

RESEARCH

Prevalence and characteristics of congenital talipes equinovarus (clubfoot) in Northern Ghana: a two year retrospective descriptive study

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Abstract

Background

Clubfoot is considered the most common birth defect of the musculoskeletal system with a reported general prevalence of 1 in 1000 live births. Literature documents that a higher prevalence exists in developing countries.

Purpose

This study aimed to determine the prevalence and phenotypic characteristics of clubfoot in the Northern Region of Ghana which is the largest and one of the most deprived regions in the country.

Methods

The researchers collected clubfoot data of children visiting the only clubfoot clinic in the region from January 2015 to December 2016. Parameters of interest included sex distribution, laterality, types of clubfoot, annual trends and overall prevalence rate of clubfoot for the region. A descriptive statistical analysis of the data using SPSS version 16 was made and the outcome represented was on tables and figures.

Results

A total of 112 clubfoot cases were recorded within the review period resulting in a prevalence rate of 0.9 per 1000 live births. The month of January recorded the highest numbers of cases for both years. In addition, twice the number of males were affected as females and bilateral clubfoot formed 65.5% of case presentation. Almost same numbers of left (n=19) feet were affected as right (n=20) in unilateral presentation of cases. Idiopathic clubfoot was the most common type of clubfoot and made up 67.9% recorded of cases.

Conclusion

The outcome suggests that the phenotypic trends of clubfoot in the Northern Region of Ghana is similar to findings in other parts of Ghana, Africa and the world. However, the prevalence rate may be an underestimation of the actual situation owing to poor birth defect surveillance and awareness education of clubfoot in the northern region and Ghana in general. Further studies are needed to investigate risk factors for clubfoot and to explain the phenotypic and annual trends in this part of the world. Management efforts also need to anticipate using greater numbers of casts per child due to the majority of bilateral clubfoot case presentations.

Introduction

Congenital talipes equinovarus (CTEV) commonly known as clubfoot 1, is reported to be the most common musculoskeletal birth defect thought to occur in early weeks of gestation 2, 3. Clubfoot is characterised by equinus of the ankle, varus of the hindfoot, as well as cavus and adductus of the forefoot with an associated atrophy of the calf muscles 4, 5. The most common form of presentation is idiopathic or primary clubfoot 5. According to the Ponseti classification 6, when clubfoot is associated with wider

neuromuscular anomalies such as spina bifida or arthrogyposis, it may be classified as secondary clubfoot.

The exact cause of clubfoot is uncertain although several genetic and environmental factors have been implicated 2, 3, 5 but perhaps the strongest risk factor equivocally documented in literature is the male sex 2-5, 7. Research also reports that first born babies are more likely to develop clubfoot 2, 3, 5 and bilateral presentation occurs in 50% of cases 4.



Research consensus suggests that clubfoot occurs in every 1 per 1000 live births 3, 5 although, variations have been found between racial ethnic groups 4, 7. For instance in the United States of America (USA) Parker, Mai 7 found an overall prevalence of 1.29 per 1000 live births out of which the white population recorded 1.38 per 1000 live births whereas Hispanics and non-Hispanic blacks recorded prevalence of 1.30 and 1.14 per 1000 live births respectively. Worldwide, over 150,000 babies are affected with more than 80% clubfoot cases reported to occur in developing countries and especially in low- resource regions 5.

Clubfoot has been inadequately researched in Ghana, a World Health Organisation recognised Low Middle Income Country (LMIC) 8 and thus the prevalence rates and trends across the country are uncertain varying from region to region. This research aims to address this knowledge gap by being the first to investigate the prevalence of clubfoot as well as the associated phenotypic trends in the Northern region, which is the largest yet third poorest region in Ghana 9. The outcome of the study apart from adding to existing knowledge about clubfoot and providing data for future research, can also help inform policies and plans regarding the treatment of Clubfoot in Ghana.

Methodology

This research set out to determine the prevalence of clubfoot per 1000 live births in the Northern region. It also sought to investigate and describe the trends of clubfoot with respect to phenotype, age group distribution as well as types of clubfoot recorded in the region using a retrospective descriptive cross sectional study design. The research used data (in retrospect) of clubfoot cases recorded from January 2015 to December 2016 in the Tamale Teaching Hospital (TTH) clubfoot clinic in Tamale, the Northern region's capital. The TTH affords tertiary healthcare to persons living in Tamale, the capital of the region, as well as the neighbouring communities and regions and it is also the only facility which runs a clubfoot clinic in the Northern region.

