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Epidemiology of Cleft Lip and Palate in Nigeria: A Data-Based Study

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ABSTRACT

Background and aim: Prevalence of cleft lip and palate varies across geographical location, races, and ethnic groups. The global prevalence is 1 per 700 live births. We aim to establish the prevalence, socio-demographic characteristics of patients with an orofacial cleft, and associated deformities in our locality.

Materials and methods: This was a retrospective study of all the patients with cleft lip (CL), cleft palate (CP), or cleft lip/palate (CLP) managed over 12 years (2009 to 2020). A uniform Smile Train® structured interviewer-administered questionnaire completed by the Surgeons was used to obtain relevant information. Data were analyzed using SPSS version 23.

Results: A total of 280 patients were managed for cleft lip and palate deformities during this period. The estimated total live births during the period were 232,168; a prevalence rate of 0.8 per 1000 live births was thus calculated. The most common type of cleft deformity in our study is CLP (61.4%), followed by isolated CP (20.7%) and then CL (17.9%). CLP was shown to be more common among males (66.1%). However, isolated CP and CL were more common among females at 67.2% and 56%. The most common associated anomalies were speech (52.8%) and growth retardation (9.6%). The least common was limb anomalies (0.7%).

Conclusion: Our study has generated new knowledge of the epidemiological distribution of orofacial cleft deformities in our subregion made possible by the Smile Train® database. It will enable more comprehensive management of orofacial clefts.

1. Introduction

Cleft lip (CL) and cleft palate (CP) are considered one of the most common congenital anomalies of the head and neck region, and collectively, craniofacial anomalies are about the most common of all congenital disabilities.[1–3] They are abnormal fissures, openings, or gaps in the upper lip, the roof of the mouth, or both, resulting from non-fusion or breakdown of embryologic structures.[4] Prevalence of cleft lip and palate varies across geographical location, races, and ethnic groups.[5] The global Prevalence is 1 per 700 live births; it is highest in Asia (1/500), intermediate in Caucasians (1/1000), and low in Africa (1/2500).[6] Studies suggest that the cases in Africa may be underestimated due to a lack of reliable data.[2, 7]

The face is the most conspicuous part of the human body; however, aside from posing aesthetic challenges, Oro-facial clefts also cause feeding difficulties by interfering with the infant's suckling and swallowing and poor growth. Speech and hearing problems may also subsist. As the child grows, Integration into the social community may be difficult due to segregation or bullying from peers; consequently, low self-esteem and poor quality of life.[4, 8–10] Ignorance and societal anxiety/embarrassment play a negative role, as patients and relations of those with cleft deformities suffer unnecessarily, whereas proper management and surgery can correct the abnormalities at no cost to the family. Smile Train® is an international children's charity organization founded in 1999, extending across over 90 countries and partnering with over 1100 hospitals and 2100 medical professionals. To date, it has sponsored over 1.5 million free clefts surgeries.[11] Management of patients with cleft deformity is multidisciplinary and may require multiple surgical interventions and revision surgeries.[5] The goal of surgery in the cleft lip is to restore normal facial architecture; however, in the cleft palate, the primary aim of surgery is to help the patient achieve a competent velopharyngeal sphincter the growing child to perform age-related functions like swallowing, hearing, and speaking.[8] Hence, the Plastic, Maxillo-Facial, Otorhinolaryngolosists (ENT), Neurosurgeons and Ophthalmologists may be involved as well as Orthodontists, Dentists, Dieticians, and Speech therapists. The etiology and pathogenesis of cleft lip and palate are not yet fully understood. However, genetic and environmental factors are known to play a

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significant role in its development.[4, 9, 12-13] The inheritance is believed to be multifactorial rather than a single gene disorder and does not follow the Mendelian pattern.[8, 13] Environmental factors like maternal alcohol consumption, smoking, and use of anticonvulsant medications, exposure to certain chemicals and radiation, maternal hypoxia, nutritional deficiencies have also been implicated.[4, 5, 9, 12] The epidemiology of cleft lip and palate is well documented in many developed countries with reliable record-keeping systems but not in developing countries like ours lacking designated congenital anomaly register.[2, 5, 7] Therefore, aim to establish the prevalence and socio-demographic characteristics of patients with orofacial cleft deformities in our locality. It will help in the proper planning and management of cleft patients.

