



## Occurrences of Idiopathic Congenital Talipes Equinovarus at the Komfo Anokye Teaching Hospital

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### Author's contribution

The sole author designed, analyzed, interpreted and prepared the manuscript.

### Article Information

DOI: 10.9734/BJMMR/2016/27304

#### Editor(s):

(1) Paulo Ricardo Gazzola Zen, Department of Clinical Medicine, Federal University of Health Sciences of Porto Alegre (UFCSPA), Brazil.

#### Reviewers:

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(2) Murat Demiroglu, Medeniyet University, Turkey.

(3) Habiba Bouhamed-Chaabouni, University Tunis El Manar, Tunisia.

Complete Peer review History: <http://sciencedomain.org/review-history/15390>

Original Research Article

Received 27<sup>th</sup> May 2016

Accepted 8<sup>th</sup> July 2016

Published 14<sup>th</sup> July 2016

### ABSTRACT

**Aim:** To investigate the occurrences and seasonal distribution of idiopathic clubfoot at the Komfo Anokye Teaching Hospital.

**Study Design:** A five – year – retrospective cross – sectional study was conducted from January 1, 2010 to December 31, 2014.

**Place and Duration of the Study:** The study was conducted at the Department of Physiotherapy of the Komfo Anokye Teaching Hospital, Kumasi – Ghana, between November 2015 and March 2016.

**Methodology:** Clinical folders of all congenital clubfoot cases over the stipulated period were reviewed, out of which 420 fell under idiopathic category of the deformity. Data on the demographic characteristics, coupled with the occurrences, in time series, of idiopathic clubfoot, were retrieved, entered into SPSS version 20, and analyzed with inferential and descriptive statistical tools.

**Results:** Four hundred and twenty (420) cases of idiopathic clubfoot were reviewed. Males recorded higher prevalence 217 (51.7%) than females, with majority of the cases (57.0%) being bilateral. Chi square testing revealed that there was no significant association between gender and number of feet affected ( $P = .95$ ), and no significant associations between the climatic season and the following; dominant gender category affected ( $P = .43$ ) and number of feet affected ( $P = .31$ ).

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Analysis of the trend line produced ( $r = -0.04$ ), confirming a negative trend of the deformity since 2010, but the slope parameter is not significant at 5% significance level ( $P = 0.09$ ).

**Conclusion:** According to the findings of the study, external climatic conditions may have very little or no influence on the occurrences and distribution of idiopathic clubfoot. Thus, the aetiology may be skewed largely to inherent pathophysiologic and or genetic events during foetal development.

*Keywords: Clubfoot; idiopathic; congenital; seasonal distribution; prevalence.*

## 1. INTRODUCTION

Congenital talipes equinovarus (CTEV), commonly called clubfoot, is a common birth defect of the foot which occurs in about one out of every thousand live births [1]. It is estimated that more than 100,000 babies are born worldwide with this deformity yearly [2]. About half of all cases are bilateral, and for the unilateral cases, the right foot is dominant [3]. The deformity physically presents like a gulf club, making it being popularly referred to as clubfoot [4]. The cause of the deformity is not completely understood, but however, linked to the combined effects of both genetic and environmental factors [3]. In developing countries, most cases of congenital talipes equinovarus are left untreated, despite the possibility of about 90% complete recovery when early and appropriate treatment is sought [5].

Epidemiological studies on the incidence of CTEV per 1000 live births have been conducted in different parts of the world, some of which are as follows; Denmark, 1.2 [6], the United States of America 1.0 [7], Australia (Caucasian) 1.11 [8], Australia (Aboriginal) 3.49 [8], Belgium 1.6 [9], Sweden 1.4 [10], Malawi 2.0 [11], Papua New Guinea 2.7 [12], Uganda 1.2 [13], Nigeria 3.4 [14], and Ghana 1.2, with an estimated eight hundred and twenty (820), babies born yearly with the deformity [15]. The dominant gender category mostly affected by this deformity is males [16-19].

