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CLEFT LIP, ALVEOLUS AND PALATE IN AFRICAN NATIVES: AN UPDATE ON DEMOGRAPHICS AND MANAGEMENT OUTCOME

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Abstract

Background—Development of craniofacial structures is a complex process and disruption of any of the numerous steps can lead to development of oro-facial clefts. This is a surgically amenable anomaly as from early life that has had conflicting pattern of demographics reported by various researchers globally. There are several factors that are critical to the surgical outcome.

Objective—Study the demographics and the management outcome of cleft lip, alveolus and palate and highlight factors responsible for improved care in recent time.

Design—Descriptive cohort study.

Setting—Tertiary health institution

Method—All consecutive patients managed for cleft lip, alveolus and palate (CLAP) over 7years and 10months were studied.

Outcome—Cleft lip, alveolus and palate repair was performed on 149 patients, January 1, 2001–December 31, 2008 with an incidence of 2.1/1000 live births. From this, 27 patients, averaging 4.5 patients per year were operated for the first 6 1/3 years while the remaining 122(81.9%) the next 1 1/2 years, averaging 81.6 patients yearly. Their ages ranged from 3 months – 60 years with 77 (51.7%) males and 72 (48.3.0%) females. Cleft lip was the main presentation in 108(72.5%) of which 72(66.7%) were left sided. Bilateral cleft lip were14 (9.4%). Five (3.4%) patients had associated anomalies out of which 3(60.0%) had CLAP while 2(40.0%) isolated cleft lip or palate.

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Declaration

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The technique for cleft lip repair was Millard's and Noordhoof's while palatal cleft was the two-flap palatoplasty with intravelar veloplasty. Success was recorded in 142(95.3%) with complication observed in 7(4.7%) patients.

Conclusion—The rarity of cleft lip, alveolus and/or palate in the African native documented previously may no longer be tenable as observe in this study. Management outcome has improved owing to the collaboration with SmileTrain, USA, along with multidisciplinary approach.

Keywords

Cleft lip; Alveolus and Palate; Demographics; African Native; Management Outcome Factors

Introduction

Cleft lip(cheiloschisis) and/or palate(palatoschisis) is the disruption of the oro- facial architecture that is an easily recognizable defect at birth. This can occur in isolation or as part of a broad range of chromosomal, mendelian or teratogenic syndromes. Cleft lip and/or palate is etiologically heterogenous and this has critical implications for understanding the biology of facial development, how environmental risks interact with genetic factors and how we can incorporate known etiologic variables to improve clinical care.[1] Research on the etiology of CLAP has over the years yielded exciting discoveries around the globe that necessitated its inclusion as part of the GENEVA(Gene, Environment, Association) cleft consortium studies which is a component of the Genome-Wide Association Study(GWAS) as a fall out of the USA-led program-Gene, Environment, health Initiative(GEI) conceived in 2006 by the National Institute of Health(NIH).[2] The GENEVA cleft consortium study confirmed IRF6(interferon regulatory factor 6), chr8q24, 10q25 with two new loci MAFB and ABCA4 not previously associated with cleft lip, alveolus and/or palate (CLAP) but now have stronger signals in Asians.[3] However, the GWAS identified fibroblast growth factor receptor2(FGFR2), SUMO1(small ubiquitin-like modifier family) gene, FOXE1(forkhead box E1 gene) as risk factors[3] while Jezewski and colleagues found mutations on MSX1(msh homeobox 1), FGFR1, FGF8 and BMP4(bone morphogenetic protein-4.[4] Oral clefts show strong familial aggregation, and the recurrence risk amongst first degree relatives is about 32 times than the general population risk for cleft lip/palate and about 56 times greater for cleft palate.[1]

The human face is a focal point for expression and interpersonal communication[5] thus a centre of attraction of all human appearances. Clefting, as it occurs at the oro-facial region, disrupts this arrangement. Individuals with clefting experience problems with feeding, speaking, hearing and social integration that can be corrected by varying degrees of surgery [1] which has generated a lot of interest across several specialties in Cleft lip, alveolus and palate (CLAP) care, moreso they could be associated with other congenital anomalies.

