

Epidemiology of Congenital Metatarsus Adductus: Experience from a Tertiary Health Institution

Phillip D. Eyimina, Richard C. Echem*

Department of Orthopaedic Surgery, University of Port Harcourt Teaching Hospital, Port Harcourt,
Post code - 500001, Rivers State, Nigeria

*Corresponding author: richy1870@yahoo.co.uk

Received March 11, 2020; Revised April 15, 2020; Accepted April 30, 2020

Abstract Background: Congenital metatarsus adductus is a common foot deformity in children. It involves adduction of the metatarsals at the LisFranc's joint. It can be primary (idiopathic) or secondary. **Aim:** To describe the epidemiology of idiopathic congenital metatarsus adductus as seen in a tertiary health institution. **Methods:** A prospective study of consecutive patients presenting with congenital metatarsus adductus in children at the University of Port Harcourt Teaching Hospital, Port Harcourt from January 2010 to December 2017. Diagnoses were made clinically. Data was obtained through a structured proforma and analysed using SPSS version 20. **Results:** Fifty children with congenital metatarsus adductus with a total of 82 feet were seen. Ages ranged from 2 weeks to 10 years. There were 27 females and 23 males. They were mostly first and second born in their families. Majority were from monogamous homes. All were Nigerians and mostly of Igbo tribe. No use of alcohol or tobacco during pregnancy. There was no consanguinity. The maternal age ranged from 24 to 36 years and paternal age from 30 to 45 years. Gestational age ranged from 34 to 40 weeks. Majority were singleton. Majority were born in public hospitals and by spontaneous vaginal delivery. Most were bilateral. Common Bleck's severity grades were mild and moderate. Most frequent flexibility grade was the partially flexible. **Conclusion:** Idiopathic congenital metatarsus adductus is commonly diagnosed within the first few months of life and commonly occurs in females and first-born. Most are either mild or moderate in severity and most frequently partially flexible.

Keywords: congenital, metatarsus adductus, epidemiology, idiopathic

Cite This Article: Phillip D. Eyimina, and Richard C. Echem, "Epidemiology of Congenital Metatarsus Adductus: Experience from a Tertiary Health Institution." *American Journal of Medical Sciences and Medicine*, vol. 8, no. 2 (2020): 54-61. doi: 10.12691/ajmsm-8-2-3.

1. Introduction

Congenital metatarsus adductus is a congenital deformity of the foot in which there is forefoot adduction relative to the hindfoot. [1,2,3,4,5] Strictly applied, this is a transverse plane deformity in which the metatarsals are medially deviated with the apex at the tarsometatarsal (LisFranc's) joints. [4,6-13] The term, metatarsus adductus, is utilized to describe forefoot adduction occurring alone or associated with supination [14-19] and hindfoot that is usually neutral but could be valgus. [1,5,6,9,10,14,17,18,19,20,] However, a varus hindfoot is possible. [10,17] The equinus deformity of the ankle that characterizes congenital clubfoot is never observed in metatarsus adductus. [17,20]

In the literature, various terminologies have been utilized to describe forefoot adduction deformities. These terms include metatarsus adductus, [1,6,9,12,14,17,18,20,21-29] metatarsus varus, [1,6,9,14,17-25,27,36] metatarsus adductus et supinatus, [16] metatarsus adductovarus, [1,20,22,24,27,28] metatarsus adductus varus, [37] metatarsus supinatus, [6,26] pes varus, [27] pes adductus, [9,20,22,24]

metatarsus internus, [1,27,28] forefoot adductus, [6] hooked forefoot, [27,28,29,38] serpentine foot, [1,28,34,37] skewfoot, [14,18,22,28,29] and z-foot. [1,28,29] Kite had also described it as "third of a club foot". [17,18] These terms that had been used have been confusing [1,28] and it had been stated that these terms are not synonymous and are used inconsistently by many [28]. Most orthopaedists prefer the term 'metatarsus adductus'. [27] The diversity of terms suggests that the most commonly used one, metatarsus adductus, may actually involve several deformities. [28] This necessitated Berg [28] to devise a radiographic classification that differentiates and acknowledges this spectrum of nomenclature. Ponseti and Becker [20] had highlighted that from their clinical experience which also confirms the viewpoint of others, [22] the two most commonly used terms, metatarsus adductus and metatarsus varus, are different degrees of the same clinical entity which some authors have called pes adductus or metatarsus adductovarus.

Metatarsus adductus is a static or dynamic medial deviation deformity of the forefoot. [29] It has been described as a deformity that is postural, [5] positional [6,7,26] or structural. [7,15,26,39] It can be primary

(isolated; idiopathic) or secondary to deformities elsewhere in the foot such as a component or sequelae of talipes equinovarus. [3,4,8,40,41,42]

The first mention of this forefoot adduction deformity was by Henke in 1863. [17,22,33,37,43,44] This was forefoot adduction with valgus heel. [22] A similar description was also given by Hueter in 1870. [22] The first detailed description and analysis of this deformity was by Cramer in 1904. [21,22,23] This was an adult case and he included photographs and radiographs. [22] He described a similar case in an infant in 1909. [22] He was the first to consider the possibility of it being of congenital aetiology. [21] The entity had been recognized in the non-English literature in the later part of the 19th and early 20th century even though it was thought to be rare. Clinicians in France, Germany and Italy were describing the entity as distinct from clubfoot. [21,35] However, Engel in 1924 suggested that the entity may not be as rare as it was assumed. [36] In Germany, it is stated that the condition was prevalent at that time. [22] In the English literature, very little was written about it in the early 20th century. The orthopaedic text books of the period either failed to describe the entity or only gave a passing reference. Whitman's text mentions it as an incomplete form of clubfoot or simple talipes varus. [30] The first reference in the English literature was by Bankart in 1921. [21,30,38] He gave a description of the condition highlighting that clinicians were familiar with the deformity and that it was a different entity from clubfoot. After this, many more publications in the English literature [2-44] have appeared describing the entity and its management.

