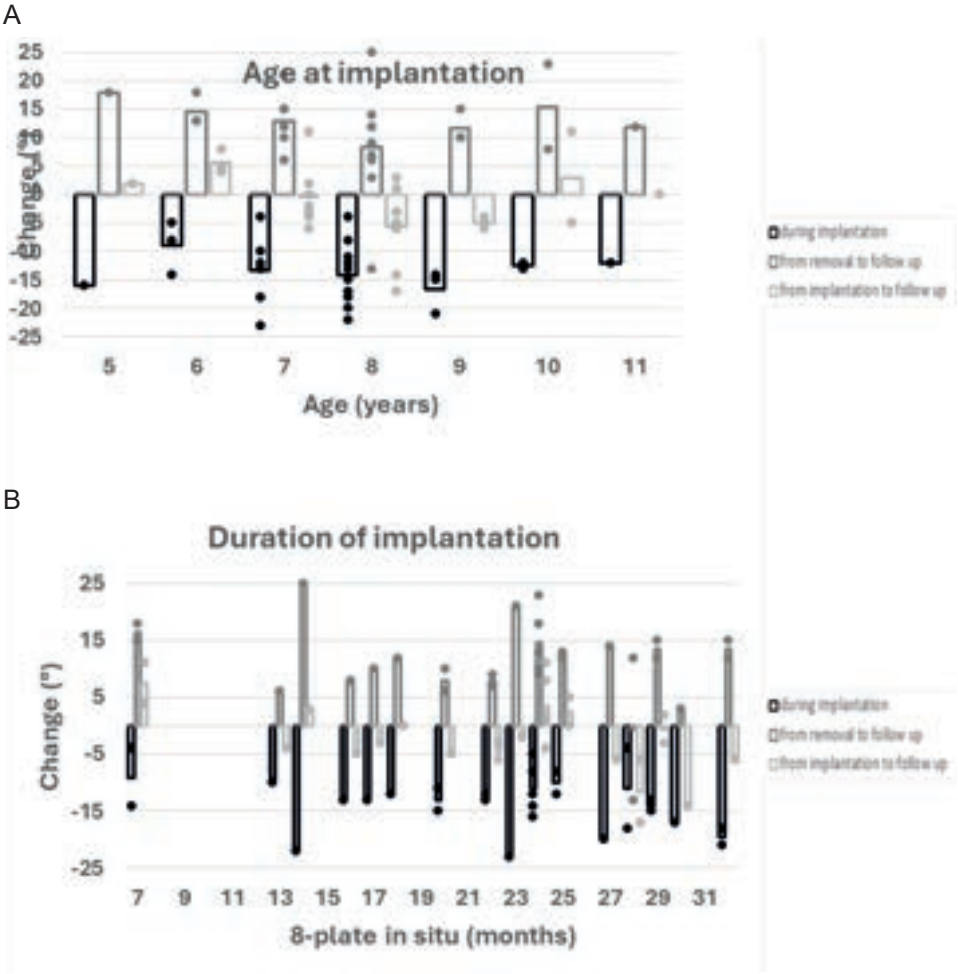

Figure 7.4 - Amount of change accomplished during implantation, after implant removal and between initial implantation of 8-plate and final follow up. Bars show mean change in ADTA for each category, dots represent results of a single patient. **A**: Change in ADTA versus age at implantation. **B**: Change in ADTA versus duration of 8-plate in situ.

Discussion

In our patient group we found a significant decrease in ADTA during temporal AHDT in relapsed, by the Ponseti method treated, clubfeet, showing that guided growth is effective in influencing the growth plate and changes the position of the tibial plafond. Remarkable is the disappearance in our patient group of this beneficial change in ADTA over time after removal of the implant. After a median

follow up of 20 months, ADTA remodeled to pre-operatively found values. When evaluating the influence of age or duration of the guided growth treatment on change in ADTA, no consistent patterns could be identified. Furthermore, clinical improvement indicated by change in dorsiflexion after guided growth could not unambiguously be quantified in the current cohort.

Radiological improvement quantified with ADTA in relapse clubfoot patients during hemi-epiphysiodesis was previously shown.[15-17,25] Although sporadic and minor rebound of ADTA after implant removal was previously reported,[15,17,25] this is the first study to show severe remodeling at least one year after removal of implants. The distal tibial growth plate closes only in late adolescence.[26] The open growth plate can correct malalignment and is mainly responsible for correcting deformities by asymmetrical growth stimulation. Growth is stimulated on the concave side and inhibited on the convex side.[27] During the hemi-epiphysiodesis, growth is influenced, but after 8-plate removal in case of open growth plates, one could expect the growth plate to act as before the treatment. This means that remodeling takes place to the old status, where forces in the growth plate are equalized by the above-named mechanism. In our opinion this phenomenon could be responsible for the rebound effect also because during treatment and removal all growth plates of the patients were still open. This finding could have implications for timing of the procedure.

The clinical improvement after guided growth in the distal tibia at longer follow up in clubfoot children could not unambiguously be quantified in our data. In the complete group a significant clinical improvement in dorsiflexion was shown till the moment of removal. However, in the smaller subgroup with also at least one year of follow up after implant removal, this improvement in dorsiflexion was not clear. Previous literature showed mostly a positive clinical effect of AHDT on the amount of ankle dorsiflexion,[15,16,25] obtaining a plantigrade foot[17] and improved quality of walking according to parents/caregivers.[25] In only one study a direct relationship is shown between the amount of change in dorsiflexion and a decrease in ADTA.[16] It should be noted that Al-Aubaidi et al. did not find a clinically significant improvement in dorsiflexion in 25 clubfoot patients with progressive equinus relapse previously treated with extensive posteromedial releases.[18] The question remains what the best method is to quantify clinical improvement. A reliable assessment of dorsiflexion is challenging in children with a severe foot deformity.[28] In the current study, all clinical investigations were performed by one senior pediatric orthopedic surgeon [AB] to minimize variation.

Small changes in the position of the foot can however have an important impact in clinical function.[16] The difference between a foot in slight equines or slight dorsiflexion is important during gait. It would possibly be more useful to determine if patients achieved a plantigrade foot position[17] or apply extensive gait analyses (e.g.[9,29]) to identify the clinical relevant treatment outcome of AHDT.

It should be noted that the described cohort consists of patients with difficult and heterogenous clubfoot appearances. In line with previous literature, patients often received relapse treatment prior to the hemi-epiphysiodesis [16,17] and/or the hemi-epiphysiodesis was combined with additional procedures.[15,18] Our group of patients consisted mostly of referred patients with severe residual clubfoot deformities and flattening of the talus. The etiology of this talar flattening remains unknown. It may be a pre-existing feature of the clubfoot or related to treatment.[30]
Before surgical interventions are considered as treatment for relapsed clubfeet, a thorough clinical assessment is necessary.[31] Furthermore a step-by-step approach, starting with repeated Ponseti casting should be considered.[7,31] When again relapses are seen in a flexible foot, according to the Ponseti-technique itself, a tibialis anterior transfer with or without a re-do Achilles tendon tenotomy should be performed.[3] For older patients with relapsed clubfoot and severe impairment of joint movement as in flat top talus deformity, less invasive surgical techniques are preferred to limit joint stiffness on the long term as much as possible.[3,32] Guided growth with its tension band mechanism, introduced with just a small incision meet these criteria. The post-operative regime after a AHDT includes immediate functional and full weight-bearing instructions, again to prevent any unnecessary joint movement impairment and burden the patient as little as possible. We indicated guided growth when severe functional impairments in combination with complaints in daily life activities existed.

Our study meets some limitations. Our patient group is heterogeneous in terms of severity of the deformity, which might influence the clinical outcome. However, the technique of minimal invasive surgery to perform AHDT is typically meant for complex cases. Still, these patient factors might complicate the interpretation of the results. Furthermore, although this is the largest cohort described in literature the sample size and available follow up is still limited. Due to the retrospective nature of this study, not all data of all patients was available for analyses. Determining the clinical outcome of hemi-epiphysiodesis purely based

on dorsiflexion remains challenging. For future research, we encourage to also take treatment impact on parents and patients into account. Furthermore, more functional and participation measurements could be important to conclude if guided growth is valuable in treating recurrent equinus deformities in clubfeet.

In conclusion, AHDT in clubfoot is effective in influencing the growth plate, save and meets criteria of a less invasive treatment with little damage as possible. Guided growth by means of hemi-epiphysiodesis should be in the armamentarium in relapse clubfoot with bony impairment after determination of an optimal timing by the treating surgeon. An important finding in this study is the rebound phenomenon. Because rebound of ADTA occurs in children with active growth plates, parents and patients should be informed about this mechanism which might result in the need for renewed hemi-epiphysiodesis when the procedure had to be used early in childhood. If possible –when indicated– the procedure could be postponed and used in the last phase of growth.

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CHAPTER 8

General discussion

General discussion

The aim of this thesis is to give an overview of recent changes in and their effect on clubfoot care in the Netherlands. Important clinical questions arose during the transition of treatment away from extensive surgical solutions to treat clubfoot. Several of these questions were addressed in this thesis to initiate clinical changes or define knowledge gaps for future research. In this general discussion, the outcome of the studies presented in this thesis will be discussed in the perspective of three main pillars related to clubfoot care and the transition of treatment in the Netherlands: organization of care, treatment outcome and relapse.