The tooth contributes significantly to the aesthetic appearance of the face. European visitors to Nigeria admired her Natives "perfect row of white teeth"[6]. The presence of CLAP affects this unique appearance.

Different pattern of demographics of CLAP and management outcome have been described in various regions of the globe, including Africa, the authors therefore undertook this study to evaluate their recent observations.

Methods

All consecutive patients who had cleft lip, alveolus and palate repair at the Federal Medical Centre, Gombe, North Eastern Nigeria, between January 1, 2001 and October 31, 2008 were studied. Particular emphasis on the last 1 ½ years was made, largely following the establishment of an organized Cleft lip and Palate Group, an affiliate of The Smile Train, USA (a Non-Governmental organization that offer assistance to patients with Cleft Lip and/or palate)in the hospital. Data was generated from the following: Age, gender, diagnosis, procedure done, nature of anaesthesia and the outcome of intervention. The operative technique for cleft lip repair was the Millard's and Noordhoof's methods while the cleft palatal repair was the two-flap palatoplasty with intravelar veloplasty. The study observed the Helsinki Declaration. Results were analyzed by simple statistical methods, tables and diagrams.

Results

A total of 149 patients were operated from January 1, 2001 to October 31, 2008. In the first 6 1/3 years (January 1, 2001–April 30, 2007), 27 patients(averaging 4.2 patients per year) were evaluated and operated, Fig 1, while 122(81.9%) for the subsequent 1 1/2 years (May 1, 2007 to October 31, 2008) averaging 81.6 patients per year were operated.

The hospital recorded 6,919 live births within the study period which translates to an incidence of 2.1/1000 live births. There were 77 (51.7%) males and 72 (48.3%) females giving M: F ratio of 1.1:1. Their ages ranged from 3months–60years (Fig. II). Mean age was 12.4years. General anaesthesia was used in 79(53.0%) patients and Local anaesthesia in 70(47.0%) patients. The presentation was majorly cleft lip in 108(72.5%). See Table I. From this, 72(66.7%) were left sided clefts and 36(33.3%) were right sided. Bilateral cleft lip were observed in 14(9.4%), combined cleft lip/palate were16 (10.7%) with 6 males(37.5%) and 10 females(62.5%) and therefore a M:F= 1:1.7. While cleft palate alone were 11(7.4%) out of which 8 were males(72.7%) and 3 females(27.3%) with M:F=3:1. Five (3.4%) patients had associated congenital anomalies from which 3(60.0%) patients had CLAP while 2(40.0%) isolated cleft lip and palate each.

Discussion

Cleft lip, alveolus and/or palate(CLAP) is the most common congenital anomaly in the head and neck region and the second commonest in the entire body, trailing behind club foot. [7] This could be non-syndromic or syndromic with over 300 associated syndromes having been described. [8] Approximately, 70% of all cases of cleft lip and/or palate and 50% of cases of cleft palate only are considered to be non-syndromic, respectively.[1]

The estimated incidence of CLAP ranges from 1:500 to 1:2500 live births with prevalence among the general population varying by race, gender, ethnic, geographical and

socioeconomic factors. [9,10] However, our series recorded an incidence of 2.1/1000 live births which is quite high contrary to previous reports that the anomaly was less prevalent in the African native.[11] The authors observed that the formation of the Cleft lip and Palate group at the institution which offer assistance for care of these patients, responsible. The previously described low incidence in Africa might largely be due to financial constraints rather than rarity of the anomaly. This could be confirmed by a 45-year old male farmer who declined operation on his cleft which he claims was his 'source of income'. (He uses it to beg for alms!). Furthermore, the increase in the number of Specialists with interest on CLAP repair in our hospital from one Cleft surgeon in 2000/2001 to nine in 2007 along with public enlightenment might have led to the increased turnover as well.

An average of 81.6 patients per year (6.8 monthly) seen in this study is higher than the yearly average of 16.5,[12] 50[13] previously reported from Northern Nigeria and 69 cases in Ghana[14] although cases done under local anaesthesia were excluded from the Ghana report. We also observed a wide patients' age range from 3 months to 60 years, this totally contrasts with the developed countries where early repair is the norm.[15] Adult cleft repair is not unusual in our practice due to ignorance and financial constraints.[16] About 26.2% of cases in this study were adults which is remarkably higher than 2.8% [12] previously recorded from the same region. From the available records, this study documented the oldest patient in cleft history.