The TTH clubfoot clinic since establishment in 2013, runs once a week and includes a team of clinical (nurses, physiotherapists and orthopaedic surgeons) and non-clinical (administrative and supportive staff). The clinic is also supported by the Ghana Clubfoot programme, which advocates the Ponseti method of treatment and supports the clinic with treatment materials such that clubfoot management is free.

Babies who are brought to TTH (or referred to TTH from other hospitals in the region) suspected to have clubfoot or other physical birth defects are referred to the clubfoot clinic after orthopaedic consultation for further management. Once at the clinic, a more detailed assessment is conducted using the Ponseti screening and classification method to confirm clubfoot and to categorise them into primary and secondary subgroups as well as determine the severity of the deformity. Laterality, sex of the babies and ages are captured in addition and babies are eventually managed with the Ponseti method which has been accepted as the gold standard for clubfoot treatment 10-13.

Operational definitions

For the purposes of this research, the following terms are defined according to the classification system for clubfoot used at the TTH clubfoot clinic:

Primary/ idiopathic clubfoot

All babies who were found to have clubfoot without any other neuromusculoskeletal anomaly were classified as having idiopathic clubfoot.

Secondary clubfoot

During assessment, babies with clubfoot who also presented with other anomalies were classified as having secondary clubfoot. This group was further classified as neuropathic or syndromic clubfoot

Neuropathic clubfoot.

Associated with a neurological disorder such as meningomyelocele, meningocele

Syndromic clubfoot.

Clubfoot presenting with other orthopaedic defects such as congenital arthrogyriposis multiplex, amniotic band syndrome, Down syndrome etc.

Postural clubfoot.

The deformity is very flexible and is thought to be due to intrauterine crowding

Neglected clubfoot.

Clubfoot that has never been treated in a child two years and over

Recurrent clubfoot.

Clubfoot that relapses after or during treatment with the Ponseti method

Treated clubfoot.

Clubfoot treated with the Ponseti method

Data collection

The researchers sought permission from the Tamale Teaching Hospital's Department of Research and Development to allow the use of patients' data for the research. Caregivers of children with clubfoot on the first day of assessment, are also informed that their children's data may be used for research to better understand the condition as part of clubfoot clinic routine.

The investigators collected and examined all folders of patients who reported to the clubfoot clinic from January 2015 to December 2016. These patients included those who had existing clubfoot but lived in their communities not seeking appropriate treatment, as well as new born babies suspected to have clubfoot and were referred to the clubfoot clinic. Once their diagnosis was confirmed, their information was included in the study. Patient folders which recorded diagnoses other than clubfoot or CTEV were excluded from the study. The parameters of research interest included details of the children such as the age of first visit, sex, unilateral or bilateral presentation of the clubfoot (laterality), foot affected (if unilateral) and the type of clubfoot were documented both in hard copy and on a Microsoft excel spreadsheet. The parameters were then analysed descriptively with the Statistical Package for the Social Sciences (SPSS) version 16 software to determine the frequencies of clubfoot cases with respect to each of the parameters during the period under review. The results of the analysis were represented in tables and figures.



Results

There was a total of 112 children diagnosed with clubfoot for the two years (see Figure 1) with a 2:1 male versus female representation (Figure 2). The children were all Ghanaian born Africans with the majority being babies of seven months and below whereas children 29 months and over formed the least frequent age group (see Table 1). The month of January recorded the most numbers of new cases at the clubfoot clinic for both 2015 and 2016, with a sharp decline in numbers especially during the middle to ends of both years (Figure 3). Bilateral clubfoot was 65.1% (n =73) of the total and 34.9% (n = 39) unilateral clubfoot cases, left and right presentations were 17% (n=19) and 17.9% (n=20) respectively of the total number of cases.

Table 1 Age distribution of participants in months. The table shows number of cases recorded for the two years under the various age brackets.

