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Research Article

Factors Associated with Recurrence after use of the Ponseti Method for the Treatment of Idiopathic Clubfeet

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Abstract

Background: Nowadays, the Ponseti technique has become the treatment of choice for clubfeet. Meanwhile, it is not devoid of recurrences. The current study aimed at describing the socio-demographic and clinical characteristics of the children having clubfeet treated by the Ponseti technique associated with rehabilitation, as well as identifying factors associated with recurrences of clubfoot.

Methods: We conducted a retrospective monocentric study between January 2012 and January 2016 among children having clubfeet treated with Ponseti associated with appropriate rehabilitation in the department of Physical medicine and Rehabilitation of Mahdia (Tunisia). We considered a recurrence any foot having a Dimeglio score > 5 and / or a Pirani score > 1.5 after a good initial correction. For each recurrence we note the age of recurrence, the corresponding Pirani and Dimeglio score, compliance to the use of orthosis and rehabilitation, the treatment of recurrence and the resulting therapeutic outcome.

Results: Fifty-four children having 81 clubfeet were retained. The recurrence rate was 13.5% of cases. The main factors incriminated in the occurrence of recurrences are: the presence of a social security ($p < 0.04$), the pudgy morphology of the foot ($p < 0.01$), the number of plaster cast ($p < 0.04$), compliance with the use of orthosis ($p < 0.006$) and length of stay ($p < 0.015$) and compliance with rehabilitation ($p < 10-3$).

Conclusion: Compliance with the use of orthosis is the best preventive measure for recurrence. The early rehabilitation and adapted complementary seems to build a new solution that could improve the therapeutic results of the Ponseti technique that deserves more attention.

Introduction

Idiopathic congenital talipes equinovarus (clubfoot) is a complex deformity which is difficult to correct. The deformity has four components: equinus, hind foot varus, forefoot adductus, and cavus. The goal of treatment is to reduce or eliminate these four deformities so that the patient has a functional, pain-free, plant grade foot, with good mobility and without calluses, and does not need to wear modified shoes. There is a universal agreement that the initial treatment of idiopathic congenital clubfoot should be non-operative, regardless of the severity of the deformity.

Historically, this treatment consisted of forcible serial manipulations with the patient under anesthesia, followed by application of a cast [1]. Today, non-operative treatment typically involves serial gentle manipulations followed by the application

of a short or long leg cast at weekly interval [1,2]. While this technique is the mainstay of non-operative intervention in North America, physiotherapy and continuous passive motion without immobilization have been successfully used in France [3]. Although all of these methods have the potential to be successful when applied correctly, most authors have reported a success rate of only 15% to 50% [4,5].

A notable exception is the Ponseti method, which involves serial manipulation, a specific technique of cast application, and a possible percutaneous Achilles tenotomy. The method has been reported to have short-term success rates approaching 90% [6], and the long-term results have been equally impressive [7]. Although this method remains the most popular with a significantly higher success rate than other techniques such as physiotherapy or surgery, the Ponseti method retains a rate of recurrence and / or surgical recourse of between 10 and 20% [8]. These unsatisfactory results of the Ponseti method have been attributed in most cases to noncompliance with the use of orthosis [9].

To our knowledge, no study has addressed the problem of noncompliance of parents to treatment and the risk factors for recurrence of clubfoot treated with the Ponseti technique associated with rehabilitation. The current study aimed at describing the socio-demographic and clinical characteristics of the children having clubfoot treated by the Ponseti technique associated with rehabilitation, as well as identifying factors associated with recurrences of clubfoot.

Material and methods

Population Study

We included in this study all children aged < 1 year with idiopathic clubfoot treated with Ponseti technique combined with appropriate rehabilitation. Children with secondary clubfoot (polymal formative syndrome, arthrogryposis) and first-line operated children were excluded.

Study Design and Data Collection

We conducted a monocentric retrospective descriptive study between January 2012 and January 2016 among children having clubfoot treated with Ponseti associated with appropriate rehabilitation in the department of Physical medicine and Rehabilitation of Mahdia (Tunisia).The technique of Ponseti as described by its author [10] is based on the manufacture of successive long leg plaster casts (renewed every week) until the disappearance of the deformation of the forefoot (cavus, adduction, supination) followed by a percutaneous tenotomy of the Achilles tendon if the dorsal flexion remains below 15° [11].