Congenital metatarsus adductus can be recognized at birth but sometimes it is not recognized until the child is older. [17,20,38] The deformity can be evaluated clinically and/or radiologically. The chief complaints include abnormal curvature of the foot, intoeing gait with stumbling or tripping, painful foot, difficulty in fitting of shoes, excessive shoe wear and cosmesis. [15,44] The physical findings include c-shaped foot with concave medial border and convex lateral border of the foot, [2,9,10,11,15,18,20,23,25,38,44-47] forefoot adduction and supination, [1,2,5,9,14,16-20,23,25,33,36,43,45,46] higher longitudinal arch than normal, [9,14,17,18,45] prominence of the base of the fifth metatarsal laterally, [2,9,10,11,17,18,20,33,44-47] medial deviation of the great toe, [14,25,30,45] increased space between the great toe and the second toe (splay between the hallux and the second toe), [9,11,18,33,45] holding the heel (posterior part of the foot) and abducting the forepart of the foot shows more resistance than normal, [17,18] observation of hyperactivity or hypertrophy of the abductor hallucis, [14,15,44] transverse crease through the medial arch, [14,25,45] and hindfoot that is neutral or in valgus [5,9,10,14,16-20,22,23,25,33,36,38,43,44,45] although rarely, varus hindfoot had been observed. [10,17] Equinus deformity is never seen. [9,17,36] Internal tibial torsion could be observed at times. [9,20,25,43] The heel bisector line is used to grade the severity and flexibility of the deformity. [5,11,12,27] The radiologic evaluation of metatarsus adductus deformity has been considered important by some authors. [9,10,15,23,28,39,42,44,46,48] Radiographs provide valuable assessment for this

deformity as a diagnostic and prognosticating tool. [46,48] However, some others consider it not imperative to evaluate or prognosticate metatarsus adductus deformity. [13,42,44] There are numerous methods described for the radiographic assessment of metatarsus adductus in the fully ossified skeleton. [42,44] But in the infant or toddler foot, the measurements are difficult to recreate with accuracy because of lack of ossification of some of the bones of the foot. [42,44]

The aim of this study is to describe the epidemiology of idiopathic congenital metatarsus adductus as seen in a tertiary health institution in the Niger Delta region of the south-south geopolitical zone of Nigeria.

2. Materials and Methods

This was a prospective study of consecutive patients aged ≤ 16 years presenting to the Orthopaedic outpatient clinic of the University of Port Harcourt Teaching Hospital, Port Harcourt, Rivers State, Nigeria with congenital metatarsus adductus. The study was carried out from January 2010 to December 2017.

The diagnoses were made clinically. Radiographs were not utilized. Clinical photographs of the children were taken. The degree of severity was graded by the Bleck's heel bisector method. This is carried out by visual examination of the plantar aspect of the foot. The heel bisector line is a longitudinal line that divides the heel pad into equal parts and when extended distally, its relationship to the toes and interspaces is noted. If the line runs through the second toe or second interspace, it is considered normal. In mild deformity, the heel bisector passes through the third toe; in moderate deformity, it passes between the third and fourth toe or through the fourth toe and in severe deformity it passes between the fourth and fifth toes. [5,11,12,27] The flexibility of the foot was classified according to the extent of passive abduction of the forefoot against the stabilized hindfoot with reference to the heel bisector line. A flexible forefoot is one which could be abducted beyond the midline heel bisector; a partially flexible forefoot is one which could be abducted to the midline heel bisector and a rigid forefoot is one which could not be abducted to the midline heel bisector. [2,5,11,12,27,44]

Included were the children aged ≤ 16 years who had idiopathic congenital metatarsus adductus. The exclusion criteria were those who had neuromuscular disease, bone dysplasias, congenital clubfoot or other foot malformations.

Data was collected through a structured proforma and included the patients age, sex, position in the family (birth order), type of family, nationality, tribe/ethnicity, any history of alcohol consumption and tobacco use during pregnancy, any consanguinity, maternal age at birth of child, paternal age at birth of child, gestational age of child at birth, number of foetuses (singleton/twin), place of birth, any complication at birth, mode of delivery, family history of clubfoot, diagnosis/laterality, Bleck's severity and flexibility grading for the feet, and associated conditions.

Data was analysed using Statistical Package for Social Sciences (SPSS) version 20 (IBM Inc., Armonk, NY,

USA). Mean, standard deviation and median where applicable were used for descriptive statistics while categorical variables were expressed in absolute frequencies. Chi - square test was used to determine association between some key variables. A p - value of ≤ 0.05 was considered statistically significant.

Ethical approval was obtained from the Research and Ethics Committee of the University of Port Harcourt Teaching Hospital.

3. Results

During the period, 50 patients having 82 feet with idiopathic congenital metatarsus adductus were seen. This constituted 5.2% of children aged ≤ 16 years that attended the orthopaedic outpatient clinic and 1.1% of all attendance to the orthopaedic clinic. Their ages at presentation ranged from 2 weeks to 10 years. The median age for the patients at presentation was 8.50 months. The most frequent ages at presentation were those less than four months (32.0%) and those greater than 12 months (30.0%) (Table 1). There were 27 females and 23 males giving a female to male ratio of 1.17: 1. The most frequent birth orders of the patients were first (46.0%) and second (38.0%) born. Almost all the patients (98.0%) came from monogamous families. All the patients were Nigerians and the most frequent tribe was Igbo (70.0%) (Table 1). The tribe/ethnicity group labelled "others" consisted of one patient each from Hausa, Ekpeye, Ibibio, Isoko and Egbema.

