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6-1-2009

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Recommended Citation

Gerlach, David J.; Gurnett, Christina A.; Limpaphayom, Noppachart; Alaei, Farhang; Zhang, Zhongli; Porter, Kristina; Kirchhofer, Melissa; Smyth, Matthew D.; and Dobbs, Matthew B., "Early results of the Ponseti method for the treatment of clubfoot associated with myelomeningocele." *The Journal of Bone and Joint Surgery*.91,6. 1350-1359. (2009).
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Early Results of the Ponseti Method for the Treatment of Clubfoot Associated with Myelomeningocele

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Investigation performed at the Department of Orthopaedic Surgery, Washington University School of Medicine, and St. Louis Shriners Hospital for Children, St. Louis, Missouri

Background: Myelomeningocele is a common birth defect that is often accompanied by clubfoot deformity. Treatment of clubfoot associated with myelomeningocele traditionally has consisted of extensive soft-tissue release operations, which are associated with many complications. The purpose of the present study was to evaluate the early results of the Ponseti method for the treatment of clubfoot associated with myelomeningocele.

Methods: Sixteen consecutive patients with myelomeningocele (twenty-eight clubfeet) and twenty consecutive patients with idiopathic clubfeet (thirty-five clubfeet) were followed prospectively while being managed with the Ponseti method. The average duration of follow-up was thirty-four months for the myelomeningocele group and thirty-seven months for the idiopathic group. Clubfoot severity was graded at the time of presentation with use of the Diméglio system. The initial correction that was achieved, casting and/or bracing difficulties, recurrences, and subsequent treatments were evaluated and compared between the two cohorts by means of appropriate statistical analysis.

Results: Eleven (39%) of the twenty-eight clubfeet in the myelomeningocele group were graded as Diméglio grade IV, compared with only four (11%) of the thirty-five clubfeet in the idiopathic group ($p = 0.014$). Initial correction was achieved in thirty-five clubfeet (100%) in the idiopathic group and in twenty-seven clubfeet (96.4%) in the myelomeningocele group ($p = 0.16$). Relapse of deformity was detected in 68% of the feet in the myelomeningocele group, compared with 26% of the feet in the idiopathic group ($p = 0.001$). Relapses were treated successfully without the need for extensive soft-tissue release surgery for all but four of the clubfeet in the myelomeningocele group and for all but one of the clubfeet in the idiopathic group ($p = 0.16$).

Conclusions: Our data support the use of the Ponseti method for the initial treatment of clubfoot deformity associated with myelomeningocele, although attention to detail is crucial in order to avoid complications. Longer follow-up will be necessary to assess the risk of late recurrence and the potential need for more extensive clubfoot corrective surgery in this patient population.

Level of Evidence: Prognostic Level II. See Instructions to Authors for a complete description of levels of evidence.

Clubfoot is a complex congenital foot deformity that is both common (occurring in one in 1000 live births¹) and difficult to correct. It is easily recognizable at birth, and it can be differentiated from common positional foot disorders on the basis of the rigid equinus deformity and its resistance to passive correction. The deformity consists of four components (equinus, hindfoot varus, forefoot adductus, and cavus) and is graded according to severity with use of the

Diméglio system². Most clubfeet occur as an isolated birth defect and are considered to be idiopathic.

Historically, treatment of idiopathic clubfoot deformity has been variable. Both nonoperative methods of manipulation³⁻⁵ and extensive soft-tissue releases⁶⁻¹⁶ have been advocated. Early methods of manipulation, such as the Kite technique³, demonstrated inconsistent results, leading to unpredictable outcomes after extended periods of casting. Extensive surgical

Disclosure: The authors did not receive any outside funding or grants in support of their research for or preparation of this work. One or more of the authors, or a member of his or her immediate family, received, in any one year, payments or other benefits of less than \$10,000 or a commitment or agreement to provide such benefits from a commercial entity (D-Bar Enterprises, Webster Groves, Missouri). No commercial entity paid or directed, or agreed to pay or direct, any benefits to any research fund, foundation, division, center, clinical practice, or other charitable or nonprofit organization with which the authors, or a member of their immediate families, are affiliated or associated.

