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Early Results of the Ponseti Method for the Treatment of Clubfoot in Distal Arthrogyryposis

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Investigation performed at Washington University School of Medicine, St. Louis; St. Louis Shriners Hospital for Children, St. Louis; St. Louis Children's Hospital, St. Louis, Missouri; and Altonaer Kinderkrankenhaus, Hamburg, Germany

Background: Clubfoot occurs in approximately one in 1000 live births and is one of the most common congenital birth defects. Although there have been several reports of successful treatment of idiopathic clubfoot with the Ponseti method, the use of this method for the treatment of other forms of clubfoot has not been reported. The purpose of the present study was to evaluate the early results of the Ponseti method when used for the treatment of clubfoot associated with distal arthrogyryposis.

Methods: Twelve consecutive infants (twenty-four feet) with clubfoot deformity associated with distal arthrogyryposis were managed with the Ponseti method and were retrospectively reviewed at a minimum of two years. The severity of the foot deformity was classified according to the grading system of Diméglio et al. The number of casts required to achieve correction was compared with published data for the treatment of idiopathic clubfoot. Recurrent clubfoot deformities or complications during treatment were recorded.

Results: Twenty-two clubfeet in eleven patients were classified as Diméglio grade IV, and two clubfeet in one patient were classified as Diméglio grade II. Initial correction was achieved in all clubfeet with a mean of 6.9 ± 2.1 casts (95% confidence interval, 5.6 to 8.3 casts), which was significantly greater than the mean of 4.5 ± 1.2 casts (95% confidence interval, 4.3 to 4.7 casts) needed in a cohort of 219 idiopathic clubfeet that were treated during the same time period by the senior author with use of the Ponseti method ($p = 0.002$). Six feet in three patients had a relapse after initial successful treatment. All relapses were related to noncompliance with prescribed brace wear. Four relapsed clubfeet in two patients were successfully treated with repeat casting and/or tenotomy; the remaining two relapsed clubfeet in one patient were treated with extensive soft-tissue-release operations.

Conclusions: Our early-term results support the use of the Ponseti method for the initial treatment of distal arthrogyryptic clubfoot deformity. Longer follow-up will be necessary to assess the risk of recurrence and the potential need for corrective clubfoot surgery in this patient population, which historically has been difficult to treat nonoperatively.

Level of Evidence: Therapeutic Level IV. See Instructions to Authors for a complete description of levels of evidence.

Clubfoot occurs in approximately one in 1000 live births¹ and is one of the most common congenital birth defects. It is easily recognizable at birth and can be differentiated from some of the more common positional foot anomalies on the basis of the rigid ankle equinus deformity and resistance to simple passive correction. Most clubfeet occur as an isolated birth defect and are considered idiopathic. The Ponseti method of serial casting to gradually correct the

deformity, combined with a percutaneous tenotomy of the Achilles tendon to correct ankle equinus followed by several years of bracing to maintain the correction, has gained widespread popularity in recent years for the treatment of idiopathic clubfoot²⁻⁵. When treatment with the Ponseti method is initiated in the first few years of life, patients with idiopathic clubfoot deformity rarely require extensive surgical treatment and commonly have excellent long-term outcomes⁶.

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Nonidiopathic clubfoot occurs in patients with additional malformations, chromosomal abnormalities, or known genetic syndromes, such as distal arthrogyriposis. Although there have been multiple reports of successful treatment of idiopathic clubfoot with use of the Ponseti method^{2,3,5}, the use of this treatment for the treatment of nonidiopathic clubfoot has not been reported, to our knowledge. On the contrary, nonidiopathic clubfoot is often treated primarily with extensive soft-tissue-release surgery because the deformities in these patients are thought to be too rigid to correct with casting alone⁷.

Arthrogyriposis has been described as a feature of >100 syndromes and is characterized by contractures of two or more different body areas⁸. Currently, ten different distal arthrogyriptic syndromes have been classified; all are characterized by hand and foot contractures, limited proximal involvement, and autosomal dominant inheritance⁹. The traditional treatment for clubfoot associated with these syndromes is either a radical soft-tissue release¹⁰⁻¹⁴ or talectomy¹⁵⁻¹⁷, both of which have been associated with mixed results. A high rate of recurrence has been reported after radical soft-tissue release for the treatment of arthrogyriptic clubfoot^{11-13,18}, with this recurrence leading to repeat soft-tissue releases or salvage procedures.