The hierarchy of scientific research places randomized controlled trials (RCT) – after meta-analysis and systematic reviews- at the top of the scientific pyramid. Clinical research involving young children is challenging. For example due to relatively small numbers of children with specific medical conditions, adaptations are required in research procedures and settings to accommodate children's emotional, cognitive and physical development. Complexities exist related to parental involvement and family decision making as there are special ethical and regulatory protections for children.[1] As a result, RCTs are difficult to conduct in pediatric orthopedics. Especially in the treatment of very young children, where once treatment is started, treatment results can be irreversible. Furthermore, due to the good outcome of the Ponseti clubfoot treatment,[2] randomization of patients to compare with other treatment methods is no longer ethically justifiable. This thesis is a combination of a guideline development based on literature review and clinical research based on prospective and retrospective – case-control– cohorts. Where high-level research must be conducted where possible, long-term registration and close follow-up of treated cohorts definitely can lead to improved care.

PART I: Organization of care

Guideline on the diagnosis and treatment of primary idiopathic clubfoot

To diminish unwanted variance in clubfoot care and inform health professionals and (parents of) patients on the best treatment of this deformity, guidance is needed. In complex care it has been shown that by centralizing care with

guideline guided treatment, optimization of care is possible.[3-5] Although the question if clubfoot care is complex, for some still is not answered definitively, reports of difficult feet and disappointing results of treatment are not scarce.[6,7]

When the Ponseti treatment became known in Europe, the discussion to choose between extensive surgery and the manipulation and casting method of Ponseti was not completely ended in the Netherlands. Consequently, treatment variation existed and patients were treated in different ways only because of their geography. This gave rise to questions from parents which were very difficult to answer. These same parents initiated an important meeting among clubfoot stakeholders. At this meeting a consensus was reached to start the development of a guideline for clubfoot. Together with parents, pediatric orthopedic surgeons and a methodologist of the Dutch Knowledge Institute of Medical Specialists (Kennisinstituut Medisch Specialisten, KMS) were assigned as part of the guideline team. The latter was indispensable to reach the highest guideline quality and gain strength to implement outcome. Goal of the guideline development was to produce a solid, scientifically based instrument that could and should be implemented in daily clubfoot care to improve treatment, give uniformized information to parents and diminish unwanted variance. As the first pediatric orthopedic guideline in the Netherlands, and to our knowledge the first guideline on clubfoot worldwide, 5 clinical topics were formulated to address. The optimal treatment, diagnosis and classification, importance of brace compliance and other patient related factors, relationship with DDH and organization of care were all subjects to study and establish with guideline advice.

Support of the Dutch Orthopedic Society and the Dutch Knowledge Institute of Medical Specialists (Kennisinstituut Medisch Specialisten, KMS) was indispensable to reach the highest guideline quality and gain strength to implement outcome.

In general, limitations of guidelines are professional conflicts of interest, narrow focus on one disease without individualization of the patient and usage of low quality of evidence.[8] The diversity in the guideline team was meant to minimize the different personal preferences and represented methodological expertise, clinical input, representation of stakeholders and affected populations.[8] There was no commercial funding for this guideline process, all costs were covered by the Dutch Orthopaedic Society, again important to prevent conflicts of interest.

During the composition of this guideline, the committee was sometimes faced with low levels of evidence. By using the G(rading of) R(ecommendations,) A(assessments,) D(evelopment and) E(valuations) method the committee could collect and grade the best available evidence but also weigh the evidence together with complementary arguments like patient values and preferences formulating so-called considerations.[9] In this way weak evidence could lead to strong recommendations, with as prerequisite that the evidence used showed all the same direction of evidence, and so direct patient care in a good way (https://bestpractice.bmj.com/info/toolkit/learn-ebm/what-is-grade/).

Our guideline posted several recommendations in organizing clubfoot care, diagnostics and treatment, prognostic factors and relationship with DDH.

At the end of the process a clear guideline was formulated, approved by the Dutch Orthopaedic Society. The main recommendation of the guideline was to treat the clubfoot with the standard Ponseti method in all details. The finding that the Ponseti method is the optimal treatment for clubfoot patients was recently verified by a meta-analysis.[10] Not only extended surgery in primary clubfoot could be abandoned also detailed description of the Ponseti method could be prescribed by the addition ‘standard’. Of course, every physician can deviate from the recommended treatment but now must argue firmly why. Almost 10 years after implementing the guideline, in discussions with Dutch Pediatric orthopedic surgeons it seems that almost no surgical treatments are used and mentioned as a first choice to treat clubfoot.

Related to the diagnosis of clubfoot, the guideline stated that physical examination must be performed, and scores must be used to classify the severity of the clubfoot. In the Netherlands, different centers used the Pirani and Dimeglio scoring system and literature could not discriminate which scoring system should be used. Complementarity was shown between both Pirani and Dimeglio scores, with different aspects being investigated (foot aspect and reducibility).[11] From unpublished analyses recently performed in our own study group, we conclude that both scoring systems show high to very high correlations (Besselaar, Boonen, van der Steen, 2023, unpublished data). As third classification system we studied the C(lubfoot) A(ssessment) P(rotocol), a validated scoring system also meant for the follow up older children with a clubfoot.[12,13] In contrast with previous used scoring systems, the CAP uses detailed measurements with passive motion values but also functional tests,

appropriate for different age groups.[14] We therefore propose to use the Pirani score until starting the brace period and the CAP score from that period on after also one initial CAP score at the start of the treatment. Experience in clinic shows combining efficacy and simplicity probably stimulates usage of scoring systems. Administrative tasks influence physicians importantly these days so attention to both is important when deciding if and which score systems are advised.[15]

The guideline highlighted the importance of bracing as part of the treatment of clubfoot patients. Parents and patients must be encouraged to use them consequently to minimize the recurrence chance. A recent systematic review and meta-analysis of the literature showed clearly brace non-compliance as one of the prognostic factors inducing recurrence.[16] Difficulties in brace wear could be due to all different factors according to foot problems, the child's behavior or parental concerns. To attest these factors is not easy and may be reason to talk about complex care when referring to the treatment of clubfoot.[17]

During the composition of the guideline literature was available that assumed a relationship between clubfoot and DDH.[18,19,20,21,22] A level 3 (low) proof level mentioned advice to screen clubfoot patients for DDH. In 2015, a systematic review and meta-analysis was published on the prevalence of DDH in idiopathic clubfoot.[23] They concluded there is no base for a relationship DDH and clubfoot but also stated that studies of better quality are needed to answer the question to routinely screen on DDH in clubfoot patients is necessary. This can possibly have implications for a guideline update: routine screening in clubfoot patients would possibly not be advised anymore.

Considering the organization of care, the guideline states that when diagnosed prenatally, parents should be referred to a clubfoot center for proper information. Here the question rises how many centers in the Netherlands are needed. In chapter 2 the incidence will be discussed in relation to the optimal (and minimal?) number of patients needed per center to serve optimal standards of care. Participation of parents' representation was important to put this question in perspective. During the establishment of the guideline, the parents initiated an online survey among parents of their association. 233 parents answered out of 248. 80% of the parents mentioned that they wanted to travel for optimalisation of care, at the inverted question who was *not* willing to travel if the care offered elsewhere had a higher quality, not one parent answered with 'yes'

(https://richtlijnendatabase.nl/richtlijn/primaire_idiopathische_klompvoet/randvoorwaarden_organisatie_van_de_zorg.html). In this perspective, as responsible physicians, there are extended possibilities to optimize clubfoot care without assumptions like parents only wanting treatment in their neighborhood. Quality of care for the clubfoot patient must be leading in this process. Experience of the treating team on different aspects as diagnoses, initial and extended treatment, complication handling, counseling, 24/7 coverage were all mentioned as prerequisites for optimal clubfoot care. Although the pediatric orthopedic surgeon is responsible for the complete clinical pathway, well trained paramedical healthcare providers (i.e. plaster technicians in the Netherlands) must be part of the treating team. Engagement for the different aspects within the treatment by the members of the clubfoot treatment team in clinic are essential. The most important aspects in my opinion are prenatal counseling, guidance during the brace period and outcome of quality-of-life measurements. Recent research emphasizes the complicity of quality of life (QOL) measurements in relation to social determinants of health and health literacy.[24] This research was provided in spine pathology, but the diversity of clubfoot patients and their parents show, in my experience, the same issues.

Evaluation of the guideline also was a concern in line with the sceptic ideas in the field. Only one third of the organizations producing guidelines, evaluate the implementation and recommendations regularly.[25] After 5 years, the NOV questioned if revision of the guideline was necessary. At that moment small differences could possibly be expected but the main message to treat clubfeet with the Ponseti method would not change. In relation to costs and manpower, the decision was made to postpone the guideline revision. With all new literature and implementation of the guideline since, it could be wise to revise and extend the guideline shortly, all above factors must direct to the intention to implement and build the situation for optimal treatment outcome. In addition to updating, it may be possible to produce guidelines for complex clubfeet and/or residual deformities. By the development of clubfoot care since the introduction and implementation of the guideline it may be possible to consider further centralizing of clubfoot care.

Incidence of congenital idiopathic clubfoot in the Netherlands

The last chapter of the guideline document mentioned to perform clubfoot treatment in appointed centers.[26] To change a care landscape, it is necessary to know how to define the extent of the patient group especially when published

figures are rare. High volumes in complex care led to a better treatment outcome.[4,5] The variation in relapse numbers to our opinion give rise to include (a part of) clubfoot treatment as complex care, till it still seems to be hard to define relapses unequivocally.[16,27] Several numbers according to the incidence of clubfoot are available and still different research groups globally are interested in incidence figures.[28-31] Different number of patients per area could also implicate that less centers are needed. High numbers per region could implement to enlarge centers and gain more expertise.