TABLE I Prevalence of Clubfoot in Patients with Myelomeningocele, 1995 to 2006

	No. of Patients (N = 67)
Level of myelomeningocele lesion	
High (L3/L4 combined lesions or above)	12 (18%)
Low (L4 or below)	55 (82%)
Foot deformity present	18 (27%)
Clubfoot deformity present	13 (19%)
High-level lesion	4
Low-level lesion	9

releases^{6,9} were proposed in order to avoid the complications and duration of the casting techniques. Despite the initial correction of the deformity, these surgical releases often led to variability in short-term outcome^{6,12}, the need for revision procedures^{8,9}, and significant limitations at the time of long-term follow-up^{15,16}.

The Ponseti method^{17,18} involves the use of serial casting to gradually correct the clubfoot deformity and a percutaneous tenotomy of the Achilles tendon to correct residual ankle equinus, followed by several years of foot abduction bracing to maintain correction. This method has gained widespread popularity and has been successful in recent years for the treatment of idiopathic clubfoot deformity, and its efficacy in achieving short-term correction has been demonstrated in several studies¹⁹⁻²³. Long-term follow-up studies also have demonstrated excellent results in association with the Ponseti method in terms of quality of life and foot function^{19,20}.

Non-idiopathic clubfoot occurs in patients with genetic syndromes, chromosomal abnormalities, or neurological disorders such as myelomeningocele. Despite the reported successful treatment of idiopathic clubfeet with use of the Ponseti method^{19,20,23}, we are not aware of any reports on the use of the Ponseti method for patients with myelomeningocele. On the contrary, clubfeet associated with myelomeningocele tradition-

ally have been treated with extensive soft-tissue release surgery, with many short-term complications having been reported, including skin complications related to the lack of normal sensation in the lower limbs²⁴, recurrent deformities^{25,26}, and the need for revision and salvage procedures^{24,26-28}. We are not aware of any long-term follow-up studies investigating the outcomes for these patients. Presumably, however, they may follow a similar, if not worse, long-term course compared with that in patients with surgically corrected idiopathic clubfeet¹⁵.

The present study was performed to evaluate whether the Ponseti method could be used to achieve initial correction of a clubfoot deformity associated with myelomeningocele. For the purposes of comparison, we examined the effectiveness of the Ponseti method in a cohort of age and sex-matched patients with an idiopathic clubfoot deformity who were managed by the same surgeon during the same time period. Given the recent data demonstrating that the use of a foot abduction brace is the most predictive factor of the maintenance of clubfoot correction^{23,29}, we also analyzed bracing tolerance in each group.

Materials and Methods

The Washington University School of Medicine Human Research Protection Committee approved the study, and written informed consent was obtained for all individuals. Eighteen consecutive patients with a clubfoot deformity and myelomeningocele were managed with the Ponseti method^{17,18,23,30} between July 2001 and July 2006. All patients were followed prospectively and were managed solely by the senior author (M.B.D.). These patients were managed at both St. Louis Children's Hospital (thirteen patients) and the St. Louis Shriners Hospital (five patients). Two patients, both of whom had undergone extensive surgical releases before presentation, were excluded from additional analysis. Thus, sixteen patients were included in the final analysis. Nine patients (56%) were female. Five patients had received some treatment for clubfoot deformity before referral to our institution; specifically, three patients had had casting and two had had physical therapy and splinting. No patient was lost to follow-up. The average

TABLE II Demographic and Clinical Characteristics

	Myelomeningocele Group (N = 16)	Idiopathic Group (N = 20)	P Value
Age at first cast* (wk)	12.4 (-1.2 to 25.9)	4.7 (2.7 to 7.6)	0.248
Female†	9 (56%; 32% to 81%)	6 (30%; 10% to 50%)	0.112
Family history†	0	2 (10%; 0% to 23%)	0.492
Full term (≥36 weeks)†	13 (81%; 69% to 100%)	15 (75%; 56% to 94%)	0.393
Cesarean section†	10 (63%; 56% to 94%)	8 (40%; 19% to 61%)	0.118
First-born child†	2 (13%; 0% to 29%)	7 (35%; 14% to 56%)	0.446
Duration of follow-up* (mo)	33.8 (25.0 to 42.7)	36.8 (30.1 to 43.4)	0.572
Bilateral clubfoot†	12 (75%; 56% to 94%)	15 (75%; 56% to 94%)	1.000

*The data for continuous variables are presented as the mean, with the 95% confidence interval in parentheses. †The data for categorical variables are given as the number of patients, with the frequency (percentage) and the 95% confidence interval in parentheses.

