Congenital idiopathic talipes equinovarus: an evaluation in infants treated by the Ponseti method

V. PAVONE, G. TESTA, L. COSTARELLA, P. PAVONE, G. SESSA

Orthopaedic Clinic, and ¹Pediatric Clinic; Polyclinic Vittorio Emanuele, Azienda Ospedaliera Universitaria, University of Catania, Catania, Italy

Abstract. – BACKGROUND: Congenital talipes equinovarus (CTEV) is a common but still not fully understood disorder of the lower limb. It is usually defined as a fixation of the foot in adduction, supination, and varus. Different treatment options exist including the Ponseti method.

AIM: We report here the results obtained in infants with CTEV treated by the Ponseti method.

PATIENTS AND METHODS: Eighty two patients (114 clubfeet) were enrolled at the Orthopaedic Clinic of Catania University during the period of March 2004 to January 2010 and followed prospectively up to February 2011: 56 patients (68.29%) were male, the anomaly was bilateral in 32 (39%) cases, unilateral in 50 (60.9%) in the right side in 28 (56%). The mean age at initiation of treatment was 14 days (range 3-81 days), severity of the club foot deformity by the Pirani Severity score was 5.56 points (range 4.3-6 points). Total numbers of Ponseti casts before tenotomy, details of tenotomy, and compliance with CTEV brace were recorded. Clinical evaluation was performed using the functional Ponseti Scoring System. Mean follow up was 4 years: range 13-83 months.

RESULTS: An average of 6.6 casts was necessary before performing the tenotomy. Tenotomy was performed by a single surgeon (V.P.) in a total of 68 patients (82.93%) always in an operating room under general anaesthesia by a percutaneous approach at a mean age of 106 days (range 45-213 days). Compliance with CTEV brace was satisfactory in 79 patients (96.3%). Functional Ponseti Scores were good/excellent in 79 (96.34%) patients (109 clubfeet; 95.61%). Only 3 patients; 3.7% (5 clubfeet; 4.4%) suffered relapse. Poor compliance with the Denis Browne splint was thought to be the main cause of failure.

CONCLUSIONS: The Ponseti method provides an excellent outcome at follow up in the treatment of congenital idiopathic clubfoot.

Key Words:

CTEV, Ponseti method, Clubfoot, Limb disorder.

Introduction

Congenital talipes equinovarus (CTEV), also known as clubfoot, is one of the most common congenital bone deformities. While some cases are associated with neuromuscular disease, chromosomal abnormalities, different syndromes or extrinsic causes, others occur in otherwise normal infants and are classified as idiopathic congenital talipes equinovarus (ICTEV). The latter is a common but still not fully understood disorder of the lower limb with birth prevalence of 1-4.5 per 10001 and incidence of 0.64-6.8 per 1000 live births²⁻⁶. It is usually defined as a fixation of the foot in adduction, supination, and varus. Three bones, the calcaneus, navicular, and cuboid, are medially rotated in relation to the talus and are held in adduction and inversion by ligaments and tendons. Although the foot is supinated, the front of the foot is pronated in relation to the back of the foot, causing cavus. Diagnosis is mainly based on clinical evidence even if prenatal diagnosis is possible through sonographic assessment.

The purpose of this study was to report our caseload in evaluating the short-midterm effectiveness of the Ponseti method^{7,8} for the treatment of ICTEV in a series of infants with this anomaly.

Patients and Methods

A total of 82 Sicilian patients (114 clubfeet) were treated by the Ponseti method^{7,8} by a single orthopaedic surgeon (V.P.) during the period of March 2004 to January 2010 at the Orthopaedic Clinic, University of Catania, and were studied prospectively up to February 2011.

All children with secondary CTEV were excluded.

The average birth weight was 3356 ± 567 g. Gestational age was 40 ± 2 weeks, maternal age 30 ± 7 years, and paternal age 33 ± 7 years. A positive family history of ICTEV was reported in 18 cases (21.95%). In 43 cases (52.44%), the child was the first-born. Forty-nine children (59.76%) were delivered by caesarean section. During the pregnancy, 30 mothers (36.58%) underwent amniocentesis, 17 (20.73%) smoked, 4 (4.88%) consumed alcohol, 3 (3.66%) were exposed to drugs, 26 (31.71%) did not take supplementary folic acid, and 3 (3.66%) experienced minor trauma. Nine of the children had other associated abnormalities not related to the clubfoot.