2. Materials and methods

Case series of all the patients with cleft lip, cleft palate, or cleft lip/palate managed over 12 years (2009 to 2020), were reviewed and analyzed. The same group of surgeons managed cases. A structured interviewer-administered questionnaire completed by the Surgeons was used to obtain relevant information about the patients from their caregivers; clinical information and interventions done were also recorded for each patient over the years. The orofacial clefts were classified according to international classification of disease (ICD9) diagnosis code into the cleft lip, cleft lip with the palate, and cleft palate; these were further subcategorized by laterality and completeness.[14] The total number of births during the ten years was estimated from the Nigeria demographic health survey fact sheet for southeastern Nigeria.[15] Prevalence rate of orofacial clefts was then calculated by dividing the total number of live births by the number of orofacial clefts and multiplying by 1000. Data were analyzed using SPSS version 23, and results were presented using frequency tables and charts. Chi-square was used to test for association between cleft types and gender.

3. Results

A total of 280 patients were managed for cleft lip and palate deformities during the period under review. The estimated total live birth for the state during the period under review was obtained as 232,168; a prevalence rate of 0.8 per 1000 live births was thus calculated. Fifty four percent (53.6%) were males and Forty-six (46.4%) females. The majority (71.43%) of the participants were aged between 0 to 3Years. Age of the participants as noted in Table 1.

Table 1. Age and Sex distribution of Cleft Lip and palate patients.

		Frequency	Percent
Sex	Female	150	53.6
	Male	130	46.4
Age	Less than one year	116	41.43
	1 to 3 years	84	30.00
	4 to 6 years	38	13.57
	7 to 9 years	20	7.14
	10 to 12 years	16	5.71
	Above 12 years	6	2.14
Total		280	100.00

The most common type of cleft deformity in our study is CLP (61.4%), followed by CP (20.7%), and then CL (17.9%), with a CLP: CP: CL ratio of 3.4:1.2:1. CLP was shown to be more common among males (66.1%), CP and CL were more common among females 67.2% and 56%, respectively. There was an equal distribution of bilateral CP in both males and females, as shown in Fig. 1 and Table 2 below. There was a significant association between gender and type of cleft deformity ($p<0.05$).

Table 2. Association between gender and type of cleft deformity.

		Gender			
	Type	Female	Male	Total	P-value
CL	-----	28 (56%)	22 (44%)	50 (17.9%)	0.039*
	Bilateral CL	6 (60%)	4(40%)	10 (3.6%)	
	Left CL	12 (54.5%)	10 (45.4%)	22 (7.9%)	
	Right CL	10 (52.6%)	9 (47.3%)	19 (6.8%)	
CLP	-----	83 (48.3%)	89 (51.7%)	172 (61.4%)	
	Bilateral CLP	20 (33.9%)	39 (66.1%)	59(21.1%)	
	Left CLP	34 (61.8%)	21 (38.1%)	55 (19.6%)	
	Right CLP	10 (52.6%)	9 (47.3%)	19 (6.8%)	
CP	-----	39 (67.2%)	19 (32.8%)	58 (20.7%)	
	Bilateral CP	3 (50%)	3 (50%)	6 (2.1%)	
	Left CP	34 (61.8%)	21 (38.1%)	55 (19.6%)	
	Right CP	30 (69.8%)	13 (30.2%)	43 (15.4%)	

