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Factors predictive of Ponseti casting for treating clubfoot: analysis of Bayesian Poisson regression model

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Abstract. – OBJECTIVE: Clubfoot is a growing public health concern in Bangladesh, with the incidence of approximately 0.64 to 6.8 in every 1000 live births. For over a decade, Ponseti method has been considered a gold standard for treating the clubfoot. Despite few studies have been estimating the number of casts required to correct the clubfoot deformities by Ponseti method, the subject of interest has always remained. Therefore, this current study aimed to investigate the significant predictive factors for the number of casts required to correct congenital clubfoot.

PATIENTS AND METHODS: In this retrospective cohort study, we used Bayesian Poisson Regression Model to investigate the influencing factors that could predict how many casts are needed to correct the clubfoot. We included 69 patients with 99 affected feet, who completed their corrective phase of treatment in the Ponseti method. For this cohort study, we integrated only pre-tenotomy casting data with no age restrictions. We used Bayesian Poisson regression analysis technique to estimate the predictive factors.

RESULTS: In Bayesian Poisson model, age was the most influencing predictive factor (24.3%) for increasing the number of castings to correct the clubfoot deformity. The clubfoot offspring of the ≤1-year-old was positive, and the incidence rate increased significantly with the casting number. The number of Ponseti casts in male clubfoot children was 28% higher than in female, and this was marginally statistically significant. There was no marked change estimated in the pattern of clubfoot, foot involvement and Pirani score of the severity.

CONCLUSIONS: We concluded that the age factor may influence the number of casts required for the correction of clubfoot and specifically ≤ 1-year-old children are highly impacted. Treating clubfoot at an early age is suggested in this study to increase the success of clubfoot treatment and decrease the risk of relapse.

Key Words:
Casting congenital clubfoot, Ponseti method, Pirani score, Bayesian Poisson regression analysis.

Introduction

Clubfoot is a common congenital foot deformity, occurring in 0.64 to 6.8 cases per 1,000 live births[1]. It causes three-dimensional foot deformities and impairment when clubfoot treatment is neglected[2]. The Ponseti method (PM) has become the gold standard for correcting the clubfoot[3]. Achieving normal gait functions, a painless plantigrade foot, and regular shoe wear are common goals among the clubfoot practitioners. The typical treatment method for clubfoot has been a surgical release, which has several long-term complications such as recurrence deformity, joint stiffness, and deformity or discomfort in the knee and/or thigh[4,5]. The Pirani score (PS) is extensively used in the clinical settings to assess the severities of clubfoot deformity. In the current clubfoot management practice, the number of casts needed to repair clubfoot is determined by the Pirani score at the outset[6-8].

With less than 5% of patients requiring substantial surgical intervention, percutaneous Achilles Tenotomy, this is commonly used noninvasive tech-

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nique provides great long-term outcomes[9,10]. Errors in casting procedures, missing the earlier treatment, the severity of the deformity, bracing noncompliance, lack of parental information, and cultural factors may contribute to the failure of conservative treatment[11-14]. The rate of percutaneous Achilles Tenotomy (pAT) is reduced with appropriate casting and progressive manipulation[15]. Manipulation and casting should therefore begin as early as possible to reduce the recurrence rate and better outcome[16-18].

Although several studies have been published to examine the factors causing the requirement higher number of casts to correct the clubfoot deformity, to the best of our knowledge, there is no study on the Bangladeshi population to determine which factors contribute the castings numbers in the low-cost setting areas. There are many demographics and clinical outcome measurement factors (age, sex, premature birth, clubfoot types and severity measured by Pirani scoring system over long-term follow-up treatment period) that can be considered as influencing factors to increase casts to achieve full correction of the clubfoot. Therefore, this current study aimed to investigate the significant predictive factors for the number of casts required to correct congenital clubfoot.

Patients and Methods

Ethical Approval

All procedures performed in the present study were in accordance with the Ethical standards of the Research Committee of the North South University (reference number: NSU/SHLS/DPH/2014-08). Prior to starting the Ponseti treatment, a written informed consent was collected from the parents of all participants.

Study Design

This research study was conducted in a retrospective cohort study design method. Given the generalization nature of the study, a systemic sampling strategy of a probability sampling approach was adopted to acquire information about individuals from the medical record. The medical record of infant with clubfoot was collected (2015-2017) from the LMRF Healthcare in a Chottogram division located in the southeastern large metropolitan city in Bangladesh.

