Factors associated with recurrence of clubfoot treated by the Ponseti method

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AIM
To assess several associated factors on the recurrence of clubfoot after successful correction by the Ponseti method.

METHODS
A total of 115 children with 196 clubfoot deformities, treated by the Ponseti method, were evaluated. Demographic data, family history of clubfoot in first-degree relatives, maternal educational level and brace compliance were enquired. Based on their medical files, the characteristics of the patients at the time of presentation such as age, possible associated neuromuscular disease or especial syndrome, severity of the deformity according to the Dimaggio grade and Pirani score, residual deformity after previous Ponseti method and number of casts needed for the correction were recorded.

RESULTS
There were 83 boys (72.2%) and 32 girls (27.8%) with a male to female ratio of 2.6. The mean age at the initiation of treatment was 5.4 d (range: 1 to 60 d). The average number of casts applied to achieve complete correction of all clubfoot deformities was 4.2. Follow-up range was 11 to 60 mo. In total, 39 feet had recurrence...
with a minimum Dimeglio grade of 1 or Pirani score of 0.5 at the follow-up visit. More recurrence was observed in non-idiopathic clubfoot deformities (\(P = 0.001\)), non-compliance to wear braces (\(P < 0.001\)), low educational level of mother (\(P = 0.033\)), increased number of casts (\(P < 0.001\)), and more follow-up periods (\(P < 0.001\)). No increase in the possibility of recurrence was observed when the previous unsuccessful casting was further treated using the Ponseti method (\(P = 0.091\)). Also, no significant correlation was found for variables of age (\(P = 0.763\)), Dimeglio grade (\(P = 0.875\)), and Pirani score (\(P = 0.624\)) obtaining at the beginning of the serial casting.

**CONCLUSION**

Using the Ponseti method, non-idiopathic clubfoot, non-compliance to wear braces, low educational level of mother, increased number of casts and more follow-up periods had more association to possible increase in recurrence rate after correction of clubfoot deformity.

**Key words:** Clubfoot; Recurrence; Cast; Talipes equinovarus; Ponseti

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Core tip: This is a retrospective study to determine factors responsible for the unsuccessful treatment of clubfoot using the Ponseti method. Recurrence of each or all major components of clubfoot deformity was seen in 39 feet among 196 feet during follow-up range of 11 to 60 mo. Recurrence was higher in patients with non-idiopathic clubfoot, non-compliance to wear braces, low maternal educational level, increased number of casts needed to correct the deformity and more follow-up periods.

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**INTRODUCTION**

Talipes equinovarus or clubfoot is one of the most common congenital deformities of the lower limb with an incidence of about 1 in 1000 live births[11]. This is a complex three-dimensional deformity of especially tarsal bones with agenesis or hypogenesis of foot and ankle essential structures. Although the exact cause of clubfoot has not been determined, several genetic and environmental factors have been suggested[12,13]. Clubfoot can occur either in normal infants, defined as idiopathic type, or in patients with neuromuscular diseases or different syndromes, referred to as non-idiopathic.

For decades, extensive complex surgical release procedures had been the main treatment of clubfoot without acceptable long-term outcomes[6,7] till Ponseti published the significant result of a gentle serial manipulation and casting method to correct the deformities of clubfoot including forefoot adduction, hindfoot varus, equinus, and cavus[2,8]. To date, the Ponseti method is widely accepted as the gold standard for treatment of all clubfeet, regardless of the type and etiology around the world in children of 1 to 9 years of age[6,10-11]. The efficiency of the popular Ponseti method in correcting clubfoot has been well established. More than 90% of the excellent results from different centers have been reported[12,13]. Despite the proper use of the Ponseti method, less than 5% of clubfeet do not respond to this method and need reconstructive procedures, all usually have an associated syndrome[14]. Moreover, the recurrence of clubfoot is a problem that requires more attention. Most times, early relapse results in equinus and varus deformities of the hindfoot[15]. The most reported cause of the recurrence of clubfoot after the Ponseti method is non-compliance of the family to use the abduction brace. In addition, several factors have been reported to have a significant effect on recurrence after the Ponseti method[16,17].

The purposes of this study were: (1) evaluation of mid-term results of the Ponseti method according to the Dimeglio grade and Pirani score; and (2) assessment of several factors on recurrence of clubfoot after successful correction by the Ponseti method.

**MATERIALS AND METHODS**

**Study design**

After approval of the study by Ethics Committee of the University and performance in accordance with the ethical standards lay down in the 1964 Declaration of Helsinki and its later amendments, consecutive infants with clubfoot treated with the Ponseti method from 2009 to 2013 in our centers were enrolled. Non-idiopathic cases were included but patients with residual deformity after surgery were excluded. In their last follow-up visit, demographic data including age, sex and family history of clubfoot in first-degree relatives, maternal educational level and brace compliance were enquired. High maternal education was defined as having diploma taken after high school education or higher educational certificates. Moreover, patients were evaluated for any recurrence using the Dimeglio grade and Pirani score[18,19]. Non-compliance was defined as lack of full time bracing in the first 3 mo and night time till 3 years of age, after correction of deformity by the Ponseti method. Recurrence was defined as any deformity in each of the major component of the clubfoot including equinus, heel varus, forefoot adduction and cavus or Dimeglio grade \(\geq 1\) or Pirani score \(\geq 0.5\).

