

Full Length Research Paper

Descriptive epidemiology and predisposing factors to idiopathic talipes equinovarus in South South Nigeria

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Talipes equinovarus (Clubfoot) is the commonest foot deformity with variable incidence across races. Exact etiology remains elusive. The objective of this study is to describe the epidemiology of clubfoot in our centre, looking for predisposing factors. This is a descriptive clinic based epidemiologic study of 106 feet belonging to 69 subjects from January 2014 – December 2015 was conducted. Structured questionnaires were administered to clinically confirmed clubfoot subject's guardian or biologic mothers. Phenotypic data from clubfoot subject were also recorded. Analysis was done using IBM SPSS version 22 for windows. Results revealed the age range of the patients as 3 days to 9 years, with a mean age of 46.17 ± 92.03 weeks. Thirty-seven (53.7%) patients had bilateral deformity while 13 (18.8%) patients had right-sided deformity, and 19 (27.5%) patients had left-sided deformity. Majority 40 (58.0%) of the patients were firstborn out of which 28 were males. Only 2 (2.89%) patients are products of multiple gestations. All mothers were none smokers and there was no reported family history of foot deformities or clubfoot. The mean maternal age was $25.81 \text{ years} \pm 4.99$ years. The average length/height of patients at presentation was 64.27 ± 23.93 cm. The average shoulder span of the patients at presentation was 23.93 ± 15.86 cm. The study thus revealed that young maternal age, gender and firstborn are predisposing factors in the etiology of clubfoot.

Key words: Clubfoot, idiopathic, predisposing factors, epidemiology.

INTRODUCTION

Clubfoot or Talipes equinovarus (TEV) is the most common developmental musculoskeletal deformity characterized by forefoot adduction, midfoot cavus, hind foot varus and rigid ankle equines (Omololu et al., 2005;

Adewole et al., 2009; Ngim et al., 2013). There are other associated deformities like, anterior bowing of the tibia and hypoplasia of the calf muscles. It is found in otherwise normal infants and has a worldwide cumulative

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incidence of 1-2 / 1000 live births. This however varies between races, ethnicities and countries. In the USA, a study reported 1.29 /1000 births, while in Africa, some studies like in Malawi reported 2/1000 births and in Nigerian, reported 3.4/1000 births (Parker et al., 2009; Mkandawire and Kaunda, 2004; Ukoha et al., 2011). There is usually an underestimation of the true incidence due to under-reporting and missing cases. Eighty-five percent of infants with clubfoot are reported to live in developing countries (Wallander et al., 2006; Saltzmanns, 2009).

The foot develops from the 9th week of gestation when the limb bud orientation is in the vertical axis. Early on, the soles of the feet normally face one another, but by the 14th week they should have rotated medially and assumed the position of Adult feet. Pre-natal ultrasound diagnosis of the condition is usually practicable by the 17th week of pregnancy. The etiology of clubfoot remains idiopathic but, it has been attributed to various unrelated factors including environmental, hereditary and pregnancy related factors. There are two types of clubfoot; "Typical" and "Atypical". The Typical variety is usually the commonest and is referred to as idiopathic or congenital clubfoot. The Atypical variety is syndromic and is associated with other congenital malformations. There are various scoring systems but the most commonly used is the Pirani scoring and its modifications, also the Dimeglio scoring system (Werler et al., 2013; Sayit and Sayit, 2015; Adegbehingbe et al., 2015; Lynn, 2009; www.global-help.org).

Epidemiologic studies have consistently reported higher prevalence of idiopathic clubfoot in males and first-born but any associations with smoking, race/ ethnicity and maternal age are not clear (Cardy et al., 2007; Dickinson et al., 2008; Kancherla et al., 2010; Pavone et al., 2012). These studies are very scanty in Nigeria and to the best of our knowledge no study has focused on idiopathic clubfoot in our country. We seek to describe the epidemiology of idiopathic clubfoot in our centre and isolate predisposing factors.