Participants

A total of 100 participants were recruited based on the complete casting record. The inclusion criteria of this study were: children with idiopathic congenital clubfoot, bilateral and unilateral clubfoot, both genders. Participants were excluded for the analysis based on the following exclusion criteria: non-adherent with casting treatment, clubfoot with associated conditions (neuromuscular illnesses or syndromes, orthopedic conditions in the knee and hip), insufficient clinic records about the number of casts. Of these 100 participants, 9 participants were excluded from this study due to insufficient clinical casting records, that is, how many Ponseti casts were required ahead of Achilles tenotomy to correct the clubfoot. Also, 22 patients were omitted due to noncompliance with the treatment schedule. If the child had any form of CC (Idiopathic *vs*. other, 84.1% *vs*. 15.9%) condition, regardless of gender (male *vs*. female, 71% *vs*. 29%), age (mean±SD 1.68±0.915 years) with single or both foot (right, left and both; 33.3%, 23.2% and 43.5%, respectively) involvement as determined by the veteran physiotherapist, participants were enrolled (Table I).

Ponseti Method and Corrective Phase

At the time of corrective phase of Ponseti method, clubfoot severity was assessed and documented by using the Pirani scoring system. The mean score of Pirani Scoring system was higher 2.45 before percutaneous Achilles Tenotomy (pAT) than the right (2.08) and left (2.05) feet, and the child needed an average of 5.09 castings (Table I). The patients were assessed by the qualified physiotherapist and tenotomy performed by the pediatric orthopedic surgeon. Ponseti cast treatment was initiated at the time of the second clinic appointment and it was applied by the skilled physiotherapist trained in the Ponseti technique. The total number of casts was calculated from the first cast to before the percutaneous Achilles Tenotomy. Many clubfeet patients received the maximum of 10 casts before tenotomy. The dynamic foot abduction orthosis was applied 3 weeks after the pAT. For patients who did not require an Achilles tenotomy, the brace was applied once the foot was fully corrected by casting and manipulation[19].

Statistical Analysis

Categorical data were expressed as numbers and percentages, and continuous data were presented as mean and standard deviation. Markov chain Monte Carlo (MCMC) is a simulation technique used to fit a model and draw samples

Table I. Baseline characteristics as per congenital clubfoot offspring at the end of follow-up.

Characteristics		N (%)	Mean ± SD
Gender	Male	49 (71)	
	Female	20 (29)	
Age (years)			1.68 ± 0.91
Premature baby	No	65 (92.8)	
	Yes	4 (5.8)	
Idiopathic Clubfoot	No	16 (15.9)	
	Yes	77 (84.1)	
Foot involvement	Right	23 (33.3)	
	Left	16 (23.2)	
	Both	30 (43.5)	
PS right foot			2.08 ± 0.77
PS left foot			2.05 ± 0.89
PS before pAT			2.450 ± 1.02
Number of castings			5.09 ± 1.60

*SD = Standard Deviation, pAT = percutaneous Achilles Tenotomy.

from the joint posterior distribution of the model parameters[20]. MCMC tests have represented the autocorrelations and trace plots in Figure 1 (A and B). We modeled the foot using data from the record including casting number before tenotomy, age, sex, early birth history, clubfoot, and severity score based on Pirani scoring system. The Bayesian Poisson regression analysis technique was used to compute the mean and standard deviation. Also, the incidence risk ratio (IRR) with a 95% credible interval (CI) was calculated for the central portion of the posterior distribution. Data in STATA were analyzed (16, StataCorp LLC, College Station, TX, USA).

Results

The results of this study showed that the marginal posterior distribution is positively associated with the mean age of sample size (0.212), and it indicates that the higher probability (95% CI: -0.004-0.420) of cast numbers increase with age. On the other hand, the mean of the preterm infant (-0.31) and atypical type of congenital clubfoot (-0.25) showed that both were associated negatively, showing decreasing values over time of the true mature infant and atypical type of congenital clubfoot ((95% CI: -1.105866 – 0.4384544, -0.7078508 – 0.1788736).