With the number of living newborns, we could define the incidence of two separate years 2013 and 2014, respectively 1.16 and 1.02 per thousand ($p>0.05$). Recent research shows an estimated, pooled birth prevalence of clubfoot of 1.18, with a range of (CI 1.00-1.36), per 1000 births over 36 countries worldwide including almost 45 million births.[32] In Europe the birth prevalence of clubfoot patients differs between 0.8 and 1.35 per thousand. Our results fit in this interval. Distribution of bilateral vs unilateral cases affected sides and sex distribution were all conform the existing literature, resp. 66% boys, bilaterality in 50% and affected sides each 50%.[33]

Our study showed around 180 newborns with clubfoot per year can be expected in our country. Considering a slight underestimation, round 200 children with approximately 300 clubfeet per year can be expected. Underestimation together with the importance of knowing incidence figures were already mentioned in literature as a base to organize care.[32]

Before this research, estimations per clinic led to implausible numbers of new clubfeet per year. Patients are seen several times per caregiver and not always it is clear how many new patients are seen by pediatric orthopedic surgeons. Since counting and registration all new patients, overestimation also was our own experience. Because of treating many new and older patients in practice, the number of new patients we experienced also were overestimated in our own minds. The current estimation of approximately 200 new clubfoot children per year is the best possible in the absence of a prospective registry. Based on these numbers with 10 centers we would serve 20 new clubfoot patients per year in the Netherlands. This seems to be a reasonable number although optimal clubfoot specific numbers, scientifically based are not yet available.

As mentioned in the previous paragraph discussing the guideline, experience within the treating team should be established but also the organization of care is more easily kept at an optimal level when at least a minimal number of patients are treated regularly. Composition of an adequately trained and motivated team and assemblage of a child-friendly surroundings with optimal facilities also forms a provision for good care. Structure and a good back-office to serve parents, also must function well. Together, many factors to structure clubfoot care, can lead to optimal treatment.

One or two centers in the country could subsequently be addressed as reference center when complex situations occur. The group difficult feet provide different types of clubfoot. Relapses need attention and specific care often, but also primarily complex or atypical feet need specialized expertise. The earlier detected, the better,[34] so determination of the diagnosis also is important.

Recently pediatric oncology and heart surgery are centralized by the Dutch government. Despite good intentions and with the intention to reach improvement of outcome of care, resistance was felt by the health care providers.[35] For a long period, different stakeholders tried to organize the change themselves but failed. Because of the need, the government took the lead not to everyone's satisfaction. It could be wise to implement these choices within the Pediatric Orthopedic Society ourselves. Often if centers are appointed to perform complex care, proud and enthusiasm arises. To advise however centers to stop complex care needs courage and open discussion because often disappointment is displayed. The Pediatric Orthopedic Society must continue to use their instruments and continue the dialogue to get to this point. When highly competent, well-functioning clubfoot centers can demonstrate their expertise, it will be an example of a good initiative where the government only needs to confirm. Simultaneously in the Netherlands a debate is held about medical care in the future. Different challenges as scarcity of personnel, high volumes of administration, experienced pressure by employees, sick leaves and of course high costs make changes and choices mandatory. In a comprehensive document, the government presented the base of which decisions with the care landscape will be made in the future (Integraal Zorg Akkoord (IZA) 2022, Rijksoverheid.nl). 'The right care on the right spot' is a one liner often used. Also in this perspective, choosing optimized, centered, well registered care should be the goal to reach.

Since 2021 a national registry for clubfoot has been implemented in the Netherlands (https://www.lroi.nl/lroi-data/kinderorthopedie). Unfortunately, the national registry system is not efficiently incorperated in the electronic patient record systems yet. Modern systems immediately converting from patient records should be available soon. Without these adjustments, the effort to register does not comprise the information gained. The technical possibilities are probably already available and are already used in implant registration systems in the Netherlands (https://www.lroi.nl/lroi-data/lir/). Providers of the patient records systems however restrict new manners by chasing huge amounts of money for system changes and new technical developments. The NOV should provide resources to solve this issue to improve registration and, indirectly, care. Without these adjustments, the effort to register does not comprise the information gained. The government or insurance companies should play a role in influencing health system providers in reducing financial barriers. This also will reduce administrative ballast, important in the care landscape nowadays (Integraal Zorg Akkoord (IZA) 2022, Rijksoverheid.nl.[36]

PART II: Outcome

Quality of life of clubfoot patients during the brace period of the Ponseti treatment

Bracing is an intense but crucial part of the Ponseti treatment. Not using the foot abduction brace has been mentioned as an important and causal factor in the creation of a relapse.[37] In clinical practice we face parental and children-related factors causing hesitation to put on the braces. On social media platforms, brace issues also are mentioned in different domains as important issue.[17] 48% Of the total amount of clubfoot related Facebook posts, were related to physical concerns related to bracing. The next most common posts handled psychological issues also related to bracing.[17] Surprisingly especially in very well corrected feet, parents think the brace is not necessary anymore. For health providers it is important to realize that these issues are in parents' perspective (too) often unsolved, leading them to ask questions on social media. Since answers on these platforms are not always controlled and sometimes only based on experiences, attention for frequent hospital visits are necessary and maybe even involvement of medical professionals in social media groups could be considered. A better understanding of bracing and health specific quality of life seems to be indicated.

Therefore, we studied the influence of this brace on the quality of life in clubfoot patients. The inability to directly ask the little children made that we used a validated instrument (TAPQOL) to ask parents about motor functions and quality of life of their children.

Our overall results showed on average a high score on the 4 domains (physical, social, emotional, and cognitive functioning) of the TAPQOL instrument corresponding with a high quality of life in clubfoot patients in the examined age group. Significant lower scores in the clubfoot group were measured in the subdomains motor functioning, sleeping, lung- and skin problems. Remarkably, there were no or only mild by the parents' perceived negative emotions related to these skin and motor functioning issues. In other words, the children did not seem to bother much. Lung and sleep problem had a bigger negative impact on the children. Childhood sleep disturbances are relatively common in young childhood. In a study to examine the relationship between measured sleep duration and parents' reported sleep habits in typically developing children between 12 and 60 months, it was shown that reports of parents consist of reliable data in relation to quantitative sleep investigation.[38] Sleeping disturbances not only have impact on the children but also on their families.[39] Impaired sleeping patterns in adults can influence working memory, attention, and behavior problems.[40] These data support that differences in sleeping patterns during brace wearing and healthy controls, needs attention during counseling and must not be underestimated even though over time these problems seem to be resolved.

The TAPQOL instrument has shown to be a valid instrument when evaluating impact of a disease comparing with healthy controls.[41] Although valid, the TAPQOL is not specific for clubfoot. With the instruments used we could not show complete causality for instance according to the skin problems. In other fields of medicine, disease specific quality of life instruments exists but sometimes have to be combined to show reliable outcomes.[42] Emphasizing the need for further research, also extension to the first year of brace wear could be important. In our experience also in the first year there are differences in acceptance and problems at night in different children. Some parents report no problems at all, for others it really is a complex issue.

We can conclude that quality of life during brace wear in clubfoot patients is high, but the individual differences need attention of treating physicians. We

must optimate the counselling and inform patients intensively about short and long-term consequences of brace wear. Primarily to optimize brace wear to get the best result in clubfoot treatment, secondly to be supportive in faced issues by the children and parents.

Participation and motor abilities in children aged 5 to 9 years with idiopathic clubfeet after treatment with the Ponseti method

At the end of the brace period the clubfoot children mostly will gain independence in their motor activities and behavior as part of functioning in life.[43] But equal to the influence of bracing, clubfoot deformities can affect these two regions of functioning in life. To reach optimal levels of health and wellbeing it has been hypothesized that adequate levels of activities and participation are necessary.[44] The International Classification of Functioning, Disability and Health (ICF) (WHO, 2001) and ICF for Children and Youth (ICF-CY) (WHO, 2007) mention activity and participation as key components of health. Activity is defined as the execution of a task or action by an individual, participation as involvement in a life situation (https://www.whofic.nl/en/node/901).[45] It is important to have knowledge about these aspects in clubfoot patients to counsel parental and children's expectations. Furthermore, identifying levels of activities and participation can serve as a tool to identify –residual– impairments and aid optimization of treatment. Therefore, we investigated participation and motor abilities in nationwide cohort of clubfoot patients.

Our overall results show that children with clubfeet in the age of 5 to 9 years overall have high levels of parents' perceived motor abilities and participation, similar to age-matched healthy controls. However, motor abilities in children with clubfeet was lower in a static and predictable environment –mostly due to poorer balance and hopping abilities–. Furthermore, a trend was observed that children with clubfeet show a higher risk for deviating motor abilities. Parameters indicative for level of participation showed only small differences between clubfoot patients and healthy controls.

One could conclude that the overall good results and relatively small differences found in our study, prove that there are no issues to solve in the clubfoot population. The wide variation in the clubfoot group however suggested differently, there are some patients that do experience problems in daily life. If this is related to possible relapse of the clubfoot it could not be addressed in this study. Because of privacy issues medical history and current status of the

clubfoot were not known. Relapsed clubfeet show one or more of the initial clubfoot features[37,46] and in practice are characterized by complaints of pain and limitations in function.

The questionnaires used in this study were not specifically designed for clubfoot patients. Some parents reported that the questions were not relevant for their children. The disease specific instrument (DSI) was specifically developed to assess the outcome of treatment in clubfoot patients. The DSI was found to be reliable, valid, and discriminative.[47-49] This questionnaire measures satisfaction of the foot appearance, teasing problems of the clubfoot child, shoe fitting and appearance, and functional outcome pain and impairment during running and physical exercise. As such in future research, the DSI could be included when assessing motor functioning and participation.