In 2007, our hospital established a partnership with Smile Train USA which funds medical care for all cleft patients. This could explain both the patient upsurge within the period and the appearance of adults for surgery.

Our M: F ratio of 1.1:1 is in agreement with similar study from Jos, North-central Nigeria.[12,17] and concurs with overall slight male preponderance in cleft lip and palate from other studies. Unilateral cleft lip accounted for 108(72.5%) of our cases which is similar to 78% of all operations reported from a study.[14] This preponderance of cleft lip is also confirmed by studies from Pakistan that recorded 42%[18] but differ sharply from other researchers where combined cleft lip and palate which constituted 60.2% was the most common type of cleft.[16] The left side was more commonly involved in unilateral cleft lip in this study (n=72, 66.7%). This observation has also been found in other epidemiologic reviews of clefts but yet to have a clear explanation.[15] Nevertheless, isolated cleft palate found in 11(7.4%) patients occurred more in males (n=8, 72.7%) than females (n=3, 27.3%) contrary to findings in the Caucasians which reported a higher female prevalence[12]. The slight male preponderance found in cases of cleft lip and palate by Adekeye in Kaduna-Nigeria is reversed in this study with a ratio of 1:1.7. [19]

Other rare clefts such as upper lip median cleft, lower lip median cleft, oblique and lateral clefts which constitute 5% of craniofacial clefts observed by some researchers [17] were not recorded in our series.

Associated congenital anomalies were seen in 5(3.4%) patients in this study which is at variance with 21% found by Milerad et al [20] in Sweden. Most of these associated anomalies observed by other studies that were right-sided, were syndromic clefts. [21] In this report,

however, cases of associated malformations were more frequent in infants who had both cleft lip and palate (n=3, 60.0%) than in infants with isolated cleft palate or infants with isolated cleft lip (n=2, 40.0%). Genetic studies are yet to be conducted to confirm this. The significance for this is to emphasize the importance of holistic approach to the care of patients of CLAP. A multidisciplinary approach has been found to be crucial in any successful CLAP care. In this series, a 3year old male child developed a cardio-pulmonary arrest post cleft lip and palate repair in which prompt evaluation revealed a bilateral choanal atresia. The ENT surgeons performed a choanoplasty. Usually, bilateral choanal atresia present as emergencies with upper airway obstruction as the paediatric age-group are obligate nasal breathers. In the index case, however, the patient might have been using the cleft for breathing and thus closure, following repair, led to respiratory embarrassment. Other anomalies were referred to their various specialties.

Our Cleft Lip and Palate Group comprises the Maxillofacial, Plastic & Reconstructive, General and ENT surgeons. The Anaesthesiologist ensures safe anaesthesia to observe the dictum “No child should die during Cleft repair” adopted by Smile Train USA. The Paediatrician, supported by dedicated nurses, social welfare workers and community health workers are part of the team that take care of the nutritional rehabilitation of such patients who often are undernourished including home visits in the event of default. This could explain the low failure rate of 4.7% compared with 14.9% reported in a study.[12.] They were successfully treated following a revision operation. Follow- up of patients is an integral part for a successful CLAP care as this spans through a long period, each having a specific intervention measure. Most of our patients who had combined cleft lip/palate often abscond from follow-up visits after repair of the cleft lip as the cosmetic appearance had improved.

Conclusion

The previously documented rarity of cleft lip, alveolus and/or palate in the African native may no longer be tenable taking into account the computed incidence in this study. Management outcome has improved owing to the collaboration with SmileTrain, USA, along with multidisciplinary approach.