The posterior distribution coefficients are associated with the following baseline covariates: gender (male) (0.22), right foot involvement (0.56), Pirani score of the right foot (0.11) and left foot (0.23). These results indicate that increase of risk of required higher number of casting when the child with congenital clubfoot have one or all of these risk factors (Figure 2).

Furthermore, this study found that the requirement of casts in male children was 28% higher than female children and this excess risk attains the marginal statistical significance (95% CI: 0.859-1.859). The results of credible interval analysis for the age showed that a significant increase in the casting number (IRR 1.24; 95% CI 0.995-1.52). Also, this study found that preterm babies require a higher number of casting (78%) than full-term babies in the follow-up period. Although the required number of castings are 79% greater in idiopathic clubfoot children than those with non- idiopathic congenital clubfoot, however, this increased risk ratio (IRR) did not meet statistically significant results (95% CI: 0.492-1.195). Interestingly this study also found that children with right side clubfoot had a 9% higher chance of needing more casting to correct fully than left side and bilateral clubfeet. The frequency of anticipated casting tends to vary by 1.13 if the Pirani score of the right foot changes by one unit. Likewise, when the left Pirani score of the foot increased by one unit, the estimated volume of casting also increased by 1.03. To find the strength of the association between the right foot involvement, Pirani score of the right and left foot with the casting number was assessed through their posterior distribution analysis and the results of the posterior distribution showed as $E(\alpha \mid D) = 2.82$ and the 95% credible intervals for right foot was 0.807-1.537 and left foot was 0.7697643-1.383 (Table II).