Out of 82 patients, 56 patients (68.29%) were male, giving male: female ratio of 2.15:1; 32 patients (39%) had bilateral involvement, while 50 patients (60.9%) had unilateral involvement, of whom 28 (56%) had right foot involvement and 22 (44%) had left foot involvement.

Age at initiation of treatment, severity of the initial clubfoot deformity measured by the Pirani Severity Score System⁹, total numbers of Ponseti casts before tenotomy, details of tenotomy, and compliance with use of a CTEV brace were all recorded.

Clinical evaluation was performed using the functional Ponseti Scoring System (with a maximum of 100 points indicating a normal foot) which includes: the incidence of residual and recurrent deformities, passive range of movement (measured by goniometer), appearance, muscle power, calf atrophy, and foot size. The results were graded as Excellent (90-100 points), Good (80-89 points), Fair (70-79 points) and Poor (less than 70 points). Poor and fair results were considered failures and needed further management for residual or recurrent deformity.

At the beginning of treatment, 76 patients (92.68%) were between 0 and 12 weeks of age (mean 14 days, range 3-81 days), 4 patients (4.88%) were between 13 and 24 weeks of age (mean 15 weeks) while 2 (2.44%) patients were between 25 and 36 weeks of age (mean 34).

weeks). Before beginning treatment, 53 children had a Pirani severity score of six, 22 children had a score of five and 7 children had a score of four. In patients with unilateral involvement, the mean Pirani score was 5.56 (range 4.3-6). Moreover, functional assessments including gait, functional limitation, shoe wear, pain and patient satisfaction were recorded. Radiological assessment was not usually performed in our study.

The patients were followed up on a weekly basis during the initial stages of treatment. After a brace was applied, the patient was seen monthly for 3 months and then once every 3 months.

Statistical Analysis

The association between categorical variables (initial severity score of babies with or without tenotomy) was examined using the chi square test. Data analysis was performed using the SPSS 16.0 for Windows (SPSS Inc., Chicago, IL, USA). *p* values of less than 0.05 were considered statistically significant¹⁰.

Results

The mean number of casts applied to obtain correction was 6.6 (range 5-10). The more severe the initial deformity, the more casts were required to obtain correction, particularly in cases in which treatment was initiated after 15 weeks of age. Sixty-eight children (82.93%) needed percutaneous tenotomy, 28 in the bilateral involvement group and 40 in the unilateral group, for a total of 96 feet (84.2%). (Table I). In all patients, 3 weeks after the final cast was removed, a Denis-Browne splint was applied 24 hours a day for 3 months and then at night only for 3 years.

Initial correction was obtained in all 114 clubfeet (100%) with the Ponseti method. Five feet (4.39%) in 3 children (3.66%) experienced a relapse of the deformity. Patient age at the time of relapse, bilateralism or unilateralism of the relapsed foot, relapsed foot deformity, treatment of-

Table I. Results of Pirani scores in the initial presentation vs need for tenotomy.

Tenotomy	Mean Pirani score	Number of feet	Standard deviation	Median	Minimum	Maximum
No	4.75	18	1.34	5.00	1.00	6.00
Yes	5.71	96	0.66	6.00	2.00	6.00
Total	5.56	114	0.87	6.00	1.00	6.00

p = 0.010

Table II. Details of relapsed cases.

Patient's age at relapse (months)	Side of relapsed foot	Relapse deformity	Treatment offered to correct the deformity	Result of treatment	Result at four year follow-up
9	Bilateral	Adductus & varus	Repeat tenotomy	Good	Good
18	Right	Equinus	Repeat tenotomy	Good	Good
18	Bilateral	All four deformities	Repeat tenotomy	Poor	Poor

fered for the relapsed foot, immediate results of the treatment offered, assessed by the Pirani Severity Score, and results at a mean 4 year follow-up assessed by the Ponseti Functional Scoring System are shown (Table II). Thus, of 3 relapsed patients, 3 clubfeet (60%) in 2 patients (66.67%) had an excellent to good functional outcome and 2 clubfeet (40%) in 1 patient (33.33%) had a poor functional outcome according to the Ponseti Functional Scoring System at a mean follow-up of 4 years. Splint compliance was compromised in all the relapsed cases due to incorrect use.

