Pattern and ENT manifestations of cleft lip and palate in Sudanese children in Khartoum State, Sudan

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ABSTRACT

Background Craniofacial anomalies, and in particular cleft lip and palate, are major human birth defects with a worldwide frequency of 1 in 700 and substantial clinical impact.

Objectives To study pattern and clinical presentations of cleft lip and cleft palate in Sudanese children in Khartoum State.

Patients and methods This is a cross-sectional hospital based study in patients who presented to two of Khartoum State Hospitals with cleft lip and palate during the period from December 2017 to September 2018. The data was collected through a validated questionnaire and clinical examination was done.

Results Seventy-one patients were included in this study. Patients' age ranged from 2 months to 15 years, (mean age 66 ± STD 59 months). The most affected age group was 2 months – 5 years. Male to female ratio was 1.15:1.00. Most of the patients (59.2 %) came from rural communities. The commonest deformity encountered was cleft palate deformity (47.9%) followed by cleft lip (29.6%), cleft lip and palate (19.7%) and submucous cleft palate deformity (2.8%). The commonest presentation was feeding difficulties (73.2 %) followed by speech disorders (47.9%), decreased hearing (8.5%) and recurrent otitis media (7.0 %). The presentation of feeding difficulty was the most common associated symptom with the cleft palate and cleft lip deformities; it was distributed among patients as follow: cleft lip only 71.4 %, cleft palate only 70.5%, cleft lip and palate 92.8 %. Hearing loss was a presentation in all patients with submucous cleft variety.

Conclusion Cleft lip and cleft palate are relatively rare conditions; cleft palate is the commonest deformity among Sudanese children with male predominance and feeding difficulty is the commonest presentation.

Recommendation High attention should be given to medical and paramedical staff for early pick up of these deformities immediately after delivery. The need for national centres for management and rehabilitation of craniofacial malformations cannot be overemphasized.

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INTRODUCTION

Based on World Health Organization report, about 3 million fetuses and infants are born each year with major malformations. Several large population based studies place the incidence of major malformations at about 2–3% of all live births.[1] Murray JC, stated that craniofacial anomalies, and in particular cleft lip and palate, are major human birth defects with a wide variability related to genetic, geographic and socioeconomic status. The worldwide frequency is 1 in 700. In general, Asian or Amerindian populations have the highest frequencies, often at 1/500 or higher, with Caucasian populations intermediate, and African-derived populations the lowest at 1/2500.[2] Specific causes have now been identified for some forms of cleft lip and palate, and we are at the beginning of a time in which the common non-syndromic forms may also have specific aetiologies identified. Mouse models' have an especially important role in disclosing cleft etiologies and providing models for environmental cotriggers

or interventions. An overview of the gene–environment contributions to non syndromic forms of clefting and their implications for developmental biology and clinical counseling is presented.[2] Leslie EJ et al declared that complications of clefting in early life are particularly devastating in developing countries where access to medical care may be limited.[3] Early detection and proper management of these conditions will improve the outcome. The objectives of this research are to study the pattern and clinical presentations of cleft lip and cleft palate in children in Khartoum State.

MATERIALS AND METHODS

This is a cross-sectional hospital-based study. All patients (aged birth-15 years) who presented with cleft lip, cleft palate and cleft lip and palate to Khartoum Ear, Nose and Throat Hospital (KENTH) and to the Maxillofacial Department, Dental Hospital, Khartoum and who gave consent were enrolled in the study. The study was conducted during the period from December 2017 to September 2018. The data is collected using questionnaire. Statistical analysis was performed via SPSS software version 22. Chi-square test was used when appropriate. A p value of < 0.05 was considered statistically significant.

Ethical clearance

The hospitals ethical committees approved the study. Informed verbal consent was obtained from the patients / care-takers after explaining the aim of the study.

RESULTS

A total of seventy-one patients were included in this study. Their ages ranged from to 2 months to 15 years, (mean age 66 ± STD 59 months). The most affected age group was 0 – 5 years (n=42, 59.2%) followed by age group 11 – 15 years (n=17, 23.9%) and 6 – 10 years (n=12, 16.9%). The number of male and females patients was 38 (53.5 %) and 33 (46.5%), respectively with male to female ratio 1.15:1.00; twenty-nine patients (40.8 %) were from urban area and forty-two (59.2 %) were from rural areas**.** As shown in Figure, the commonest deformity encountered was cleft palate only (n=34, 47.9%) followed by cleft lip only (n=21, 29.6%), cleft lip and palate (n=14, 19.7%) and submucous cleft palate (n=2, 2.8 %). The commonest presentations were feeding difficulties in 52 patients (73.2 %) followed by speech disorders in 34 patients (47.9%), decreased hearing in 6 patients (8.5%) and recurrent attacks of otitis media in 5 patients (7 %). Regarding the cleft palate only, there were 34 patients: 19 (26.8 %) patients had complete cleft whereas 15 (21.1 %) patients had incomplete cleft**.** The patients of cleft lip and palate were divided into bilateral and unilateral. Bilateral cleft lip were 4 patients (1 patient was of complete variety and 3 patients were of incomplete variety) and unilateral were 10 patients (3 patient were of complete variety and 7 patients were of incomplete variety)**.**

The presentation of feeding difficulty was the most commonly associated symptom with the cleft Palate and cleft lip deformities; it was distributed among patients as follow: cleft lip only 15/21 (71.4 %), cleft palate only 24/34 (70.5%), cleft lip and palate in 13/14 (92.8 %) patients (Table). All patients with submucous cleft palate have had recurrent otitis media (OM). This finding was found in small percentage (7.6 % and 5.8 %, respectively) in cleft lip and palate as well as cleft palate only; but no patients with cleft lip only had recurrent OM.