Despite the complexity with the nature of its aetiology, family history has proven to be common with most cases [20], but with heritable variations in different populations such as 24-30% in Caucasian population [21], as against 54% in Polynesian population [20]. The mode of inheritance, however, does not conform to the classic Mendelian pattern [22], and complex segregation analyses carried out showed that a single gene major effect operating against polygenic background could be the most likely pattern of inheritance [20,22]. Although maternal smoking and family history have separately been associated with the cause of the deformity, their

combined effect showed greater influence, indicating the interaction between genetic factors and tobacco exposure [23]. Amniocentesis trials linked amniocentesis, especially during the first trimester, to the occurrence of clubfoot, and is attributed to amniotic fluid loss [24]. When the vasculature at the level of the sinus tarsi of 12 clubfoot cases were examined by Atlas et al., there were blocking of one or more of the branches of the vascular tree of the foot of the fetuses [25]. Reduced perfusion through the anterior tibial artery was then directly linked to the muscle wasting at the ipsilateral calf region commonly seen with idiopathic clubfoot cases [25].

Barker and Macnicol in 2001 investigated the relative occurrence of idiopathic clubfoot with respect to the season of the year in a Scottish population, recording more cases in March, and October recording the least [21]. It is, however, unclear whether the seasonal climatic factors influenced the outcomes of the study or not. Moreover, global generalisation of the results of the study may not be very appropriate by virtue of the different climatic conditions occurring in different geographical locations. Historically, from 1778 to 1940, birth rates in the Northern climates peak in late winter (March), and are lower in summer (June, July, August) [26]. Possible relationship between temperature and seasonal birth rates was studied by Lam and Miron [27], whose results showed decreased conception with extreme higher environmental temperatures, emphasising on the study by Levine [28] who reported of the concentration (quality) and quantity of sperm per single ejaculation being significantly lower in the summer than any other season.

Clubfoot may be classified based on the cause and the treatment stage of the deformity. Classification based on cause groups the deformity under either primary (of unknown cause) or secondary (associated with other conditions) [29]. The second mode of classification, which is based on the treatment stage, takes into account a method of conservative management introduced by

Dr. Ignacio V. Ponseti in the 1950's, a method which was later made popular in 2000 by Steve Mannio Wildon (Physiotherapist) in Africa, and by Dr. John Herzenberg in the United States and Europe [30]. Severity of the deformity is assessed with the Pirani severity grading system which is based on six clinical contracture scores which are graded 0 (no deformity), 0.5 (moderate), and 1.0 (severe) [31]. The scores assess and quantify anatomical deviations in hind and mid-foot. The hind-foot contracture scores consist of posterior crease (PC), rigidity equinus (RE), and emptiness of heel (EH), while the mid-foot contracture scores (MFCS) comprise of the medial crease (MC), curvature of lateral border (CLB) and lateral head of the talus (LHT) [31].

Clubfoot may be managed either conservatively or by surgery depending on severity [32], with the goal of achieving a plantigrade, pain-free and functional foot [5]. The Ponseti method serves as the gold standard of clubfoot management, as it presents better outcomes than other conservative methods such as the Kite's [33]. About 90% of clubfoot cases are successfully treated with this method, producing optimal foot function and appearance [34]. Surgical correction of clubfoot is generally recommended for severe cases, but the lasting effects are generally not desirable, as some of the effects include weakness, stiffness, and early arthritic changes [35]. And again, in the long term, a significant proportion of adults who had surgical treatment for their clubfeet may require additional surgeries, which are not only expensive but also associated with several complications as well [36].

Birth defects pose global threats as, for example, global estimate of 510000 deaths occurred in the year 2010 as a result of congenital disorders [37]. Idiopathic clubfoot, just like any other congenital abnormality, presents economic, psychosocial, emotional, and health significance or implications [2,38]. There have been isolated studies which report sharp contrasting results on clubfoot such as unilateral cases being larger than bilateral ones [39,40], a scenario which is uncommon.

Despite the knowledge about the incidence rate of congenital talipes equinovarus in Ghana being 1.2 per thousand live births [15], little is understood of the pattern of distribution and occurrences across the year. A study by Osei et al. [41] reported of more births occurring in the rainy season in a part of Ghana from 2010 to

2014. Since generalisation of the findings of Barker and Macnicol [21] may not aptly represent the Ghanaian perspective due to climate differences, the study sought to investigate whether or not the climatic conditions of a particular season could influence the distribution of idiopathic clubfoot. Again, the study intends to expand the body of knowledge regarding the deformity in Ghana, particularly with respect to the idiopathic type. Despite the variations in the climatic seasons between Ghana and Scotland, the trend of occurrence of the deformity would be better understood. Thus, there would be enormous benefit for the scientific discourse community and the nation at large.