Table 1. Sociodemographic characteristics of the study patients

Variables	Frequency (n=50)	Percentage (%)
Age at Presentation		
<4 months	16	32.0
4-6 months	7	14.0
7-9 months	4	8.0
10-12 months	8	16.0
>12 months	15	30.0
Mean (\pmSD), Median	14.40 (\pm22.05), 8.50	
Female	16.41 (\pm29.14), 4.75	
Male	12.04 (\pm8.28), 11.40	
Range	2weeks - 10years	
Sex		
Female	27	54.0
Male	23	46.0
Position in Family(Birth order)		
1 st	23	46.0
2 nd	19	38.0
3 rd	4	8.0
4 th	4	8.0
Family Type		
Monogamy	49	98.0
Polygamy	1	2.0
Nationality		
Nigerian	50	100.0
Tribe/Ethnicity		
Igbo	35	70.0
Ijaw	3	6.0
Ogoni	3	6.0
Ogba	2	4.0
Yoruba	2	4.0
Others	5	10.0

None of the mothers admitted to taking alcohol or using tobacco during the pregnancy. There was no case of consanguineous marriage.

Table 2. Maternal, paternal and delivery characteristics of the study patients

Variables	Frequency (n=50)	Percentage (%)
Maternal Age at Birth		
24-26years	8	16.0
27-29years	15	30.0
30-32years	20	40.0
>32years	7	14.0
Mean (\pmSD), Median	29.76 (\pm2.94), 30.00	
Range	24 - 36years	
Paternal Age at Birth		
30-34years	11	22.0
35-39years	28	56.0
40-44years	10	20.0
45-49years	1	2.0
Mean (\pmSD), Median	36.72 (\pm3.23), 36.50	
Range	30 - 45years	
Gestation Age at birth		
≤ 36 weeks	3	6.0
38weeks	18	36.0
≥ 39 weeks	29	58.0
Mean (\pmSD), Median	38.98 (\pm1.35), 40.00	
Range	34 - 40weeks	
Number of foetuses		
Singleton	49	98.0
Twin	1	2.0
Place of birth		
Public Hospitals	39	78.0
Private Clinics	9	18.0
Home	1	2.0
Maternity Home	1	2.0
Complication at Birth		
Yes	0	0.0
No	50	100.0
Mode of Delivery		
SVD	43	86.0
Emergency C/S	5	10.0
Elective C/S	2	4.0
Indications for Emergency C/S (n=5)		
Foetal Distress	4	80.0
Breech Presentation	1	20.0
Indications for Elective C/S (n=2)		
Previous C/S	2	100.0

Key: SD = Standard deviation, SVD = Spontaneous vaginal delivery, C/S = Caesarean section.

The age at which the mothers gave birth ranged from 24 to 36 years with a mean of 29.76 ± 2.94 years. The most common maternal age bracket at delivery was 30-32 years (40.0%). The paternal ages at birth of the children ranged from 30 to 45 years with a mean of 36.72 ± 3.23 years. The most frequent paternal age bracket at birth of the children was 35-39 years (56.0%) (Table 2). The difference in the mean age of the parents was 6.98 years. The gestational age at the birth of the children ranged from 34 to 40 weeks. Most were born at gestational age ≥ 39 weeks (58.0%).

There were three preterms (6.0%). One was born at 34 weeks and two at 36 weeks. The average for the three preterms was 35.3 weeks. There was only one twin gestation, with the rest being singleton. The twin was an identical twin and the second twin did not have any congenital foot deformity. Majority of the children were born in public hospitals (78.0%). The parents did not admit to any complications at birth for the children or their mothers. Majority of the children were delivered by spontaneous vaginal delivery with vertex presentation (86.0%). Caesarean sections were performed for mostly emergencies and foetal distress was the indication in the majority (Table 2).

Only one patient (2.0%) gave a family history of clubfoot in two first degree relatives (Table 3). None gave any history suggestive of metatarsus adductus. Most of the children (64.0%) had bilateral metatarsus adductus. In the unilateral, the right and left were equally affected. The most frequent Bleck's severity grading was moderate (50.0%). The most frequent flexibility grade was the partially flexible (50.0%). The most common associated conditions were congenital talipes equinovarus (put together 10.0%), internal tibial torsion (6.0%) and congenital hydrocele (6.0%) (Table 3). There was no case of developmental dysplasia of the hip demonstrated clinically.

Table 4 shows the association between Bleck's severity grading and the age and sex of the patients. There was a significant association between the severity and the age and sex of the children.

Table 5 shows the association between Bleck's flexibility grading and the age and sex of the patients.

There was a significant association between the flexibility and the age and sex of the children.

Figure 1 is a clinical photograph of a child with bilateral metatarsus adductus.