Because of the young age (5 to 9 years old) of the children, our study focused on the parents' perception of activity and participation This indirect manner of questioning has its limits, ideally the children themselves are questioned. To question a 5-year-old can be quite different from a child at the age of 8. Also, children among the same age can really differ in expressions at all and of well-being specifically. Visual parts of a questionnaire or digitalization by modern technology may be in the future provide new insights in combination with disease specific, age-related formats. Presumably in an older child, quality of life measurements can be done directly by asking the child itself but when in a younger age, difficulties can possibly be solved using modern technologies. Attention for studying Q(uality) O(f) L(ife) in youngsters therefore still is needed. Because of our experience of cognitive differences between children in the used age group, indirect questionnaires probably will stay as most applicable. Also in other fields of QOL research, parents' perceived questionnaires are used as a standard.[50]

To reach optimal levels of health and wellbeing it has been hypothesized that adequate levels of activities and participation are necessary.[44] In the latter mentioned study important remarks are made about environmental aspects influencing participation. The specific diagnosis seems less important where age, sex and functional abilities just are. The complexity of participation as phenomenon, as Law states, makes understanding of personal, environmental, and family factors influence on daily activities of the children very important. Functional abilities, as investigated too in our study, are mentioned as important

facet influencing participation. The authors advise to move beyond diagnosis to focus on activity limitations as a major predictor of participation. This vision strengthens our study design where we combine motor ability and participation. Future studies should focus even more on functional abilities and other aspects in the clubfoot group to better judge participation. Besides questionnaires also other research methodology such as movement analyses could be employed to investigate functioning in children.[51,52] If we could define the determining factors that influence clubfoot patients' functioning in life better, we may focus differently during treatment protocols. Priority should be given by stimulating this type of new research rather than trying to re-emphasize already known issues, such as the efficacy of Ponseti treatment in general.

PART III: Relapse

Identification and treatment of residual and relapsed idiopathic clubfoot in 88 children

Although the Ponseti treatment generates good to excellent results in most clubfeet, a substantial number shows persistent deformities.[37,53]. For identification and treatment purposes it is important to have information on the presentation of problems. This will support the choice how frequently and prolonged follow-up should be. Furthermore, it supports thorough physical examination of more difficult feet to identify problems. Within clubfoot related problems a deviation should be made between residual and relapse clubfeet. In our study we defined residual deformities as deformities that underwent primary Ponseti treatment but never were fully corrected. Relapse deformities were defined as clubfeet showing any of the features of an untreated clubfoot after initially successful treatment, which needs additional treatment. To structure this residual and relapsed deformities we performed this study. We found several relapse combinations, mostly with equinus or decreased dorsiflexion. Also, adduction and active supination were often seen in relapses, therefore we concluded that attention to these deformities after complete correction is important. According to age we found that in young children mostly single deformities were seen, in older children mostly more complex combinations were seen. The age when relapses occur would be important to encounter. Dietz stated that relapses mostly occur before 7 years of age.[54] Our findings, where we also faced older children identified as relapses, are more in line with the

review of Rastogi and Thomas. Therefore, follow up to skeletal maturity is advised.[55,56]

Relapses after initially satisfying results and residual deformities that have never been corrected completely, are seen in different amounts varying between 3.7 and 67%.[53] These numbers differ significantly, explanations could be found in the diversity in definitions and descriptions of rest-deformities, another could be the follow-up described in different studies. As a complicating factor, studies do not always add the applied definition of relapse.[56] Ponseti stated that a fully corrected clubfoot, can relapse, showing 'old' clubfoot deformities again. Mostly two deformities are present: hind foot equinus and varus.[45] Thomas et al.,[56] identified four types of relapse definitions used in literature. These definitions were based on reappearance of physical qualities, the need for additional cast, surgical or any treatment after initial corrections. Furthermore, combinations of these categories are used. Recently in the BSCOS Consensus group the definition 'Relapse implies a reappearance of any of the clubfoot deformity elements in a foot that has previously fitted easily into the FAB' was used.[57] A uniform definition is important to enable comparisons between cohorts and outcomes of additional treatment.

The definition "a relapse is the recurrence of any component of deformity after a complete correction of clubfoot"[58] is clear and useful in clinical practice. In addition, different types of relapses could be divided anatomically (forefoot, midfoot, hindfoot), according to the deformity direction (varus, adduction, equinus) or a combination of these two. Important other factor is the cause of the deformity.[59] Dynamic relapses due to muscle imbalance must be differentiated from rigid equinus deformities as in flat top talus. I would propose to define the relapse in an anatomical location and direction (i.e. hindfoot varus, forefoot adduction) and mention if the deformity is rigid or dynamic. The latter shows dynamic supination as most important finding with direct implications for the treatment. But also, a springy end-feeling in dorsal flexion can be a sign of a short Achilles tendon where a hard end-feeling leads more towards a flat top talus deformity. Of course, the treatments indicated can obviously differ.

Treatment of residual and relapse clubfeet should follow a step-by-step treatment.[46] In the cohort presented, we showed that under 4, the Ponseti protocol is dominant. Over 9, bracing is not possible anymore and therefore older children are mostly represented in the surgery group. The choice of

treatment is based on multiple factors. The age of the patient, the deformities detected and of course the intensity and frequency of complaints of the patient are important factors.[46] Muscle imbalance could lead to recurrence of the clubfoot deformity. Interestingly Zionts, showed the likeliness to undergo a A(nterior) (T)ibialis (T)endon (T)ransfer at all. They state a chance of almost 30% in clubfoot children at an age of 6 years. Noteworthy is the distinction between families reliably using bracing (19% likelihood for an ATTT) and families who reported nonadherence (42% likelihood for ATTT).[60] This implicates the importance of brace adherence but also support the complexity of treatment decision. Also, talar flattening remains an unsolved cause in clubfoot relapse that may be pre-existing or iatrogenic.[61] Further research is necessary to produce answers according to causes and possible treatment.

In the context of the organization the intensity and continuity of follow-up have implications for clubfoot center appointments and will influence the practice of treating clubfeet. If a relapse deformity is noticed, at least check-up frequencies must rise till a definitive treatment decision is made. To decide in clinic when to intervene thoroughly can only be done if the indication is made as early as possible and so unwanted deformity progression is prevented.

After the brace period children often want to develop their motor skills and are less concerned than their parents. In clinic often the physician is concerned while the parent experiences a non-complaining child but understands the surgeon. At the same time the child experiences no pain or impairments. Still, it is difficult to predict the influence of a mild deformity for development in the future. Early recognition simplifies treatment and generate better outcome.[62] It is advised to intervene early. In a Delphi protocol-based study[63] the panel agreed on that once diagnosed, dynamic supination should be treated quickly by transferring the tibialis anterior muscle.

Anterior Distal Tibia Epiphysiodesis in relapsed Ponseti treated clubfoot patients: prolonged radiological and clinical effects

In residual or relapsed deformities, different treatment options are available.[64-66] Current literature still differs not only how to treat residual and relapsed clubfeet, also debate about outcome after extensive surgery in clubfeet is present.[64,66-69] Scaring, stiffness and pain are complaints after extensive clubfoot surgery which of course is detrimental. Unfortunately, also without extensive surgery, clubfeet already show stiffness compared to healthy controls.[70] Restriction in ankle and

subtalar movement due to differences in subtalar articular surfaces have been described earlier.[71] Because especially impaired dorsal flexion importantly restricts walking,[72] we examined correction of this deformity to more extent. To minimize the articular impairment after surgery and the chance of post-operative stiffness and minimize the impact for the patient, hemi-epiphysiodeses was our first-choice treatment when faced with persistent equinus deformity.

We found statistically significant changes after hemi-epiphysiodesis of the A(nterior) D(istal) T(ibial) A(ngle) of the distal tibia before 8-plate removal. To our surprise one year after removal a remodeling took place with again significant changes but now in the direction of the pre-treatment values. These latter changes, implicate the importance of good counselling when treatment is indicated in young children. Delay of the treatment if possible or information about possible re-implantation are logical consequences. When searching for clinical effects as a result of radiological changes, we could not draw firm conclusions. This could be due to the small follow-up group but also to the fact that detailed clinical examination in the pediatric ankle-foot region is not yet well described and defined. It is also difficult to perform a detailed functional examination of the foot in our young age group.

Figure 8.1 - Schematic drawing showing force distribution for a deformation compensated by asymmetric growth in the growth plate.

In hindsight remodeling taking place after removal of implants in young children with open growth plates, could be expected theoretically. In pediatric deformities in fracture care, we also face remodeling due to different forces acting on the growth plate.[73]

Already Hueter and Volkmann stated the mechanism behind asymmetrical growth as a result of differences in pressure on the growth plate. More pressure stimulates growth, less pressure inhibits growth.[74-77]

In an attempt to understand the reason of remodeling after removal of the 8-plate it seems logical to assume that most pressure during gait would be on the anterior part of the growth plate in the distal tibia. According to the above mentioned theory, the anterior growth plate should than be inhibited and the with the 8-plate obtained correction should be stimulated. Our results however, suggest differently. Indicating that the effects of forces over the anterior growth plate are more complex than just explained or that another mechanism is working. Hypothetically, the tether at the posterior side of the tibia, compensated by the hemi-epiphysiodesis, still exists after removal of the plate and causes the rebound mechanism. Ground for further research.