TABLE III Outcome Measurements

	Myelomeningocele Group (N = 28)	Idiopathic Group (N = 35)	P Value
Diméglio grade*	3.3 (3.0 to 3.6)	2.7 (2.4 to 2.9)	0.003
Diméglio grade IV†	11 (39%; 26% to 66%)	4 (11%; 1% to 26%)	0.014
Number of casts*	5.0 (4.6 to 5.7)	5.1 (4.7 to 5.5)	0.862
Number of casts ≤5†	16 (57%; 39% to 75%)	25 (71%; 56% to 86%)	0.237
Tenotomy†	24 (86%; 73% to 99%)	33 (94%; 87% to 100%)	0.250
Initial correction†	27 (96%; 90% to 100%)	35 (100%; 100%)	0.444
Recurrence†	19 (68%; 51% to 85%)	9 (26%; 11% to 40%)	0.001
Noncompliant recurrence†	7 of 19 (37%; 15% to 59%)	6 of 9 (67%; 36% to 97%)	0.139
Recurrence resolution†	15 of 19 (79%; 61% to 97%)	8 of 9 (89%; 68% to 100%)	1.000
Initial dorsiflexion* (deg)	21.2 (18.3 to 24.1)	25.3 (23.4 to 27.2)	0.084
Final dorsiflexion* (deg)	18.3 (15.7 to 20.8)	21.7 (19.1 to 24.2)	0.065
Successful†	24 (86%; 73% to 99%)	34 (97%; 92% to 100%)	0.162
Extensive soft-tissue release†	4 (14%; 1% to 27%)	1 (3%; 0% to 8%)	0.162

*The data for continuous variables are presented as the mean, with the 95% confidence interval in parentheses. †The data for categorical variables are given as the number of feet, with the frequency (percentage) and 95% confidence interval in parentheses.

duration of follow-up was thirty-four months (range, eight to sixty-eight months).

A second cohort, consisting of twenty patients with idiopathic clubfoot deformities who were managed with the Ponseti method by the senior author (M.B.D.), was prospectively gathered during this same time period. This cohort of patients with idiopathic clubfoot was matched with the myelomeningocele group on the basis of age at the time of the initiation of treatment. All patients in the idiopathic group were managed at St. Louis Children's Hospital. Two patients had received casting before referral, with one of these patients also having had a tenotomy of the Achilles tendon. This latter patient was diagnosed with complex clubfoot deformity³¹ at the time of presentation to our clinic. The average duration of follow-up was thirty-seven months (range, fourteen to sixty-eight months).

The severity of the foot deformities was classified according to the grading system of Diméglio et al.² at the time of presentation. With this system, clubfeet are categorized into four grades according to the severity of the deformity. Grade-I feet have a mild deformity that is >90% passively reducible, grade-II feet have a moderate deformity, grade-III feet have a severe deformity, and grade-IV feet have a very severe deformity and are nearly immobile to passive manipulation.

Charts were reviewed with regard to demographic data (including sex, bilaterality or unilaterality of the clubfoot deformity, and age at the time of the onset of treatment), previous treatments (including manipulations and castings, physical therapy, and surgical releases), the level of myelomeningocele, and medical comorbidities. During the course of treatment, the number of castings required for initial correction, the need for a percutaneous Achilles tendon tenotomy to correct residual equinus contracture after castings, and tolerance with bracing protocols were noted. Reports by the family

regarding the use of the foot abduction brace were used to ascertain tolerance. Complications such as skin blistering, cast slippage, and tibial and fibular fractures were recorded. 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 initial correction of the deformity was obtained, passive ankle dorsiflexion and plantar flexion as well as heel varus-valgus deformity were measured by the senior author (M.B.D.) with a handheld goniometer. These measurements were all extracted from the medical record for analysis. Recurrent deformities were documented with regard to their severity, the age of the patient at the time of recurrence, compliance with treatment protocols before recurrence, and any additional treatments needed to regain correction.