Table 3. Clinical characteristics of study patients

Variables	Frequency (n=50)	Percentage (%)
Family history of clubfoot		
Yes	1	2.0
No	49	98.0
Diagnosis/Laterality		
Left metatarsus adductus	9	18.0
Right metatarsus adductus	9	18.0
Bilateral metatarsus adductus	32	64.0
Patients Bleck's severity grading for the feet (n=82)		
Mild	35	42.7
Moderate	41	50.0
Severe	6	7.3
Patients Bleck's flexibility grading for the feet (n=82)		
Flexible	35	42.7
Partially flexible	41	50.0
Rigid	6	7.3
Associated conditions		
Internal tibial torsion	3	6.0
Right congenital talipes equinovarus	3	6.0
Left congenital talipes equinovarus	2	4.0
Congenital hydrocoele	3	6.0
Umbilical hernia	1	2.0
None	38	76.0

Table 4. Association between Bleck's Severity Grading and some Sociodemographic Characteristics

Variables	Bleck's Severity Grading			χ^2	p-value
	Mild	Moderate	Severe		
Age of presentation					
<4 months	21 (80.8)	3 (11.5)	2 (7.7)	39.942	<0.001
4-6 months	7 (63.6)	2 (18.2)	2 (18.2)		
7-9 months	2 (28.6)	5 (71.4)	0 (0.0)		
10-12 months	3 (21.4)	11 (78.6)	0 (0.0)		
>12 months	2 (8.3)	20 (83.3)	2 (8.3)		
Sex					
Female	25 (61.0)	14 (34.1)	2 (4.9)	11.217	0.004
Male	10 (24.4)	27 (65.9)	4 (9.8)		

Table 5. Association between Bleck's Flexibility Grading and some Sociodemographic Characteristics

Variables	Bleck's Flexibility Grading			χ^2	p-value
	Flexible	Partially Flexible	Rigid		
Age of presentation					
<4 months	21 (80.8)	3 (11.5)	2 (7.7)	39.942	<0.001
4-6 months	7 (63.6)	2 (18.2)	2 (18.2)		
7-9 months	2 (28.6)	5 (71.4)	0 (0.0)		
10-12 months	3 (21.4)	11 (78.6)	0 (0.0)		
>12 months	2 (8.3)	20 (83.3)	2 (8.3)		
Sex					
Female	25 (61.0)	14 (34.1)	2 (4.9)	11.217	0.004
Male	10 (24.4)	27 (65.9)	4 (9.8)		



Figure 1. Clinical photograph of a child with bilateral congenital metatarsus adductus

4. Discussion

This study has shown that children with congenital metatarsus adductus present to the hospital within the first months of life, females are most commonly affected, and the children are mostly first born with their mothers being in their third and fourth decades while their fathers are mostly in their fourth decades of life. Majority of the children were born at term, were singleton and born mainly in public hospitals by spontaneous vaginal delivery. The deformity was mostly bilateral, moderate in severity and partially flexible.

Idiopathic congenital metatarsus adductus constituted 5.2% of children aged ≤ 16 years attending the hospitals orthopaedic outpatient clinic and 1.1% of all the orthopaedic clinic attendance. This is higher than the 0.6% of the total patients treated or examined in an orthopaedic hospital in Denmark reported by Petersen [36] more than 75 years ago. From the period when the deformity was first recognized and thought to be rare, there has been an increase in the frequency of occurrence of this condition [17,20,21,22,23,25,30,31,35]. Ponseti and Becker [20] had highlighted some of the reasons for the increase which include the fact that medical consultation became more popular and available and the medical profession was better instructed in the recognition of even the mildest forms of foot deformities in infants. Wynne-Davies [49] had also noted an increase in hospital attendance after the start of the National Health Service in United Kingdom.

Children with these deformities may not be referred to the hospital early and the condition may be overlooked or ignored at birth, considered normal with perinatal paediatric assessment or the deformity does not manifest until later. [49,50,51] It has been regarded as a common foot deformity in the newborn by some authors [5,12,31,40,45,52] while others regard it as the most common foot deformity in the newborn [1,2,13,25,44,46]. There is diverging information in the literature relating to the incidence of congenital metatarsus adductus. [53]

Incidence from as low as 0.02% [54] to 13% in term infants have been reported. [1,2,6,14,15,44,46-53]. Higher rates of up to 25% have been reported in preterm [53,55] although a lower figure of 0.2% had been reported by others [56] in this group of patients. The incidence is higher in twin/multiple births. [44,48,53]

In the present study, the most frequent age at presentation is within the first three months of life (32.0%). In the series by Kite, [17] the condition was recognised at birth in 31.3% while Ponseti and Becker [20] reported two-thirds of their patients were recognized with the deformity at birth. Rushforth [38] reported 30% of their series presented at birth. In the series by Martos-Mora et al [1] those who presented within one month made up 31% while those presenting within 3 months constituted almost 75%. Chong [5] in their series had almost 60% of their patients being within the first three months of life. Those within four months in Bleck's series [27] made up 43% while almost 72% were seen within eight months of life. In the study by Fedele et al, [57] 35% of the diagnosis of this deformity was made in the first month with 86% of their patients being seen within the first eight months of life.

Different authors have reported varying sex distribution. While some report equal or almost equal proportion, [1,16,21,46] some report a male preponderance [5,6,12,17,18,20,27,28,33,36,38,43,44,52,53,57,58] while others report a female preponderance. [14,15,23,49,54] In the present study, there was a female preponderance. It is worthwhile highlighting that overall, there are more series with male preponderance than female preponderance.

First born children (46%) were most frequent in the present study followed by those who were second born (38%). In the series by Rushforth [38], half of their patients were second born while Berg [28] reported 59% of the patients as first born children. The studies by Wayne-Davies [49] and Wynne-Davies et al [54] did not have any evidence to show any connection between the deformity and birth order. However, the study by Wynne-Davies [49] had more of those in the study as first and second born with the second born being more. It has been suggested that compression could be a causative factor for conditions in which there were more first born due to the tight uterus and good abdominal muscles of the primigravida. [49]

Almost all the patients in the present study were from monogamous families. Monogamy is a common form of marriage in Nigeria. [59] All the patients in the present study were Nigerians and they were majorly of the Igbo tribe (70%). The study by Rushforth [38], though English also included eight West Indians and one Asian. The study by Hunziker et al [53] included newborns of Swiss origin only.