## **2. MATERIALS AND METHODS**

### **2.1 Procedure**

Notification on the proposed study was sent to the Research and Development Unit of the Komfo Anokye Teaching Hospital (KATH), through the head of the Department of Physiotherapy. Certification from the Research and Development Unit facilitated the seeking and acquisition of ethical clearance from the Committee on Human Research Publications and Ethics (CHRPE) of the Kwame Nkrumah University of Science and Technology (KNUST) and KATH. Following ethical clearance and permission from the head of the study site (Department of Physiotherapy), the study commenced.

### **2.2 Participants**

All cases of clubfoot reported to the Clubfoot Clinic of the Department of Physiotherapy for possible management from January 1, 2010 to December 31, 2014 served as the study population out of which participants were selected. There were 524 folders available for review at the clubfoot clinic of the physiotherapy department. These clinical folders have diagnosis sections where physiotherapists document the diagnosis following a detailed physical assessment. The physical examination, which is based on the Pirani grading system, is coupled with syndromic and or neurologic findings or presentations that aid differential diagnosis. Following the review process, there were cases of 420 idiopathic clubfoot, 39 postural clubfoot, 42 secondary clubfoot, and the remaining 23 composed of other congenital orthopaedic cases such as aarthrogryposis congenita, metatarsus adductus and other foot defects. Having determined the various diagnosis

in the clinical folders, the idiopathic clubfoot cases were all included in the study. Thus, secondary clubfoot (neuropathic and syndromic), postural clubfoot cases, and non-clubfoot conditions were excluded thereof.

### 2.3 Study Design and Data Collection

A cross-sectional study plan was implemented. Clinical folders of clubfoot cases which satisfied the inclusion criteria of the study were retrieved. Data capturing sheets were used to collect the relevant data to the study and reviewed thereof. Demographic data and information on seasonal characteristics and distribution were recorded on structured data capturing sheets.

### 2.4 Data Analysis

Data obtained were analysed using the Statistical Package for Social Sciences, SPSS version 20, along with Microsoft excel, 2010. Descriptive and inferential statistical methods were used for the analysis. Bar charts, frequencies, tables, trend lines, chi square, and regression for time series constituted the core statistical methods used for analysing data obtained from the study.

## 3. RESULTS AND DISCUSSION

### 3.1 Results

Clinical folders of four hundred and twenty (420) cases of idiopathic clubfoot, out of 524 folders, from January 2010 to December 2014 were considered for the study. The prevalence of idiopathic CTEV, according to the study, is higher in males 217 (51.7%) than in females 203 (48.3%). The gestational period of majority of cases 384 (91.7%) were full term, while 23 (5.5%) and 13 (3.1%) were preterm and post term respectively. It can be observed that for full term and post term deliveries, majority within each group recorded higher bilateral affectation than unilateral, (57% for full term; 54% for post term) (Fig. 1). A chi square test revealed that the proportions of number of feet affected across the various gestational periods are not significantly different from one another ( $P = .659$ ). Higher bilateral cases of idiopathic clubfoot 238 (56.7%) than unilateral 182 (43.4%) were recorded in the study. For all unilateral cases reviewed, the right foot recorded higher prevalence 99 (54.4%) than did the left foot 83 (45.6%). The prevalence of idiopathic clubfoot in the rainy season of Ghana was 231 (55.0%) whilst that of the dry season, 189 (45.0%). Chi square test showed that prevalence of idiopathic CTEV in the rainy

season is significantly higher than that of the dry season ( $P = .04$ ).