In order to assess the prevalence of clubfoot deformity in patients with myelomeningocele, we reviewed the available medical records for all patients with myelomeningocele who had been managed at St. Louis Children's Hospital between 1995 and 2006.

Treatment Method

All of the patients who were enrolled in the study were managed with use of the Ponseti method according to a published protocol^{17,18,23,30}. When possible, treatment is initiated within the first few weeks of life; however, for premature infants, the initiation of casting is delayed until the baby reaches a normal birth weight and size. Correction of the clubfoot deformity is not, in itself, inherently difficult in these very small patients; however, it can be difficult to find a foot abduction brace that is small enough to fit. If <10° of ankle dorsiflexion is present, a percutaneous tenotomy of the Achilles tendon is performed in the clinic for all patients who are less than twelve months of age, with use of a local anesthetic, before the application of the

final cast. Measurements for the foot abduction orthosis are performed at the time of tenotomy. Patients with myelomeningocele who had both clubfeet and congenital knee dislocations were managed with a modified casting protocol to address both deformities simultaneously³².

According to the date on which the patient was first managed with a foot abduction orthosis, one of two bracing systems was used. For patients who were seen prior to late 2004, the standard foot abduction brace consisting of open-toed, high-top, straight-last shoes (Markell Shoe Company, Yonkers, New York) attached to a Denis Browne bar was used. For more recently managed patients, a newly designed dynamic foot abduction orthosis (D-Bar Enterprises LLC, Webster Groves, Missouri) was used. Each brace was prescribed to be worn full time (for twenty-three hours per day) for the first three months, followed by part-time use (for fourteen to sixteen hours per day, mostly during naps and at night) until the age of four years. Affected feet were placed in 50° to 70° of external rotation, depending on the amount of external rotation achieved in the last cast. Normal feet were placed in 30° of external rotation. The width of the bar was set to approximately the shoulder width of the patient so that, when worn, the center of each heel lined up with the outer edge of each shoulder. Once they reached walking age, the patients with myelomeningocele were managed with either a solid ankle-foot orthosis or a stander and a hip-ankle-foot orthosis, depending on the level of motor function.

Patients were closely followed throughout the course of their treatment. Intolerance with brace wear is most frequently encountered early in the bracing protocol. A dedicated nurse educator (K.P.) instructed parents on expectations, issues, and protocols. In addition, the nurse educator made direct follow-up telephone calls to parents during the first weeks of brace wear to ascertain compliance and to identify potential complications. These duties of the nurse educator have been shown to improve parental compliance²³. Parents were instructed on how to effectively perform range-of-motion exercises for the ankle when the foot was out of the brace. The exercises were described in detail in a previous publication²³. They are performed at the time of diaper changes and have been found to improve the ability to maintain the ankle motion that is achieved at the time of tenotomy²³.

Statistical Analysis

Demographic data, clinical characteristics, and outcomes were compared between groups with use of unpaired t tests for continuous variables and chi-square tests (or Fisher exact tests when appropriate) for categorical variables. Continuous variables are presented as the mean and the 95% confidence interval, and categorical variables are presented as the frequency (percentage) and the 95% confidence interval of the percentage. For patients who underwent treatment on both feet (including twelve patients in the myelomeningocele group and fifteen patients in the idiopathic group), the outcomes from each foot were analyzed individually. All statistical tests were two-tailed, and the level of significance was set at $p \leq 0.05$.

Source of Funding

No external funds were received in support of the current study. One of the authors (M.B.D.) has a royalty agreement with D-Bar Enterprises, Webster Groves, Missouri. This agreement did not play any role in the investigation.

Results

Prevalence of Clubfoot in Patients with Myelomeningocele

The records of sixty-seven patients with myelomeningocele who were managed with neurosurgical closure at St. Louis Children's Hospital between 1995 and 2006 were available for review (Table I). Eighteen (27%) of these patients had a diagnosed foot deformity; the deformities included clubfoot deformity (thirteen patients; 19%), metatarsus adductus (three patients; 4%), vertical talus (two patients; 3%), and calcaneovalgus deformity (two patients; 3%). Of the thirteen patients with a clubfoot deformity, nine (69%) had low-level lesions (L4 or below).

