



## Association of maternal age and order of pregnancy with prevalence and severity of clubfoot- A single-center observational study

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### Abstract

**Background:** Congenital talipes Equinovarus (CTEV) is one of the most common congenital anomalies of the lower extremity with an incidence rate of 1-2 per 1000 live births. Diagnosis is mostly by history and clinical evaluation. The Ponseti method of clubfoot management is accepted worldwide as the most ideal treatment for CTEV. Findings regarding maternal age and parity of the mother have been inconsistent and relate differing associations with idiopathic CTEV.

**Objectives:** To evaluate the factors like maternal age and order of pregnancy and their association with the prevalence and severity of CTEV in a geographically defined population

**Methods:** This was an observational descriptive cross-sectional study done among 173 children with CTEV deformity. The subjects were selected as per inclusion criteria. Data collected were analyzed using SPSS.

**Results:** The prevalence of CTEV was predominant in male children (67.1%). CTEV child born from mothers in the age group of 18-25 years constitutes 63% of all. The majority of CTEV babies (60%) were born in the first birth order of mother. No association between birth order and the association between isolated/multiple congenital defects with CTEV has been determined in our study setting ( $p=0.30$ )

**Conclusion:** Early maternal age-related to the high prevalence of children born with clubfoot deformity. The severity of clubfoot was assessed with the Pirani score for both feet when compared with the birth order of the child, the results were statistically not significant.

**Keywords:** Congenital Talipes Equinovarus, Clubfoot, Pirani score, Ponseti method.

### Introduction

Congenital talipes equinovarus (CTEV), otherwise called clubfoot is one of the most common congenital anomalies of the lower extremity with an incidence rate of 1-2 per 1000 live births<sup>(1)</sup>. It may occur in isolation as an idiopathic type or as a part of a syndrome with associated other

malformations. The classical deformity comprises the plantar flexed ankle (equinus), an inverted (varus) heel, and inverted and adducted (varus) mid foot and forefoot<sup>(2)</sup>. (Figure 1) It has been reported that about half the cases of talipes equinovarus have bilateral involvement and that the right leg is more likely to be affected than the

left in unilateral cases<sup>(3)</sup>. Being a clinically diagnosed condition, normally identified and managed as early as possible. The Ponseti method of clubfoot management is accepted worldwide as the most ideal treatment that includes serial manipulation and plaster casting, which starts as early as 1<sup>st</sup> week of birth<sup>(4)</sup>. Although the incidence of clubfoot surgery is reduced considerably, still some children undergo surgical treatment and disability can occur despite such treatment<sup>(5)</sup>.

Although the exact etiology of CTEV in most instances is not known, there is aetiologic heterogeneity for idiopathic CTEV and a combination of genetic, maternal, and environmental factors are hypothesized<sup>(6)</sup>. However, the reported findings of many of these studies are inconclusive. Historically increases in intrauterine pressure with fetal malposition were the mechanical forces thought to be the cause of this foot anomaly. The research on embryological development and genetic analysis has countered this hypothesis<sup>(7)</sup>. Findings regarding maternal age and parity of the mother have been inconsistent and project differing associations with idiopathic clubfoot<sup>(8)</sup>. A multistate study of the epidemiology of clubfoot in the USA showed the highest incidence (38%) among primigravida. The common age group of mothers having clubfoot children was between 23-34 years(57.6%)<sup>(9)</sup>. The kinds of literature available on aetiological factors associated with CTEV show conflicting findings. The objective of the current study was to evaluate the factors like maternal age and order of pregnancy and their association with the prevalence of CTEV in a geographically defined population of Odisha. The secondary aim was to find out their association with the severity of foot deformity.

### Material and Methods

The present study is an observational descriptive cross-sectional study conducted at a National rehabilitation institute, in Odisha, India within a period of one year from September 2020 to

January 2022. The study was performed in agreement with the Declaration of Helsinki and informed consent was taken from the parents. All the children with CTEV deformity who came for treatment at the outpatient department between the age group of 0-10 years of age were included in the study population. The inclusion criteria for recruitment to the study include Children of both sexes with idiopathic CTEV or CTEV with other anomalies, and children whose parents gave informed consent for the study procedure. Elderly neglected clubfoot and children with improper & inconsistent history were excluded from the study. Independent variables such as the age of the mother, order of birth of the CTEV child, and other associated factors like smoking and alcohol intake history of the mother were recorded. The severity of the foot deformity was assessed by using Pirani score and the number of casts required for correction.

