Initial treatment of congenital idiopathic clubfoot: Prognostic factors

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A B S T R A C T

Background: The initial treatment of congenital idiopathic clubfoot (CIC) is nonoperative. Either the French physiotherapy method or the Ponseti casting method may be used. Whether either method is superior over the other remains unclear. However, the method used initially is not the only determinant of the final outcome.

Objective: The primary objective was to identify determinants of the final outcome as evaluated based on the need for surgical treatment and on the Ghanem-Seringe score.

Hypothesis: Factors associated with the final outcome can be identified.

Methods: Between 2004 and 2011, 100 CICs in 79 patients were treated in two centres, 47 using the French method and 53 the Ponseti method. The Diméglio grade was determined at baseline and the Ghanem-Seringe score at last follow-up. Surgical procedures (if any), splinting duration, and rehabilitation therapy duration were recorded. The two groups showed no statistically significant differences for Diméglio grade distribution, time from birth to treatment initiation, or mean follow-up.

Results: Factors significantly associated with a poor outcome by univariate analysis were use of the Ponseti method ($P=0.0027$), older age at last follow-up ($P=3 \times 10^{-4}$), initial Diméglio grade ($P=7 \times 10^{-5}$), and need for surgery ($P=10^{-5}$); no significant effect was found for splinting duration, rehabilitation duration, bilateral involvement, or antenatal diagnosis. By multivariate analysis, factors independently associated with a poor prognosis were older age at last follow-up, Diméglio grade, and need for surgery.

Conclusion: This study confirms the major prognostic significance of initial severity (Diméglio grade) on the final outcome. The data do not firmly establish that one method is superior over the other. Nevertheless, the need for percutaneous Achilles tenotomy with the Ponseti method leads us to prefer the French physiotherapy method.

Level of evidence: IV, retrospective study.

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1. Introduction

Congenital idiopathic clubfoot (CIC) affects one to two infants in every 1000 live births. The initial treatment relies on nonoperative methods, i.e., either a sequence of casts (Ponseti method) [1] or physiotherapy (French method) [2]. Whether either method is superior over the other remains unclear [3]. Chotel et al. [4] reported better outcomes with the Ponseti method in patients with Diméglio grade 3 or 4 CIC but no difference between the methods for grade 2 CIC. In contrast, Richards et al. [5] found higher recurrence rates with the Ponseti method. Clearly, in addition to the initial treatment method, other factors also influence the final outcome [6].

We therefore conducted a study in two paediatric orthopaedic centres to identify factors associated with treatment outcomes in CIC. We assessed outcomes based on the Ghanem-Seringe score at last follow-up [7] and the proportion of patients requiring surgery.

2. Patients and method

This was a retrospective study, for which ethics committee approval was not required. Patients from two paediatric orthopaedic centres were studied. Consecutive patients born during the 10-year period from 2000 to 2010 with unilateral or bilateral CIC Diméglio grade 2 or higher [8] were included if they had learned to walk at least 6 months before the last follow-up visit. Of the 93 patients who met these inclusion criteria, 17 were excluded, because they transferred to another centre during the treatment ($n=9$), were lost to follow-up ($n=8$), or refused the follow-up visit. One centre (47 feet in 37 patients) used the French method (FM) and the other (53 feet in 39 patients) the Ponseti method (PM).
FM [2] involved passive mobilisation and muscle stimulation by specially trained physiotherapists. Between sessions, the foot was maintained in the corrected position in a Denis Browne splint. If foot dorsiflexion was less than 10° when the infant was 6 months of age, percutaneous Achilles tenotomy was performed. The PM [1] consisted of 5 weeks of casts that were changed weekly and fashioned by an orthopaedic surgeon, followed by percutaneous Achilles tenotomy then by casting for 1 month. Maintenance physiotherapy sessions and a Denis Browne splint to be worn at night were then prescribed.

In both groups, surgery was performed if the deformities persisted or recurred. Recurrence of all three deformities was managed by posterior-medial release and isolated residual deformities by limited adjutant procedures (muscle transfer, Cahuzaq osteotomy [9], or tibial derotation osteotomy).

The study involved two phases. First, an independent observer abstracted the following data from the medical charts: demographics, side affected, Dimeglio grade, whether the diagnosis was established antenatally, and how well the parents adhered to the prescribed treatment. Then, the same observer evaluated all patients during a clinic visit, which included history taking, a physical examination, and radiographs of the affected foot or feet to allow determination of the Ghanem-Seringe score [7]. In addition to the parameters used for this score, we recorded the duration of nighttime splinting and the duration of rehabilitation therapy. Based on the results of the evaluation, the feet were classified according to Ghanem-Seringe [7] and the outcomes were divided into four groups: excellent (85–100 points), good (70–84 points), fair (60–69 points), and poor (<60 points).