Alcohol consumption by mothers during pregnancy has been implicated in the development of congenital anomalies. [60,61] Although there has been an association of this with congenital talipes equinovarus, [60,61] there has so far been no study on this in relation to congenital metatarsus adductus. In the present study, none of the mothers admitted to taking alcohol during pregnancy.

Maternal smoking during pregnancy has also been associated with congenital anomalies of the foot. This has been more studied with congenital talipes equinovarus. [60,62,63] Kite [18] in his series, indicated that only 20% of mothers of the patients with congenital metatarsus

adductus admitted to smoking and therefore felt that this was not a causative factor for the deformity. In the present study, none of the mothers of the patients in the series admitted to using tobacco during pregnancy.

In the present study, there were no consanguineous marriages found among the parents of the patients. Studies by Wynne-Davies [49] and Wynne-Davies et al [54] did not reveal any consanguineous marriages in the two studies.

The mean maternal age at birth of the children in the present study was 29.76 ± 2.94 years with a range of 24 years to 36 years. In Rushforth's study [38] the maternal age at birth of the children ranged from 15 years to 36 years with a mean of 27 years. The average maternal age in the study by Wynne-Davies [49] was 26.9 years. These mean maternal ages are comparable. No evidence of any connection between maternal age at birth and congenital metatarsus adductus has been demonstrated. [49,54]

The average paternal age at birth of the children in the study by Wynne-Davies [49] was 29.4 years. The mean age of the fathers in the present study was 36.72 ± 3.23 years, which is higher than that of Wynne-Davies. [49] This may be because the fathers in our environment marry later. The difference in the parental age of 2.5 years reported by Wynne-Davies [49] is lower than that of the present study (6.98 years). The difference in parental age in the Wynne-Davies [49] study was not markedly different from that expected for normal families in United Kingdom at the time of the study. No evidence of any connection between paternal age at birth of the children and congenital metatarsus adductus has been demonstrated. [49,54].

Majority of the children in the present study were born at term (94%) with a mean of 38.98 ± 1.35 weeks. There was none that was postdate and there were 6% preterm children. However, Rushforth [38] reported that nearly half of the patients were born at term with equal proportion of premature and post mature deliveries. Ponseti and Becker [20] highlighted that all the patients in their series were products of normal pregnancies. Martos-Mora et al [1] reported six preterms in their series of 87 patients. In the study by Hunziker et al [53] the mean for both the male and female term patients was 40.1 weeks. Our figures are comparable to this. The mean for the male preterm was 33.7 weeks while that for the female preterm was 33.1 weeks. This is lower than the average of 35.3 weeks for our three preterm. However, this difference needs to be interpreted with caution as our preterm numbers are few.

It had been reported that congenital metatarsus adductus is higher among twins and multiple births. [48,53] Martos-Mora et al [1] had one twin in their series of 87 patients while Ponseti and Becker [20] had two twins in their series of 57 patients. Wynne-Davies et al [54] had two dizygotic twins in their series of 10 metatarsus adductus patients. These were discordant. They did not report any instances of monozygotic twins. In the present study, there was only one twin in the series of 50 patients and they were identical twins. The twin of the patient in the present study did not have any congenital foot deformity. It has been suggested that twin pregnancy might contribute to the deformity through intrauterine constraint. [53,54]

The majority of the patients in the present study were born in public hospitals followed by private clinics. This corroborates a study in North Eastern Nigeria in which

most of the mothers who had antenatal care in a public teaching hospital delivered in the public hospital, [64] although a study in South Western Nigeria [65] and another in South Eastern Nigeria [66] revealed women who preferred delivery in private hospitals.

In the present study, the parents did not admit to any complications at birth for the children or their mothers. However, foetal distress was indication for caesarean section in four patients. Hunziker et al [53] highlighted that in term infants on their study, pregnancy and labour were free of complications, but that for the preterm infants they are high risk for perinatal complications such as foetal distress, infections, metabolic problems and respiratory difficulties. Martos-Mora et al [1] also reported perinatal complications such as foetal distress, respiratory distress, neonatal jaundice, etc. and also reported gestational diabetes as well as arterial hypertension amongst other complications in the mothers.

Ponseti and Becker [20] reported that all their series were products of normal deliveries. For the term infants, Hunziker et al [53] reported spontaneous vaginal deliveries with vertex presentation. Martos-Mora et al [1] in their series had 12 caesarean deliveries. In the present study, majority of the deliveries were by spontaneous vaginal delivery with vertex presentation and there were seven caesarean sections with five being emergencies.

Heredity has been associated with occurrence of congenital metatarsus adductus. [6,15] Wynne-Davies [49] reported 4.4% of siblings (first degree relatives) being affected. It was further stated that in a family, if a sibling is affected, the risk for a second child having the same deformity is one in 20. Several authors have highlighted the familial disposition to congenital metatarsus adductus in their series. [1,17,18,20,36,38,54,57] Several authors have also reported a family history of congenital talipes equinovarus among patients with congenital metatarsus adductus. [1,38,49] In the present study, in only one patient was there a family history of clubfoot in two first degree relatives but none gave a family history of congenital metatarsus adductus.