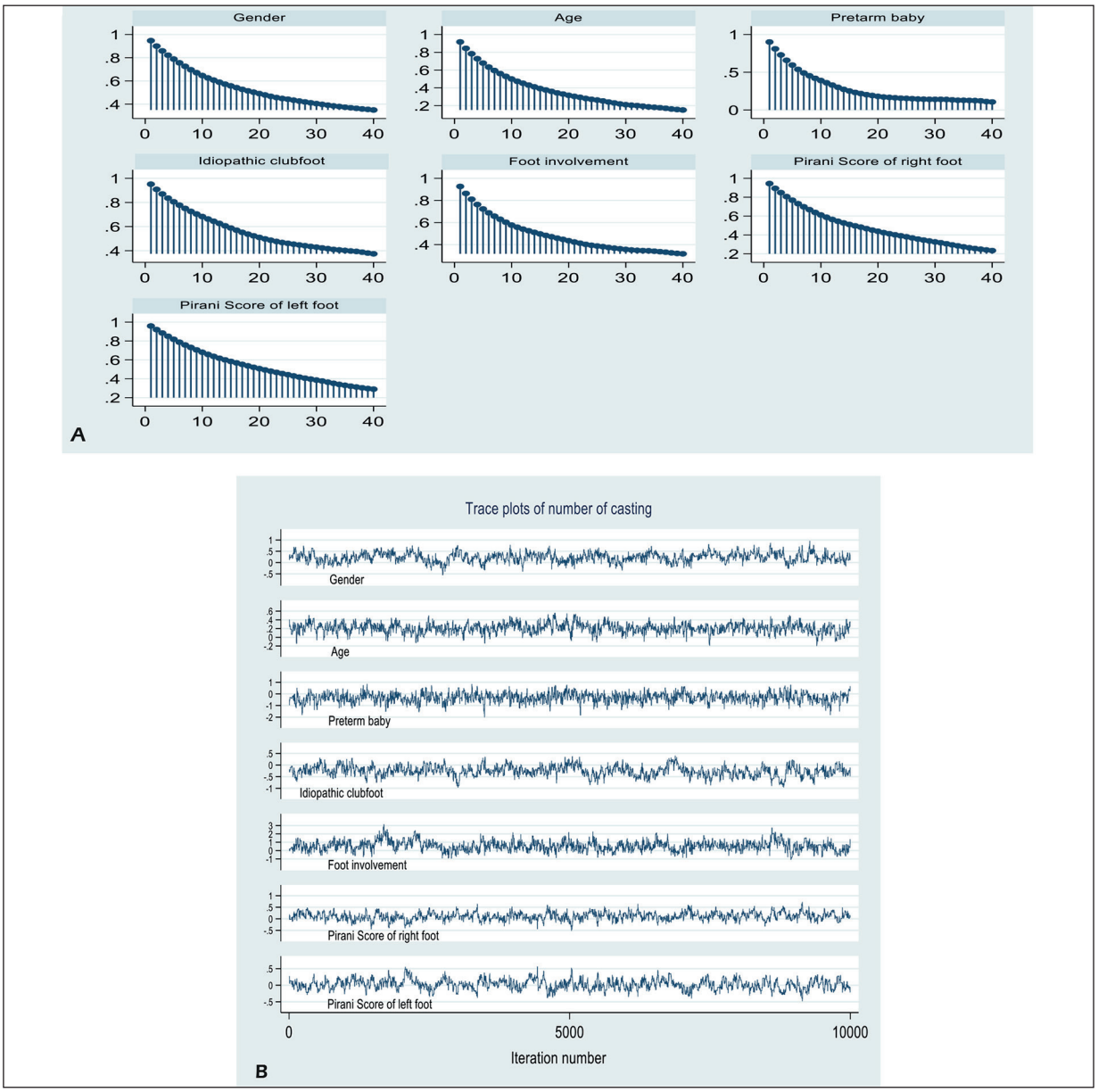

Figure 1. A, Degree of correlation between the Ponseti cast and observed variable. **B,** Trace plots predicted values by Ponseti cast from each iteration.

Furthermore, our study analyzed the influence of age of the sample size on casting number (≤1 year=33. *vs.* >1 year=36) With the casting number, the posterior distribution had the positive association (0.223; 95% CI: 0.01-0.431) with children under the age of one year, indicating that children with over one year of age needed higher number of castings. The credible interval for the incidence rate ratio of age suggested a strong significant increase with the casting number (IRR 1.25; 95% CI 1.011-1.555) (Table III).

Discussion

This study found that the requirement of casting was very less among children with clubfoot under the age of one. Many studies investigated the application of Ponseti method for children with congenital clubfoot at their earliest age for having a better result. The Ponseti treatment should begin as soon as possible, like within a week of life. Therefore, we can take advantage of the favorable fibroelastic properties of the con-

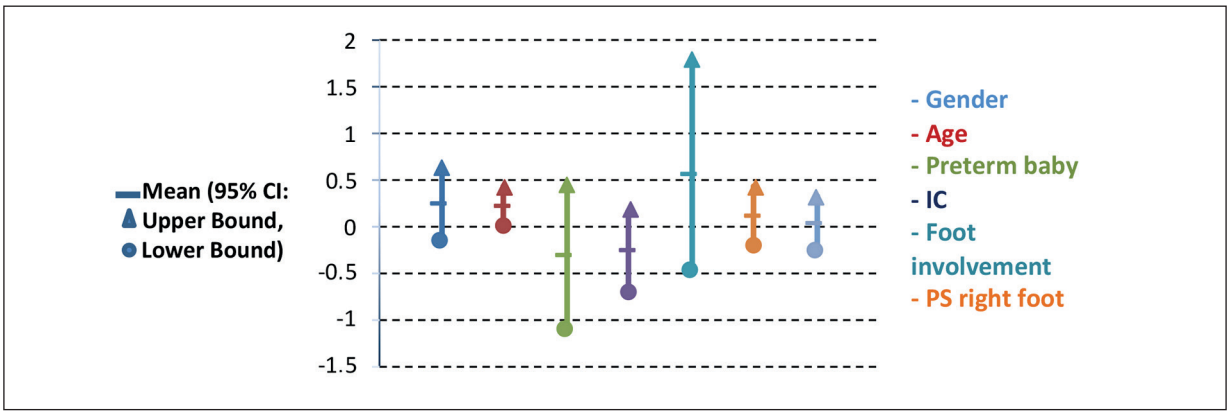

Figure 2. Variable's credible intervals for normal mean of the congenital clubfoot offspring by casting number.

nective tissue which forms the ligaments, joint capsules, and tendons. In contrast to the casting treatment, early surgical interventions for clubfoot correction can create various complications such as fibrosis, scarring and stiffness[21-23]. According to a recent study, Ponseti casting should begin at 1-2 months of age to get better clubfoot results than other younger age groups[24]. Also, adopting precise manipulation techniques followed by the insertion of well-molded plaster casts of Ponseti method is the best and safest way to treat all types of clubfeet deformities in infants[25]. Improper casting methods can cause various complications such as pain, pressure related mild infection, swelling of the forefoot and toes, erythema, midfoot-hyper abduction, pseudoaneurysm, slippage of casting, and rocker-bottom deformity[23,26].