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ADDENDUM

Impact paragraph

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Clubfoot is a common condition worldwide. An estimated 175,000 children are born with clubfoot worldwide, with an estimated incidence of 1.18 per 1000.[1] Treatment is necessary and – fortunately – possible. Untreated, this deformity leads to severe physical restrictions.[2] Treatment is possible by means of the Ponseti method with (very) good results in most patients. After an initial good correction, there is still a significant group of patients who experience a relapse. Fortunately, the vast majority of these can be treated with re-casting or soft tissue procedures. Importantly, most patients are functionally performing well.[3] Recently, the Ponseti method has again been shown to be the method of choice for clubfoot patients.[4] With this knowledge, the Ponseti method is established as the preferred treatment worldwide. In some low- and middle-income countries, large projects have been registered to organize this clubfoot treatment for every child with clubfoot with good results.[5,6] In Western Europe, as in the Netherlands, the Ponseti method was introduced at the end of the last century. Initially, there was quite some resistance from orthopedic surgeons who used to solve the clubfoot pathology surgically. Remarkably in some African low income countries, this resistance seemed lower, resulting in relatively fast acceptance of the Ponseti method and accordingly good treatment results.[5,6] To resolve this discussion in the Netherlands, a scientifically based guideline was initiated, chapter 1 in this thesis. Not only did medical care change, but so did the organization of care and the hospital set-up. Not immediately or in a short time, but gradually, the care of children with clubfoot has changed for the better. Unfortunately, there is still some debate about details of treatment where surgeons or paramedics want to deviate from the 'standard' Ponseti method. This is where research again is needed. Randomized Controlled Trials (RCTs) - often considered the highest level of scientific evidence - are however relatively hard to initiate in clubfoot treatment. Aware of recent research where the Ponseti method shows superior results,[3,7] there is no need to conduct new studies with different approaches where the outcome may also be less favorable. Parents are already reluctant to blindly expose their baby to different treatment study protocols, especially when the known method shows good results. New gaps in scientific knowledge therefore need to be defined well in advance.

The focus of further improvement in clubfoot care should be to get rid of unwanted variation. Where improvements in the treatment itself are mentioned in the above paragraph, the organization of care can also be further optimized.

The Ponseti treatment was introduced in the Netherlands more than 20 years ago. Despite centralization, improved training and standardization of clubfoot treatment since its introduction, there is still unwanted variation. In the Western context, political, personal, financial and logistical barriers challenge the smooth introduction of new insights that contribute to optimal care. Parents need to be able to rely on the advice of the care provider, regardless of who is treating them or where. By using long-term registration, modern follow-up strategies and solving logistical problems with modern technology, we can help to further optimize clubfoot care. These registries should besides information on initial severity, treatment details and clinical measurements on outcome of treatment –such as range of motion– also incorporate aspects reflecting patient and parental perspectives. Chapters 4 and 5 showed that aspects such as a patient's function and quality of life give important insights that can be leading in changes within clubfoot care which may be necessary.

Issues to standardize care, complexity in defining residual deformities and the different options how to solve them discussed in chapter 2, 6 and 7 in this thesis, result in the assumption that clubfoot care should be considered as complex care. If one agrees on this, centralization improves the outcome.[8] Identifying the incidence of clubfoot in the Netherlands (chapter 2) was an important first step. To guide care in the future and before government regulations direct care for non-medical reasons, Dutch pediatric orthopedic surgeons should organize the number of clubfoot centers in relation to the number of treated clubfoot patients in a center. To overcome logistic problems in the future, two types of clubfoot centers could be established: some regular centers for the treatment and follow-up of uncomplicated clubfeet. These centers should closely work together with one or two highly specialized centers of last resort for complicated cases. All centers need to gain the necessary experience to detect relapse clubfoot patients (chapter 5) knowing when to refer these often complex pathologies to experienced specialists with special armamentaria of treatments (chapter 6).

Internationally, the problem of unwanted variance between centers is also real. If we could be an example to the world on how to further optimize clubfoot care and establish this with government and regulatory bodies, it would be of great value, especially for the patients and their parents.

Of course, detailed, uniform treatment protocols are essential to implement. Initiatives are now underway to use modern technology to improve care. Two

members of our study group are involved in an international expert panel to develop an application on a mobile phone to guide the treatment of clubfoot with the Ponseti method (Ponseti Clubfoot Navigator, PCN). The application is supported by algorithms coupled with artificial intelligence, based on available research or, if not available, on consensus of expert opinions. The application can guide colleagues in different hospitals in the region, but also far away in low-income countries. It can visually show patients their improvement and can guide as data storage. Privacy issues are of course important to solve. Integration in existing data or electronic patient files is also important in order not to generate new unnecessary effort needed to register. After the development phase, of course, implementation depends on financial resources, which could be an issue too. It is very important that initiatives like the PCN project get at least the possibility to show the strength where new technology can improve care and –if approved– get financial support to be implemented.

Another important pillar is the organization of unified education. Again, the parents' association gathered in Clubfoot World, together with clubfoot experts and supported by educational specialists, now organizes training courses including practical sessions in high-volume areas. All these aspects will ultimately improve the care of clubfoot and can serve as an example to other parts of the medical community.

The use of data will continue to be important. Although sometimes retrospective, we can evaluate treatments and identify areas for improvement. An easy-to-use, well-structured database system should be implemented, at least nationally but ideally internationally. Avoidance of duplicate registration where electronic patient records are also used is essential. Registration must take as little time as possible away from patient care. Scientific organizations, government and hospitals need to invest quickly in optimizing the data systems currently in use. The data will allow us to compare care and outcomes with other colleagues, even in different countries.

Although local settings and possibilities will differ, in clubfoot care the Ponseti method seems to be a common ground. For example, the guideline in the United Kingdom has a different base, but is very useful as comparison.[9] By using a Delphi process method to establish a solid consensus to set a benchmark for the best quality of care a different approach was used. The Delphi approach produces outcome with optimal support among parties involved, otherwise

sometimes consensus cannot be reached and/or is based more on consensus and less on scientific bases. The similarities with our scientific guideline were however remarkable. Grossly, the same advice was produced.

Of course, the well-being and functioning of the clubfoot patient must be paramount. Not only in young children, but also in adolescents. Chapters 3 and 4 were inspired by questions proposed by parents on the impact and outcome of the Ponseti treatment. Detailed research methods must be used to further investigate the quality of life and functioning, especially for clubfoot patients. From there, we need to identify patients' needs and deficiencies. Only then the final stage of solving these problems in clubfoot care can be tackled. National and international collaboration is needed to address some of the disabling deficiencies such as talar flattening or muscle imbalance. Also in this field technology may be useful. Movement analyses are widely used nowadays and identified clinically relevant impairments in clubfoot patients, such as limited ankle power and plantarflexion.[10,11] Although relevant, movement analyses also pose challenges for clinical practice. Gait laboratories of course are not always available. Not to mention the time needed to perform, analyze and interpret movement analyses. Clinical assessment such as the Clubfoot Assessment Protocol might be used to identify patients for who additional analyses might be worthwhile.[12] Further understanding of the relation between impairments identified in gait laboratories and patients' functional impairment and quality of life –discussed in chapter 4– is necessary. An important assignment where clinicians and scientists have to work closely together.

In conclusion, the treatment of clubfoot in the Netherlands has improved considerably over the last decade. This thesis presents a compilation of these changes and a guidance for further improvement. Articular surgical solutions have been abandoned and recurrent and residual deformities have been corrected with mostly soft tissue solutions. The guideline described in this thesis has been a tremendous help that caregivers could not neglect. Further centralization and optimization of clubfoot care and the search for improvements in care guidelines and training should be a priority. Unwanted and unnecessary variation should be completely reduced with the aim of improving the quality of life and functioning of children with clubfoot from infancy to maturity. If this thesis, which addresses important scientific questions about clubfoot, also forms an inspiration for further optimization of clubfoot care, then an important goal has been achieved.

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ADDENDUM

English summary

Enghlish summary

Clubfoot is an important congenital foot deformity. Not only its frequency, but also its disabling nature when untreated or treated with unsatisfactory results makes it a deformity that deserves all attention. This thesis deals with the changes in the treatment of clubfoot in the Netherlands. Soon after the introduction of the Ponseti method, a new form of manipulative treatment of congenital clubfoot, in the Netherlands, it became clear that a scientific approach was needed to implement this new treatment method.

Following a general introduction, this thesis can be divided into 3 main topics related to clubfoot care in the Netherlands, namely (1) organization of care, (2) treatment outcome and (3) recurrence detection and treatment. In the final chapters the main findings and implications for clinical practice of the studies presented in this thesis are discussed.

Organization of care

Since the introduction of the Ponseti method as a non-surgical method of treating clubfoot, it took until the 1990s for its popularity to grow worldwide. Around 2005, the treatment was also introduced in the Netherlands, but for some medical professionals who had been using surgical solutions for years, the choice between the two modalities was not obvious. Unwanted variation in provided care resulted in variable outcome and uncertainty by parents creating an urgent need for a scientifically based guideline for the initial treatment of patients with a clubfoot in the Netherlands, described in **Chapter 2**. The development of the guideline was initiated by the Parents' Association and supported by the Dutch Orthopaedic Association (NOV). The guideline committee consisted of medical professionals, members of the parents' association and a methodologist. The guideline was developed using an evidence-based approach, in which clinical questions were first formulated after in-depth discussion with all stakeholders. For all clinical questions, a systematic literature search was performed, the relevant results were evaluated and discussed in detail within the guideline committee. The frequent lack of high-quality evidence during the development of this guideline made the use of the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) methodology essential. The GRADE method considers not only the available scientific evidence, but also incorporate factors such as patient values and preferences and resource use. The guideline panel assessed the strength of

the recommendation. Several recommendations were formulated where the strength of the recommendations reflect the extent to which the guideline panel was confident that desirable effects outweigh the undesirable ones. The most important recommendation was to use the standard Ponseti treatment as the preferred treatment for primary idiopathic clubfoot. Also, different methods and materials were defined within the treatment strategy. Secondly, recommendations were formulated on patient-related factors (brace use, severity of the clubfoot deformity and registration). Classification preferences and diagnostic tools were described. The relationship between clubfoot and DDH was defined in a separate chapter. Finally, the organization of care was defined to provide the highest chance of an optimal treatment outcome.