Few complications were encountered (Table III). Two children (2.44%) experienced a phlebostatic syndrome that healed without any medication when the cast was removed for 5 days. One child (1.22%) had a plaster sore on the lateral aspect of the skin overlying the talar head. This healed with a local dressing only. The mean time to heal the score was 7 days. Two children (2.44%) presented

Table III. Report of complications in 82 young patients.

Complications	Rate
Recurrences Splint sore Phlebostatic syndrome Plaster sore	3.66% 2.44% 2.44% 1.22%

with minor heel sores related to the use of the D-B splint (one case bilaterally and one involving only the unaffected foot) that needed local dressing and discontinued use of the brace for 10 days. No complications following tenotomy and correction of equinus, including serious bleeding following tenotomy or any wound problems with percutaneous incision were encountered.

At a mean four year follow-up, we found a nearly normal passive range of motion in 81 patients (98.78%) representing 112 clubfeet (98.25%). The parents of 45 patients (54.88%) accepted the appearance of the clubfoot as nearly normal and the parents of 36 patients (43.90%) accepted the appearance of the clubfoot as normal. Following the functional Ponseti Scoring System, good to excellent results were obtained in 79 patients (96.34%) representing 109 clubfeet (95.61%) at a mean 4 year follow up.

In the Table IV our results are compared to other published studies.

Discussion

There are different methods available for the treatment of clubfoot, regardless of the severity of the deformity. The treatment which has been shown to have the best long-term success rate is

Table IV. Results of the comparison regarding use of the Ponseti Method in three studies.

	Pavone et al	Porecha et al ²³	Bor et al ²⁴
Patients	82	49	74
Clubfeet	114	67	117
Male (Sex Ratio)	56 (2.1)	39 (3.9)	48 (1.8)
Bilaterality	32 (39.02%)	18 (36.73%)	43 (58.11%)
Mean Pirani severity score	5.56	5.83	5.09
Mean follow-up years	4	5	6.3
Mean number of casts	6.6	6.8	6.3
Tenotomy feet	96 (84.2%)	65 (97.0%)	108 (92.3%)
Good to excellent outcome	95.61%	86.56%	89.2%
Relapses	3 (3.7%)	14 (28.6%)	24 (32.4%)
Complications	6 (7.3%)	2 (4.1%)	

the Ponseti method^{7,8}, which includes serial corrective manipulation, a specific technique of cast application, a possible percutaneous Achilles tenotomy and a specific brace.

Recently, several studies of the Ponseti method have shown excellent results^{11,12}. The Ponseti technique, available for more than 50 years, has become accepted worldwide because extensive open surgery is commonly associated with long-term stiffness and weakness which is avoided by the Ponseti technique¹³⁻¹⁶. However, the literature contains primarily short-term follow-up studies^{14,17} while long-term follow-ups are scarce^{18,19}. Incorrect casting technique, improper tenotomy, under-corrected deformity, ill-fitting splints, lack of understanding and poor compliance of patients' parents due to a poor socio-economic situation can all affect a successful outcome and are the most common problems²⁰⁻²².

Poor splint compliance was a major issue especially in children of parents whose education level was inadequate. In our investigation, out of 3 relapses we found that in 2 patients, the Denis-Browne splint was used infrequently due to a lack of understanding and poor compliance of the parents due to poor socio-economic status.

Several Authors have studied whether the initial age at presentation impacts the results of treatment. Abdelgawad et al¹⁴ reported a 6.6% failure rate in patients who presented late for treatment (mean age, 36.3 weeks). Other reports have suggested age at presentation does not affect the end result of treatment; 19% of patients were older than 6 months in a study by Morcuende et al^{21,23}. All 17 patients who presented for treatment after walking age achieved full correction in the Lourenco et al work²⁴, and good results were achieved in a previous study that consisted of babies whose average age was 5 months. We found no relation between the final range of motion and the patients' age at presentation for treatment even if an increased period of casting was needed (mean number of 8 casts).

The number of casts per foot in our study was five to ten (average 6.6). In a series by Ponseti et al^{15,16}, the number of casts per foot was also five to ten (average 7.6). In another study by Laaveg et al¹⁹, the mean number of casts during their treatment was seven. Over time, as experience of this technique increases, clinicians have started to change the plaster casts at shorter intervals²⁵⁻²⁷.

In our series, tenotomy was required in 84.2% of cases and in all cases the initial Pirani score was > 5. This means that tenotomy was required

in those patients who initially had a severe deformity. Our percentage is lower than that found in other series. Porecha et al²⁸ performed tenotomy in 97.0% of cases while Bor et al²⁹ performed tenotomy in 92.3% of subjects (Table IV).