Data analysis: SPSS version 23.0. and Microsoft Excel has been used for statistical analysis. The proportion of subjects with different clinical profile was presented as frequency and percentages along with Clopper–Pearson 95% CI. Mean with standard deviation, and median with interquartile range was calculated to describe continuous data. Proportions were calculated to present the categorical data (age groups, sex, dichotomous outcome, etc.). Appropriate tests of significance have been used to determine the statistical significance of differences in proportions or means. A p-value of less than 0.05 has been considered statistically significant.

### Results

The mother or the legal guardian of the child with CTEV with or without other congenital anomalies was interviewed and the data collected were analyzed. A total of 173 cases were included in the study, out of which 116(67.1%) were male and 57(32.9%) were female clubfoot children. The mean age of mothers was 24.34years with a standard deviation of 10.2 years.[Table 1]. CTEV child born from mothers in the age group of 18-25

years constitute 63% (109 cases) and age group of 26 to 32 years and >33 years age group were 34.10%(59 cases) and 2.89%(5cases) respectively. With reference to the side of involvement, bilateral affection in 100 (57.9%) cases, the left side in 21(12.1%), and the right side affected in 52(30%) children. The average number of cast applications done was 5 ranging from 3-14 cast applications. The majority of CTEV babies were born in the first order i.e., 60% followed by 2nd order birth at 34%, 3rd order at 4.6%, and 4th order at 1.3%. Previous history of congenital anomalies either in the family or a previous child affected with congenital anomalies was seen in 8(4.6%) cases.[Table 2] In our study, no history of smoking was found associated with mothers of

CTEV babies. Only one mother has a history of drug intake during the first trimester. The prevalence of CTEV associated with other congenital defects in our study was found to be 18(10.4%). No association between birth order and the association between isolated/multiple congenital defects with CTEV has been determined in our study setting (p=0.30)[Table 3] Comparing the mean Pirani score in left and right foot in isolated CTEV and CTEV with other congenital anomalies, the difference was not statistically significant when compared in different birth order groups among the idiopathic CTEV group as well as in CTEV with other congenital anomalies.[Table 4, Table 5, Figure 2]

**Table 1:** Demographic profile of study group

Biosocial characteristics	Number (n=173)	Percentage (%)
<b>Gender</b>		
Female	57	32.9%
Male	116	67.1%
Mean age of mothers	24.34 with an SD of 10.2 in years	

**Table 2:** Obstetrics history of mothers of CTEV children

Obstetrics parameters	Number (n=173)	Percentage (%)
<b>Birth Order</b>		
1st order	104	60%
2nd order	59	34.1%
3rd order	8	4.6%
4th order	2	1.3%
<b>Family H/o of congenital anomalies</b>		
Present	8	4.6%
Absent	165	95.4%

**Table 3:** Association between birth order and idiopathic CTEV and associated other congenital defects

Birth order	Isolated CTEV (n=155)	CTEV with another congenital defect (n=18)	Chi-square Test, p-value
First	92	11	3.61 0.30
second	54	5	
Third	8	1	
Fourth	1	1	

**Table 4:** Comparing Mean Pirani Score in of left and right foot in Isolated CTEV patients

Birth order	Mean Right-sided Pirani Score	Mean Left-sided Pirani Score	P-value <sup>#</sup>
1st	5.54	5.73	0.08
2nd	5.47	5.37	0.58
3rd	5.88	5.92	0.73
4th	5.5	5.5	1