The incidence of clubfoot varies worldwide. The incidence of clubfoot patients is an important factor for centralization of care, one of the recommendations of the guideline. As no central register of clubfoot deformities existed in the Netherlands, in **Chapter 3** a cross-sectional nationwide multicenter study on the incidence of clubfoot is presented. The 21 accredited clubfoot treatment centers were visited for data collection. To minimize sampling bias, all centers were visited in person and the records obtained were accurately reviewed. Children born between January 2013 and December 2014 with congenital clubfoot were included in the study described.

Using official reports from Statistics Netherlands (Central Bureau of Statistics), we obtained the number of 346 522 live births in the Netherlands. After excluding 16 duplicate patients, we found 377 unique patients with idiopathic clubfoot, born between January 2013 and December 2014. There was no significant difference in the incidence between the two years studied, 1.16 and 1.02 per thousand live births, respectively. We found more affected boys (66%), bilaterality in 50% of patients, and equally affected sides.

To verify the accuracy of our data, results were compared with the databases of Perined and EUROCAT-NNL. Based on the current study we found an incidence of 1.09 patients per thousand live births. This means that approximately 200 new clubfoot patients are born each year in the Netherlands.

Treatment outcome

Bracing is an essential part of Ponseti's treatment for clubfoot. At the same time, it can be intense for parents and their child. In the clinical setting, parents often

ask questions about the impact of the brace on the child's quality of life. In **Chapter 4** we studied the health-related quality of life (HRQoL) of clubfoot patients during the brace period. The TNO-AZL Preschool Children's Health related Quality of Life (TAPQOL) questionnaire was presented to parents of clubfoot patients during and after the brace period. A reference sample was available through the Netherlands Organization for Applied Scientific Research (TNO). The TAPQOL consists of 12 subscales comprising the subscales physical, social, emotional and cognitive functioning.

Responses of 80 parents of clubfoot patients and 238 parents of healthy controls was available for analysis. On average both the healthy controls and clubfoot patient groups showed high scores on all domains of the TAPQOL questionnaire. The highest scores were recorded for positive mood and liveliness, and the lowest for problem behavior and sleep. However, clubfoot patients scored significantly lower in the overall group on the subscales motor functioning, sleeping, lung problems and skin problems. Especially, young clubfoot patients (1 and 2 years old) showed significantly lower scores on the sleeping subscale compared to their aged-matched healthy controls while the 2-3- and 3–4-year-old clubfoot patients scored significantly lower on the motor functioning subscale. During the first year after the end of the brace period, no differences were found compared to healthy controls.

The results of this study can be used to counsel parents on what they might expect during the brace phase of the Ponseti treatment. Considering the overall high level of HRQoL in clubfoot patients, the findings may alleviate some of the concerns that parents have.

The functional outcome of treatment can be defined in different ways. Relevant although underexposed aspects are participation levels and motor abilities in children with clubfoot. Therefore, **Chapter 5** describes the level of parents' perceived motor ability and participation of 86 Ponseti treated children with clubfeet compared with 62 age-matched healthy controls. Two questionnaires were sent to the parents: the Assessment of Life Habits for Children (LIFE-H), developed and validated to assess social participation and the Dutch translation of the Movement Assessment Battery for Children-2 (MABC-2) checklist to assess motor abilities. In general, children with clubfoot and controls scored high on the LIFE-H and MABC-2. In terms of perceived motor abilities within the clubfoot group we found a significant difference in movements in a dynamic

and/or unpredictable environment. Higher levels of participation proved to be correlated with better motor abilities. No significant differences were found between children with unilateral or bilateral clubfoot according the by parents perceived motor abilities.

This study of perceived participation and motor ability differs substantially from previous studies in which health professionals assessed ability at a point in time. We conclude that although the differences in perceived motor ability and participation are small and the overall scores are high, the large variation in the scores of clubfoot patients indicates that some patients have problems. We suggest that there may be a relationship with relapse or severity of clubfoot in general.

Relapse

Although the Ponseti method is widely accepted as the treatment of choice for clubfoot, residual deformities and recurrences occur. Clinical practice and literature is however faced with a variety of definitions, identification and treatment strategies. **Chapter 6** describes our study of 88 patients with residual or relapse deformity. We performed a retrospective dossier study and included patients with idiopathic clubfoot with residual or recurrent feet who required additional treatment. Residual feet were defined as deformities that were never fully corrected after initial treatment, whereas relapses were defined as any feature of clubfoot that recurs after successful initial treatment.

Not only static deformities such as adduction, equinus, varus and cavus but also dynamic patterns such as active supination were recorded in recurrent clubfoot. The combination decreased dorsiflexion and adduction occurred most often. In more than half of the patients a combination of deformities was present. Although half of the residual and relapsed clubfoot were identified before the age of 4 years, relapses occurred in all stages of treatment and follow up. This leads to the recommendation to follow clubfoot patients until the end of growth.

Three treatment groups were composed: patients who were treated with (1) the extended Ponseti protocol (2) an extra brace adaptation or (3) additional surgery. The Ponseti protocol was mainly applied in residual and relapsed feet up to the age of 5. The proportion of brace adaptations increased up to 6 but was not advised anymore in patients over 9 years of age. Additional surgery was not influenced by age.

Overall, this study showed that early recognition of defined residual and recurrent deformities is essential and attention to the sometimes complex combination of deformities is necessary. Due to the heterogeneous nature of residual and relapsed clubfeet, treatment should be individualized with a focus on the extended Ponseti treatment.

When treatment options within the extended Ponseti protocol are insufficient to treat severe recurrent or residual deformities, additional surgical procedures might be necessary. Considering the risk of prolonged rehabilitation and complications such as join-stiffness, extra-articular solutions such as distal tibial hemi-epiphysiodesis are advised. In **Chapter 7** we describe 31 affected children with 45 relapsed clubfeet. A subgroup of 23 feet were followed at least one year after removal of the small plate used. Treatment effect was assessed both radiologically –by means of the Anterior Distal Tibial Angle (ADTA)– and clinically –by means of ankle dorsiflexion.

The presence of flatting of the talus and the heterogenous treatment history of the patients following initial Ponseti treatment, indicate the complexity of the patient group.

During hemi-epiphysiodesis the ADTA significantly decreased. This implies a sagittal slope change of the distal tibia that places the foot indirectly more in a dorsiflexed position. However, there was a notable rebound effect after at least one year of follow-up, where the ADTA returned significantly to pre-implantation levels. The median dorsiflexion measured at the time of plate removal showed a significant change ($p<0.01$), but when measured after at least one year of follow-up, no significant change was found. We conclude that hemi-epiphysodesis of the distal tibia in recurrent clubfoot has its effect on the distal tibia, but if growth remains there is an important rebound phenomenon. Although not quantified, many children in clinic, after hemi-epiphysiodesis and before removal of the 8-plate, report experiencing more heel strain - and sometimes even experiencing temporary discomfort from this. They also sometimes report being better at certain activities such as climbing stairs. Factors to consider in follow-up research. This rebound phenomenon should be considered when deciding the timing of treatment. If the child needs surgery at a young age, parents and patients should be informed of this mechanism, which may result in the need for a repeat hemi-epiphysiodesis.

Overall, the aim of this thesis was to outline the changes in clubfoot treatment since the introduction of the Ponseti method in the Netherlands and to share the scientific evidence needed to improve care and implement changes. All to improve patient care, where correct treatment of a severe foot deformity will in most cases result in an excellent functional outcome. I hope that this thesis will motivate fellow (pediatric-)orthopedic surgeons to continue research into clubfoot until all questions are answered and problems solved.

ADDENDUM

Dutch summary, Nederlandse samenvatting

Dutch summary, Nederlandse samenvatting

De klompvoet is een belangrijke congenitale voetdeformiteit. De afwijking komt niet alleen frequent voor, ook de invaliderende effecten wanneer deze niet of niet afdoende wordt behandeld, maakt het een aandoening die aandacht verdient. Dit proefschrift beschrijft de veranderingen in de behandeling van de klompvoet in Nederland. Na de introductie van de Ponseti methode, een destijds nieuwe (gips) redressie techniek voor de behandeling van de aangeboren klompvoet, in Nederland, bleek een wetenschappelijke onderbouwing nodig om deze behandelmethode ook daadwerkelijk te implementeren.

Na een algemene introductie, is deze thesis ingedeeld in 3 onderdelen, gerelateerd aan de klompvoet zorg in Nederland. Achtereenvolgens worden behandeld: (1) organisatie van zorg, (2) behandelresultaten en (3) het diagnosticeren van terugval en de behandeling daarvan. In de laatste hoofdstukken worden de belangrijkste resultaten van de diverse studies en de betekenis voor de klinische praktijk besproken.