It has previously been shown that relapses occur in severe clubfeet whether these are treated surgically or nonsurgically ³⁰. The Ponseti technique is flexible in that it provides an opportunity to recast patients who lose their corrections. The relapsing cases were related to a delay in procurement and use of fabricated abduction foot braces³⁰. However, relapses were not related to the patient's age at presentation nor to the number of casts required for correction²⁷. The relapse rate in our series was comparable (3.66%) to that reported by Ponseti²⁷ in noncompliant patients using the straight-laced shoe and abduction bar protocol (7%). Our results and those of Ponseti and others suggest that the importance of maintaining correction with the foot abduction bar is paramount for successful treatment^{15,17,31}. There was no previously untreated clubfoot patient treated with the Ponseti method who relapsed. All the patients with relapsed clubfeet were successfully treated with further manipulations and recastings for two to six weeks with or without tendo-Achilles tenotomy/lengthening and foot abduction bar regimens.

In our research, the Ponseti method proved successful, with 95.61% of cases (109 clubfeet) achieving an excellent to good outcome when evaluated by the functional Ponseti Scoring System. Other Authors, after different follow-up periods, have also reported the outcome of the Ponseti technique. Porecha et al²⁸ reported an excellent to good outcome in 86.56% of cases (58 clubfeet). In a report by Bor et al²⁹, the Ponseti method proved largely successful, with 89.2% (99 clubfeet) achieving a good outcome. Ippolito et al²⁰ compared babies treated with different protocols (Ponseti and the Marino-Zuco method). In the Ponseti group, 78% of the feet achieved excellent or good results compared with only 43% of feet in the non-Ponseti group.

Radiographic data are of little value and we agree with Roye et al²⁶ concerning its poor utility in evaluation of clubfoot outcome. Indeed despite radiographically imperfect feet, most patients demonstrate excellent levels of function³¹. A German work noted the wide distribution of the talocalcaneal angle in both normal and clubfeet and reasoned that drawing conclusions based on changes in this number with a large standard deviation made little sense³². Considering that there was no relationship

between radiographic results and function, we preferred to use clinical evaluation to assess the outcome of congenital talipes equinovarus.

The aim of obtaining a straight, painless, plantigrade flexible, normal-looking foot, which allows the child to live a regular daily life, is achieved by the Ponseti Method which remains the best way to treat the ICTEV anomaly.

Acknowledgements

We wish to thank International Science Editing Co, Shannon Ireland, for editing the manuscript.

Conflict of Interest

The Authors declare that they have no conflict of interests.

References

- CAREY M, BOWER C, MYLVAGANAM A, ROUSE I. Talipes equinovarus in Western Australia. Paediatr Perinat Epidemiol 2003; 17: 187-194.
- LOCHMILLER C-L, JOHNSTON D, SCOTT A, RISMAN M, HECHT JT. Genetic epidemiology study of idiopathic talipes equinovarus. Am J Hum Genet 1998; 79: 90-96.
- Wallander H, Hovelius L, Michaelsson K. Incidence of congenital clubfoot in Sweden. Acta Orthop 2006; 77: 847-852.
- CARTLIDGE IJ. Observations on the epidemiology of clubfoot in Polynesian and Caucasian populations. J Med Genet 1984; 21: 290-292.
- DE ANDRADE M, BANHOLTZ JS, AMOS CL, LOCHMILLER C, SCOTT A, RISMAN M, HECHT JT. Segregation analysis of idiopathic talipes equinovarus in Texan population. Am J Med Genet 1998; 79: 97-102.
- DANIELSSON LG. Incidence of congenital clubfoot in Sweden: 128 cases in 138.000 infants 1946-1990 in Malmo. Acta Orthop Scand 1992; 63: 424-426.
- 7) PONSETI IV. Treatment of congenital clubfoot. J Bone Joint Surg Am 1992; 74: 448-454.
- 8) PONSETI IV. The treatment of congenital clubfoot. J Orthop Sports Phys Ther 1994; 20: 1.
- DYER PJ, DAVIS N. The role of the Pirani scoring system in the management of club foot by the Ponseti method. J Bone Joint Surg Br 2006; 88: 1082-1084.
- 10) PLACKETT RL. Karl Pearson and the Chi-Squared test. International Statistical Review (International Statistical Institute ISI) 1983; 51: 59-72.
- ARONSON J, PUSKARICH CL. Deformity and disability from treated clubfoot. J Pediatr Orthop 1990; 10: 109-119.
- Herzenberg JE, Radler C, Bor N. Ponseti versus traditional methods of casting for idiopathic clubfoot. J Pediatr Orthop 2002; 22: 517-521.
- 13) LEHMAN W-B, MOHAIDEEN A, MADAN S, SCHER DM, VAN BOSSE HJ, IANNACONE M, BAZZI JS, FELDMAN DS. A method for the early evaluation of the Ponseti (Iowa) technique for the treatment of idiopathic clubfoot. J Pediatr Orthop 2003; 12: 133-140.