A post-tenotomy plaster is made and kept for 3 weeks. Then the feet are immobilized in 60° external abduction-rotation and 15° dorsal flexion using the abduction orthosis described by Steenbeek Foot Abduction Brace (SFAB). This orthosis was worn 23 hours / day for 3 months (full time) then only at night or during naps until the age of 3 years (part time). The rehabilitation program was undertaken as soon as the last cast was removed at 3 sessions per week for 1 to 3 years depending on the condition of the corrected foot. The protocol involves mobilizations of the foot after derotation of the calcaneo-pedis block, stretching of the Achilles tendon, stimulation of the peroneus muscles as well as exercises of "neuro-motor" rehabilitation to stimulate the eversion muscles.

Regarding Data Collection

We gathered socio-demographic information (age, gender, presence or absence of social security, educational level of parents) and clinical characteristics (the affected side, the overall morphology of the foot, the number of plaster and the indication of tenotomy). We evaluated the severity of deformation by the Pirani score [12] (annex I) and Dimeglio score [13] (annex II).

The feet were then classified according to the Dimeglio score into four categories with regard 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, the most common category, indicates 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, and an arthrogrypotic appearance.

We appreciated the compliance of children with the brace and the rehabilitation (Table 1).

	Compliant	Non-compliant
Full time (First 3 months)	> 22 hours / day	< 22 hours / day
Part-time (From 3 months - 3 years)	8 hours / day And / or for a period > 18 months	8 hours / day And / or for a period < 18 months

Table1: Compliance criteria for children wearing braces.

Furthermore, we considered a "recurrence" as a foot having a Dimeglio score > 5 and/or a Pirani score > 1.5 after a good initial correction. For each identified case of recurrence, we noted the age, the corresponding Pirani and Dimeglio scores, compliance to the use of orthosis and rehabilitation, the treatment of recurrence and the resulting therapeutic outcome.

Statistical Analysis

Data were entered and analyzed using the SPSS 16.0 software. We performed a univariate analysis to examine association between

recurrence (our dependent variable) and different variables particularly the age, the corresponding Pirani and Dimeglio score, compliance to the use of orthosis and rehabilitation. Thus, we used the Chi^2 test for qualitative variables and the Student t test (or the Mann Whitney test when appropriate) for quantitative variables. p values <0.05 were considered statistically significant.

Results

Initially, 66 patients with 99 clubfeet were included in our study. Then, we excluded 12 children because they were lost to follow-up. Consequently, 54 children having 81 clubfeet were retained (Figure 1).

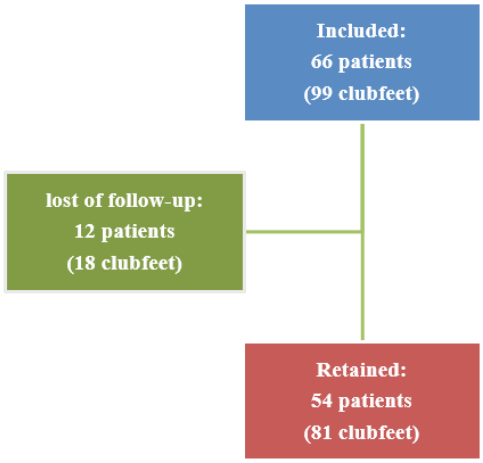

Figure 1: Flowchart of the study sampling Socio-demographic and clinical characteristics of the patients.

Socio-Demographic and Clinical Characteristics of the Patients

There were 40 boys and 14 girls with a sex-ratio of 2.86. Forty-three children (79.6%) had social security. The parental educational level was high in 35.1% of cases. Among the 81 included clubfeet, 51.8% of them were grade IV. Table 2 summarizes socio-demographic and clinical information's related to our population study.

	Mean (range)	Number	Percentage (%)
Age (days)	50 (1-364)		
Gender			
Male		40	74
Female		14	26
social security			
Yes		43	79.6
No		11	20.3
educational level of parents			
high school		19	35.1
less than high school		35	65.8
Age at the first plaster (days)	61 (1-364)		
Morphology of the foot			
Pudgy		14	17.2
Fine		67	82.7
Affected side			
Bilateral		26	48.1
Right		13	24
Left		15	27.7
Peroneus muscles testing < 3		81	100
Pirani score	5,1 (2-6)		
Dimeglio score	15 (8-20)		
Grade			
I		4	4.9
II		12	14.8
III		23	28.3
IV		42	51.8
Number of plaster	4,63		
Indication of tenotomy			
Yes		64	79
No		17	21

Table 2: Socio-demographic and clinical characteristics of the patients.