METHODS

This is a hospital-based prospective Epidemiologic study conducted among patients attending the foot deformity clinic of a tertiary hospital located in the South-South region of Nigeria, offering specialized services in all aspect of Medicine. It is a 500 bed hospital with Pediatric Orthopedic Surgery as one of the subspecialties of Orthopedics practiced in the Centre. Nigeria is divided into six geopolitical regions (North-East, North-West, North-Central, South-West, South-East and South-South) for ease of administration. Patients are received from neighboring states and Republic of Cameroun.

Though the Ponseti method of treatment of clubfoot was introduced into Nigeria in 2009, its application in our centre started fully in late 2012 with the creation of a special clinic (Clubfoot clinic) in early 2013. Approval for this study was obtained from the institutional Health Research and Ethics Committee.

The study recruited consecutive patients attending the foot

deformity out-patient clinic from January 2014 – December 2015. Those who met the inclusion criteria were recruited into the study; this consisted of patients with only idiopathic talipes equinovarus. The exclusion criteria include those with other foot deformities such as acquired clubfoot, congenital talipes calcaneo-valgus, metatarsus adductus and congenital vertical talus amongst others. They were excluded to increase the likelihood of detecting only predisposing factors associated with clubfoot.

A structured questionnaire was employed, which elicited the following variables, patients' sociodemographic data/clinical features, maternal age, position in the family, mode of delivery and multiple gestations etc. All patient parents or guardian gave informed consent.

Statistical analysis was done using Statistical Package for Social Sciences software version 22 for windows (SPSS 22 Trademark of IBM Corporation). P-value of 0.05 was considered statistically significant. The mean age, prevalence among gender, birth order, laterality (foot affected) and gestational factors amongst other variables were determined.

RESULTS

Sixty-nine (69) patients with one hundred and six (106) feet who met the inclusion criteria were recruited for this study. The prevalence is 3 per 1000 live births in our centre. Age ranged from 3 days – 9 years with "mean SD" of 46.17 - 92.03 weeks. There were forty-three males (62.3%) and 27 females (37.3%) with male to female ratio of 1.7: 1 (Table 1).

Thirteen (18.8%) patients have left foot deformity out of which 8 were males and 5 were females, 19 (27.5%) patients have right foot deformity of which 13 were males and 6 were females. Bilateral feet involvement was in 37 (53.7%) patients out of which 22 were males and 15 were females (Table 2). Relationship between laterality of foot affectation and gender was not statistically significant.

Majority of the patients 40 (58%) were first-born of their mother, while 24 (34.8%) were second born of their mother. Within the first-born group, 28(40.53%) out of 40 were males (Figures 1 and 2). This finding was not statistically significant. Median maternal age was 26 years with a "mean SD" of 25.81 - 4.99 years (Table 1).

Two of the patients were products of multiple gestations but no breech presentations were recorded. All mothers in the study sample were none smokers. There was no positive maternal or paternal family history of clubfoot deformity.

DISCUSSION

The prevalence of clubfoot in our centre is 3 per 1000 live births which is similar to some studies that reported 3.4 and 2 per 1000 live birth respectively (Mkandawire and Kaunda, 2004; Ukoha et al., 2011).

