

DOES LATERALITY OF DEFORMITY INFLUENCE THE SEVERITY OF THE IDIOPATHIC CLUBFOOT?

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ABSTRACT

BACKGROUND: The idiopathic clubfoot has been widely reported as the commonest congenital deformity of the lower limbs with incidence of approximately 1-2 in 1000 live births. Its exact aetiology is not known. The deformity is bilateral in more than half of the cases. This study was designed to compare the severity of bilateral and unilateral idiopathic clubfoot, and to determine if there is any correlation between the left and right feet of bilateral cases.

METHODS: Records of all patients with idiopathic clubfoot who presented at our clubfoot clinic between January 2013 and November 2018 were reviewed. Patients were categorized into two groups, the 'Bilateral Idiopathic Clubfoot' and 'Unilateral Idiopathic Clubfoot' groups. The degree of deformity of each affected foot was categorized based on the Pirani score at presentation as 'Less Severe' for scores less than or equal to 4.0. Statistical analysis and tests for associations and correlation between variables were done using IBM SPSS version 22.0.

RESULT:

Records of 185 idiopathic clubfoot cases belonging to 120 (65 bilateral, 55 unilateral) patients who met the inclusion criteria were studied. The age range of the patients at presentation was 3 days to 24 months. The degree of deformity was proportionately more severe in the 'Bilateral Idiopathic Clubfoot' group compared to the 'Unilateral Idiopathic Clubfoot' group ($p < 0.0001$). The left and right feet of the bilateral cases were highly correlated ($r = 0.861$, $p < 0.0001$).

CONCLUSION:

Bilateral idiopathic clubfoot cases were more severely deformed than unilateral idiopathic clubfoot cases. Left and right feet of bilateral cases were highly correlated.

KEYWORDS: Severity, bilateral, idiopathic, clubfoot, correlation.

NigerJmed2019: 148 - 151

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INTRODUCTION

The idiopathic clubfoot has been widely reported as the commonest congenital deformity of the lower limbs with incidence of approximately 1-2 in 1000 live births.¹⁻⁶ However, the incidence varies across ethnicities, countries and races, and is reported to be as high as 3-3.4 per thousand births in Nigeria.⁵ The deformity is bilateral in more than half of the cases, and is commoner in males.^{1, 2, 5, 6, 8} The exact aetiology of this condition is not known. However, several theories supporting different aetiologies have been proposed.^{2, 9}

Evidences suggest that different subgroups of idiopathic clubfoot exist with differences in severity and treatment outcomes.^{9, 10} In cohort

studies of idiopathic clubfoot, analysis of correlated feet of bilateral cases as individual feet could lead to artificial inflation of the sample size, resulting in false significant findings.^{11, 12} A few measures to avoid artificial inflation of data and the resultant false significant findings in clubfoot studies have been described.

Richards et al¹³ analyzed their data in 2 phases. In the first phase, a single foot was randomly selected from each of the bilateral cases and analyzed together with the affected foot of all the unilateral cases, while the second phase comprised of analysis of both feet of all bilateral cases together with the affected foot of all unilateral cases. Other measures such as using the mean score of both feet in bilateral cases, randomly selecting one foot of bilateral cases for analysis or separate analysis of bilateral cases have all been described.^{11, 14} However, selecting only one foot from bilateral

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cases for analysis could lead to under representation of the study population, thereby influencing the outcome of the study.^{11, 13} Therefore, in studies involving a mixture of unilateral and bilateral cases, sample size adjustment as well as stratification of unilateral and bilateral cases during randomization should be considered.¹¹

Presently, few studies have compared the severity of unilateral and bilateral idiopathic clubfoot.^{10,11,15} Similarly, any correlation between the left and right feet of bilateral cases is yet to be widely established.¹¹ Therefore, the aim of this study was to compare the severity of bilateral and unilateral idiopathic clubfoot, and to determine if there is any correlation between the left and right feet of bilateral cases.

METHODS

We reviewed the records of all patients with idiopathic clubfoot who presented at our clubfoot clinic between January 2013 and November 2018. We excluded patients who were more than 2 years of age at presentation, as well as those with any established co-morbidity. Patients who had any form of treatment for their clubfoot prior to presentation were also excluded from the study. Records of patients' socio-demographic characteristics as well as laterality of deformity and Pirani scores of affected feet at presentation were extracted.

For the purpose of this study, patients were categorized into two groups, the 'Bilateral Idiopathic Clubfoot' and 'Unilateral Idiopathic Clubfoot' groups. The degree of deformity of each affected foot was categorized based on the Pirani score at presentation as 'Less Severe' for scores less than or equal to 4.0, and 'More Severe' for scores greater than 4.0. Statistical analysis was done using IBM SPSS version 22.0.