The purpose of the present study was to evaluate the early results of the Ponseti method for the treatment of clubfoot associated with distal arthrogyriposis.

Materials and Methods

Twelve consecutive infants with bilateral clubfoot deformity (twenty-four clubfeet) associated with distal arthrogyriposis were managed with the Ponseti method¹⁹, and the cases of these patients were retrospectively reviewed. Ten patients were managed at St. Louis Shriners Hospital for Children by one orthopaedist (M.B.D.), and two patients were managed at Altonaer Kinderkrankenhaus (Hamburg, Germany) by another orthopaedist (M.F.S.). All patients underwent evaluation by a clinical geneticist to confirm the diagnosis of distal arthrogyriposis. Five patients were male, and seven patients were female. The average age (and standard deviation) at the time of presentation to our clinics was 3.7 ± 4.2 months. Eight patients were managed at our institutions from the start, and four patients (33%) had had plaster-cast treatment elsewhere before their initial visit; the precise techniques of manipulation or casting that had been used for the latter four patients are unknown. No patient was lost to follow-up. The average age at the time of the latest follow-up was 32.3 ± 10.6 months. Institutional review board approval was obtained for this study.

The four patients who initially had been managed by other orthopaedists had been managed with a mean of 14.5 ± 4.2 plaster casts before referral. Some of these casts were below-the-knee casts, whereas others were a combination of below-the-knee and above-the-knee casts. No patient had had a percutaneous tenotomy of the Achilles tendon before presentation to us. All four patients experienced partial or complete cast slippage resulting in a complex clubfoot, an entity recently described by Ponseti et al.²⁰.

The severity of the foot deformity was classified according to the grading system of Diméglio et al.²¹ at the time of presentation. In this grading system, four parameters are assessed on the basis of reducibility with gentle manipulation as measured with a handheld goniometer: (1) equinus deviation in the sagittal plane, (2) varus deviation in the frontal plane, (3) derotation of the calcaneopedal block in the horizontal plane, and (4) adduction of the forefoot relative to the hindfoot in the horizontal plane. A score is given to each of the four parameters on a 4-point scale, with 4 points indicating reducibility from 90° to 45° ; 3 points, reducibility from 44° to 20° ; 2 points, reducibility from 19° to 1° ; 1 point, reducibility from 0° to -20° ; and 0 points, reducibility of less than -20° . The sum of these parameters constitutes a 16-point scale. Four additional points, consisting of 1 point each for a posterior crease, a medial crease, the presence of forefoot cavus, or poor muscle condition (such as fibrous, hypertonic, or contracted triceps, tibial, or peroneal muscles and the absence of voluntary dorsiflexion in eversion and pronation) can be added, for a total of 20 points. Atrophy of the calf, a short foot, and decreased limb length are not scored as they are considered part of the natural history of clubfoot deformity. The foot can then be classified into four categories with respect to the severity of the deformity. Grade-I feet have a mild deformity that is >90% reducible, with a score of 0 to 5 points. Grade-II feet have a moderate deformity, with a score of 6 to 10 points. Grade-III feet have a severe deformity, with a score of 11 to 15 points. Grade-IV feet have a very severe deformity, with a score of 16 to 20 points.

Charts were reviewed for demographic data, including gender, bilaterality or unilaterality of clubfoot, and age at the time of onset of treatment. The number of infants who received treatment before referral and the type of treatment performed were recorded. Other data that were noted include the number of casts required for initial correction, complications (cast slippage), the need for a percutaneous Achilles tenotomy to obtain correction of the residual equinus contracture, and compliance with foot abduction bracing. Reports by the family with regard to the use of the brace were used as a measure of compliance. Correction was defined as a plantigrade foot with no residual forefoot adduction, forefoot cavus, or hindfoot varus and a minimum of 5° of passive ankle dorsiflexion. After correction was obtained, passive ankle dorsiflexion and plantar flexion as well as varus-valgus heel deformity were measured by a single examiner (M.B.D. and M.F.S.) at each institution with use of a handheld goniometer. Recurrent deformities were documented with regard to the age at the time of recurrence and any additional treatment necessary to regain correction.