In the literature, majority of the congenital metatarsus adductus were bilateral. This has been reported by several workers with rates ranging from 51% to 80%. [1,5,12,15-18,20,21,23,27,33,36,46,52-54,57,58] The series by Farsetti et al [43] and Hunziker et al [53] among the preterms reported more unilateral cases than bilateral. The unilateral cases more commonly affect the left foot [1,5,16,17,18,43,46,54,57,58]. However, Ponseti and Becker [20] reported equal occurrence for both feet while Peterson [36] and Eamsobhana et al [52] reported preponderance of the right foot. In the present study, more of the patients had bilateral involvement and the right and left feet were equally affected.

The present study utilized the Bleck's severity grading for the patients and the most common was moderate severity in half of the patients. Studies that utilized the Bleck's severity grading commonly reported the mild grade as the most common with the severest grade being the least. [12,52] The present study also had the severest grade as the least. However, Bleck [27] in the patients treated reported the mild grade as 12% and the moderate and severe grades constituting 44% each. In the present study, there was a significant association between the

severity grade and the age and sex of the children. Rushforth's series [38] which utilized visual grading using photographs of patients in standing position, reported the moderate grade as the most common (52%) with the severe grade being the least (20%). Martos-Mora et al [1] using their grading system based on the presence of adduction, supination, presence of transverse grooves and extent of correction of the deformity reported their mildest grade (grade 1) as the most common with grade 3 (severe) being the least. Also, Fedele et al [57] utilizing a grading system based on what they described as angle of adduction, the axes of the forefoot relative to the hindfoot measured on the sole and the degree of passive and active reducibility, reported their mildest group (Type I) as the most common and the severest (Type III) as the least.

In terms of flexibility of congenital metatarsus adductus the present study had the partially flexible constituting half of the feet and the least being the rigid variety (7.3%). Bleck [27] had the partially flexible group as the most common (65%) while the rigid group is the least. However, Bohne [12] using the heel bisector as reference classified his patients into flexible (88%) and rigid (12%). The flexible and partially flexible were lumped together in the classification. In all the series, the rigid variety is the least occurring. In the present series, there was a significant association between the flexibility of the deformity and the age and sex of the children.

Congenital metatarsus adductus can be found in association with other congenital anomalies. Various congenital anomalies have been reported [1,17,20,28,33,36,38,42,49,50,54,57,58,67]. The commonly reported are internal tibial torsion, [1,20,28,42,57] congenital talipes equinovarus, [17,33,36,57] developmental dysplasia of the hip, [1,28,36,38,42,51,57,58,67] and congenital talipes calcaneovalgus [17,33]. In the present study, the common associated anomalies were congenital talipes equinovarus, internal tibial torsion and congenital hydrocele.

The present study has a number of limitations. Only clinical assessment was utilized for the study. Radiographs would have also provided valuable assessment. However, radiographs would have exposed the children to radiation and also the lack of ossification of some of the bones of the foot limits its application. [42,44,52]. This was a hospital-based study and may not give a complete picture of what obtains in the general population. Also, the responses obtained in the proforma were from the parents mostly and there could be recall bias.

5. Conclusion

Congenital metatarsus adductus is commonly diagnosed within the first few months of life and occurs commonly in females and first born. Majority were from monogamous families. They were all Nigerians and most frequently of Igbo tribe. There was no admittance of alcohol or tobacco use during pregnancy. The parents were mostly in their third and fourth decades of life at the time of the birth of the children. Majority were born at term and were singleton. Majority were born in public hospitals and by spontaneous vaginal delivery with vertex presentation. There was no family history of congenital

metatarsus adductus. Most of the deformities were bilateral. The common severity grades were mild and moderate while the most frequent flexibility grade of the feet was the partially flexible. The commonly associated conditions were congenital talipes equinovarus, internal tibial torsion and congenital hydrocele.

Statement of Competing Interest

The authors have no competing interest

Financial Support Statement

The study was self-funded.

Acknowledgements

The authors acknowledge the assistance of Viome Showers in the data analysis. The authors also acknowledge Dr. U. Ajoku for assistance with some of the fulltext articles used for the study.

References

- [1] Martos-Mora C, Gentil-Fernandez J, Conejero-Casares JA, Ramos-Moreno R. [Congenital metatarsus adductus: a clinical classification and therapeutic attitude]. *Rehabilitacion(Madr)* 2012; 46(2): 127-134.
- [2] Williams CM, James AM, Tran T. Metatarsus adductus: development of a non-surgical treatment pathway. *J Paediatr Child Health* 2013; 49(9): E428-E433.
- [3] Feng L, Sussman M. Combined medial cuneiform osteotomy and multiple metatarsal osteotomies for correction of persistent metatarsus adductus in children. *J Pediatr Orthop* 2016; 36(7): 730-735.
- [4] Knorr J, Accadbled F, Jegu J, Abid A, de Gauzy JS, Cahuzac JP. Behavior of the first cuneiform in the surgical correction of metatarsus adductus. *Rev Esp Cir Ortop Traumatol* 2008; 52(2): 84-93.
- [5] Chong A. A new device for the treatment of metatarsus adductus. *J Prosthet Orthot* 1990; 2(2): 139-148.
- [6] Nadji H, Mourbes D, Makhour F, Dimassi A, Jawish R. Forefoot adduction in children. Management and treatment. *J Med Liban* 2016; 64(3): 134-141.
- [7] Yu GV, Johng B, Freireich R. Surgical management of metatarsus adductus deformity. *Clin Podiatr Med Surg* 1987; 4(1): 207-232.
- [8] Cahuzac JP, Laville JM, de Gauzy JS, Lebarbier P. Surgical correction of metatarsus adductus. *J Pediatr Orthop B* 1993; 2(2): 176-181.
- [9] Galluzzo AJ, Hugar DW. Congenital metatarsus adductus: clinical evaluation and treatment. *J Foot Surg* 1979; 18(1): 16-22.
- [10] Harley BD, Fritzhand AJ, Little JM, Little ER, Nunan PJ. Abductory midfoot osteotomy procedure for metatarsus adductus. *J Foot Ankle Surg* 1995; 34(2): 153-162.
- [11] Connors JF, Wernick E, Lowy LJ, Falcone J, Volpe RG. Guidelines for evaluation and management of five common podopediatric conditions. *J Am Podiatr Med Assoc* 1998; 88(5): 206-222.
- [12] Bohne W. Metatarsus adductus. *Bull N Y Acad Med* 1987; 63(9): 835-838.
- [13] Marshall N, Ward E, Williams CM. The identification and appraisal of assessment tools used to evaluate metatarsus adductus: a systematic review of their measurement properties. *J Foot Ankle Res* 2018; 11: 25.
- [14] Bost FW. Pediatrics - epitomes of progress: Treatment of metatarsus adductus in infants. *West J Med* 1982; 136(4): 327-328.