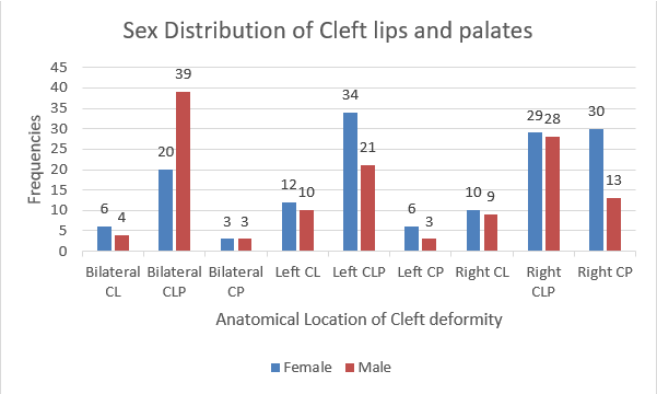

Fig. 1. Distribution of orofacial clefts sex and laterality.

Most of the participants were referred to us by Hospitals and Physicians (68.9%), other means through which they learnt about Smile Train® were friends and relatives (17.1%), Charity Organizations (5.4%), newspaper and television (4.6%), radio (2.9%), internet (1%). Others, including Churches, seminars, markets, constituted 2.9%.

Table 3. How patient caregivers heard about Smile Train®.

	Frequency	Percent
Charity Organization	15	5.4
Friends and Relatives	48	17.1
Hospital/Physicians	193	68.9
Internet	1	0.4
Newspaper and Television	13	4.6
Others	2	0.7
Radio	8	2.9
Total	**280**	**100.0**

The most common associated anomaly observed in our study involved speech (52.8%), retarded growth (9.6%), Nose (9.3%), eyes (6.4%), and mental retardation (3.5%). Other less common ones are skull anomalies (2.8%), mandibles (1.79%), fingers and toes (1.08%), ears (1.79%), limbs (0.71%), and Genito-urinary system (1.07%).

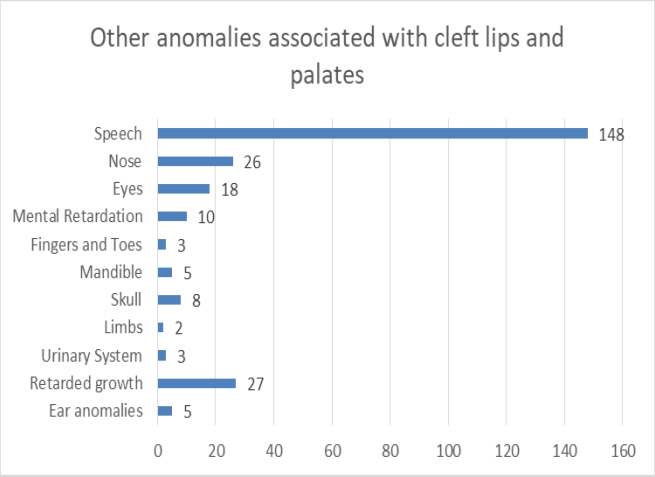

Fig 2. Other congenital anomalies associated with orofacial cleft deformities.

The most common surgical procedures done were primary unilateral lip repair in 37.5%, primary cleft palate repair (30.7%), and fistula repair (10.7%); others were fistula repair (10.7%), Lip/Nose revision (7.5%), and the least common was secondary cleft palate repair in 5% of the patients. Millard Rotation/Advancement and Variants are the most common procedures used in unilateral and bilateral lip repair; Langenbeck Variants was more commonly used in cleft palate repair, while local palatal flaps are used for fistula closure.