Statistics: the statistical tests were performed by an independent observer using XLSTAT (Microsoft Excel®, Microsoft, Redmond, WA, USA).

To compare distributions, Student’s t-test was chosen for quantitative variables and Fisher’s exact test for qualitative variables. Pearson’s coefficient was computed to evaluate correlations between quantitative variables. Multivariate analyses were performed using logistic regression for quantitative dependent variables and linear regression for quantitative dependent variables. Results are reported as mean (range).

3. Results

Tables 1A, 1B and 2 list the main features in the two groups. The distribution of the Dimeglio grades was not significantly different between the two groups. Mean age at last follow-up was 5 years (range, 2–12 years) in the FM group and 6 years (range, 3–10 years) in the PM group (P > 0.05). Both feet were affected in 9/37 patients in the FM group and 15/39 patients in the PM group.

The overall Ghanem-Seringe score indicated an excellent outcome, with a mean of 87 (62–98). The final score was 89 (69–98) when surgery was not required and 79 (46–96) otherwise. Percutaneous Achilles tenotomy was performed on 4 (8.5%) feet (4 patients) in the FM group and 50 (94%) feet (37 patients) in the PM group.

3.1. Univariate analysis

3.1.1. Association between outcome and type of treatment

The final overall Ghanem-Seringe score was 90 ± 6.2 (75–98) in the FM group and 84 ± 11.2 (62–97) in the PM group, a statistically significant difference (P = 0.0027) (Table 3). After exclusion of the patients who wore a splint for less than 2 years (Table 4), the results remained significantly better in the FM group (P = 0.025).

Tables 5 and 6 detail the values of the various Ghanem-Seringe criteria in the two groups. In both groups, the patients engaged similarly in age-appropriate everyday and sports activities, with no restrictions.

3.1.2. Association between outcome and age at last follow-up

Older age at last follow-up was significantly associated with a worse outcome (P = 3 × 10–14) and a greater need for surgery. Fig. 1 shows the distribution of surgical procedures according to year of management.

Table 1A

<table>
<thead>
<tr>
<th>Feature</th>
<th>PM group (39 patients)</th>
<th>FM group (37 patients)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bilateral clubfoot</td>
<td>15</td>
<td>9</td>
</tr>
<tr>
<td>Males</td>
<td>30</td>
<td>29</td>
</tr>
<tr>
<td>Antenatal diagnosis</td>
<td>25</td>
<td>18</td>
</tr>
</tbody>
</table>

The data are numbers of patients.

PM: Ponseti method (casting); FM: French method (physiotherapy).

Table 1B

<table>
<thead>
<tr>
<th>Feature</th>
<th>PM group (53 feet, 39 patients)</th>
<th>FM group (47 feet, 37 patients)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Side involved: right/left</td>
<td>25/28</td>
<td>21/26</td>
</tr>
<tr>
<td>Initial Dimeglio grade</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>3</td>
<td>21</td>
<td>21</td>
</tr>
<tr>
<td>4</td>
<td>28</td>
<td>28</td>
</tr>
<tr>
<td>5</td>
<td>6</td>
<td>6</td>
</tr>
</tbody>
</table>

The data are numbers of feet.

PM: Ponseti method (casting); FM: French method (physiotherapy).

Table 2

<table>
<thead>
<tr>
<th>Feature</th>
<th>PM group (53 feet)</th>
<th>FM group (47 feet)</th>
<th>P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Number of feet with surgery</td>
<td>19 (35%)</td>
<td>9 (19%)</td>
<td></td>
</tr>
<tr>
<td>other than PAT intervention</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Postero-medial release</td>
<td>12</td>
<td>6</td>
<td>0.08</td>
</tr>
<tr>
<td>Other</td>
<td>7</td>
<td>3</td>
<td></td>
</tr>
<tr>
<td>Surgical revision</td>
<td>5</td>
<td>1</td>
<td></td>
</tr>
</tbody>
</table>

The data are numbers of feet.

PM: Ponseti method (casting); FM: French method (physiotherapy); PAT: percutaneous Achilles tenotomy.