Age groups	Number of children	Percentage distribution/%
7 months or less	76	67.9
8 – 14 months	17	15.2
15 – 21 months	8	7.1
22 – 28 months	7	6.3
29 months and above	4	3.6
Total	112	100.0

Figure 1 Annual distribution of clubfoot cases. The bar charts display the total numbers of clubfoot cases recorded for the years 2015 and 2016, in the Northern region of Ghana, at the clubfoot clinic.

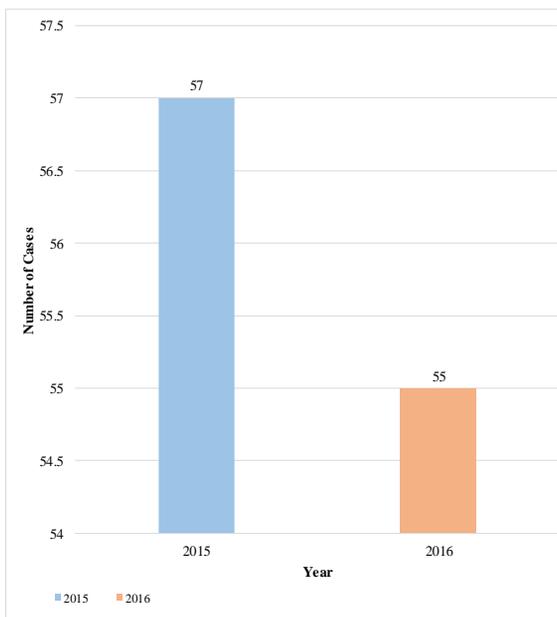


Figure 2 Sex distribution of Participants. The pie chart shows the total number of male and female babies and children with clubfoot, recorded for the years 2015 and 2016 at the clubfoot clinic in the Northern region of Ghana.

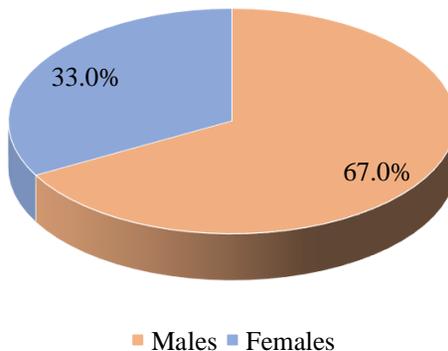
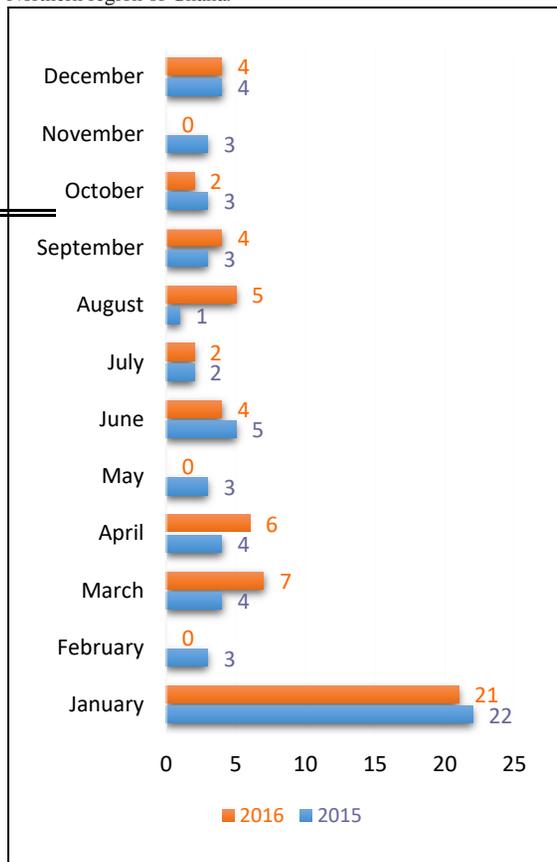


Figure 3 Clubfoot Cases by Monthly Attendance. The figure below represents the number of clubfoot cases recorded for each month of years 2015 and 2016 at the clubfoot clinic of the Northern region of Ghana.





The largest classification subgroup was idiopathic clubfoot whereas postural and neuropathic clubfoot presented least frequently. Details of subgroup results are captured in Table 2.