Demographic Characteristics

With the numbers available, no significant differences were noted between the myelomeningocele and idiopathic groups with regard to sex, laterality, familial association, breech presentation, cesarean section delivery, or term birth. Two of the sixteen patients in the myelomeningocele group were first-born, compared with seven of the twenty patients in the idiopathic group ($p = 0.45$) (Table II). In cases of unilateral deformity, both the myelomeningocele group (four of four patients) and the idiopathic group (four of five patients) had a predominance of right foot involvement. For the myelomeningocele group, the mean age at the time of initial casting was twelve weeks (range, one to 103 weeks); the mean age at the time of presentation decreased to six weeks when a single patient, in whom casting was initiated at 103 weeks of age, was excluded (see Appendix). For the idiopathic group, the mean age at the time of initial casting was five weeks (range, one to nineteen weeks) ($p = 0.25$).

Medical Comorbidities

Before undergoing treatment for clubfoot deformity, fifteen of sixteen patients had undergone both myelomeningocele closure and the placement of a ventriculoperitoneal shunt. Eleven of the patients were classified as having a low-level spinal lesion (L4 or lower), whereas seven patients had a higher-level lesion (includes L3/L4 combined lesions and higher). Medical problems associated with myelomeningocele were common: urethral reflux was encountered in ten patients, musculoskeletal problems such as congenital knee dislocation were encountered in three patients, and congenital hip dislocation was seen in nine patients. Medical comorbidities were rare in the idiopathic group: one patient was diagnosed with a benign brain tumor, and another had a congenital trigger thumb.

Outcomes

Data from each foot were analyzed individually (Table III). Twenty-eight feet from the myelomeningocele cohort and



Fig. 1-A



Fig. 1-B

Figs. 1-A, 1-B, and 1-C Clinical photographs of the feet of a patient in the myelomeningocele group (Case 1; see Appendix). **Fig. 1-A** Photograph made at the time of presentation, demonstrating a very severe right-sided clubfoot deformity. **Fig. 1-B** Photograph made when the patient was eighteen months old, demonstrating recurrent hindfoot varus and equinus deformities on the right side.

thirty-five feet from the idiopathic cohort were included in the analysis. Eleven clubfeet (39%) in the myelomeningocele group had Diméglio grade-IV (very severe) deformities at the time of presentation to our clinics (Fig. 1-A). The mean Diméglio grade for the entire myelomeningocele cohort was 3.3 (range, 2 to 4). In contrast, the idiopathic cohort had a mean Diméglio grade of 2.7 (range, 2 to 4) ($p = 0.003$), with only four (11%) of thirty-five idiopathic clubfeet having grade-IV deformities ($p = 0.014$). Patients with low-level lesions (L4

and below) had a mean Diméglio grade of 3.3 (range, 2 to 4), whereas patients with high-level lesions had a mean Diméglio grade of 3.4 (range, 3 to 4) ($p = 0.11$).

Initial correction was achieved in 96% (twenty-seven) of twenty-eight clubfeet in the myelomeningocele group and in 100% (thirty-five) of thirty-five clubfeet in the idiopathic group ($p = 0.44$). The mean number of casts was 5.0 (range, two to eight) in the myelomeningocele group and 5.1 (range, four to eight) in the idiopathic group ($p = 0.86$). In the my-



Fig. 1-C

Photograph made when the patient was forty-five months old, after the recurrent clubfoot deformity had been treated with repeat casting and tenotomy of the Achilles tendon.

elomeningocele group, patients with high and low-level lesions were similar with regard to the mean number of casts (4.9 compared with 5.2), initial ankle dorsiflexion (20.6° compared with 22.4°), and final ankle dorsiflexion (17.7° compared with 18.4°).