RESULTS

Records of 185 idiopathic clubfoot cases belonging to 120 (65 bilateral, 55 unilateral) idiopathic clubfoot patients which met the inclusion criteria were studied. The study population was predominantly male, with a Male: Female ratio of 1.3: 1. The age range of the patients at presentation was 3 days to 24 months. Majority of the patients (N=77, 64.2%) were 3 months of age or less at presentation. See Table I.

Table 1: Distribution of age, gender and laterality of deformity.

Variable	Frequency	Percentage
Age (Months)		
0 - 3	77	64.1
4 - 6	22	18.3
7 - 12	13	11.0
13 - 18	2	1.6
19 - 24	6	5.0
Total	120	100.0
Gender		
Male	67	55.8
Female	53	44.2
Total	120	100.0
Laterality of Deformity		
Unilateral		
Right	28	23.2
Left	27	22.5
Bilateral	65	54.3
Total	120	100.0

The median total Pirani score and mean total Pirani score of the affected feet at presentation were higher for the 'Bilateral Idiopathic Clubfoot' group. See Table 2. The degree of deformity was proportionately more severe in the 'Bilateral Idiopathic Clubfoot' group compared to the 'Unilateral Idiopathic Clubfoot' group ($p < 0.0001$). See Table 3. The left and right feet of the bilateral cases were highly correlated ($r = 0.861$, $p < 0.0001$).

Table 2: Distribution of median and mean total Pirani scores

Laterality of Deformity	Median Total Pirani Score	Mean Total Pirani Score (SD)
Unilateral		
Right	4.0	4.01 (1.499)
Left	4.5	4.33 (1.500)
Bilateral		
Right	5.0	4.57 (1.569)
Left	5.0	4.55 (1.503)

Table 3: Degree of severity of deformity.

Laterality	Less Severe (%)	More Severe (%)	Total (%)
Unilateral			
Right	14 (50.0)	14 (50.0)	28 (100.0)
Left	13 (48.1)	14 (51.9)	27 (100.0)
			<i>P</i> <0.0001
Bilateral			
Right	22 (33.8)	43 (66.2)	65 (100.0)
Left	24 (37.0)	41 (63.0)	65 (100.0)

DISCUSSION

The incidence of idiopathic clubfoot varies across ethnicities, countries and races.^{5,7} Several studies on idiopathic clubfoot have reported predominance of bilateral deformity.^{1, 2, 5, 6, 8} Although the exact aetiology of the deformity is not known, there are theories supporting different aetiologies and the existence of subgroups of the disease.^{2, 9, 10} It is quite possible for unilateral clubfoot cases as well as individual feet in bilateral clubfoot cases in cohort studies to belong to different subgroups.

Zionst et al¹⁵ reported that on the average, bilateral clubfoot patients did not have increased severity, but presented with a larger range of severity than patients with unilateral clubfoot. Agarwal et al¹⁰ reported a higher mean pre-treatment Pirani score (5.4±0.6) for the bilateral clubfoot group compared to the unilateral clubfoot group (4.9±0.7) in their study. Deformity was more severe in a higher proportion (86.4%) of feet of the bilateral clubfoot group compared to the unilateral clubfoot group (60.0%).

However, they noted that despite higher severity of bilateral clubfoot at presentation and the need for more casts to correct the deformity in these cases, outcome of treatment was similar compared to unilateral cases. Similarly, Gray et al¹¹ reported a mean pre-treatment Pirani scores of 5.2 for bilateral clubfoot cases, compared to 4.7 and 4.9 for the left and right unilateral clubfoot

cases respectively. Also, a higher proportion of feet of the bilateral clubfoot cases (71.0%) were very severe, compared to the unilateral clubfoot cases with 56.0% as very severe. They also found a large correlation between the left and right feet of bilateral clubfoot cases ($r=0.68$, $p<0.001$).

In our study, bilateral idiopathic clubfoot constituted majority of the study population (N=65, 54.2%). This is quite close to 53.2% reported by Gray et al.¹¹ However, lower figures were reported by Agarwal et al¹⁰ (40.9%) and Wijayasinghe et al¹⁶ (48.0%), while Azarpira et al¹⁷ (70.5%) reported a much higher figure. The median and mean total Pirani scores were higher for the bilateral idiopathic clubfoot group at presentation, suggesting that the bilateral clubfoot cases were more severe compared to the unilateral cases. The degree of deformity was more severe in a higher proportion of the bilateral clubfoot cases. This is the same pattern reported by Agarwal et al¹⁰ and Gray et al.¹¹ Similar to the report of Gray et al¹¹, we found the left and right feet of bilateral cases to be highly correlated ($r=0.861$, $p<0.0001$).

CONCLUSION

The bilateral idiopathic clubfoot cases were associated with increased severity compared to unilateral cases at presentation. The left and right feet of bilateral cases were highly correlated.

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