Table 3

<table>
<thead>
<tr>
<th>Outcome</th>
<th>PM group (53 feet)</th>
<th>FM group (47 feet)</th>
<th>P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Excellent</td>
<td>32 (59%)</td>
<td>36 (76.5%)</td>
<td>0.0027</td>
</tr>
<tr>
<td>Good</td>
<td>16 (31%)</td>
<td>11 (23.5%)</td>
<td></td>
</tr>
<tr>
<td>Fair</td>
<td>3 (6%)</td>
<td>0</td>
<td></td>
</tr>
<tr>
<td>Poor</td>
<td>2 (4%)</td>
<td>0</td>
<td></td>
</tr>
</tbody>
</table>

The data are numbers of feet.

PM: Ponseti method (casting); FM: French method (physiotherapy).

Table 4

<table>
<thead>
<tr>
<th>Outcome</th>
<th>PM group (n = 29 feet)</th>
<th>FM group (n = 33 feet)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Excellent</td>
<td>14 (48%)</td>
<td>27 (82%)</td>
</tr>
<tr>
<td>Good</td>
<td>12 (41%)</td>
<td>6 (18%)</td>
</tr>
<tr>
<td>Fair</td>
<td>1 (5%)</td>
<td>0</td>
</tr>
<tr>
<td>Poor</td>
<td>2 (6%)</td>
<td>0</td>
</tr>
</tbody>
</table>

The data are numbers of feet.

PM: Ponseti method (casting); FM: French method (physiotherapy).
3.1.3. Association between outcome and initial Diméglio grade

The initial Diméglio grade correlated significantly with the final Ghanem-Seringe score (< 0.4; \( P = 1 \times 10^{-4} \)) (Table 7). A worse initial Diméglio grade was significantly associated with the need for surgery: surgery was performed in 2/7 Diméglio 2 patients, 8/61 Diméglio 3 patients, and 18/32 Diméglio 4 patients (\( P = 7 \times 10^{-5} \)). In contrast, no association was found between the initial Diméglio grade and the type of surgical procedure performed.

3.1.4. Association between outcome and need for surgery

Percutaneous Achilles tenotomy was not considered for this analysis (Table 8). On average, the need for other surgical procedures was halved in the FM group compared to the PM group, although the difference fell short of statistical significance (\( P = 0.08 \)) (Table 2). The type of initial treatment (FM or PM) was not significantly associated with the type of surgery performed (postero-medial release or other). Neither was the type of surgical procedure associated with the final outcome. Among patients who had surgery, those in the PM group were significantly less satisfied with their scar (regardless of type of procedure) compared to those in the FM group (\( P = 0.0017 \)).

All patients with fair or poor outcomes had had surgery. There was a highly significant difference in outcomes in favour of the feet that did not require surgery (\( P = 1 \times 10^{-5} \)).

3.1.5. Associations between outcome and other parameters

The instruction to use the splint at night for more than 2 years was followed more often in the FM group (70% of feet) than in the PM group (55% of feet) (\( P = 0.0001 \)). Duration of rehabilitation therapy was not significantly different between the two groups (3.7 years in the FM group and 4 years in the PM group). Outcomes were not significantly different in the group with a duration of splinting and rehabilitation longer than 2 years compared to the group with a shorter duration.

No significant differences in outcome were found between the groups with and without an antenatal diagnosis of CIC or between the groups with unilateral vs. bilateral CIC.

The final outcome was significantly better in females compared to males (\( P = 0.0061 \)).

3.2. Multivariate analysis

By multivariate analysis, factors independently associated with a worse outcome were older age at last follow-up (confidence interval [CI]: −0.66 to 0.09; \( P = 0.008 \)) and need for surgery (CI: 0.56 to 3.08; \( P = 0.004 \)). Furthermore, the need for surgery was significantly associated with older age at last follow-up (CI: −0.88 to 0.18; \( P = 0.003 \)) and the initial Diméglio grade (CI: 0.03 to 0.46; \( P = 0.02 \)).

4. Discussion

The objective of this work was to identify factors associated with the outcome of CIC as assessed by the Ghanem-Seringe score [7] and proportion of feet requiring surgery. The main original feature of
our study is that prognostic factors were evaluated separately for outcomes after treatment with the FM and with the PM. The main limitation is that the final assessment of outcome was performed before the end of growth.