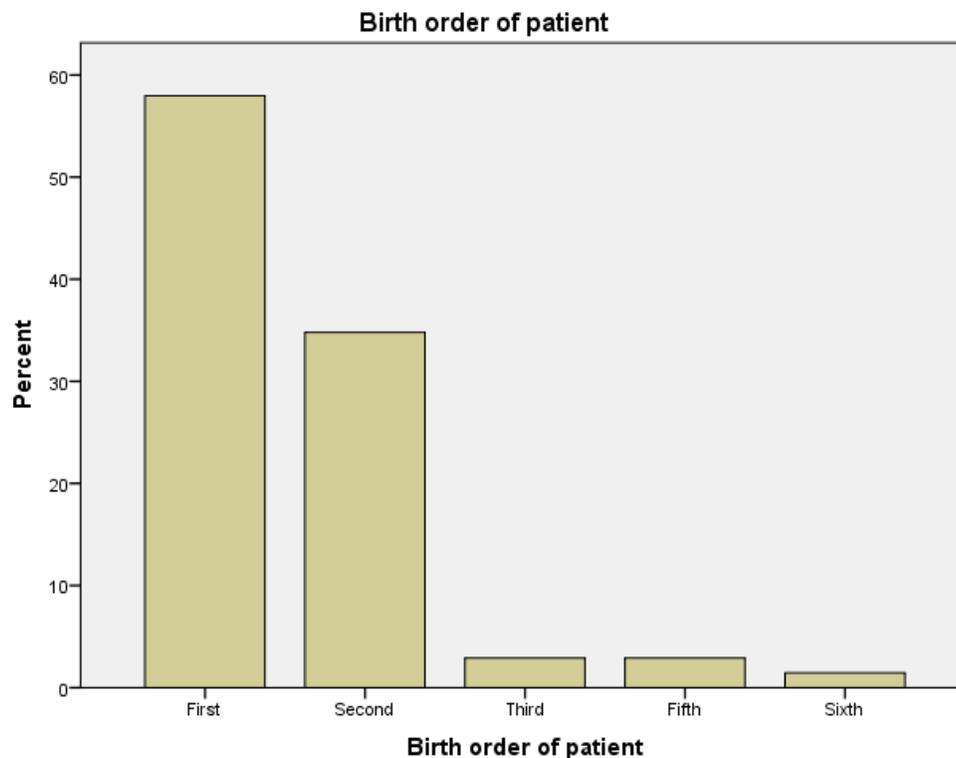
The age of presentation ranges from 3 days – 9 years, which was similar to other Nigerian studies which reported 7 days – 4 years and 1 day – 2 years (Mkandawire and Kaunda, 2004; Adewole et al., 2014).

Table 1. Descriptive statistics.

Parameter	Minimum	Maximum	Mean	Standard deviation
Age of patients in weeks	1	468	46.17	91.444
Height of patients in cm	20	126	62.891	18.334
Shoulder span of patients in cm	7	108	25.16	11.879
Maternal age in years	18	35	25.81	4.992

Table 2. Showing laterality among gender and sample.

Gender		Foot affected			Total count (%)	df	P-value
		Right	Left	Bilateral			
Male	Count	8	13	22	43	2	0.805
	Percentage	18.6	30.2	51.2	100		
	Percentage in sample	11.6	18.8	31.9	62.3		
Female	Count	5	6	15	26		
	Percentage	19.2	23.1	57.7	100		
	Percentage in sample	7.2	8.7	21.8	37.7		
Total	Count	13	19	37	69		
	Percentage	18.8	27.5	53.7	100		

**Figure 1.** Bar chart showing the percentage birth orders of the patient.

The mean age in months was approximately 11.5 months which was still in comparison with other studies in Australia, Bangladesh and India that reported 12 months,

22 months and 4.5 months respectively (Brewster et al., 2008; Chaudhry et al., 2012; Ford-Powell et al., 2013). In the UK and US, some studies have reported mean ages

