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The Idiopathic Clubfoot: Relationship between Patient's Gender and the Degree of Severity of the Deformity

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Authors' contributions

This work was carried out in collaboration among all authors. Author COA designed the study, performed the statistical analysis, wrote the protocol and wrote the first draft of the manuscript. Authors IEA and JEA managed the analyses of the study and reviewed the initial draft of the manuscript. Authors COA and PUA managed the literature searches. All authors read and approved the final manuscript.

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ABSTRACT

Background: The prevalence of the idiopathic clubfoot has been largely reported to be higher in male patients. However, the existence of any relationship between the gender of patients and the degree of severity of the deformity is yet to be established. This study was designed to determine if there is any relationship between the gender of idiopathic clubfoot patients and the degree of severity of deformity.

Methods: Records of all idiopathic clubfoot patients aged 2 years and below who presented at the clubfoot clinic of the University of Calabar Teaching Hospital between January 2013 and March 2019 were reviewed. The degree of deformity of each affected foot based on the ascribed Pirani score at presentation was graded as 'Less Severe' for scores less than or equal to 4.0, and 'More Severe' for scores greater than 4.0. For the purpose of data analysis, patients were categorized into 2 groups, 'Male Idiopathic Clubfoot' and 'Female Idiopathic Clubfoot' groups. Statistical analysis was done using IBM SPSS version 22.0.

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Results: Median total Pirani score and mean total Pirani score were higher for male patients. The degree of severity of deformity for unilateral cases was evenly distributed irrespective of gender, while for bilateral cases, the degree of severity was proportionately more severe for male patients, and evenly distributed for female patients (p=0.514).
Conclusion: In this study, the researchers did not find any significant relationship between gender and the degree of severity of the idiopathic clubfoot.

Keywords: Gender; severity; idiopathic; clubfoot.

1. INTRODUCTION

Idiopathic clubfoot is a congenital deformity of the foot and distal part of the leg which is characterized by equinus and varus of the hindfoot, as well as adductus and cavus of the forefoot [1-6]. It is the commonest congenital deformity of the extremity [1-3]. The Incidence of the deformity ranges from 1 to 3.4 per thousand live births, the value depending on patient's tribe, ethnicity or race [4,7-10]. The definitive cause of idiopathic clubfoot is not known, but its development has been attributed to many factors including environmental, hereditary and pregnancy-related factors [1-6].

A number of reports from studies based on segregation analysis have suggested the presence of a single major gene in the development of clubfoot [11-14]. Also, observation of greater concordance among monozygotic twins compared to dizygotic twins, as well as affectation of a significant proportion of pedigrees of families with multiple affected members both suggest the existence of a potential genetic mechanism [1,15,16].

Several authors have reported a higher prevalence of idiopathic clubfoot in males, with Male: Female ratios ranging from 1.36: 1 to 2.88: 1 [1,4,17-23]. Although a single gene causing idiopathic clubfoot is yet to be identified, the lower prevalence in females may be attributed to an inherent difference in the susceptibility to the deformity, females requiring a higher number of susceptibility genes than males [1].

Zionst et al. [17] found no significant difference in the severity of deformity due to gender. However, any relationship between gender and severity of idiopathic clubfoot is yet to be widely established. This study was therefore designed to determine if there is any relationship between the gender of idiopathic clubfoot patients and the severity of deformity.

2. METHODS

2.1 Study Area

This study was carried out in the University of Calabar Teaching Hospital, Calabar, located in Cross River State, in the South-South region of Nigeria. It is a tertiary facility providing healthcare for residents of Cross River as well as other states of the region and parts of Southern Cameroon.

2.2 Study Design

Records of all patients with idiopathic clubfoot who presented at the clubfoot clinic of the University of Calabar Teaching Hospital between January 2013 and March 2019 were reviewed. Documented records of patients' socio-demographic characteristics, laterality of deformity as well as the degree of severity of each affected foot based on the Pirani scoring system were studied.