The mean follow-up was 26.7 months (range: 2-42 months). The mean full-time immobilization was 2 months 3 weeks (range: 0-3 months) and only 93.8% of children having 76 clubfeet were compliant with the use of orthosis (Table 3). The duration of wearing/day was 22h/day in 91.3% of the cases and the orthosis were used for 3 months in 97.5% of the cases (Table 3).

Orthosis SFBA made for 81 clubfeet		
Duration of wearing/ day		
22h/d	74	91.30%
<22h/d	3	3.70%
Use of orthosis		
for 3 months	79 clubfeet	97.50%
<3months	2 clubfeet	2.40%
Mean duration of immobilization 2months 3 weeks(0-3months)		
Complaint	76 clubfeet	93.80%
Non complaint	5 clubfeet	6.10%

Table 3: Full-time immobilization phase.

On a part-time basis, the number of children compliant with the use of orthosis was 49 (74 clubfeet) (Table 4). The mean duration of immobilization was 20.15 months (range: 1-38 months). It was between 18 and 36 months in 89% of the cases (Table 4). The duration of wearing/day was more than 8 hours in 93.8% of the cases (Table 4).

Part -time wearing orthosis for 81 clubfeet		
Duration of immobilization	20.15 months	1 to 38 months
<18 months	5 clubfeet	6.10%
18-36 months	66 clubfeet	89%
>36 months	3 clubfeet	4%
Duration of wearing/ day		
<8h	5 clubfeet	6.10%
>8h	76 clubfeet	93.80%
Complaint	74 clubfeet	91.30%
Non complaint	7 clubfeet	8.60%

Table 4: Part-time immobilization phase.

During the full-time immobilization phase, 52 children with 78 clubfeet (96.2% of cases) benefited from a rehabilitation and they continued in the phase of part-time immobilization with a mean duration of 18.61 months (Table 5). The mean number of sessions per week was 1.76 (range: 1-3).

	Full time	Part time
Rehabilitation	78 clubfeet (52 children) 96.2%	78 clubfeet (52 children) 96.2%
Duration of rehabilitation	3months	18.61 months (1 to 36months)
Number of sessions	2.7 sessions/week	1.76 sessions / week

Table 5: Rehabilitation during the two immobilization phases.

Overall, the recurrence rate was 13.5% (11 clubfeet); 90.9% of them (10 clubfeet out of 11) occurred in the first year and 18.2% in the first 3 months. The mean age of recurrence was 11.33 months. The mean Pirani score during recurrence was 2.45. Recurrence was bilateral in 66.7% of cases. Furthermore, all recurrent feet received another treatment attempt using the Ponseti technique.

Factors Associated with Recurrence of Clubfeet

Univariate analysis Table 6 showed that factors significantly associated with clubfeet recurrence were the presence of social security ($p < 0.04$), the pudgy morphology of the foot ($p < 0.01$), the number of plaster cast ($p < 0.042$), compliance with the use of orthosis ($p < 0.006$) and length of stay ($p < 0.015$) and compliance with rehabilitation ($p < 10^{-3}$). However, recurrence was not related to the educational level of parents, the age at first plaster, the gender, the affected side, the pre-treatment, the initial severity of deformity nor to the performance of tenotomy.

		Recurrence		p value
		Yes (n=11)	No (n=70)	
Social Security	Yes	3	8	0.04
	No	64	6	
Educational level of parents	High school	2	26	0.055
	Less than high school	9	44	
Gender	Male	6	48	0.103
	Female	5	22	
Affected side	Unilateral	4	25	0.36
	Bilateral	7	45	
Mean age at the first plaster (Months)		63.2	47.2	0.78

Morphology of the foot	Fine	2	65	0.01
	Pudgy	9	5	
Pre-treatment	Yes	2	9	0.44
	No	9	61	
Mean Initial Pirani score		5.53	5.03	0.52
Mean Initial Dimeglio score		15.5	14.72	0.95
Mean Number of plaster		4.93	4.07	0.04
Tenotomy	Yes	5	59	0.07
	No	6	11	
Compliance with the use of orthosis	Yes	5	69	0.006
	No	6	1	
Mean Duration of use of orthosis (months)		7.45	18.45	0.015
Mean Duration of rehabilitation (months)		7.6	17.89	$<10^{-3}$

Table 6: Factors associated with clubfeet recurrence.