Twenty-four (86%) of the twenty-eight clubfeet in the myelomeningocele group required a percutaneous Achilles tendon tenotomy to correct a residual equinus deformity. The remaining four clubfeet (two patients) that did not require an Achilles tendon tenotomy were the only clubfeet in the myelomeningocele group with Diméglio grade-II deformities. Thirty-three (94%) of the thirty-five clubfeet in the idiopathic group required an Achilles tendon tenotomy. There was no significant difference between the two study cohorts with regard to the need for a percutaneous tenotomy ($p = 0.25$). Details on the patients with myelomeningocele can be found in a table in the Appendix.

All of the patients in the idiopathic group and fifteen of the sixteen patients in the myelomeningocele group were managed with a foot abduction orthosis after the final cast was removed. The one patient who was not managed with a foot abduction orthosis had ongoing medical problems that prohibited the use of a brace. Seven patients (44%) in the myelomeningocele group and seven patients (35%) in the idiopathic group were managed with the dynamic foot abduction orthosis ($p = 0.23$), whereas the remaining patients were managed with a traditional Denis Browne bar with straight-last, open-toed shoes.

Clubfoot correction was measured after the Achilles tendon tenotomies were performed and before the bracing period. The mean amount of ankle dorsiflexion that was acquired after the Achilles tendon tenotomy was 21.2° (range, 5° to 30°) in the myelomeningocele group and 25.3° (range, 15° to 35°) in the idiopathic group ($p = 0.08$). At the time of the latest follow-up, the mean amount of ankle dorsiflexion was 18.3° (range, 0° to 35°) in the myelomeningocele group and 21.7° (range, 10° to 35°) in the idiopathic group ($p = 0.065$).

Relapse and Treatment

The rate of clubfoot relapse after initial successful treatment was 68% (nineteen of twenty-eight feet; eleven patients) in the myelomeningocele group, compared with 26% (nine of thirty-five feet; five patients) in the idiopathic cohort ($p = 0.001$). Relapse was defined when $\geq 5^\circ$ of hindfoot varus and $< 10^\circ$ of ankle dorsiflexion were both present (Fig. 1-B). The mean time from correction until the development of relapse was 7.1 months (range, one to twenty months) for both cohorts.

Intolerance with the bracing protocol after an initial period of successful brace use was identified in three (60%) of the five patients (six of nine clubfeet) in the idiopathic group who had a clubfoot relapse, compared with four (36%) of the eleven patients (seven of nineteen clubfeet) in the myelomeningocele group who had a clubfoot relapse. The difference in compliance was not significant ($p = 0.60$). The remaining relapses in both cohorts occurred despite use of the brace as



Fig. 2
Anteroposterior radiograph of the tibia and fibula of a patient in the myelomeningocele group (Case 6; see Appendix), demonstrating distal tibial and fibular shaft fractures that occurred after the child was placed into the foot abduction brace at twenty weeks of age.

prescribed. In the myelomeningocele cohort, there were recurrences in six of seven patients (eleven of thirteen feet) with high-level lesions, compared with five of nine patients (seven of fifteen feet) with low-level lesions ($p = 0.042$).

In the myelomeningocele group, the original correction was re-achieved with repeat casting alone in eleven (58%) of the nineteen feet that had a relapse, with use of a mean of 3.7 casts (range, one to nine casts). Of the other eight feet in that group that had a relapse, three feet required four sets of casts each, followed by a repeat Achilles tendon tenotomy (Fig. 1-C); one foot required a single cast, followed by a repeat Achilles tendon tenotomy; and four feet (in two patients with bilateral involvement) required extensive soft-tissue release operations. High-level lesions were present in both of the patients with myelomeningocele who required extensive soft-tissue release operations. Overall, fifteen (79%) of the nineteen recurrences in the myelomeningocele group were successfully treated without an extensive soft-tissue release operation.

In comparison, eight (89%) of the nine recurrences in the idiopathic group were successfully corrected without the need for an extensive soft-tissue release operation. Two feet in the idiopathic group were treated with repeat casting alone (twelve casts); four feet were treated with six sets of casts, followed by an Achilles tendon tenotomy; and two feet with a dynamic supination deformity were treated with two sets of casts each, followed by a tibialis anterior tendon transfer to the third cuneiform. The one patient (one clubfoot) in this group who required an extensive soft-tissue release procedure had a long history of brace intolerance due to ongoing treatment of a brain tumor.