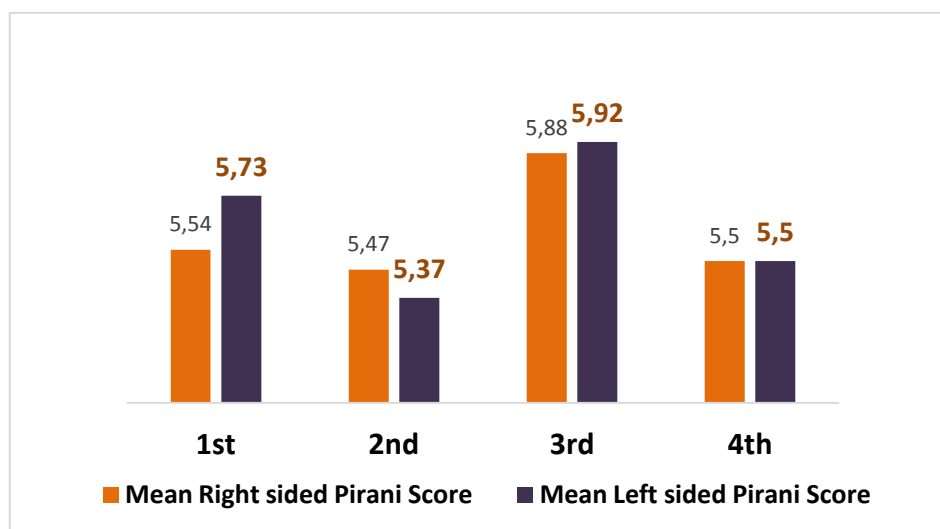
# Mann Whitney U test

**Table 5:** Comparing Mean Pirani Score in of left and right foot in CTEV with other congenital anomalies.

Birth order	Mean Right-sided Pirani Score	Mean Left-sided Pirani Score	P-value <sup>#</sup>
1st	5.92	5.94	0.91
2nd	5.9	6	0.98
3rd	6	6	1
4th	5.5	NA	---



**Figure 1:** Congenital talipes equinovarus (CTEV) foot deformity



**Figure 2:** Comparison of Mean Pirani Score with Birth orders in idiopathic CTEV group

**Discussion**

In the current study, out of 173 cases of CTEV, 18 patients were found to have been associated with other congenital defects. The prevalence of CTEV associated with other congenital defects in our study came to be 10.4%. The study by Wang H et al showed that the majority of cases were isolated congenital clubfoot (82%) and 11% had

associated major congenital anomalies which are similar to our findings<sup>(10)</sup>. Our study includes 57 (32.9%) female and 116 (67.1%) male CTEV children. The proportion of cases with bilateral CTEV was similar to those previously reported. Our study result supported the male sex as a strong risk factor for clubfoot. There was no significant association between different gender

with Isolated CTEV and associated other congenital defects with CTEV. The study conducted by Stoll C et al showed that the male sex (OR=1.68; 95% CI, 1.48-1.94) has a significant association with clubfoot<sup>(11)</sup>. Epidemiological studies have consistently found a higher prevalence of idiopathic clubfoot in males and first-born children<sup>(8)</sup>. The majority of CTEV babies in our study were born in first order i.e., 60% followed by 2nd order birth at 34%, 3rd order at 4.6%, and 4th order at 1.3%. The results support the old hypothesis that the mother of first-order pregnancy has a poorly developed uterus with an expected increase in intrauterine pressure. The ischemia develops from abnormal pressure and affects the malposed fetus resulting in CTEV deformity. However, the hypothesis does not stand true as the fetal position is not constant in the mother's uterus.

Higher parity and higher gravidity were associated with decreased odds of CTEV reported by Chen C et al.<sup>(12)</sup>. The study by Parker S E et al observed that parity was moderately associated with clubfoot, with multiparous mothers having a decreased risk child with CTEV deformity. Young maternal age (<23 years) was weakly associated with an increased risk compared to older maternal age (OR: 1.14, 95% CI: 1.08, 1.21)<sup>(9)</sup>. Liu YB et al. reported that maternal age at conception between 20-24 years of age appears to be an increased risk of having a clubfoot child<sup>(13)</sup>. A similar result was observed in the current study where 63% of cases of a mother having a clubfoot baby were within the 18 to 25 years age group. A reverse result was observed by Hollier et al. reporting a higher incidence of anomalies in advanced maternal age beyond 25 years<sup>(14)</sup>. However, Pavone V et al. did not find any correlation between the prevalence of CTEV with respect to maternal age<sup>(15)</sup>.

The difference in Pirani score of the right and left foot was not significant when compared in different birth order groups among the CTEV children. Birth order does not affect Pirani's score in our study setting. The difference in the number

of casts applied for treating the CTEV condition was highly significant among the CTEV with other anomaly groups (median = 12) when compared to idiopathic CTEV (median = 5) in our study

### Conclusion

Although the published literature shows inconsistent results, our single-center observational study reveals early maternal age related to the high prevalence of children born with clubfoot deformity. The severity of clubfoot was assessed with the Pirani score for both feet, when compared with the birth order of the child, the results were statistically not significant.

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