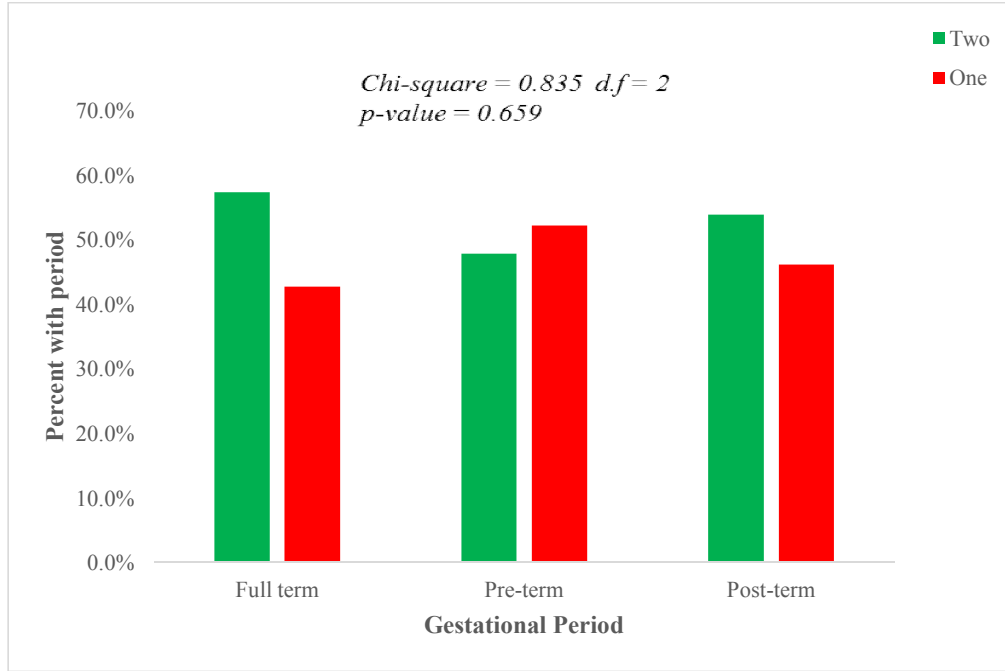
Annual trend lines of cases of idiopathic clubfoot (Fig. 2) revealed that January 2010 recorded the highest number of cases under the years of review, while August 2012 had the least. With the exception of the years 2012 and 2013, the rest of the years appear to exhibit a decreasing trend from January to December. Findings for gender and number of feet affected showed that for all bilateral cases, both males and females accounted for 199 (50.0%). Similarly, for the unilateral cases, the proportion of males and females were 99 (53.8%) and 84 (46.2%) respectively. In testing for the significance of the association between gender and number of feet affected, a chi square test for independence revealed that there is no significant association between gender and number of feet affected in idiopathic clubfoot. ( $P = .95$ ). With respect to the significance of the association between the climatic season and dominant gender category affected, a chi square was used to examine this relationship at 5% significance level. The test results showed a non-significant association between the two variables ( $P = .43$ ). Again, analysis of the association between climatic season and number of feet affected showed no significant association ( $P = .31$ ).

As a result of the data being in a time series, trend analysis was conducted. First, the study looked at comparing the number of cases across the years, each month across the years, and estimation of the trend line. The year 2010 recorded the highest number of cases 101 (24.0%), while the year 2014 recorded the least number of cases (66 cases (16.0%)). Generally, there is a decreasing trend from 2010 to 2014 (Fig. 3). A chi square test revealed that the proportion of cases recorded in each year are not significantly different ( $P = .06$ ).