At the time of the latest follow-up, successful treatment of the clubfoot deformity, with avoidance of major soft-tissue releases or transfers, was achieved in twenty-four (86%) of the twenty-eight clubfeet in the myelomeningocele group and in thirty-four (97%) of the thirty-five clubfeet in the idiopathic group ($p = 0.16$).

Complications

Nine clubfeet (32%) in the myelomeningocele group sustained blistering in the standard foot abduction orthosis that was limited to superficial blisters about the heel. Six of these nine clubfeet subsequently had a recurrence following the discontinuation of brace use, which was required for blister healing; all were successfully treated with repeat casting alone. Five clubfeet (14%) in the idiopathic group sustained blistering while in the standard foot abduction brace, but none had a recurrence. No blisters were deep, and none led to infection. Three patients (three clubfeet; 11%) in the myelomeningocele group had cast slippage leading to a complex clubfoot deformity that required correction with a modified casting technique³¹. No patient in the idiopathic group experienced cast slippage. No patient in either group had development of skin complications during casting.

Iatrogenic distal tibial fractures occurred during treatment in two patients in the myelomeningocele group. In one, a healed distal tibial fracture was diagnosed incidentally on radiographs made after clubfoot correction was achieved. In the second, the patient's parents noticed leg swelling twenty-four hours after the application of the standard foot abduction brace. Subsequent radiographs revealed distal tibial and fibular fractures, which were treated with casting (Fig. 2). There were no iatrogenic fractures in the idiopathic cohort.

Discussion

The Ponseti method has been widely shown to be an effective means of obtaining both short-term and long-term correction of idiopathic clubfoot deformity^{19,20,23}. However, the use of the Ponseti method to address clubfoot deformity in patients with myelomeningocele has not been reported, to our knowledge. Currently, the recommended treatment for these patients, who generally present with stiffer, more recalcitrant clubfeet, has been extensive soft-tissue releases performed either primarily or following a period of serial casting^{24-27,33-35}. This type of surgery has been shown to be associated with a

high rate of both short-term and long-term complications³⁶⁻⁴³. The justification for surgical management in this patient population is based on previous reports that have shown serial casting to be ineffective and complication-prone^{25,28}. However, data from the current study support the use of the Ponseti method for the management of patients who have myelomeningocele, for whom the short-term success rate approached that for the patients with idiopathic clubfoot.

Although non-idiopathic clubfoot deformity has been reported to be more rigid than idiopathic clubfoot deformity^{28,44,45}, there is a precedent for the successful use of the Ponseti method to achieve initial correction of clubfoot associated with distal arthrogyposis⁴⁵ as well as clubfoot associated with a variety of other genetic syndromes^{44,46}. The only reported difference has been the greater number of casts required to achieve correction in patients with non-idiopathic as compared with idiopathic clubfeet⁴⁵. In the current study, we were able to utilize the Ponseti method successfully to achieve initial correction in twenty-four (86%) of the twenty-eight clubfeet in the myelomeningocele group, a rate similar to that in the idiopathic group.

Despite the achievement of initial correction with the Ponseti method in both cohorts, the recurrence rate was significantly higher in the myelomeningocele cohort as compared with the idiopathic cohort. Within the myelomeningocele group, there was an increased risk of recurrence in the patients with higher-level lesions as compared with those with lower-level lesions. This finding is consistent with the reported risk of recurrence in patients with myelomeningocele who have been managed with extensive soft-tissue release operations^{26-28,36}. The higher risk of recurrence in patients with high-level lesions has been attributed to a more severe initial deformity, lack of full correction of the deformity, and the absence of weight-bearing in this patient population²⁷.

Another critical factor leading to clubfoot relapse is failure to use the foot abduction brace as prescribed, or brace intolerance^{23,34,35,45}. Many factors can lead to brace intolerance, including heel and foot blistering, which has been reported in association with traditional foot abduction bracing²³, as well as the gradual inability to fit in the brace because of a clubfoot that is recurring or that was not fully corrected before being treated with the brace. In the current study, the majority of recurrent clubfeet in both the myelomeningocele cohort and the idiopathic cohort were treated successfully with casting and a repeat Achilles tendon tenotomy when $>5^\circ$ of ankle dorsiflexion could not be achieved with casting alone. After correction of the clubfoot relapse, the patients in both cohorts were again managed with a foot abduction brace for nighttime wear. The brace was then worn until the child turned four years of age. Brace tolerance was improved in this cohort through frequent teaching and telephone follow-up by a dedicated nurse educator, and no secondary relapses had developed by the time of the latest follow-up.