Prevalence rate

A total of 127,910 live births were recorded in the Northern region of Ghana within the two year period thus the prevalence for the region was 0.9 per 1000 live births.

Table 2 Classification of Clubfoot cases. The table displays the number of clubfoot cases recorded for each type of clubfoot and the associated sex distribution.

Type of clubfoot	Number		Percentage distribution/%
	Males	Females	
Idiopathic clubfoot	54	22	76
Syndromic clubfoot	14	11	25
Neuropathic Clubfoot	1	0	1
Postural clubfoot	0	1	1
Neglected Clubfoot	3	1	4
Treated clubfoot	1	2	3
Recurrent clubfoot	2	0	2
Total		112	100.0

Discussion

Clubfoot is the most common birth defect of the musculoskeletal system⁵. However, there is a dearth of research investigating clubfoot in Ghana. To the best of our knowledge, this is the first clinic-based study in Ghana to determine the prevalence of clubfoot and specifically for the northern region. Generally, findings of this study with regard to phenotypic trend of clubfoot were similar to existing evidence. The results will be discussed under subheadings to cover the prevalence rate, seasonal variation and phenotypic characteristics of clubfoot in the Northern region as follows:

Prevalence of clubfoot in the Northern region

The 1.2 per 1000 live births prevalence reported by previous Ghana based studies^{13, 14} was a figure obtained from the global clubfoot initiative⁸ which in turn used the 'widely accepted' 1.2/1000 to estimate prevalence for low and middle income countries (LMICs) including Ghana. The current study found that clubfoot occurred in every 0.9 per 1000 live births for the 2015 to 2016 period which is lower than the reported prevalence for Ghana⁸ as well as the frequently reported 1 per 1000 global prevalence³⁻⁵. Research has found no association of African (black) ethnicity as a risk factor for developing clubfoot and indeed, prevalence among babies of African descent was consistently lower than that of Caucasians in previous studies^{3, 15, 16}. Our 0.9 prevalence is consistent with the relatively lower occurrence of clubfoot African descent in the United States¹⁵⁻¹⁷ and also falls within the 0.96 to 1.26 range of pooled prevalence for Africa².

Interestingly, LMICs such as Ghana, are said to record about 80% of total clubfoot cases worldwide^{2, 11}. Studies conducted in other African countries such as Uganda and Nigeria found a clubfoot incidence of 1.2¹⁸ to 3.4¹⁹ per 1000 live births respectively. In

Malawi a clubfoot incidence of 2/1000 was found in a clinic based study²⁰ whereas literature documents a report of 3.5 incidence per 1000 live births among black South Africans²¹. Such high incidence figures have enormous implications for prevalence of clubfoot in these countries. These figures also suggest that perhaps not primarily ethnicity, but rather multifactorial causative factors comprising both genetic and environmental components² peculiar to LMICs, could account for the higher prevalence reported for developing countries. There is a chance that the lower prevalence found in our study may be an underestimation of the actual situation, attributable to poor birth defect surveillance of new births and thus an underreporting of clubfoot cases in the Northern region.

Our study captured 112 babies with clubfoot whereas the other studies in Ghana^{13, 14} and Africa^{11, 18-20} have investigated clubfoot in a greater number of children. For instance in Ghana, Boakyee, Afriyie¹³ in a six year retrospective study and Abebre¹⁴ in a five year review recorded 271 cases 420 cases of idiopathic clubfoot respectively in the same clubfoot clinic. Boakyee, Afriyie¹³ included only babies between 0 and 6 months who did not default treatment. An Ethiopian, study²¹ found 258 clubfoot cases (in two-year review), whereas Ukoha, Egwu¹⁹ found 72 within one year in a Nigeria-based research. A multicentre study in Uganda, one of the least developed countries found 872 cases within two years¹⁸ and a study in a Malawi hospital based also found 64 cases in a two-year retrospective study²⁰. It should be noted that Malawi is also reported to be a very poor country and although their population is approximately half Ghana's, it recorded clubfoot incidence that was almost twice the prevalence found in the current study²⁰.