Organisatie van zorg

Na de introductie van de Ponseti methode, als niet-operatieve methode om een klompvoet te behandelen, duurde het tot 1990 voordat de behandeling wereldwijd aandacht kreeg. Rond 2005 werd de behandeling geïntroduceerd in Nederland. Voor enkele gevestigde kinder-orthopedisch chirurgen, die gewend waren de klompvoet chirurgisch te behandelen, was de overgang naar de Ponseti techniek niet vanzelfsprekend. Er ontstond een ongewenste variatie in behandeling die leidde tot wisselende resultaten en onzekerheid bij ouders. Hierdoor ontstond er een sterke vraag naar een wetenschappelijk onderbouwde richtlijn voor de initiële behandeling van de idiopathische klompvoet in Nederland, beschreven in **Hoofdstuk 2**. De ontwikkeling van deze richtlijn werd geïnitieerd door de oudervereniging van ouders van kinderen met klompvoetjes (Stichting Klompvoet Nederland) en ondersteund door de Nederlandse Orthopaedische Vereniging (NOV). De richtlijn commissie bestond uit (kinder-) orthopedisch chirurgen, leden van de oudervereniging en een methodoloog van het Kennisinstituut van de Federatie Medisch Specialisten.

Bij het ontwikkelen van de richtlijn werd gebruik gemaakt van een evidence-based methode waarbij in eerste instantie klinische vragen worden geformuleerd, voortkomend uit een discussie met een vertegenwoordiging van

stakeholders. Voor alle vragen werd een literatuuronderzoek uitgevoerd waarna deze systematisch werd geanalyseerd. De relevante uitkomsten van literatuuronderzoek werden vervolgens in detail besproken binnen de richtlijn commissie. Het regelmatig ontbreken van hoogwaardig wetenschappelijk bewijs, maakte het gebruik van de Grading of Recommendations Assessment, Development, and Evaluation (GRADE) methode, essentieel. Deze systematische en transparante methode beoordeelt niet slechts de beschikbare wetenschappelijk bewijskracht, maar incorporeert ook factoren als de mening en voorkeuren van de patiënt, kosten en organisatie van zorg. Het beschikbare wetenschappelijk bewijs samen met overwegingen die voortkomen uit de beoordeelde factoren leidt dan tot aanbevelingen, gegradeerd in sterkte door de richtlijn commissie. Deze sterkte reflecteert het vertrouwen van de richtlijn commissie dat de gewenste effecten van de interventie opwegen tegen de ongewensten. De belangrijkste aanbeveling van de in dit proefschrift beschreven richtlijn, betreft het advies de standaard Ponseti behandeling te gebruiken voor de primaire idiopathische klompvoet. Verder worden er verschillende technieken en materialen gedefinieerd binnen de behandeling. Er worden aanbevelingen gedaan betreffende patiënt-gerelateerde factoren zoals het brace gebruik en classificatie van de ernst van de klompvoet. Voorkeuren ten aanzien van registratie systemen en het gebruik van aanvullend diagnostisch onderzoek worden beschreven. De relatie tussen de klompvoet en heupdysplasie wordt in een apart hoofdstuk beschreven. Tot slot wordt de organisatie van de klompvoetzorg beschreven waarin de voorwaarden worden gedefinieerd om een optimaal behandelresultaat te behalen.

De klompvoetincidentie varieert wereldwijd. Wanneer men centralisatie van de klompvoetzorg nastreeft, een aanbeveling uit de richtlijn in hoofdstuk 1 beschreven, is het essentieel te weten hoeveel nieuwe klompvoet patiëntjes er überhaupt in Nederland geboren worden. Omdat er geen centraal register voor klompvoet patiëntjes in Nederland voorhanden was, beschrijft **Hoofdstuk 3** een cross-sectionele nationale multicenter studie, opgezet ter bepaling van de incidentie van nieuwe patiëntjes in Nederland. De 21 klompvoetcentra in Nederland werden bezocht voor datacollectie. Om sampling bias te voorkomen werden alle centra persoonlijk bezocht door dezelfde persoon die de patiëntendossiers stuk voor stuk bestudeerde. Kinderen met congenitale klompvoeten, geboren tussen januari 2013 en december 2014 werden geïncludeerd in de studie.

Volgens het Bureau van de Statistiek (CBS) werden er in Nederland tijdens de studieperiode 346 522 kinderen levend geboren. Na exclusie van 16 dubbel geregistreerde patiënten vonden we 377 unieke patiënten met idiopathische klompvoeten tussen januari 2013 en december 2014. Er werd geen significant verschil gezien tussen de twee bestudeerde jaren, respectievelijk 1.16 en 1.02 klompvoet patiënten op 1000 levendgeborenen. Jongens (66%) waren vaker aangedaan dan meisjes, dubbelzijdig voorkomen van de klompvoet werd in 50% gevonden en er was geen verschil in frequentie tussen links- en rechtszijdigheid. Om de nauwkeurigheid van de gevonden data te verifiëren, zijn de verzamelde data vergeleken met de gegevens van Perined en Eurocat-NNL.

Gebaseerd op deze studie stelden we een incidentie vast van 1.09 idiopathische klompvoet patiënten per 1000 levendgeborenen. Dit betekent dat er elk jaar ongeveer 200 nieuwe klompvoet patiëntjes in Nederland geboren worden.

Behandel uitkomsten

Het gebruik van een voet-abductiebrace is een onmisbaar onderdeel van de Ponseti behandeling. Tegelijkertijd kan de brace behandeling belastend zijn voor zowel patiënt als ouders. In de behandelkamer vragen ouders vaak naar de impact van de brace op het functioneren van het kind. In **Hoofdstuk 4** worden de resultaten beschreven van een studie naar de kwaliteit van leven bij klompvoet patiëntjes behandeld met een voet-abductie brace tijdens de Ponseti methode. De TNO-AZL Preschool Children's Health related Quality of Life (TAPQOL) vragenlijst werd gebruikt om de gezondheid gerelateerde kwaliteit van leven te meten. De TAPQOL-vragenlijst beschikt over 12 onderdelen die verschillende aspecten van het kind onderzoekt en wordt door de ouders ingevuld. Zo worden fysieke klachten, motorisch functioneren, sociaal functioneren, cognitief functioneren en psychologisch functioneren bevraagd. De resultaten van patiëntjes met een klompvoet werden vergeleken met de Nederlands referentiegegevens beschikbaar via TNO (Nederlandse Organisatie voor toegepast-natuurwetenschappelijk onderzoek). Ouders van 80 klompvoet patiënten en 238 ouders uit de controlegroep vulden de vragenlijst in. In het algemeen scoren zowel de klompvoet patiënten als de controlegroep kinderen hoog op alle onderdelen van de TAPQOL-vragenlijst. De hoogste scores werden gezien bij de onderdelen positieve emoties en levendigheid. De laagste sores werden gezien bij probleemgedrag en slaapproblemen. In de groep als geheel, scoorden klompvoet patiënten lager in de subgroepen motorisch functioneren, slaap-, long- en huidproblemen. Met name jonge klompvoet patiëntjes (leeftijd 1-

2 jaar) lieten lagere scores zien bij het onderdeel slaap in vergelijking met de leeftijd genoten uit de controlegroep. De groep van de 2-3 en 3-4-jarigen scoorden significant lager op de motorisch functioneren sub schaal. Tijdens het eerste jaar na het staken van de brace therapie, werden er geen verschillen meer gezien tussen de klompvoet en de controle kinderen.

Deze resultaten kunnen worden gebruikt om ouders te informeren omtrent hun verwachtingen ten aanzien van het brace gebruik van hun kind binnen de Ponseti behandeling. Wanneer men de, overall hoge, scores binnen de gezondheids-gerelateerde kwaliteit van leven in ogenschouw neemt, kunnen deze mogelijke zorgen van ouders deels weggenomen worden.

De functionele uitkomsten van de klompvoet behandeling zijn op verschillende manieren te definiëren. Belangrijke, maar nog onderbelichte aspecten van deze functionele uitkomsten zijn het niveau van participatie en motorische vaardigheden van kinderen met klompvoeten.

Dit was de reden om de, in **Hoofdstuk 5** beschreven, studie te starten naar de door de ouders ervaren motorische vaardigheden en participatie van hun kind. Data van 86 klompvoet patiëntjes, behandeld met de Ponseti methode werd vergeleken met data van 62, even oude, gezonde kinderen als controlegroep.

Er werden twee vragenlijsten naar de ouders verstuurd: de Nederlandse versies van Assessment of Life Habits for Children (LIFE-H), ontwikkeld en gevalideerd om sociale participatie te meten en de Movement Assessment Battery for Children-2 (MABC-2) checklist die motorische vaardigheden evalueert. Zowel kinderen met een klompvoet als die uit de controlegroep, scoorden hoog op de LIFE-H en MABC-2 meetinstrumenten. Ouders scoorden kinderen met een klompvoet significant slechter op bewegingen in een dynamische en/of onbekende omgeving. Binnen de totaalscores in de gehele studiegroep werd een toegenomen participatie geassocieerd met beter ervaren motorische vaardigheden. Of een kind enkel- of dubbelzijdige klompvoeten had, maakte geen significant verschil ten aanzien van, de door ouders ervaren, motorische vaardigheden en participatie.

Deze studie naar de door ouders ervaren participatie en motorische vaardigheden van hun kinderen met klompvoeten verschilt aanzienlijk van eerder uitgevoerde studies waarbij zorgverleners op een bepaald tijdstip de

mogelijkheden van het kind onderzochten. Ook al zijn de in de studie gevonden verschillen in motorische vaardigheden en participatie klein en de overall scores hoog, de grote variatie in de scores van de klompvoet groep geeft aan dat er wel degelijk problemen bij deze patiënten voorkomen. Een mogelijke verklaring zou kunnen liggen in het vóórkomen van relapses in de klompvoet groep en/of de ernst van de klompvoet in het algemeen.