4.1. Association between outcome and type of treatment

By multivariate analysis, the FM was associated with better Ghanem-Seringe scores compared to the PM. However, given the study design, this difference might reflect bias due to a centre effect. The need for surgery was not significantly different between the two treatment methods. Furthermore, maintenance physiotherapy was provided in the PM group after the initial treatment phase. This modification to the treatment method as described by Ponseti may be open to criticism. However, according to a recent study [10], the Ponseti method used was described in detail in only half the published studies, and in 17% of articles, this description indicated that a variant of the PM was applied. By multivariate analysis of our data, the type of treatment was not associated with the outcome.

An original feature of our study is the detailed description of each of the parameters used in the Ghanem-Seringe score. In keeping with data reported by Xu [11], persistent hindfoot varus was less marked with the FM method. The range of dorsiflexion was significantly smaller in the PM group. This finding may be ascribable to the more widespread use in the PM group of extensive release, a procedure found by Van Gelder et al. [12] to be associated with stiffness. In contrast, Gottschack et al. [13] reported that ankle range of motion, particularly dorsiflexion, was more often normal with the PM than the FM (82% vs. 48%). These authors predicted a change in outcomes in the near future due to the recent addition of percutaneous Achilles tenotomy to the FM. Jumping on one leg was more difficult in the PM group than in the FM group. Our results are consistent with those reported by Rampal et al. [14] (triceps deficiency in 2% of cases). The most likely explanation is that percutaneous Achilles tenotomy is usually performed in patients managed with the PM, sometimes on several occasions, and may weaken the triceps. On the other hand, in our study, talar dome appearance and talo–calcaneal range of motion were similar in the two groups. In contrast, Rampal et al. [14] found that talar dome flattening was more common with the PM. This result may be related to the addition of physiotherapy to casting. The level of sports activities was good and similar in both our groups, although other studies obtained conflicting results on this point [15,16].

4.2. Association between outcome and age at last follow-up

Outcomes were significantly worse in the patients with the longest follow-ups in our study. This finding may indicate gradual worsening of clubfoot deformities as growth progresses [17], with a particularly high risk of deterioration at puberty. Another possibility is a learning curve effect, with worse outcomes in the patients treated first. Finally, studies showing unfavourable outcomes after surgery [18] has prompted a gradual restriction in surgical indications over time, as described in a recent publication [19].

4.3. Association between outcome and initial Dimeglio grade

In both centres, the Dimeglio system was used to classify the clubfeet. Inter-observer reproducibility of this system is excellent [20,21]. The ability of the Dimeglio grade to predict the outcome of CIC is debated. In keeping with early data [22], recent publications have confirmed that initial CIC severity is associated with an increased need for surgery [23], a greater number of sequential casts [24], or a greater need for percutaneous Achilles tenotomy [25]. Despite these data, the link between initial severity and management challenges is controversial [2,13,26]. In our study, greater initial severity as indicated by a higher Dimeglio grade was associated with treatment failure by both univariate and multivariate analysis.

4.4. Association between outcome and need for surgery

The adjunctive surgical procedures performed in our patients were too diverse for an analysis of potential associations between the outcome and specific procedures. The small proportion of feet for which percutaneous Achilles tenotomy was performed in the FM group (8%) does not allow conclusions about the efficacy of this procedure added to the FM.

In contrast, our data establish that a need for surgery is associated with significantly worse outcomes [17].

4.5. Associations between outcome and other parameters

Our results are at variance with earlier studies [1,8,27] in that neither duration of splinting nor duration of rehabilitation was associated with the outcome.

As reported previously [28], whether CIC was diagnosed antenatally or at birth was not associated with the outcome.

In a recent study by Zions et al. [29], CIC severity was not significantly different between males and females. Similarly, Cosma et al. [30] and Dobbs et al. [6] reported that gender was not associated with outcome. In our study, in contrast, female gender was significantly associated with a better outcome.

CIC is traditionally believed to be more severe when bilateral rather than unilateral [29]. Nevertheless, in our study, this factor was not significantly associated with the outcome.

5. Conclusion

Our work confirms the prognostic significance of the initial Dimeglio grade. We did not obtain incontrovertible proof that one nonoperative method was superior over the other. Nevertheless, given the need for early percutaneous Achilles tenotomy when using the PM, we indicate to parents that we have a preference for the FM. Furthermore, as suggested by Richards et al. [5], Dimeglio [8], and Seringe [17], percutaneous Achilles tenotomy or casting can be incorporated into the FM to obviate the need for more extensive surgery, which is associated with poorer outcomes.

Disclosure of interest

The authors declare that they have no competing interest.

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References