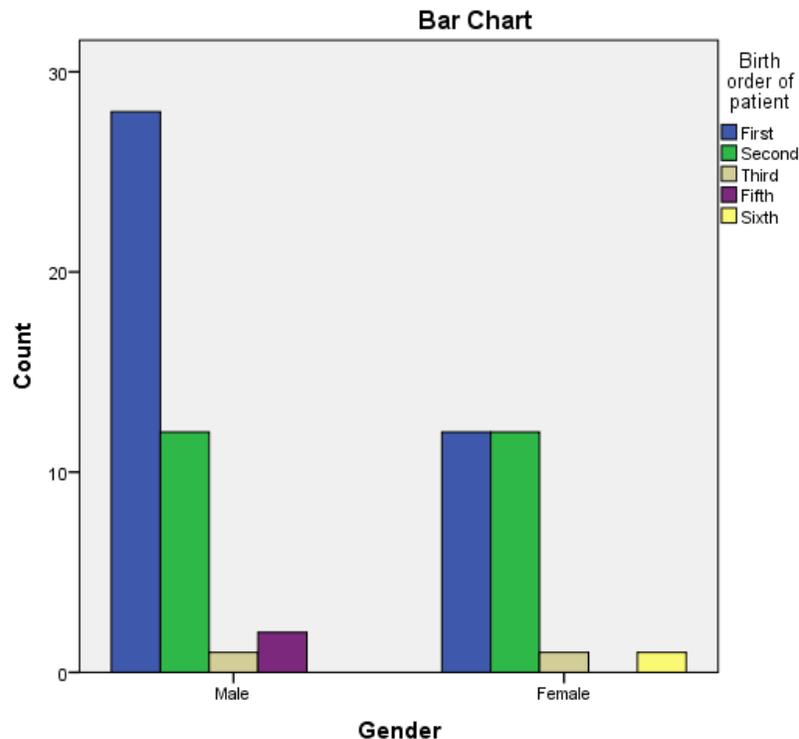


Figure 2. Bar chart showing birth order within the gender.

of 4.5 weeks and 60 days at presentation (Dickinson et al., 2008; Kancherla et al., 2010). The highest age ever reported in Nigeria at presentation was 18 years (Adegbehingbe et al., 2010). This is a reflection of the difference in health awareness of the society to the deformity.

The study shows a strong male preponderance which was similar to other studies in various countries across the world (Werler et al., 2013; Nguyen et al., 2012; Mathias et al., 2010). This confirms the observation by epidemiologic studies that males are more likely to have clubfoot than females. No hypothesis or genetic findings have been reported to fully explain the high prevalence in males.

The findings regarding laterality in our study was not different from other epidemiologic studies but a study by Sayit and Sayit (2015) reported the incidence of foot affected amongst and within the gender, their findings was similar to our study. The tendency that the foot affected would be right or left was not statistically significant. None of the local studies in Nigeria showed this.

The findings that majority of the children affected were first-born (57.9%) of their mother was not different from results of other studies in which the association with primiparity was described (Werler et al., 2013; Cardy et al., 2007; Kancherla et al., 2010). However, this study went a step further to show the number of males and females involved even though this was found not to be

statistically significant. This supports to some extent the theory of fetal constraint as a possible etiological factor in the occurrence of clubfoot (Carey et al., 2005).

In our study, none of the mothers was a smoker. This differs from some studies that reported increased risk of clubfoot in women who smoke cigarettes before and during pregnancy (Dickinson et al., 2008; Werler et al., 2015; Skelly et al., 2002). In our culture, smoking by women is a taboo not to mention the married and those bearing children. We took extra effort to probe into their pre-marital social life but could not extract the information. Also, none of the subjects had a family history of congenital clubfoot and foot deformities. This has been proposed as an etiologic factor. The mode of inheritance does not follow any classic pattern as reported by epidemiological observational studies but has been linked to a gene that directly regulates skeletal muscle protein (Engell et al., 2006; Kruse et al., 2008; Wang et al., 2008). However, both genetic and environmental factors are probably involved (Wynne-Davies, 1972). This finding in our study supports the sporadic nature of the deformity.

In our study majority of the mothers were of young age, although "maternal age has been shown to be both inversely and positively associated with clubfoot" (Cardy et al., 2007; Dickinson et al., 2008; Kancherla et al., 2010). Some studies differ in opinion in that they found no association (Werler et al., 2013; Pavone et al., 2012; Carey et al., 2005; Skelly et al., 2002).

Conclusion

Male gender, first-born males and young maternal age are predisposing factors to the development of clubfoot. Multiple gestations were not significant. All of our cases were sporadic in occurrence. There is need for the establishment of an electronic birth registry and surveillance system in my country.

Limitations

- i) It is a hospital based study which is not reflective of the entire population. A large cohort study is needed to give an analytical epidemiology.
- ii) All of the study subjects have idiopathic clubfeet; therefore certain statistical analysis could not be done.

Conflict of Interests

The authors have not declared any conflict of interests.

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