However, the common questions among parents after first casting was that how many casts are required more to correct the feet, and the cost of the treatment. This is especially important in underdeveloped nations, where the length of treatment has a significant impact on parental compliance and treatment outcome. In the literature, there were conflicting reports on the use of age and its predictive usefulness for the number of casts required for correction[27].

Although our study found a strong link between age and casting number, that differs from the work of Agarwal and Gupta[27] that reported a weak but positive correlation. This may be due to a wider number of age variations among the groups. A positive age average in this test indicates that actual age values are increasing over time, which is more likely to be in the age range indicated by the increasing number of castings. The average age of our participants was 1.68 years, with a casting average of 5.20. The association between Pirani score and casting was not confirmed by the results. According to a study conducted on an Indian population, the number of casts required to repair idiopathic clubfoot was influenced by both initial Pirani score and age. However, that study reported that there was no linear link

Table II. Incidence rate ratio of required casting number among CC children.

Casting	IRR	Std. Dev.	MCSE	Median	95% Credible Interval
Gender	1.283	0.256	0.024	1.263	0.859-1.859
Age	1.243	0.131	0.009	1.241	0.995-1.523
Preterm baby	0.782	0.297	0.017	0.733	0.330-1.55
IC	0.794	0.171	0.0168	0.776	0.492-1.195
Foot involvement	2.096	1.537	0.154	1.695	0.615 -6.031
PS right foot	1.132	0.186	0.013	1.112	0.807-1.537
PS left foot	1.036	0.158	0.012	1.024	0.769-1.383
Constant	2.822	7.892	0.461	0.719	0.014-20.487

*IRR = Incidence Rate Ratio, MCSE = Monte Carlo Standard Error, Std. Dev. = Standard deviation, IC = Idiopathic clubfoot.

Table III. Posterior distribution of categorical age and Ponseti casts among CC children.

Casting	Mean (95% CI)	Std. Dev. (Mean, IRR)	MCSE (Mean, IRR)	Median (Mean, IRR)	IRR (95% CI)
Age (≤ 1 year *vs.* > 1 year)	0.223 (0.014-0.431)	0.107 0.136	0.002 0.004	0.221 1.243	1.257 (1.011-1.555)
Constant	1.277 (0.917-1.608)	0.177 0.637	0.004 0.019	1.284 3.617	3.655 (2.537-5.016)

*CI = Credible Interval, IRR = Incidence Rate Ratio, MCSE = Monte Carlo Standard Error, Std. Dev.= Standard deviation.

between initial Pirani score and the number of corrective casts[27].

The Ponseti procedure was found to require fewer casts, a shorter time to achieve the correction, and a lower chance of relapses than other methods in a study of children under the age of two[11]. The average initial presentation of casting would influence the treatment's outcome[28]. In our investigation, the incidence rate of more than one-year-old CC offspring increased significantly with the casting number. Results indicated that the number of casts in young infants was lower than in older children (one to two years, six to ten casts; two to three years, nine twelve casts). When it came to older children, the casts were on the rise. The average age of the children at the start of treatment was 10.7±6.28 weeks (1–23 weeks), with an average of 4.88 ±0.88 casts required for full correction[29]. A similar result was reflected by Khan and Kumar[30], where the average number of casts was 12.1 in children aged 8.9 years.

Strength and Limitations

This study had several strengths and limitations. A long-term and thorough follow-up was available to ensure that the initial correction was obtained. All the subjects in this study had achieved the initial correction of clubfoot by utilizing Ponseti method[19]. The main limitation of this study is that it's a single center study and data were collected from only one center in Bangladesh. Another limitation of this study was the small sample size.

Conclusions

Age factor is still a significant predictor of Ponseti casting and is critical to the Ponseti method's success. The number of casts in clubfoot is influenced by age and specifically ≤ 1-year-old child is highly impacted. To increase the chances of success and decrease the risk of relapse, parents and guardians should be encouraged to start treatment at an early age. A further study is required to indicate the exact number of casts as per child's age from 0 days to 12 months.

Conflict of Interest

The Authors declare that they have no conflict of interests.

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