The Northern region of Ghana is the largest but one of the poorest regions of Ghana²². In fact, the three northern regions (Northern, Upper East and Upper West regions) have been documented to inhabit 50% of Ghanaians living in poverty and up to 80% of the Ghanaian population living in extreme poverty²². The poverty in Northern Ghana has been closely linked with the consequential poor health and transportation access and well as poor sanitation and housing conditions [Ibid]. It is interesting therefore, that only 112 cases of clubfoot were recorded within two years in our study. It has been documented that LMICs usually lack thorough birth defect surveillance programmes which make determination of prevalence challenging¹¹.

The clubfoot clinic used in this study is a relatively newly established referral center within the Tamale Teaching Hospital which is a Tertiary health facility. Looking at the age group categories in our studies, the participants were relatively young with the largest age group being children seven months and below (67.9%) followed by children 8 to 14 months (17%). The four children in the '29 months and above' category also happened to be the neglected cases of clubfoot (according to the clinic records). In Nigeria, the high clubfoot prevalence was attributed to their large numbers of neglected clubfoot cases¹¹. It is possible that there are more neglected and untreated clubfoot cases in Ghana's Northern region communities which have not sought medical attention simply because they are unaware that clubfoot is treatable and freely available at the clubfoot clinic. In addition, if health workers in primary and secondary healthcare facilities or traditional birth attendants are not trained to identify clubfoot or know that treatment



is possible, they will likely not refer cases to centres of treatment. Closely related to case identification in rural settings is treatment seeking behaviour of the rural population which is also found to be strongly influenced by poverty and challenges with physical access to treatment facilities¹⁸. These situations mentioned can potentially result in the underreporting of clubfoot cases² and the eventual under-estimation of the Northern Region's prevalence rates.

On the other hand, the relatively lower prevalence in this study compared to other African countries may also be a reflection of the variation in prevalence across different developing African countries and not due to underreporting of cases per say. It is worth noting that the majority of these Africa-based studies were conducted in individual hospitals and may not reflect the true prevalence of clubfoot nationally for each country. However, future research which will investigate the predictors of variation in clubfoot prevalence across different African countries could make an important addition to understanding clubfoot in Africa's LMICs. Likewise, promotion of clubfoot awareness and robust birth defect surveillance of newborns could massively improve research in clubfoot and other congenital deformities in Ghana. Such surveillance database can also be the evidence for underreporting of congenital birth defects like clubfoot to medical facilities.

Seasonal variation in the annual distribution of clubfoot cases

Treatment seeking behaviour of our study population could have also influenced this study's findings with respect to the monthly trend of new cases in the clubfoot clinic within each year. January recorded the most numbers of new cases for both 2015 and 2016 and the numbers significantly declined in the middle and ending of the year (Figure 3). The two seasons in Ghana (rainy and dry seasons) may have influenced this observation due to the associated economic activities during these periods of the year. The indigenous population of the Northern Region are mainly farmers with the majority farming on a small scale. Research²² indicates that due to the poverty in Northern Ghana compounded by rainfall variations (experienced in this region) in recent years, most northern farmers (male and female) migrate to the south of Ghana during the rainy season (March/ April to October) in anticipation of poor harvest and acute food shortage.

Investigating the reason for high number of clubfoot cases in January (dry season) and the sharp decline thereafter was beyond the scope of this study as was including the occupations of parents or caregivers of the participants. However, it is possible that caregivers of children with clubfoot did not seek treatment at other times in the year due to seasonal farming activities during the rainy season period. In January when the harvest period was over, most caregivers may have found it more suitable to seek medical attention for their children with clubfoot. This explanation requires further research to be established as a likely reason for the seasonal variation in reported clubfoot cases in the Northern region of Ghana.

Phenotypic characteristics of clubfoot in the Northern region of Ghana

Idiopathic clubfoot was found to be the most frequently presenting type of clubfoot, as was observed in 76 children, making up 67.9% of the 112 cases recorded within the review period. This observation is in agreement with existing studies^{2-4, 13, 14} which have

reported that idiopathic clubfoot is the commonest form of the condition. The cause of idiopathic clubfoot is still uncertain but has been associated with some sociodemographic and pregnancy related risk factors including maternal age (at conception of affected child), smoking, marital status, education and diabetes^{4,5}.