In the present study, the Ponseti method was associated with complications, including skin blistering during the bracing phase as well as two known cases of iatrogenic tibial

and fibular fractures. Although skin blistering can occur in association with both idiopathic and non-idiopathic clubfoot, it has the potential to be more problematic in patients with a myelomeningocele because of the sensory deficits in that group. Such deficits can lead to a late diagnosis and a more advanced skin complication requiring the cessation of either casting or bracing for a period of time to allow the skin to heal. The tibial and fibular fractures were only seen in the myelomeningocele group. Both underlying osteopenia⁴⁷ and a lack of sensation in the leg may be risk factors for iatrogenic fracture with use of the Ponseti method. The lack of sensation makes it easy to be overly aggressive with manipulation because the child does not cry or resist. This overly aggressive manipulation in combination with osteopenia can lead to fracture. The fractures, when recognized, can be readily treated with casting.

In reviewing the type of foot deformities in our patients who had myelomeningocele, we found that the type of deformity could not always be predicted on the basis of the level of paralysis. Other studies have demonstrated this finding as well, making it difficult to attribute the occurrence of different foot deformities to the simple imbalance of voluntary muscles^{48,49}. If the foot deformities were strictly due to muscle imbalance, then a clubfoot deformity would be predicted to occur only in patients with a mid-lumbar-level lesion, whereas congenital vertical talus and calcaneovalgus deformities would occur in patients with low-lumbar and sacral-level lesions. However, these patterns are rarely found, suggesting the possibility that other factors may contribute to the etiology of these foot deformities. The foot phenotype, whether dependent on the level of myelomeningocele or other factors, does appear to be genetic as we have managed two sets of identical twins who have had both a myelomeningocele and nearly indistinguishable bilateral clubfoot deformities. Additional genetic studies are warranted in this patient population to investigate the factors that contribute to the etiology of these foot deformities.

We have found that a team approach involving a skilled pediatric orthopaedic surgeon, a nurse educator, and an orthotist is essential for the successful treatment of clubfoot deformities in patients with myelomeningocele. Achieving correction of clubfoot with use of the Ponseti method in patients with myelomeningocele is possible in the vast majority of cases, but attention to detail is crucial. Care should be taken to manipulate the foot gently to avoid fractures in the distal parts of the tibia and fibula. A well-molded cast is crucial to reduce the risk of skin sores. The use of a foot abduction orthosis that is soft on the skin as well as having the caregiver monitor the skin frequently by removing the brace will greatly reduce the number of skin complications resulting from bracing. A skilled orthotist is essential to monitor the fit of the foot abduction orthosis and to make adjustments as necessary. Having a nurse educator who can spend time with the family to demonstrate correct brace application, to answer questions, and to contact the family during the first week after the initiation of brace wear in order to ensure that no problems have arisen has proven to be very helpful for improving bracing tolerance and treatment outcomes^{23,45,46}.

Although the clubfeet in the myelomeningocele group remained well corrected at two years, longer follow-up is necessary in order to assess the continued risk of recurrence and to give more accurate recommendations with regard to the necessary duration of foot abduction bracing. Quality-of-life outcomes also will be important for assessing the long-term success. To date, relapses in our patients with myelomeningocele have been treated with repeat manipulations, castings, and tenotomy of the Achilles tendon, as is the case for patients with idiopathic clubfoot. However, depending on the level of motor involvement, some patients with myelomeningocele may be more prone to continued relapses. This select group of patients may benefit from tendon-balancing procedures to maintain foot correction over the long term.

Appendix

eA A table showing details on the patients with myelomeningocele is available with the electronic versions of this article, on our web site at jbsj.org (go to the article citation and click on "Supplementary Material") and on our quarterly CD/

DVD (call our subscription department, at 781-449-9780, to order the CD or DVD). ■

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