- [15] Theodorou DJ, Theodorou SJ, Boutin RD, Chung C, Fliszar E, Kakitsubata Y, Resnick D. Stress fractures of the lateral metatarsal bones in metatarsus adductus foot deformity: a previously unrecognized association. *Skeletal Radiol* 1999; 28(12): 679-684.
- [16] Ghali NN, Abberton MJ, Silk FF. The management of metatarsus adductus et supinatus. *J Bone Joint Surg Br* 1984; 66(3): 376-380.
- [17] Kite JH. Congenital metatarsus varus. Report of 300 cases. *J Bone Joint Surg Am* 1950; 32(3): 500-506.
- [18] Kite JH. Congenital metatarsus varus. *J Bone Joint Surg Am* 1967; 49(2): 388-397.
- [19] Reimann I, Werner HH. The pathology of congenital metatarsus varus. A post-mortem study of a newborn infant. *Acta Orthop Scand* 1983; 54(6): 847-849.
- [20] Ponseti IV, Becker JR. Congenital metatarsus adductus: the results of treatment. *J Bone Joint Surg Am* 1966; 48(4): 702-711.
- [21] Peabody CW, Muro F. Congenital metatarsus varus. *J Bone Joint Surg Am* 1933; 15(1): 171-189.
- [22] McCormick DW, Blount WP. Metatarsus adducto-varus: skewfoot. *J Am Med Assoc* 1949; 141(7): 449-453.
- [23] Sirbu AB, Colloff B. Early treatment of metatarsus varus (adductus). *Pediatrics* 1949; 4(6): 810-819.
- [24] Heyman CH, Herndon CH, Strong JM. Mobilization of the tarsometatarsal and intermetatarsal joints for the correction of resistant adduction of the fore part of the foot in congenital club-foot or congenital metatarsus varus. *J Bone Joint Surg Am* 1958; 40(2): 299-310.
- [25] McCauley J Jr, Lusskin R, Bromley J. Recurrence in congenital metatarsus varus. *J Bone Joint Surg Am* 1964; 46(3): 525-532.
- [26] Rothbart BA. Metatarsus adductus and its clinical significance. *J Am Podiatry Assoc* 1972; 62(5): 187-190.
- [27] Bleck EE. Metatarsus adductus: classification and relationship to outcomes of treatment. *J Pediatr Orthop* 1983; 3(1): 2-9.
- [28] Berg EE. A reappraisal of metatarsus adductus and skewfoot. *J Bone Joint Surg Am* 1986; 68(8): 1185-1196.
- [29] Asirvatham R, Stevens PM. Idiopathic forefoot-adduction deformity: medial capsulotomy and abductor hallucis lengthening for resistant and severe deformities. *J Pediatr Orthop* 1997; 17(4): 496-500.
- [30] Bankart AS. Metatarsus varus. *Br Med J* 1921; 2(3174): 685.
- [31] Baker F. Metatarsus varus. *Calif Med* 1948; 69(5): 346-348.
- [32] Thomson SA. Hallux varus and metatarsus varus: a five-year study (1954-1958). *Clin Orthop Relat Res* 1960; (16): 109-118.
- [33] Reimann I, Werner HH. Congenital metatarsus varus: on the advantages of early treatment. *Acta Orthop Scand* 1975; 46(5): 857-863.
- [34] Browne RS, Paton DF. Anomalous insertion of the tibialis posterior tendon in congenital metatarsus varus. *J Bone Joint Surg Br* 1979; 61(1): 74-76.
- [35] Kane R. Metatarsus varus. *Bull N Y Acad Med* 1987; 63(9): 828-834.
- [36] Petersen KE. Congenital metatarsus varus. *Acta Orthop Scand* 1944; 15(1): 30-48.
- [37] Lloyd-Roberts GC, Clark RC. Ball and socket ankle joint in metatarsus adductus varus (s-shaped or serpentine foot). *J Bone Joint Surg Br* 1973; 55(1): 193-196.
- [38] Rushforth GF. The natural history of hooked forefoot. *J Bone Joint Surg Br* 1978; 60(4): 530-532.
- [39] Votta JJ, Weber RB. A nonsurgical treatment regimen for metatarsus adductus utilizing orthoses. *J Am Podiatry Assoc* 1981; 7(2): 69-72.
- [40] Gordon JE, Luhmann SJ, Dobbs MB, Szymanski DA, Rich MM, Anderson DJ, Schoenecker PL. Combined midfoot osteotomy for severe forefoot adductus. *J Pediatr Orthop* 2003; 23(1): 74-78.
- [41] Lichtblau S. Section of the abductor hallucis tendon for correction of metatarsus varus deformity. *Clin Orthop Relat Res* 1975; (110): 227-232.
- [42] Hutchinson B. Pediatric metatarsus adductus and skewfoot deformity. *Clin Podiatr Med Surg* 2010; 27(1): 93-104.
- [43] Farsetti P, Weinstein SL, Ponseti IV. The long-term functional and radiographic outcomes of untreated and non-operatively treated metatarsus adductus. *J Bone Joint Surg Am* 1994; 76(2): 257-265.