Syndromic clubfoot was seen in 20% of the cases which is quite significant and clinically important considering that this form is more resistant to treatment^{6,18} likely due to the underlying genetic-linked risk factors for this type²³. Bacino and Hecht²³ also documented 20-25% of syndromic clubfoot although they included neuropathic forms of clubfoot in their definition of 'syndromic'. Researchers have postulated that variation in PITX1 as well as TBX4 genes cause syndromic clubfoot and this gene variation has not been observed in non-syndromic forms of clubfoot²³. Ultimately, maternal smoking and family history is said to increase risk of clubfoot up to about twenty times.

Genetic factors have also been associated with sex^{3, 23} and laterality distribution²⁴ in clubfoot cases. Our study found that male children made up 67% of the babies and thus were twice the number of female clubfoot cases. This result is consistent with reports of higher numbers of male clubfoot cases in existing literature in Africa^{13, 14, 20, 25} and other parts of the world^{3, 5, 7, 23, 24} which strongly suggests that the male sex as the most consistently reported risk factor for developing clubfoot. For the various subtypes of clubfoot as well, males had higher numbers than females according to the current study (Table 2). There is no specific reason for the male dominance in clubfoot prevalence; other congenital malformations such as spina bifida, orofacial cleft, neural tube defects and some cardiac anomalies have also demonstrated male preponderance for reasons uncertain²⁶. Possible attributing factors could be that more male children are generally born than females, thus a higher number of birth defects such as clubfoot could be controlled by sex-specific survival mechanisms²⁶. Other theories suggest that there may be sex-specific vulnerability to disease associated teratogenic factors in favour of female babies resulting in the fewer manifestations of such malformations relative to males exposed to similar teratogens^{3, 27}. Also, a threshold effect of these clubfoot-related genes has been suggested such that, females will need a greater genetic load in order to express clubfoot²⁸.

Similarly, bilateral clubfoot may result from an increased load of genetic linked factors for clubfoot with resultant manifestation in both feet rather than one. The current study documented twice as many bilateral clubfoot (65.1%) as against unilateral clubfoot (34.9%). The greater bilateral composition of clubfoot in the current study though consistent with literature, is relatively higher than existing findings within^{13, 14, 19} and outside Africa,^{2, 5} perhaps second only to Malawi where 71% of bilateral clubfoot was documented. In addition, the current study found approximately the same number of right (17.9%) clubfoot as left¹⁷ in the unilateral cases. The side most affected by clubfoot is commonly reported to be the right foot^{14, 17, 20, 21} even though slight dominance of the left clubfoot has been found by some researchers⁵.



Our study did not include sub group analysis to determine laterality distribution between the sexes or their respective severities of the condition in order to lend support or otherwise to the proposed gene threshold theory. This notwithstanding, the postulation that clubfoot in females, bilateral clubfoot and syndromic clubfoot presentations may result from a relatively high clubfoot-associated gene load has important clinical implications for associated severity²⁸ and thus possible resistance to treatment. Thus, clinicians who manage clubfoot in these sub categories of children could consider the possibility of these groups requiring more casts or having more cases of recurrence attributable to the underlying genetic factors.

Strengths and limitations of the study

This study is the first research in Ghana to calculate clubfoot prevalence and to describe associated phenotypic trends in the Northern region and thus it contributes to existing literature in this regard. Also, the findings are generally unanimous with existing data in terms of bilateral, male and idiopathic preponderance of clubfoot presentations. However being purely descriptive, the study did not set out to conduct risk factor investigations for the study population in this research. Also, being a clinic based study, our data was limited to the cases that were identified and referred to the clubfoot clinic and thus the outcome may not be generalisable as the actual

reflection of clubfoot characteristics for the entire northern region or for Ghana as a whole. In the absence of a birth defect surveillance database for the region however, there is no objective evidence of 'missed' cases.

Conclusion

This study found a 0.9 per 1000 live births prevalence of clubfoot in the Northern region of Ghana with twice as many males than females having the condition. In addition, idiopathic clubfoot and bilateral manifestations were the most common disease presentations for the 2015 to 2016 period. The significant numbers of bilateral and syndromic presentations may have important clinical and financial implications for management of clubfoot in this setting.

There remains the possibility that the prevalence rate may be an underestimation of the actual situation and this warrants further research in order to build a case for thorough national congenital defect surveillance as well as mass clubfoot education programmes for interest groups nationwide.

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