Based on their medical files, the characteristics of patients such as age, possible associated neuromuscular disease or especial syndrome, severity of the deformity according to the Dimeglio grade and Pirani score,
residual deformity after previous Ponseti method, and number of casts needed for the correction were recorded.

**Statistical analysis**

Statistical analysis was done using SPSS, version 18.0 for windows (SPSS Inc. Chicago, IL, United States). Chi-squared test was used for non-parametric variables such as gender, type, brace compliance, residual deformity, maternal educational level, and positive family history. Using ANOVA test, Dimeglio grade and Pirani score at presentation, age of the patient at the initiation of treatment, and number of casts were compared between recurrence group patients and non-recurrence group subjects. The results were considered to be significant at the level $P < 0.05$.

**RESULTS**

From 2009 to 2013, 115 consecutive infants with a total of 196 clubfeet (right: 100, left: 96) had been treated with the Ponseti method. There were 83 boys (72.2%) and 32 girls (27.8%) with male to female ratio of 2.6. Thirty four subjects (29.5%) suffered from unilateral involvement. Interestingly, bilateral involvement was seen in about 70% of cases. Recurrence occurred in 26.5% of unilateral deformities and 18.5% of cases with bilateral involved feet. Analysis with $\chi^2$ test revealed no significant difference between recurrence in unilateral and bilateral cases ($P = 0.084$). One hundred cases had idiopathic clubfoot deformities while the others (13.0%) had an associated disease including 12 cases of arthrogryposis, a myelomeningocele patient, and one case with diagnosis of caudal regression syndrome. Twenty three cases (20%) were referred due to residual deformity after previous Ponseti method, and number of casts needed for the correction were recorded.

The mean age at the initiation of treatment was 5.4 d (range: 1 to 60 d). The mean Pirani score at the beginning of treatment for all subjects was 5.4 with a minimum of 1 and maximum of 6. The average number of casts applied to achieve complete correction of all clubfoot deformities was 4.2. The follow-up range was 11 to 60 mo with a mean of 30.5. Although, about 66% of mothers had a high level of education, 70.4% of families reported complete compliance with the brace and stretching exercises.

In total, 39 feet had recurrence with a minimum Pirani score of 0.5 or Dimeglio grade 1 at the follow-up visit. Tables 1 and 2 show a comparison of recurrence group and non-recurrence group cases with regards to type, residual deformity at the initial stage of treatment, family history, Brace and stretching compliance of families, and educational level of the mothers. Non-idiopathic clubfoot deformities had more recurrence with $P$ value of 0.001. There was no increase in the possibility of recurrence when previous unsuccessful casting was further treated with the Ponseti method ($P = 0.091$). Maternal educational level and compliance of brace usage were considered as important factors with regards to the reduction of the recurrence of clubfoot after the Ponseti method. Table 3 shows the mean of follow-up time in months, age of the children at the initiation of treatment, Pirani score and Dimeglio grade at the beginning of the serial casting, and number of casts needed for correction. Statistically significant difference was observed between the recurrence group and non-recurrence group with regards to follow-up duration and number of casts. It means that the longer the follow-up duration, the more the probability of recurrence. No significant correlation was found for variables of the age and Pirani scores or Dimeglio grade at the beginning of the serial casting.

**DISCUSSION**

Having a functional mobile plantigrade foot without pain is the main goal of clubfoot treatment. This goal can be acceptably achieved using the effective Ponseti method of serial casting. Although, the failure rate of the Ponseti method was reported to be about 3% to 5%[14], recurrence after correction occurred in 20% to 41% of cases[22]. It is believed that the majority of recurrences after the Ponseti method are due to the insufficient correction rather than the actual recurrence[21]. However, the possibility of late recurrence after complete correction of the clubfoot deformity by the Ponseti method was stated[22]. Recurrence became very rare 8 years after correction by the Ponseti method and this may be the only sign of non-idiopathic clubfoot[23]. Brace intolerance was determined as the leading cause of recurrence ranging from 30% to 45%[15].

### Table 1  Number of clubfeet with recurrence or non-recurrence and $P$ value of different factors

|                      | Recurrence | Non-recurrence | $n$ | $P$ value |
|----------------------|------------|----------------|-----|-----------|
| Idiopathic           | 28 (16.3%) | 144 (83.7%)    | 172 | 0.001     |
| Non-idiopathic       | 11 (45.8%) | 13 (54.2%)     | 24  |           |
| Residual deformity at the initiation of treatment | 12 (29.3%) | 29 (70.7%) | 41  | 0.091     |
| Non-residual deformity at the initiation of treatment | 27 (17.4%) | 128 (82.6%) | 155 |           |
| Positive family history | 9 (19.6%) | 37 (80.4%) | 46  | 0.094     |
| Negative family history | 30 (20.0%) | 120 (80.0%) | 150 |           |