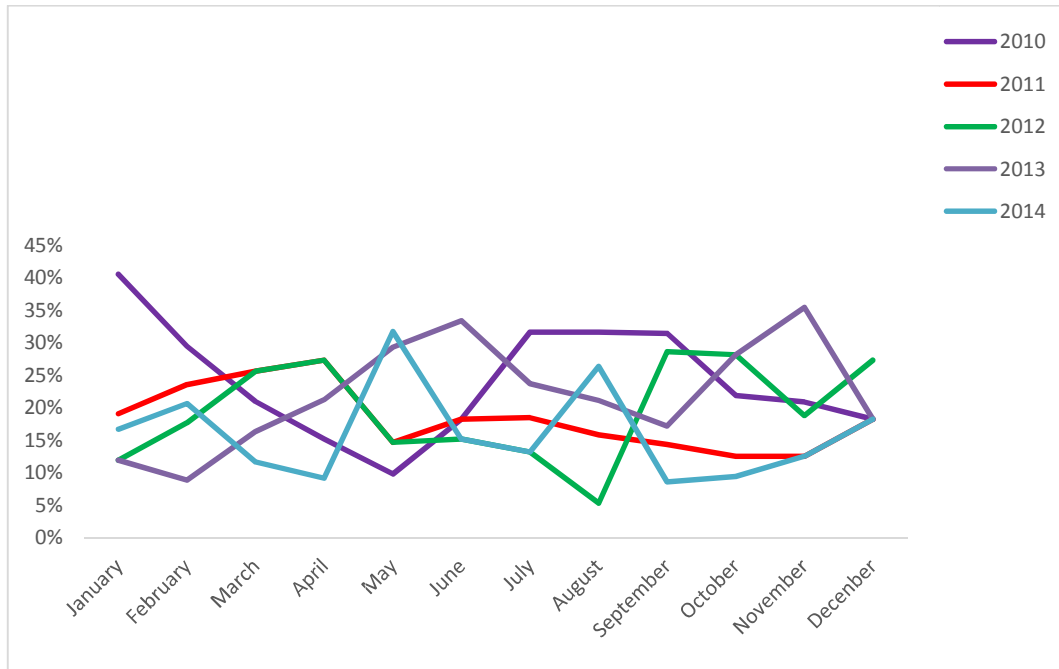
From Table 1, only the month of January exhibits significantly not the same proportion of cases across the years. ( $p = .019$ ). January recorded the highest in 2010 (40.0%) and the lowest in both 2012 and 2013 (12.0%). The remaining months did not significantly exhibit different patterns across the years. For the month of February, it recorded its highest and lowest proportion of cases in 2010 and 2013 respectively. Details from the other months are summarized in (Table 1). It can be observed from Fig. 4 that the overall trend line of cases of idiopathic CTEV exhibits a decreasing trend. A

regression technique was used to estimate the trend line. The slope of the line is negative ( $r = -0.04$ ) confirming the negative trend of cases of idiopathic CTEV since 2010 January.

However, the slope parameter is not significant at 5% significance level ( $p\text{-value} = 0.090$ ). The regression estimate of trend line is shown in Table 2.



**Fig. 1. Gestational period's relationship with number of feet affected**



**Fig. 2. Yearly trend lines of cases of idiopathic clubfoot**

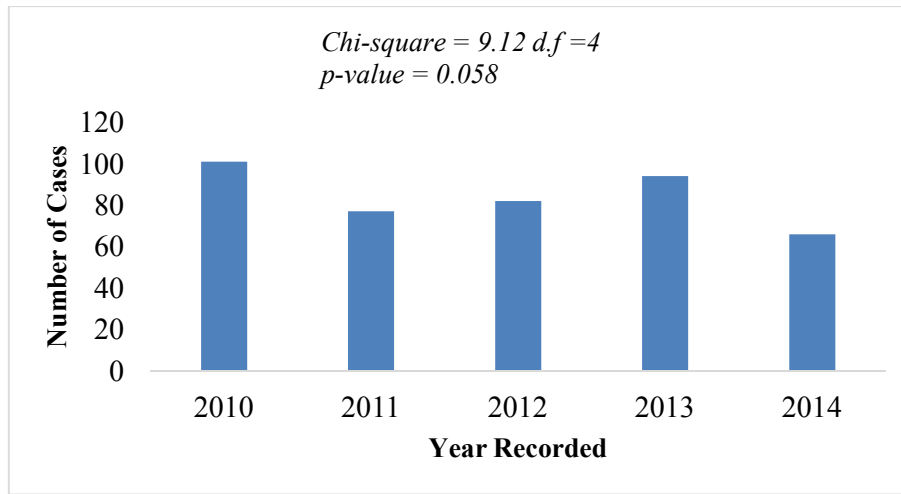


Fig. 3. Number of cases across years

Table 1. Monthly comparison of cases across years

Month	Year of delivery					Chi-square	Df	P-value
	2010	2011	2012	2013	2014			
January	40%	19%	12%	12%	17%	11.81	4	0.019*
February	29%	24%	18%	9%	21%	3.94	4	0.414
March	21%	26%	26%	16%	12%	3.16	4	0.531
April	15%	27%	27%	21%	9%	4.12	4	0.390
May	10%	15%	15%	29%	32%	7.90	4	0.095
June	18%	18%	15%	33%	15%	3.82	4	0.431
July	32%	18%	13%	24%	13%	4.63	4	0.327
August	32%	16%	5%	21%	26%	3.90	4	0.420
September	31%	14%	29%	17%	9%	6.57	4	0.160
October	22%	13%	28%	28%	9%	4.88	4	0.300
November	21%	13%	19%	35%	13%	8.46	4	0.078
December	18%	18%	27%	18%	18%	0.73	4	0.976

### 3.2 Discussion

The study sought to investigate, primarily, how, in time series, seasonal climatic conditions of Ghana could influence the occurrence and pattern of distribution of idiopathic congenital talipes equinovarus. Since idiopathic clubfoot constitutes the majority (about 90%) [15] of all cases of clubfoot combined, and with aetiology which is not completely understood, investigating this subject was an important area or aspect of clubfoot to tackle. A related study conducted in Scotland by Barker and Macnicol [21], who investigated the seasonal pattern of the deformity, considerably influenced the commencement of this study.