Classification of Arthrogyriposis

An attempt was made to classify all twelve patients into a known subtype of distal arthrogyriposis on the basis of clinical characteristics and the results of blood tests when appropriate. All patients were evaluated by a pediatric geneticist, and seven of the patients were also seen by a pediatric neurologist. Seven patients (58%) were classified as distal arthrogyriposis type 1

(DA1A), which is characterized by clubfeet, adducted thumbs, ulnar deviation of the metacarpophalangeal joints, normal facies, and normal intelligence. Three patients were classified as distal arthrogyposis type 2B (DA2A), a variant of Freeman-Sheldon syndrome, which is characterized by clubfeet, a distinct facies, and flexion and ulnar deviation of the fingers. Of the remaining two patients, one was classified as distal arthrogyposis type 4 (DA4) and one was classified as distal arthrogyposis type 5 (DA5). DA4 is characterized by the presence of progressive scoliosis, short stature, camptodactyly, and clubfeet. DA5 is characterized by clubfeet, limited ocular motility, and flexion deformities of the interphalangeal joints.

All patients had motor delays typical of patients with distal arthrogyposis. The mean age at which patients started walking was eighteen months (range, thirteen to twenty-two months). In addition, all patients were receiving occupational and physical therapy. Although a formal grading of motor strength in the lower extremities was not done in this patient cohort, generalized muscle weakness was noted. At the time of the latest follow-up, three patients had no detectable active function of the muscles in the anterior compartment of the leg bilaterally. These patients were managed with a solid ankle-foot orthosis for walking in addition to the foot abduction bracing at nighttime.

Treatment Method

Eight (67%) of the twelve patients were managed with the Ponseti method according to the published protocol^{12,22-24}. Four (33%) of the twelve patients were managed with a modified Ponseti method recommended for the treatment of complex idiopathic clubfoot²⁰. In general, the Ponseti method of treatment is started within the first few weeks of life, if possible. However, in the case of a premature infant with clubfoot, the initiation of casting is delayed until the patient reaches a normal birth weight and size because, while it usually is not difficult to achieve correction in such a patient, it can be very challenging to find a foot abduction brace small enough to maintain the correction. A percutaneous tenotomy of the Achilles tendon is performed in the clinic with the patient under a local anesthetic before the last cast is applied if $<10^\circ$ of ankle dorsiflexion is present. Brace measurement, either for the Smeda clubfoot brace (Smeda Medizinische Instrumente, Hamburg, Germany) (two patients), the standard Foot Abduction Brace consisting of Open Toe, Straight-Last Boots (Markell Shoe Company, Yonkers, New York) attached to a Denis Browne Bar Splint (two patients), or a new dynamic foot abduction orthosis (Dobbs Brace; Orthotic and Prosthetic Lab, St. Louis, Missouri) (eight patients), was performed at the time of tenotomy. The dynamic foot abduction orthosis has been described in detail in a previous publication²⁵.

Braces were prescribed to be worn full-time (approximately twenty-three hours per day) for three months followed by part-time use (at nighttime and naptime, approximately fourteen to sixteen hours a day) until the age of four years on the basis of the current recommendations of Ponseti for patients with idiopathic clubfeet. Affected feet were placed in 50°

to 70° of external rotation, depending on the amount of external rotation of the foot achieved in the last cast.

A dedicated clubfoot nurse educator in our clinics instructed parents on brace wear and made follow-up telephone calls to the parents during the first week of brace wear to ascertain whether there were compliance issues with the brace. If there were concerns that could not be addressed over the phone, the child was brought back to the clinic to make brace adjustments or to provide further instructions².

The nurse educator also instructed the parents and provided them with a handout demonstrating how to effectively perform range-of-motion exercises for the ankle at every diaper change when the patient was out of the brace. These exercises were described in detail in a previous publication²⁵.

Statistical Methods

Continuous variables (the number of casts required to achieve correction) are expressed as the mean and standard deviation, and the rest of the variables are presented as percentages (frequencies). A two-tailed t test was used to compare the number of casts required to achieve correction of the clubfoot in our cohort with the number of casts required to achieve correction in a group of patients with idiopathic clubfoot who were managed by the senior author (M.B.D.) during the same time period. The level of significance was set at $p < 0.05$.

Results

Twenty-two clubfeet in eleven patients were graded as Diméglio grade IV (very severe) at the time of presentation to our clinics (Figs. 1-A and 1-B), and two clubfeet in one patient who had been managed with casting prior to presentation were classified as Diméglio grade II. Treatment in both groups began in the first six months of life. Initial correction was achieved in all clubfeet after treatment with a mean of 6.9 ± 2.1 casts (95% confidence interval, 5.6 to 8.3 casts) (see Appendix). All twelve patients (twenty-four clubfeet) underwent a percutaneous Achilles tendon tenotomy to correct a residual equinus deformity. The clubfeet in the present study required significantly more casts to achieve initial correction as compared with a cohort of 219 idiopathic clubfeet that were treated during the same time period by the senior author (M.B.D.) with use of the Ponseti method (mean, 4.5 ± 1.2 casts; 95% confidence interval, 4.3 to 4.7 casts) ($p = 0.002$).