4. Discussion

The present review involved patients who received treatment from Smile Train® network, a global charity organization that sponsors comprehensive management of orofacial clefts and has also opened portals for research and creating public awareness to address social and cultural issues associated with cleft deformities. Our study estimated a prevalence rate of 0.8 per 1000 live births, comparable to figures obtained by other studies in Nigeria,[7, 16] South Africa,[2] and North Eastern brazil.[10] Higher rate has been observed in Asia (1/500) and Caucasians (1/1000).[6] Studies suggest that the values in Nigeria and other African countries may be underestimated due to poor records keeping.[2, 7] The majority (71.43%) of our participants were between 0 to 3 Years of age, with a male to female ratio of 1:1.2. Onah in Enugu[17] noted the early presentation of patients for surgery. Conway et al. 6 pulled data from 33 African countries over ten years and observed a male to female ratio of 1:1.46. However, they recorded a relatively late presentation as most of their patients presented after four years of age with an average age at surgery of 9.34 years.[6] They attributed the delay at presentation in their study to lack of access to media and public enlightenment and the fact that their study was multi-center based with a larger number of participants. Smile Train® Inc is currently partnering with the agencies in Nigeria with resultant improved awareness through various communication media and churches. It may have influenced the relatively early presentation observed in our study. The sex distribution of 1:1.2 noted in our study is comparable to other studies[6, 10] and slightly different from others.[18, 19] Ajike's finding[19] may be explained by the fact that adult females have better health-seeking behaviors and are more concerned about aesthetics than their male counterparts and would have had a repair earlier.

The most common type of cleft deformity in our study was CLP (61.4%), followed by CP (20.7%), and then CL (17.9%), with a CLP: CP: CL ratio of 3.4:1.2:1. It agrees in most parts with other studies.[4, 20-22] but differed completely with a study in Tanzania that reported isolated CL (49.2%) to be the most common, followed closely by CLP (39.2%) and then CP (11.7%).[23] They attributed the low cases of isolated CP to a possible higher mortality rate in this group from functional difficulties with feeding and malnutrition. Studies have also suggested that isolated CP has more association with other congenital anomalies and poorer prognosis than other cleft types.[9, 24] CLP was shown to be more common among males (66.1%), whereas isolated CP and CL were more common among females 67.2% and 56%, respectively, in our study. Most of the patients were referred to us from the hospitals and maternities where they were born or first presented, friends and relatives (17.1%), and charity organizations (5.4%). However, in Ajike's study, friends and relatives (41.8%), radio (22.8%), and charity organizations (16.5%) predominated. Again, this study was conducted amongst adults compared to ours in which the children predominated. The most common associated anomaly observed in our study involved speech (52.8%), retarded growth (9.6%), Nose (9.3%), eyes (6.4%), and mental retardation (3.5%). In Conway's survey, six growth retardation (30.8%) was the most common associated anomaly, followed by eyes (16.4%), Fingers/Toes (8.2%), skull (7.4%), limbs (6.8%), Ears (6.6%), Mental Retardation (5.8%). The speech was not evaluated. Approximately 30% of orofacial cleft anomalies are syndromic and have different epidemiological patterns from those without any associated anomalies.[6, 24-25] Geographical variation may thus explain the subtle differences in the distribution of associated anomalies in our study from Conway. On the other hand, it may also be an emerging trend that needs to be further elucidated via more studies. The majority of our patients had primary unilateral lip repair, most commonly through rotation/advancement variant. Bilateral lip repair was commonly done using the Millard type variant, cleft

palate repair was commonly done using Langenbeck's variant, and all fistula repairs were done using local palatal flaps. it is in keeping with reports from similar studies.[6, 9, 26]

5. Conclusion

We recorded a prevalence of 0.8 per thousand live births of orofacial cleft deformities in our locality, as well as a relatively early presentation of our patients. Our study's most common type of cleft deformity was cleft lip with cleft palate(CLP), followed by isolated cleft palate(CP). Cleft lip with cleft palate(CLP) was more common among males, whereas isolated cleft palate(CP) and isolated cleft lip(CL) were more common among females; These findings were in keeping with few cited literature and partly in variance with most others, which may represent an emerging trend in the pattern of clefting. Further studies are, however, required to either elucidate or refute this claim. Haven established the prevalence and socio-demographic characteristics of patients with orofacial cleft deformities in our locality, we have added to the body of knowledge, and this will enable more comprehensive management and care for cleft patients, as well as instigate other studies in this area with Smile Train® database, reliable statistics are now easily available for African cleft studies and several myths about orofacial clefts, and some other congenital anomalies has mainly been demystified.

Conflict of Interest

The authors declared that there is no conflict of interest.

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