Discussion

In this study, we have shown that the occurrence of recurrence during treatment of clubfoot by the Ponseti technique coupled with rehabilitation is related to compliance with the use of orthosis and rehabilitation, to the morphology of the foot as well as to the number of plaster made. Like any treatment, the Ponseti technique has a certain rate of recurrences. In our study, it was 13.5% of cases, lower than that found by Panjavi (18%) [14] and Dobbs (31%) [9]. This rate of recurrence found during the treatment of clubfoot by the Ponseti technique is still lower than that reported after surgical treatment. Indeed, Jose A and Ponseti noticed that the recurrence rate went from 46% to 11% for a series of 256 clubfeet treated with the Ponseti technique [15]. Contrary to what is believed, recurrences do not occur after an incomplete correction of the deformity, but rather, they are due to the same initial physio pathological mechanism of clubfoot [5].

In fact, the clubfoot, a congenital malformation occurring in the 2nd trimester of pregnancy [4], is due to an excess of synthesis and accumulation of collagen in the tendons and ligaments [16]. This accumulation of collagen is at its maximum the first year of life and continues until the 3rd or 4th year of life [5]. Relapses occur therefore, because the factors inducing deformation are still active. They become rare after four years, regardless of whether the deformity is corrected in whole or in part [5]. This explains two important points: The first point, the high rate of recurrence during the first year. Indeed, in our study, 90.9% of recurrences occurred during the first year. The mean age of recurrence was 11.33 months, higher than that found by Dobbs (6 months).

The second point, the importance of immobilization with the orthosis for 3 to 4 years to prevent recurrence. It is for this reason that noncompliance with the use of orthosis has always been incriminated in the genesis of recurrences. In our series, of the 7 non-compliant clubfeet, 5 recurred ($p = 0.006$). This rate is close to that found by Geoffrey ($p = 0.0009$). Similarly, Matthew and Dobbs have shown that a non-compliant child with the use of orthosis is 183 times more likely to reoffend [9]. Rehabilitation is not indicated in the treatment of clubfoot by the technique of Ponseti and the peculiarity of our study lies in the prescription of early and adapted physiotherapy associated with immobilization by the orthosis. We showed that compliance to rehabilitation and its duration were related to recurrence ($p < 10^{-3}$). In addition, we found that children without social security are more likely to reoffend ($p < 0.04$). This can be explained by the fact that without social security, children cannot buy the orthosis or continue the rehabilitation sessions.

On the other hand, and contrary to what is found by Changulani and Dobbs [9,17], we did not find any relation between the occurrence of recurrence and the educational level of the parents. Moreover, although several studies recommend the Ponseti technique at an early age [18], we did not find any relationship between the recurrence rate and the age of the first plaster ($p > 0.05$). Our results were consistent with those found by Ponseti and Dobbs. Indeed, they showed that the relapses are not related to the age at the time of the diagnosis nor to the age of the 1st plaster.

Although there is a tendency towards an increased risk of recurrence in children with the most severe deformity, our results demonstrated that the occurrence of recurrence is not dependent on the severity of the initial deformity. Our results are similar to those found by Dobbs and Ponseti [5,6]. Similarly, we did not find a correlation between the risk of recurrence and the age at the start of treatment, gender, the side affected, the bilaterality and the previous treatment although it was recommended by some teams in the severe clubfeet to soften the feet before treatment with Ponseti but without objective results. Our results are consistent with those of the literature [5,9,15].

Meanwhile, we noticed that pudgy feet are more likely to reoffend ($p < 0.01$). Our results are similar to those found by Rakotonirina [19]. In addition, the influence of the number of plasters made on recurrences has been discussed in several studies, with controversial results. In our study, we objectified a relationship between the number of plaster and recurrences ($p <$

0.042). In fact, the average number of plasters for recurrent feet was statistically higher (4.97 plasters) than that of well corrected feet (4.07 plasters). Consistent with our findings, Panjavi noted that the duration and number of plasters required for complete correction is strongly correlated with recurrence [14]. On the other hand, Changulani and Jobe found that recurrence was independent of the number of plasters performed to obtain a good correction [9,17].