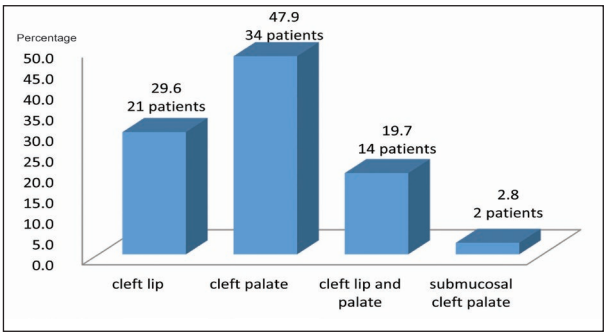

Figure. Distribution of cleft deformities among patients in the study group

Table. The association between cleft deformities and feeding problems in the study group

Deformity	Feeding problem		Total
	Present	Not present	
Cleft lip	15	6	21
Cleft palate	24	10	34
Cleft lip & palate	13	1	14
Submucouscleft palate	0	2	2
Total	52	19	2

P value 0.03

DISCUSSION

Deformities of cleft lip and palate create a challenge for the health care professional involved in their management. The need for an accurate database for cleft registrations as well as systematic record keeping is essential. This is of particular importance when limited resources exist for planning and developing multidisciplinary teams for treating children with cleft lip, cleft palate, or both[4].

In the present study the male gender was dominant (53.5 %) which is in line with reports from the literature. Al Johar et al in KSA and Fakhruddin from Sudan showed higher affection of male gender.[5,6] The study done by Goudy et al reported a high difference in gender distribution (males: 65 and females: 36) which would suggest that a genetic and geographic influence may play a role.[7]

In this study, the majority of patients were from rural area (59.2 %), this correlates well with the study done by Messer et al in Texas,[8] but contradicts with the study of Fakhruddin.[6] This difference may be due to the fact that most our patients were referred from rural area for surgical correction.

In our study, we encountered cleft palate only and cleft lip only to occur in 47.9 % and 29.6 %, respectively; this result differs from a similar local study done by Suleiman who reported respective rates of 30% and 54% for cleft palate and cleft lip.[9] This contradiction is obviously because isolated cleft lip is easier to diagnose while that of cleft palate is easier to miss by the community.

Again, our study contradicts the study done by Al Omari et al in Jordan;[4] it showed most of the patients have clefts of the lip and palate (48%) followed by cleft lip (30 %) and cleft palate (22 %), this could be explained by the fact that the Jordanian study is retrospective with large number of cases, in different times of history, so the significance would not be statistically reliable.

In this study, feeding difficulties was the most common presentation (73.2 %) compared to speech (47.9%) or hearing (8.5%) disorders. This may be because the majority (59%) of our study group was under 5 years, where the feeding problems are more noticeable than the speech or hearing problems. This finding is in line with the study done by de Vries IA et al, where patients with feeding difficulty represented 67 % of the series.[10]

Moreover, in this study, only 6 (8.5%) patients presented with hearing loss, while in the Sheahan study this was a common presentation in 28%.[11] The likely explanations is that the majority of cases of cleft lip/palate malformations encountered in this study were from the maxillofacial department, where hearing assessment is not routinely done (in our set up), as in the Sheahan study. as it was done in ENT department, where the tools for hearing assessment were available.

The recurrent attacks of otitis media in our study was found in 5 patients (7 %), which is lower than the study done by Goudy who found otitis media in 19% of patients;[7] the higher incidence in Goudy study may be due to his larger sample of patients.

Submucous cleft palate in our study was present in only 2 patients; both of them were from KENTH due to familiarity with the condition, both patients presented with hearing loss and recurrent attack of otitis media. This result is not in agreement with Reiter R et al in his study of 439 patients, where 45% of them had problems with hearing loss; the difference may be due to large sample size in that series.[12]

CONCLUSION

Cleft lip and palate are relatively rare conditions with cleft palate being the commonest deformity among Sudanese children and it is more common in rural communities. Feeding difficulty is a common presentation among affected patients. Complete cleft palate is the commonest variety within patients with cleft palate. Therefore in otolaryngology practice, more attention should be paid to the palate especially in children with recurrent otitis media. Early case detection will provide prompt management and prevent further life-long suffering.

RECOMMENDATION

Raising the awareness of the community about antenatal care among pregnant women to take adequate food supplements may guard against foetal deformities. High attention should be given by medical and paramedical staff for early pick up of these deformities immediately after delivery. An expanded program of research for management and rehabilitation protocols should also be established. Not to mention the need for national centres for craniofacial malformations.

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