Terugval (relapse)

Hoewel de Ponseti methode algemeen geaccepteerd is als behandeling van keuze bij klompvoeten, komen persistente afwijkingen en terugval voor. Deze deformiteiten zijn echter in de literatuur niet uniform omschreven met verschillende definities en behandelstrategieën. **Hoofdstuk 6** beschrijft onze studie waarin 88 klompvoet patiënten worden beschreven met persistente deformiteiten of terugval. We voerden een retrospectieve dossier studie uit waarbij patiënten met idiopathische klompvoeten werden geïncludeerd met persistente klompvoet afwijkingen en klompvoet patiënten die een terugval vertoonden. De klompvoeten die altijd afwijkingen waren blijven vertonen ondanks behandeling, werden gedefinieerd als residu klompvoeten. Klompvoeten die aanvankelijk wel volledig werden gecorrigeerd maar later terugval vertoonden waarbij een of meerdere klompvoet kenmerken werden vastgesteld, definieerden we als relapse voeten.

In de relapse voeten werden niet alleen statische afwijkingen als adductie, equinus, varus en cavus gezien, ook dynamische afwijkingen als actieve supinatie werden gevonden. De combinatie beperkte dorsaalflexie/equinus en adductie werd het meest gezien. Hoewel de meeste relapse en residu klompvoeten voor de leeftijd van 4 jaar werden gedetecteerd, werden relapses in alle behandelstadia en op elke leeftijd gezien. Vandaar de aanbeveling de klompvoet patiënt te vervolgen tot het einde van hun (voet)groei.

Er werden 3 behandelgroepen gedefinieerd: kinderen behandeld volgens (1) een uitgebreid Ponseti protocol (inclusief re-achillespees tenotomie en/of M. Tibialis Anterior transpositie), (2) een verlengd of hernieuwd brace protocol of (3) chirurgische correctie. Optie (1) werd vooral toegepast bij kinderen tot 5 jaar. Soms werd het (hernieuwd) dragen van de voet-abductie brace geadviseerd bij kinderen tot 6 jaar. Boven de leeftijd van 9 jaar werd het dragen van de brace niet meer geadviseerd. Chirurgische correcties konden op alle leeftijden uitgevoerd worden.

Concluderend laat deze studie zien dat vroege herkenning van klompvoet afwijkingen bij residu of relapse klompvoeten essentieel is en dat aandacht voor de soms complexe combinaties van afwijkingen nodig is. Door het heterogene aspect van residu of relapse klompvoeten, moet de behandeling afgestemd worden op de individuele patiënt met, zo mogelijk, de nadruk op het uitgebreide Ponseti protocol.

Als de behandelingen volgens het uitgebreid Ponseti protocol onvoldoende toereikend zijn kan het noodzakelijk zijn additionele chirurgie uit te voeren. Met in het achterhoofd het langdurige herstel en mogelijke complicaties zoals stijfheid bij intra-articulaire chirurgie, worden bij voorkeur extra-articulaire chirurgische opties zoals hemi-epifysiodese uitgevoerd. In **Hoofdstuk 7** wordt de studie beschreven waarin 31 relapse kinderen met 45 klompvoeten een hemi-epifysiodese hebben ondergaan. Een subgroep met een minimale follow-up van 1 jaar na het verwijderen van het epifysiodese plaatje, werd gevolgd om het lange termijneffect na verwijdering te meten. Het behandeleffect werd zowel radiologisch (met behulp van de Anterior Distal Tibial Angle (ADTA) als klinisch gemeten.

De onderzochte populatie bestond uit een complexe groep klompvoet patiënten, zich uitend in afvlakking van het sprongbeen (talus) die aanwezig bleek bij alle patiënten. Tevens was er een grote diversiteit aan reeds toegepaste behandelingen bij deze patiëntengroep.

Gedurende de hemi-epifysiodese nam de ADTA significant af. Deze correspondeert met een sagittale slope verandering van het tibia plafond waarbij indirect de voet in meer dorsaalflexie komt te staan. Belangrijke bevinding echter was het reboundeffect in de follow-up groep waarbij de mediane ADTA terugging naar de uitgangsituatie. Concluderend stellen we dat een groei beïnvloeding middels distale hemi-epifysiodese van de distale tibia zijn effect heeft maar dat bij de resterende groei na het verwijderen van het implantaat, een ongewenst rebound fenomeen optreedt. De mediane dorsaalflexie toonde eenzelfde beeld waarbij tijdens de hemi-epifysiodese een significante verandering werd gevonden. In de follow up groep kon ten aanzien van de dorsaalflexie geen verschil worden aangetoond. Hoewel niet gekwantificeerd, vertellen veel kinderen in de spreekkamer na een hemi-epifysiodese en voor het verwijderen van de 8-plate, meer belasting te ervaren van de hiel (en soms hier zelfs last van te ervaren). Ook melden ze soms bepaalde activiteiten zoals

traplopen beter te kunnen uitvoeren. Factoren om in vervolg-onderzoek mee te nemen.

Hemi-epifysiodese bij relapse klompvoeten leidt tot een verandering van de ADTA. Het rebound fenomeen na verwijdering van het materiaal bij patiënten met open groeischijven dient wel in ogenschouw genomen te worden. Het kan invloed hebben op de timing van de ingreep en/of op de voorlichting richting ouders waarbij een mogelijk hernieuwd plaatsen van een groeiplaatje kan worden besproken.

Samenvattend beschrijft dit proefschrift de veranderingen binnen de klompvoetzorg, sinds de introductie van de Ponseti methode in Nederland. Zij beschrijft de wetenschappelijke onderbouwing die nodig was om de klompvoet zorg in Nederland te verbeteren en de veranderingen voorgesteld, te onderbouwen. Met altijd als doel de patiëntenzorg te optimaliseren zodat door een correctie behandeling in de meeste patiënten een uitstekend functioneel resultaat kan worden bereikt. Ik hoop dat dit proefschrift collega (kinder)orthopedisch chirurgen kan motiveren het wetenschappelijk onderzoek naar klompvoet problemen voort te zetten totdat alle vragen beantwoord zijn en alle problemen zijn opgelost.

ADDENDUM

Dankwoord

Dankwoord (Acknowledgement)

Hoewel ik enorm trots ben op de inhoud van dit proefschrift en het oprecht voor velen lezenswaardig vind, is de realiteit dat het dankwoord toch erg populair onder vele lezers zal zijn. Gaat u er dus maar even rustig voor zitten. Een andere realiteit is dat aan een proefschrift dat zo lang onderweg is geweest, heel veel mensen, al dan niet bewust, hebben meegewerkt met het risico dat ik incompleet in mijn dankwoord zou kunnen zijn. Bij voorbaat excuses daarvoor en alsnog heel veel dank!

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As often mentioned by me: I have two lives within the orthopedic world. The clubfoot world and the pediatric trauma world. Although these two worlds often live separately, for me many friends in the pediatric trauma (AO) world are a huge inspiration to combine clinical care with research and education. Especially my big friend Unni Narayanan from Toronto, Canada and all members of the pediatric trauma groups in Davos, Leeds and of course Oisterwijk are so important that I want to mention them here. Co-chairs in AO courses as Michiel Verhofstadt, Rogier Simmermacher, Job Doornberg and especially my soul mate Taco Bijlsma gave such a huge amount of energy I never would have missed and still will enjoy in the future! I am very honored to have Prof. Unni Narayanan form the Hospital of Sick Children in Toronto, as an inspirer, friend but also as a representative of this live in the Corona. Thank you so much for sharing this great moment!

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ADDENDUM

Curriculum vitae

Curriculum vitae

Arnold Timotheus Besselaar was born in Eindhoven on 30 December 1964. He graduated from high school (Eindhovens Protestant Lyceum) in 1984 and was trained as a physiotherapist at the Academie Fysiotherapie 'Thim van der Laan' Utrecht in 1984-1988. He finished his medical study at the University of Utrecht in 1996 and started his training as a general practitioner in Utrecht in 1997 but switched after a short but very inspiring period at the orthopedic department in ‘s-Hertogenbosch, to be trained in orthopaedic surgery in 1999 (1999-2001: general surgery under supervision of dr. Ton Hoofwijk, 2001-2005: Orthopedic Surgery under supervision of dr. Jan van Mourik). He then completed a fellowship in pediatric orthopedic surgery at the Wilhelmina Children's Hospital in Utrecht under supervision of Prof. dr. René Castelein.

In 2006 he started as a consultant in (pediatric) orthopaedic surgery and traumatology at the Catharina Hospital Eindhoven. After the merger of the orthopedic groups of the Máxima Medical Centre and the Catharina Hospital, he continues to work with great pleasure in the 'Orthopedie Groot Eindhoven' group.

In 2014 (after 3 years of leading the guideline group on clubfoot in the Netherlands) he didn't know that the publication of the clubfoot guideline would be the start of this thesis. This was partly due to the many different roles and activities that made his life in pediatric orthopedics and trauma so interesting. He published in various trauma books, was faculty and chair in many different AO courses (national and international) and was chair of the AO Principles and Pediatric course in Oisterwijk for many years. With a special interest in education, he joined the AO Netherlands Board as Education Officer. He had the opportunity to lecture on various topics all over the world and in recent years at the Medical University of Maastricht. He has led several clubfoot courses in the Netherlands with great support from the plaster physician groups in both hospitals.

He is part of an enthusiastic group of researchers looking for answers in clubfoot pathology led by dr. Marieke van der Steen and dr. Wai-Yan Liu and is part of a large international clubfoot expert group initiated by Prof. Shafique Pirani.

In his spare time, he likes to ski as much as possible and back home he enjoys playing tennis, cycling and sailing on the 'Zeeuwse wateren', all activities preferably with his family and/or good friends.

He is happily married to Eveline and together they have two wonderful children: Lotte and Tobias.


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