According to the study, the male to female prevalence ratio of idiopathic clubfoot is 1.1:1. This result is consistent with other clubfoot-

related studies including Boakye et al. [16], Agarwal and Gupta, [17], Lavy et al. [18], Wallender et al. [19]. However, it must be stated that these referenced studies did not consider only the idiopathic type of clubfoot. Thus, the other types of clubfoot contributed to the ratios obtained thereof. The record of higher bilateral than unilateral cases according to this study were consistent with previous findings of Boakye et al. [16] and Honein et al. [23], but however, inconsistent with Wallender et al. [19], Byron-Scott et al. [39] and Rasit et al. [40] whose findings produced higher unilateral than bilateral cases. Right unilateral cases were higher in the unilateral category, and is consistent with studies conducted by Boakye et al. [16], Agarwal and Gupta, [17], Wallender et al. [19], and Byron-Scott et al. [39], but inconsistent with that of Rasit et al. [40] who recorded more left unilateral cases than right. Genetic and environmental variations

among different populations could have accounted for some of the variations of findings of these different studies.

Results on the gestational periods of all the cases of idiopathic clubfoot indicated a 91.7% for those who completed full term gestational period. Despite the proportion of full term delivery cases being significantly larger than the other categories, it is not suggestive that full term babies are more likely to suffer idiopathic clubfoot since the study considered only the cases without the controls.

Regardless of the gestational period of the cases, chi square test revealed that having a bilateral or unilateral affectation are not significantly different among the gestational period categories. ( $P = .66$ ). Therefore, it can be implied that the factors causing idiopathic clubfoot, perhaps, do so inherently with little or no influence on the length of time spent in the womb, regardless of the nature of the external climatic factors around the mother. Also, the strong familial evidence of the deformity in 54% in Polynesian population [20] and 24-30% in Caucasian population [21] support the extent of the inherent nature of the aetiology of idiopathic clubfoot. However, studies on genetic connectedness to idiopathic clubfoot have not been conducted in many parts of the world to strengthen the globalisation of such findings,

owing to the fact that there could even be greater possible variations in family history of the deformity in different populations as, for instance, reported by Chapman et al. [20] and Barker and Macnicol [21].

Higher prevalence was recorded in the rainy season which is typified by lower temperatures than it did for the dry season. However, this is not conclusive since there are strong global evidences that suggest that quality (concentration) and quantity of sperm [28], conception, and birth rates [26] are themselves favoured by lower temperatures. The linear trends and seasonality of births, a study conducted by Osei et al. [41] in Ghana covering the years 2010 to 2014, reports of higher birth rates in the rainy season than for the dry season. Hence, the higher prevalence of idiopathic clubfoot in the rainy season occurred as a result of the corresponding rise in birth rates usually recorded during that season. Moreover, in other populations, temperature, in itself, has been regarded as unimportant in the trend of birth rates [27]. Similarly, considering the study by Ehrenkranz [26], it may be suggested that increased birth rates in March could have naturally resulted in a corresponding increase in the number of idiopathic clubfoot cases in March in the study conducted by Barker and Macnicol [21] in a Scottish population in 2001.