Conclusion

The Ponseti method allowed a quick correction of the clubfoot. However, recurrence is the main risk of this treatment and their recognition and the early initiation of appropriate treatment remains a challenge. In our study, the main factors incriminated in the occurrence of recurrences are: the presence of a social security, the pudgy form of the foot, the number of plaster achieved, the compliance with the use of orthosis and duration of wearing as well as compliance with rehabilitation.

Failure with the Ponseti method has been frequently attributed to noncompliance with the use of the orthosis after correction has been obtained. This is not a trivial issue, given the expense and the time (two to four years) that is required, as well as the psychosocial factors, such as the stigma of prolonged use of an orthosis, which have an impact on compliance. The use of orthosis is undoubtedly the best preventive, but once installed another therapeutic attempt by Ponseti can be considered. The early rehabilitation and adapted complementary seems to build a new track that could improve the therapeutic results of the Ponseti technique that deserves more attention.

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Annex I : Pirani score

LOOK

Curvature of lateral border | **Medial crease** | **Posterior crease**

0 = Normal

0.5 = Moderate

1 = severe

FEEL

Lateral part of head of talus

0 = Complete reduction

0.5 = Partial reduction

1 = Fixed subluxed

Emptiness of the heel

0 = Tuberosity palpable

0.5 = Tuberosity partially palpable

1 = Tuberosity not palpable

MOVE

Rigidity of equinous

0 0.5 1

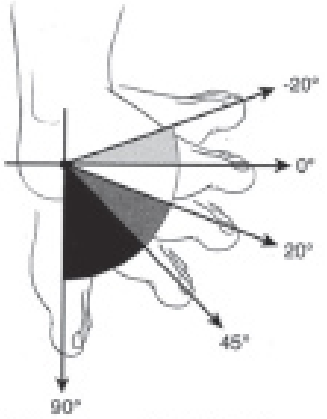
Catterall / Pirani (Normal: 0 points; Most abnormal: 1.0 points)

Hindfoot contracture (HFCS)	Points	Midfoot contracture (MFCS)	Points
a. Posterior crease: 0, 0.5 or 1.0 points		a. Curvature of lateral border: 0, 0.5 or 1.0 points	
b. Empty heel: 0 or 1.0 points		b. Medial crease: 0, 0.5 or 1.0 points	
c. Rigid equinus 0, 0.5 or 1.0 points		c. Lateral head of talus: 0, 0.5 or 1.0 points	
HFCS sub-total		**MFCS sub-total**	**Total Score (HFCS and MFCS)**

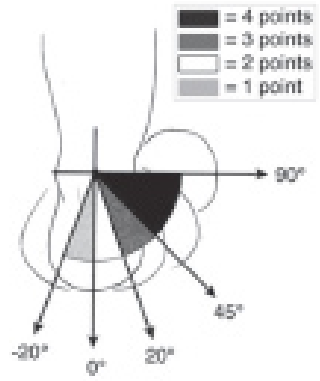
Annex II : Dimeglio score

Classification

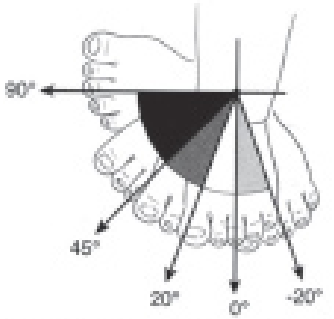
Classification Grade	Type	Frequency (%)	Score
I	Benign	20	(<5)
II	Moderate	33	(=5<10)
III	Severe	35	(=10<15)
IV	Very severe	12	(=15<20)



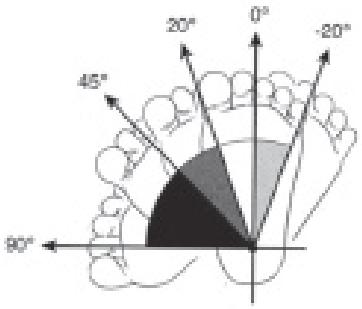
Sagittal plane evaluation of equinus.



Frontal plane evaluation of varus.



Horizontal plane evaluation of derotation of the calcaneopedal block.



Horizontal plane evaluation of forefoot relative to hindfoot.

Assessment of Clubfoot by Severity Scale

Characteristics: Reproducibility	Points	Characteristics: Other parameters	Points
90°-45°	4	Posterior crease	1
45°-20°	3	Medial crease	1
20°-0°	2	Cavus	1
<0° to -20°	1	Poor muscle condition	1