Relapse ($\geq 5^\circ$ of hindfoot varus and $<10^\circ$ of ankle dorsiflexion) was detected in three patients (six feet; 25%) after initial successful treatment. The average time from correction until the diagnosis of relapse was 6.3 ± 1.2 months (95% confidence interval, 3.5 to 9.2 months). One patient had a second relapse in both feet that was observed six months after the first relapse. The parents of all three patients who had a relapse (including one patient who had been managed with the Smeda brace and two patients who had been managed with the standard foot abduction brace) reported noncompliance with brace wear. There were no reports of noncompliance with brace wear among the patients who did not experience a re-



Fig. 1-A



Fig. 1-B

Figs. 1-A through 1-D Case 3 (see Appendix). **Figs. 1-A and 1-B** Clinical photographs made at the time of presentation, demonstrating all components of the clubfoot deformity.

lapse. In all cases of noncompliance, the parents reported problems with slippage of the foot and the development of blisters on the heel and/or dorsum of the foot.

The original correction in all three patients who had a relapse was recovered with use of serial manipulations and castings. This process required two above-the-knee casts in one patient and four above-the-knee casts in each of the remaining two patients. The casts were changed at weekly intervals. Before the application of the last cast, a repeat percutaneous Achilles tenotomy was performed on one foot in one patient

because 10° of ankle dorsiflexion had not been obtained. After successful treatment of the relapses, the two patients who initially had been managed with the standard foot abduction orthosis were managed with the Dobbs brace, and the remaining patient continued to be managed with the Smeda brace. Additional education was provided to all three families on the importance of brace wear. The families of the two patients being treated at St. Louis Shriners Hospital for Children then received weekly telephone calls by the nurse educator to address any concerns about the brace and to bring the families



Fig. 1-C



Fig. 1-D

Clinical photographs made at the time of the latest follow-up, when the patient was three years old.

back to the clinic early if problems had arisen; both of these families subsequently reported compliance with the brace regimen, and no further relapse had been detected in either patient more than a year after the treatment of the first relapse. The remaining family, who did not receive the weekly telephone calls, reported continued noncompliance with bracing, and the child had a second relapse that required an extensive soft-tissue-release operation bilaterally to regain correction.

The Ponseti method was used successfully for eleven (92%) of the twelve patients with distal arthrogyposis in the present study (Figs. 1-C and 1-D). At the time of the latest follow-up, the deformities in these eleven patients (twenty-two

clubfeet) were well corrected, with a mean ankle dorsiflexion of $15^\circ \pm 5.75^\circ$ (95% confidence interval, 3.7° to 26.3°) bilaterally and with no evidence of hindfoot varus as measured with a handheld goniometer. At the time of the latest follow-up, the one patient who had been managed with an extensive soft-tissue-release operation bilaterally after a second relapse had plantigrade feet, a neutral heel position, and 0° of ankle dorsiflexion bilaterally.

Discussion

Excellent outcomes have been documented in association with the use of the Ponseti method of serial casting for

children with idiopathic clubfoot^{2-4,6}, but to our knowledge no studies have evaluated the results of this treatment method when used for patients with distal arthrogrypsis. Common practice has been to treat clubfoot arising from complex etiologies such as distal arthrogrypsis with extensive soft-tissue-release surgery¹⁰⁻¹⁴ or a talectomy¹⁵⁻¹⁷, an assumption based on the severe rigidity often associated with these conditions. The data from the current study, however, support the use of the Ponseti method for patients with distal arthrogrypsis on the basis of the short-term success approaching that for idiopathic clubfoot.