### Table 2  Numbers of cases with recurrence or non-recurrence and $P$ value of different factors

|                      | Recurrence | Non-recurrence | $n$ | $P$ value |
|----------------------|------------|----------------|-----|-----------|
| Brace compliance     | 6 (7.5%)   | 74 (92.5%)     | 80  | < 0.001   |
| Brace noncompliance  | 18 (51.4%) | 17 (48.6%)     | 35  |           |
| High educational level of mothers | 11 (14.1%) | 67 (85.9%) | 78  | 0.033     |
| Low educational level of mothers | 13 (35.1%) | 24 (64.9%) | 37  |           |
Dobbs et al\(^{[16]}\) reported a 183-fold increased risk of recurrence due to poor compliance of parents to use braces after correction by the Ponseti method. Although, the direct effect of brace wearing and stretching exercise after correction to reduce recurrence was approved by the presented study \((P < 0.001)\) and others \(^{[2,20,23,24]}\), Gelfer et al\(^{[23]}\) did not find a significant relationship between the rate of recurrence and the compliance of brace wearing. In our study, 7.5% of cases with good compliance to wearing of brace had recurrence deformities; hence, the possibility of recurrence after successful brace wear should be considered.

Parental status with regards to their educational level, income level and insurance were explained to be important factors in recurrence of clubfoot after the Ponseti method \(^{[16]}\). Of the cases in our study, there was a statistically significant difference in the recurrence of clubfoot with regards to maternal educational level \((P = 0.033)\), but some controversies were observed on these variables. Although, unmarried parents with no insurance support, low parental education and low income were reported as significant risk factors for recurrence \(^{[17]}\). Dobbs et al\(^{[16]}\) reported no significant relationship for recurrence after the Ponseti method between parental marital status, income, and their medical insurance.

According to our results, recurrence was observed in 16.3% of cases with idiopathic clubfoot and 45.8% of non-idiopathic clubfoot subjects \((P = 0.001)\). These results are in line with the report of Gelfer et al\(^{[23]}\) and contrary to the study by Funk et al\(^{[11]}\) which showed no significant difference in recurrence rate between idiopathic and non-idiopathic congenital clubfoot treated using the Ponseti method. In that study, treatment of non-idiopathic cases started later and took longer than idiopathic clubfoot patients.

The severity of deformity at the beginning of serial casting according to the Dimeglio grade \((P = 0.875)\), age of the patients at the initiation of treatment \((P = 0.763)\) and any residual deformity due to unsuccessful previous serial casting \((P = 0.091)\) did not have a major effect on the possibility of recurrence. These variables were also declared by others as insignificant risk factors for clubfoot recurrence \(^{[16,23]}\).

The strengths of the present study are adequate number of cases, comparing idiopathic and non-idiopathic subjects, comparing unilateral and bilateral cases, and acceptable average follow-up period.

Finally, in order to prevent the recurrence of clubfoot after the Ponseti method, we insisted on complete correction of the deformity using castings as much as possible. Moreover, asking and teaching the mothers about the usage of brace wear after correction of the deformity could lead to low recurrence rate.

In conclusion, non-idiopathic clubfoot deformities, non-compliance to brace wear after correction of deformities, low educational level of mothers, increased number of casts necessary to achieve complete correction, and more follow-up periods had more association with possible increase in recurrence rate after correction of clubfoot deformity using the Ponseti method.

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**COMMENTS**

**Background**

Talipes equinovarus or clubfoot is a complex three-dimensional deformity of especially tarsal bones with agenesis or hypogenesis of foot and ankle essential structures. The exact cause is undefined. Clubfoot can occur either in normal infants, defined as idiopathic type, or in patients with neuromuscular diseases or different syndromes, referred to as non-idiopathic. Gentle serial manipulation and casting method of Ponseti is accepted as the gold standard for treatment of all clubfeet, regardless of the type and etiology around the world, in children of 1 to 9 years of age. In this study, the factors that resulted in recurrence of the clubfoot deformity after achieving correction of the deformity using the Ponseti method was evaluated.

**Research frontiers**

In recent years, using the Ponseti method to correct clubfoot deformity worldwide was with excellent clinical results. Determining the factors that led to the recurrence of the components of this deformity could help in the improved management of clubfoot deformity.

**Innovations and breakthroughs**

The present study was carried out to determine factors related to the recurrence of clubfoot deformity after the Ponseti method correction. Non-idiopathic clubfoot, non-compliance to wear braces, low educational level, increased number of casts to achieve perfect correction and more follow-up periods should be considered as important factors with regards to reducing the recurrence of clubfoot deformity after the Ponseti method.

**Applications**

The data in this study recommended being careful with the treatment of clubfoot in children with non-idiopathic types, low compliance to wear especial braces after correction, maternal low educational level and high cast frequency during serial casting. In addition, it is important to follow the patients for a longer period of time.

**Terminology**

Clubfoot is a congenital anomaly including forefoot adduction, hindfoot varus, equinus, and cavus. Ponseti method is a gentle serial manipulation and casting.
method used to correct the deformities of clubfoot.

**Peer-review**
The paper is well conducted. It’s an interesting paper and suitable for publication.

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