- [44] Lincoln TL, Suen PW. Common rotational variations in children. *J Am Acad Orthop Surg* 2003; 11(5): 312-320.
- [45] Crawford AH, Gabriel KR. Foot and ankle problems. *Orthop Clin North Am* 1987; 18(4): 649-666.
- [46] Lepow GM, Lepow RS, Lepow RM, Hillman L, Neville R. Pediatric metatarsus adductus angle. *J Am Podiatr Med Assoc* 1987; 77(10): 529-532.
- [47] Dawoodi AI, Perera A. Radiological assessment of metatarsus adductus. *Foot Ankle Surg* 2012; 18(1): 1-8.
- [48] Wan SC. Metatarsus adductus and skewfoot deformity. *Clin Podiatr Med Surg* 2006; 23(1): 23-40.
- [49] Wynne-Davies R. Family studies and the cause of congenital club foot. Talipes equinovarus, talipes calcaneo-valgus and metatarsus varus. *J Bone Joint Surg Br* 1964; 46(3): 445-463.
- [50] Widhe T, Aaro S, Elmstedt E. Foot deformities in the newborn - incidence and prognosis. *Acta Orthop Scand* 1988; 59(2): 176-179.
- [51] Paton RW, Choudry Q. Neonatal foot deformities and their relationship to developmental dysplasia of the hip. An 11-year prospective, longitudinal observational study. *J Bone Joint Surg Br* 2009; 91(5): 655-658.
- [52] Eamsobhana P, Rojjananukulpong K, Ariyawatkul T, Chotigavanichaya C, Kaewpornsawan K. Does the parental stretching programs improve metatarsus adductus in newborns? *J Orthop Surg(Hong Kong)* 2017; 25(1): 1-5.
- [53] Hunziker UA, Largo RH, Duc G. Neonatal metatarsus adductus, joint mobility, axis and rotation of the lower extremity in preterm and term children 0-5 years of age. *Eur J Pediatr* 1988; 148(1): 19-23.
- [54] Wynne-Davies R, Littlejohn A, Gormley J. Aetiology and interrelationship of some common skeletal deformities. (Talipes equinovarus and calcaneovalgus, metatarsus varus, congenital dislocation of the hip, and infantile idiopathic scoliosis). *J Med Genet* 1982; 19(5): 321-328.
- [55] Widhe T. Foot deformities at birth: a longitudinal prospective study over a 16-year period. *J Pediatr Orthop* 1997; 17(1): 20-24.
- [56] Katz K, Naor N, Merlob P, Wielunsky E. Rotational deformities of the tibia and foot in preterm infants. *J Pediatr Orthop* 1990; 10(4): 483-485.
- [57] Fedele JM, Aubrespy P, Seriat-Gautier B, Derlon S, Baiada A. [Congenital metatarsus varus treated by external low temperature thermoplastic splint]. *Ann Kinesither* 1991; 18(10): 489-497.
- [58] Kumar SJ, MacEwen GD. The incidence of hip dysplasia with metatarsus adductus. *Clin Orthop Relat Res* 1982; (164): 234-235.
- [59] George IN, Ukpong DE, Imah EE. Cultural diversity of marriage sustainability in Nigeria: strengths and challenges. *Sociology and Anthropology* 2014; 2(1): 7-14.
- [60] Werler MM, Yazdy MM, Kasser JR, Mahan ST, Meyer RE, Anderka M, et al. Maternal cigarette, alcohol, and coffee consumption in relation to risk of clubfoot. *Paediatr Perinat Epidemiol* 2015; 29(1): 3-10.
- [61] Mesquita MA, Segre CA. Congenital malformations in newborns of alcoholic mothers. *Einstein* 2010; 8(4Pt1): 461-466.
- [62] Skelly AC, Holt VL, Mosca VS, Alderman BW. Talipes equinovarus and maternal smoking: a population-based case-control study in Washington state. *Teratology* 2002; 66(2): 91-100.
- [63] Honein MA, Paulozzi LJ, Moore CA. Family history, maternal smoking and clubfoot: an indication of a gene-environment interaction. *Am J Epidemiol* 2000; 152(7): 658-665.
- [64] Ekele BA, Tunau KA. Place of delivery among women who had antenatal care in a teaching hospital. *Acta Obstet Gynecol Scand* 2007; 86(5): 627-630.
- [65] Lamina MA, Sule-Odu AO, Jagun EO. Factors militating against delivery among patients booked in Olabisi Onabanjo University Teaching Hospital, Sagamu. *Niger J Med* 2004; 13(1): 52-55.
- [66] Nwosu BO, Ugboaja JO, Obi-Nwosu AL, Igwegbe AO. Attitude of women towards private and public hospitals for obstetric care in South-East Nigeria: implications for maternal mortality reduction. *Orient J Med* 2012; 24(1-2): 1-6.
- [67] Jacob JE. Metatarsus varus and hip dysplasia. *Clin Orthop Relat Res* 1960; (16): 203-213.