Available reports on the surgical treatment of arthrogrypotic clubfoot typically have not distinguished patients with distal arthrogrypsis from patients with amyoplasia (arthrogrypsis multiplex), which is associated with a more rigid clubfoot. This lack of distinction makes it difficult to clearly describe the results of clubfoot surgery in patients with distal arthrogrypsis alone. In general, surgical treatment of arthrogrypotic clubfoot is often complicated by high recurrence rates leading to salvage procedures¹¹⁻¹⁵. Extensive soft-tissue-release surgery as a primary treatment for arthrogrypotic clubfoot has been most successful when performed for patients under the age of one year¹⁰. Repeated soft-tissue releases have been unreliable for achieving correction because of extensive postoperative scarring. Talectomy has been suggested by some authors as a primary procedure and by others as a salvage procedure to address relapses. While this procedure often provides good initial correction^{11,12,26,27}, a high percentage of patients experience osteoarthritic changes at the tibio-calcaneal joint when followed over the long term¹⁶. Our initial experience with the use of the Ponseti method for the management of several patients who had clubfoot associated with arthrogrypsis multiplex suggests that it is more difficult to treat than either idiopathic clubfoot or distal arthrogrypotic clubfoot.

The arthrogrypotic clubfeet that were treated in the present study represented a severe form of clubfoot as all but two were considered to be grade IV according to the Diméglio criteria. As a surrogate measure of the severity of the clubfoot, we counted the number of serial casts required for correction with the Ponseti method. Although a significantly greater number of casts were required for the initial correction of distal arthrogrypotic clubfeet as compared with idiopathic clubfeet, treatment with the Ponseti method was successful for all patients in terms of the achievement of initial correction. The clubfoot recurrence rate of 25% in the present study is similar to that reported in the literature following the treatment of idiopathic clubfoot with the Ponseti method², and the percentage of patients requiring extensive soft-tissue-release surgery was small (<10%). Before referral, four of the twelve patients in the present study had had previous cast treatment that had been complicated by cast slippage, resulting in a complex clubfoot²⁰. Modifying the casting technique as recently recommended by Ponseti for the treatment of complex idiopathic clubfoot allowed us to achieve correction in those feet as well. The results of the present study support the use of the Ponseti method, either as a primary treatment or following a failed casting


protocol, for patients with clubfoot associated with distal arthrogrypsis.

Noncompliance with brace wear is a critical factor leading to clubfoot relapse following successful initial correction with the Ponseti method^{2,3,28}. Previous reports have cited heel and foot blistering as a common problem associated with traditional foot abduction bracing, resulting in noncompliance rates of 30% to 40%^{2,29}. However, on the basis of those reports as well as the current report, it is likely that compliance may be influenced by a gradual inability to adequately or comfortably brace the recurring clubfoot deformity, such that noncompliance with brace wear does not cause the recurrent deformity but rather reflects other intrinsic factors related to recurrence. Noncompliance with brace wear also may result from attempts to brace a clubfoot that is not fully corrected. The undercorrected clubfoot is likely to fit poorly in the brace.

In the current study, clubfoot relapse occurred in the first six months following the initiation of brace wear. This is the most common time to see recurrent deformities in patients with idiopathic clubfoot as well, and it is usually related to noncompliance with brace wear². If the problems with noncompliance can be solved during the first six months, continued compliance with the brace is likely^{2,28}. It has been shown that the relapse rate in patients with idiopathic clubfoot is extremely low if compliance with brace wear is maintained up to the age of four years²⁸. Although longer follow-up will be necessary to ensure maintenance of correction of the distal arthrogrypotic clubfoot, we recommend brace wear until the age of four years in this patient population. Noncompliance with brace wear may have been reduced in our patient cohort through the more aggressive use of a nurse educator for teaching and follow-up conversations, which has been shown to improve compliance in other cohorts of patients with clubfoot^{2,25}.

The ultimate measure of success associated with the use of the Ponseti method for the treatment of distal arthrogrypotic clubfoot will be the long-term maintenance of correction and quality-of-life outcomes. Several authors have reported late clubfoot relapses in arthrogrypotic patients who initially were managed with extensive soft-tissue-release operations as infants³⁰⁻³², and these relapses have been reported to occur until skeletal maturity³⁰. Although the clubfeet in the present study were well corrected after two years of follow-up, longer follow-up is necessary to assess the continued risk of recurrence and to allow for more accurate recommendations regarding the length of time necessary for brace wear. Because of the ability to avoid extensive soft-tissue-release surgery with the Ponseti method, negative long-term consequences in terms of foot pain, ankle and foot osteoarthritis, and quality of life³³ may be avoided in this group of patients who often have been difficult to treat.

Appendix

 A table showing clinical details on all patients is available with the electronic versions of this article, on our web site at jbjs.org (go to the article citation and click on "Supplementary Material") and on our quarterly CD-ROM (call

our subscription department, at 781-449-9780, to order the CD-ROM). ■

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