- 14) ABDELGAWAD AA, LEHMAN WB, VAN BOSSE HJ, SCHER DM, SALA DA. Treatment of idiopathic clubfoot using the Ponseti method: minimum 2-year followup. J Pediatr Orthop B 2007; 16: 98-105.
- 15) PONSETI IV. Clubfoot Management. Editorial. J Pediatric Orthopaedics 2000; 20: 699-700.
- 16) PONSETI IV. Relapsing clubfoot: causes, prevention, and treatment. Iowa Orthop J 2002; 22: 55-56.
- COLBURN M, WILLIAMS M. Evaluation of the treatment of Idiopathic clubfoot by using the Ponseti Method. J Foot Ankle Surg 2003; 42: 259-267.
- 18) COHEN-SOBEL E, CASELLI M, GIORGINI R, STUMMER S. Long-term follow-up of clubfoot surgery: analysis of 44 patients. J Foot Ankle Surg 1993; 32: 411-423.
- LAAVEG SJ, PONSETI IV. Long-term results of treatment of congenital clubfoot. J Bone Joint Surg Am 1980; 62: 23-31.
- IPPOLITO E, FARSETTI P, CATERINI R, TUDISCO C. Longterm comparative results in patients with congenital clubfoot treated with two different protocols. J Bone Joint Surg Am 2003; 85: 1286-1294.
- MORCUENDE JA, DOLAN LA, DIETZ FR, PONSETI IV. Radical reduction in the rate of extensive corrective surgery for clubfoot using the Ponseti method. Pediatrics 2004; 113: 376-380.
- Goksan SB. Treatment of congenital clubfoot with the Ponseti Method. Acta Orthop traumatol Turc 2002; 36: 281-287.
- MORCUENDE JA, ABBASI D, DOLAN LA, PONSETI IV. Results of an accelerated Ponseti protocol for club-foot. J Pediatr Orthop 2005; 25: 623-626.
- 24) LOURENCO AF, MORCUENDE JA. Correction of neglected idiopathic club foot by the Ponseti method. J Bone Joint Surg Br 2007; 89: 378-381.
- COOPER DM, DIETZ FR. Treatment of idiopathic clubfoot. A thirty year follow-up note. J Bone Joint Surg 1995; 77: 1477-1489.
- ROYE BD, VITALE MG, GELINS AC, ROYE DP Jr. Patient-based outcomes after clubfoot surgery. J Pediatr Orthop 2001; 21: 42-49.
- PONSETI IV, SMOLEY EN. Congenital clubfoot: the results of treatment. J Bone Joint Surg (Am) 1963;
 45: 2261-2275.
- 28) PORECHA MM, PARMAR DS, CHAVDA HR. Mid-term results of Ponseti method for the treatment of congenital idiopathic clubfoot (A study of 67 clubfeet with mean five year follow-up). J Orthop Surg Res 2011; 12: 3.
- Bor N, Coplan JA, Herzenberg JE. Ponseti treatment for idiopathic clubfoot: minimum 5-year follow up. Clin Orthop Relat Res 2009; 467: 1263-1270.
- BOR N, HERZENBERG JE, FRICK SL. Ponseti management of clubfoot in older infants. Clin Orthop Relat Res 2006; 444: 224-228.
- HUTCHINS PM, FOSTER BK, PATERSON DC, COLE EA. Long term results of early surgical release in club feet. J Bone Joint Surg Br 1985; 67: 791-799.
- Herbsthofer B, Eckardt A, Rompe KD, Küllmer K. Significance of radiographic angle measurements in evaluation of congenital clubfoot. Arch Orthop Trauma Surg 1998; 117: 324-329.