Patients who were 2 years of age and below at the time of presentation were included in the study. Patients who had any form of treatment for their clubfoot before presenting to the clinic and those with any established co-morbid conditions were excluded from the study.

2.3 Analysis

The degree of deformity of each affected foot based on the ascribed Pirani score at presentation was graded as 'Less Severe' for scores less than or equal to 4.0, and 'More Severe' for scores greater than 4.0. For the purpose of data analysis, patients were categorized into 2 groups, 'Male Idiopathic Clubfoot' and 'Female Idiopathic Clubfoot' groups. Statistical analysis was done using IBM SPSS version 22.0.

3. RESULTS

A total of 161 idiopathic clubfoot patients who presented between January 2013 and March

2019 were studied. Thirty seven patients which did not meet the inclusion criteria were excluded, leaving the study with 192 clubfoot cases, belonging to 124 (68 bilateral, 56 unilateral) idiopathic clubfoot patients. At presentation, the age range of patients was 3 days to 24 months, with majority presenting at 3 months of age and less. See Table 1.

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

Variable	Frequency	Percentage
Age (Months)		
0 – 3	80	64.5
4 – 6	23	18.5
7 – 12	13	10.5
13 – 24	8	6.5
Total	124	100.0
Gender		
Male	69	55.6
Female	55	44.4
Total	124	100.0
Laterality of deformity		
Unilateral Right	28	22.6
Left	28	22.6
Bilateral	68	54.8
Total	124	100.0

The total Pirani scores of affected feet at presentation ranged from 0.5 to 6.0. Majority (63.8%) of feet with total Pirani Scores of 5.5 and above belonged to male patients. This figure represents 48.6% of the total number of feet studied. See Fig. 1. Male patients had higher median total Pirani score and mean total Pirani score compared to female patients. Irrespective of gender, the mean total Pirani score of the affected feet was marginally higher for the left feet (p=0.497). See Table 2.

The degree of severity of deformity for unilateral cases was evenly distributed irrespective of gender. For bilateral cases, degree of severity was proportionately more severe for male patients, and evenly distributed for female patients (p=0.514). See Table 3.

4. DISCUSSION

Several studies on the inheritance pattern of clubfoot have been inconclusive, even though their findings are suggestive of the existence of a single major gene responsible for the development of clubfoot [1]. In a study by Rebbeck et al. [12], complex segregation analysis suggested the existence of an incompletely dominant gene, leading to incomplete penetrance of the disease, while de Andrade et al. [13] reported the possibility of the existence of a mixed model in their own study. Also, complex segregation analysis of Pacific and Maori people suggesting the existence of a single dominant gene with 33% penetrance has been reported [11].