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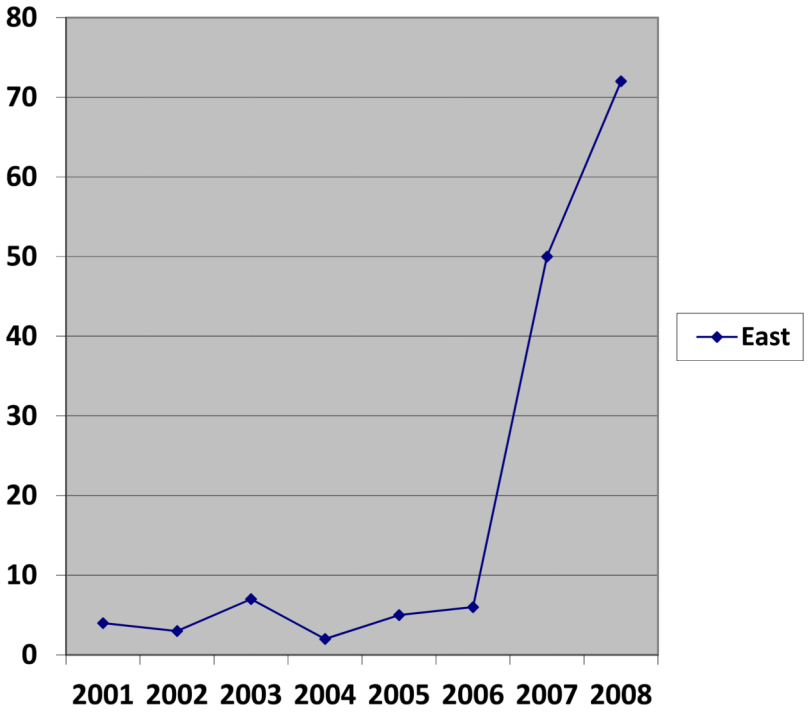

Figure I.
Yearly Distribution of Cleft Lip and Palate Operation

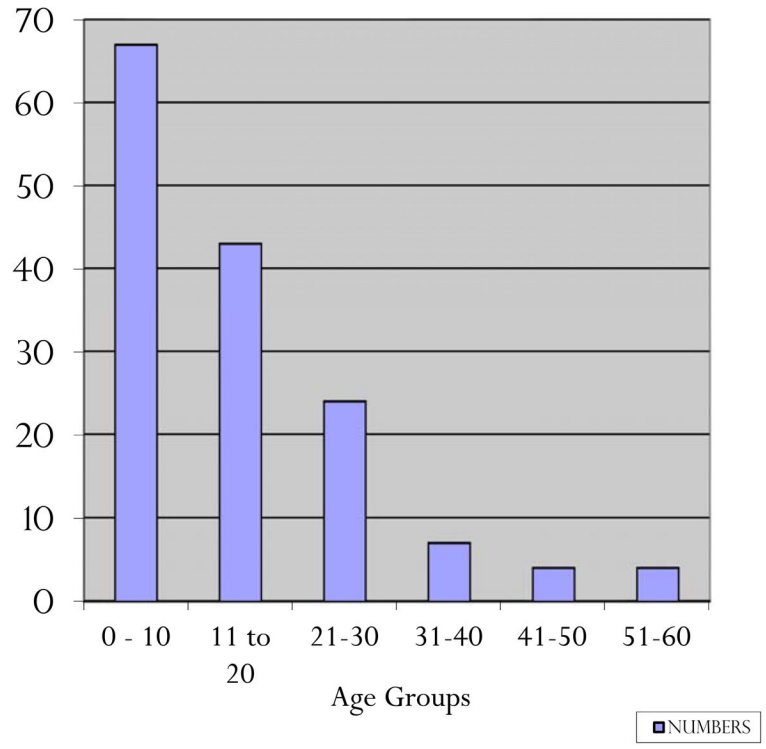

Figure II.
Distribution of Population By Age Groups

Table I

showing Diagnosis by affected side

	Right	Left	Total (%)
CLEFT LIP			
Unilateral *complete*	*22*	*52*	*74(49.7)*
Unilateral incomplete	*14*	*20*	*34(22.8)*
Total Unilateral	***36***	***72***	***108(72.5)***
Bilateral complete	-	-	08(5.4)
Bilateral incomplete	-	-	06(4.0)
Total Bilateral			**14(9.4)**
CLEFT LIP/PALATE	-	-	**16(10.7)**
CLEFT PALATE	-	-	**11(7.4)**
GRAND TOTAL			**149(100.0)**