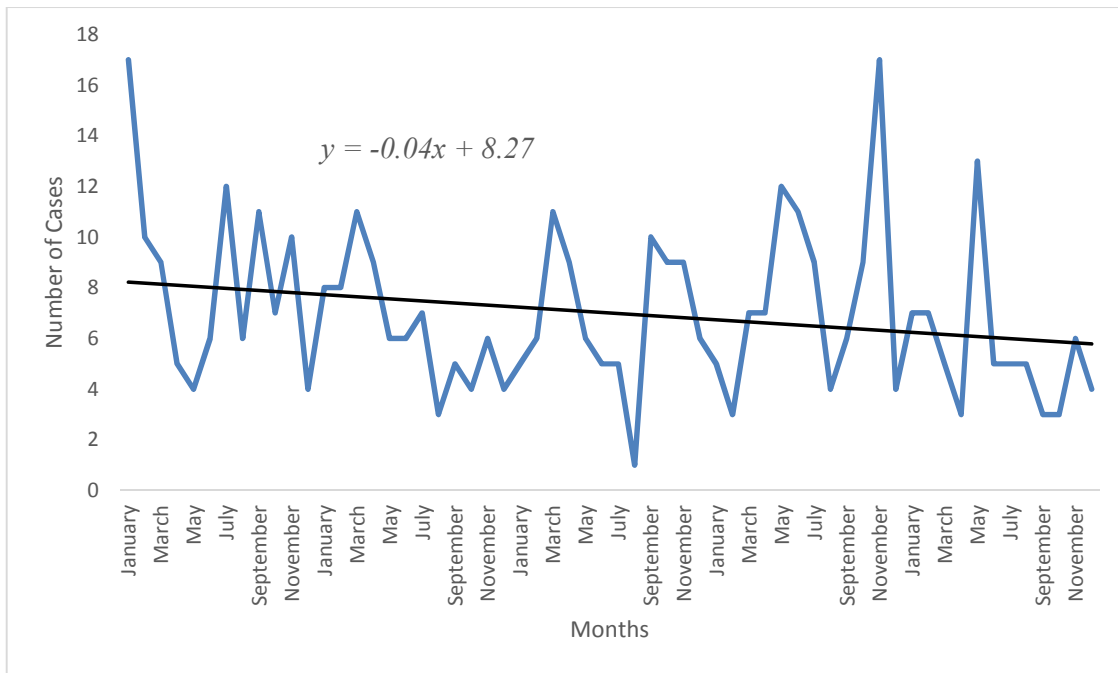


Fig. 4. Overall trend line for the cases recorded

**Table 2. Regression estimate of trend line**

Regression parameters	Coefficients	Standard error	P-value	Lower 95%	Upper 95%
Intercept	8.27	0.84	<0.001	6.58	9.96
X variable (time)	-0.04	0.02	0.090	-0.09	0.01

The influence that the gender of a baby could have on the number of feet that could suffer this deformity did not yield a significant association. Thus being born a male or female has no bearing on whether one or both feet will suffer the deformity. Having idiopathic clubfoot based on gender, and or suffering bilaterally or unilaterally with respect to the climatic season established no significant associations when the chi square test was used. Thus, external climatic conditions may have very little or no influence on the occurrences and distribution of idiopathic clubfoot. Over the five-year-period for which the study considered, there was a statistical decrease in the prevalence of idiopathic clubfoot as indicated by the slope of the trend line ( $r = -0.04$ ). However, the slope parameter is not significant at 5% significance level ( $P = 0.09$ ). Therefore, despite the decrease in the prevalence over the years, the decrease in itself is not statistically significant. Potentially, with the training of more physiotherapists and establishment of newer physiotherapy centers in both private and public health facilities in Ghana, the rate of referrals to the major clubfoot management point in Ghana, KATH, could have gone through gradual reduction resulting in the decreasing trend of the prevalence of idiopathic clubfoot in KATH.

Despite the available evidence of the combination of both environmental and genetic factors being linked to the cause of clubfoot [3,20,22], it is important to note that constituents of the environmental factors, per the findings of this study, depend not on external climatic conditions, but on factors such as maternal smoking [23], amniocentesis [24], and or any other lifestyle or medical procedure that may influence the occurrence.

#### 4. CONCLUSION

Idiopathic clubfoot, as part of the general types of clubfoot pose socioeconomic burden on the society. Understanding the intricacies of its aetiology is very key in the medical perspective, socioeconomic and political viewpoint at large. The climatic seasons of a locality cannot single handedly explain the cause of the deformity, thus requiring further studies on the concerted

influence from other possible causes. Evidence from genetics, maternal smoking, amniocentesis trials among others discussed in this study could provide firmer grounds for further researches aimed at understanding the aetiology of idiopathic clubfoot.

#### CONSENT

It is not applicable.

#### COMPETING INTERESTS

Author has declared that no competing interests exist.

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