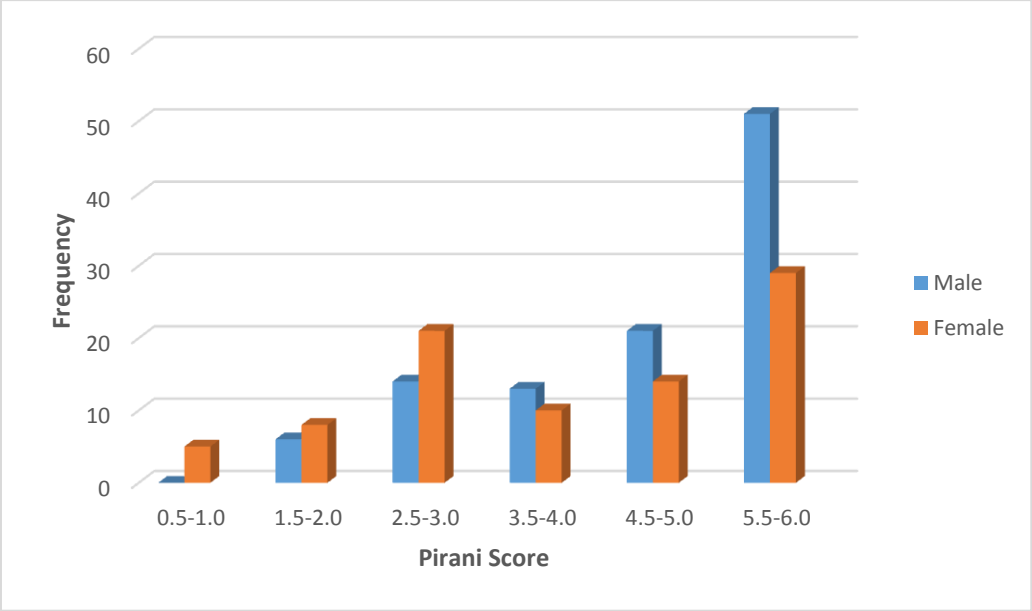

Fig. 1. Distribution of total Pirani scores across gender at presentation

Table 2. Distribution of mean and median total pirani scores across gender

Gender and laterality of deformity	Median total Pirani score	Mean total Pirani score (SD)
Male		
Right	5.00	4.673 (1.3785)
Left	5.50	4.837 (1.2926)
Female		
Right	4.50	4.036 (1.7158)
Left	3.75	4.076 (1.6226)

Table 3. Distribution of severity of deformity across gender

Gender / Laterality	Severity	
	Less severe	More severe
Male (Unilateral)		
Right	10	9
Left	5	10
Male (Bilateral)		
Right	9	26
Left	9	26
Female (Unilateral)		
Right	4	5
Left	8	5
Female (Bilateral)		
Right	15	18
Left	17	16

A polygenic inheritance model with a dynamic sex threshold where females require a greater number of, or more potent susceptibility genes than males to inherit clubfoot has been proposed by Kruse et al. [1]. In their study, they observed that females with clubfoot were 5.55 times more likely than affected males to transmit clubfoot to their children. The prevalence of clubfoot was lowest in daughters of affected males (33%) and highest in sons of affected mothers (85%). They highlighted the fact that the rate of transmission of clubfoot by affected mothers to sons was higher than would be expected with a fully penetrant autosomal dominant condition, which would be transmitted 50% of the time. They therefore submitted that the high transmission rate suggests several possible mechanism, including multigenic, mitochondrial or other environmental factors.

There is no question as to whether there is any relationship between the prevalence of the idiopathic clubfoot and gender as several studies have already established a higher prevalence of idiopathic clubfoot in males over the years, with Male to Female ratios ranging from 1.36: 1 to 2.88: 1 [1,4,17-22]. However, till date, any relationship between patient's gender and severity of the idiopathic clubfoot is yet to be widely established. In a study of 240 infants with idiopathic clubfoot over a period of 8 years, Zionst et al. [17] found no significant difference in the severity of deformity due to gender.

In this study, majority of the affected feet with total Pirani scores of 3.5 and above belonged to male patients, while those with scores of 3.0 and less were predominantly those of female patients. Similarly, the median total Pirani score and mean total Pirani score of affected feet of male patients were higher than those of female patients. Irrespective of gender, the degree of severity of deformity was evenly distributed for unilateral cases but not so for bilateral cases. For the bilateral cases, the degree of severity of deformity was proportionately more severe for male patients and evenly distributed for female patients, a relationship which was found not to be statistically significant ($p=0.514$). This observed pattern may be explained by the fact that the bilateral deformity which is known to be associated with a wider range of deformity was found more in the male patients in this study. The fact that the degree of severity for the unilateral clubfoot cases was evenly distributed irrespective of gender lends further credence to the suggestion that laterality rather than gender is associated with any observed pattern of distribution of severity of deformity.

5. CONCLUSION

The higher incidence of idiopathic clubfoot in male patients as well as the predominance of bilateral disease are well documented. However, the existence of any relationship between gender and the severity of the disease is yet to be established. In this study, the researchers did not find any significant relationship between gender and the degree of severity of the disease. However, it is the opinion of the researchers that a Multi-Centre regional or national study would give further validation to the existence or otherwise of any relationship between gender and severity of deformity.

CONSENT

It is not applicable.

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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