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Descriptive Epidemiology of Orofacial Clefts in Ethiopia

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Abstract

Background—The prevalence of birth defects including orofacial clefts (OFC) in Ethiopia is not known and there is no established birth defects registration system.

Objectives—To investigate the prevalence and incidence of OFC in Ethiopia.

Design—Retrospective hospital based descriptive study.

Methods—We obtained data from the Smile Train database on Ethiopian patients with OFC who underwent surgical treatment from June 2007 - December 2013 at 31 hospitals distributed throughout the country. Data related to live births in Ethiopia during the mentioned period was obtained from the Federal Ministry of Health database for estimates of the incidence and prevalence rates.

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Dr Butali participates in the conception of the study, data cleaning, statistical design, and critical revision of the final draft.

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Drs Ibrahim Mohammed, Yohannes Demissie, Milliard Deribew, Muluaem Gessese and Paul Egil Gravem participate in reading and providing valuable comments

Results—The total number of live births during the study period was 18,811,316. During this same period, 18,073 cleft patients approximately ranging from 1 to 75 years old were examined and treated at the hospitals mentioned above. The incidence rate estimated from the total number of affected children during the study period (N=8232) is 0.44/1000 live births. The prevalence rate is 0.20/1000 and this was estimated using the number of total population in 2013 (N= 88,703,914). There is a significant difference in frequency between bilateral CLP (26.9%) versus unilateral CLP (73.1%) ($P<0.0001$). There is also a significant difference in frequency between bilateral cleft lips only (15.4%) versus unilateral cleft lip only (84.6%) $P<0.0001$.

Conclusion—It is obvious that the findings in this study cannot be representative of the true picture but provides a previously unavailable national estimate of incidence and prevalence of orofacial clefts in Ethiopia. It can also be used as comparison for future community based studies.

Keywords

Cleft Lip; Cleft Palate; Cleft Lip and Palate; Epidemiology; Ethiopia

Introduction

Ethiopia is the second most populous country in Africa with a population of more than 96 million people. The average growth rate is about 2.89%, with 50.3% females and 49.7% males. Only 19% of pregnant women have 4+ visits during their pregnancy and only 10% of the deliveries were attended by skilled health care professionals (1). This means that about 90% are seen by either unskilled persons or traditional birth attendants with the risk of adverse birth outcomes and unrecorded births / birth defects. Clefts of the lip and/or palate (CLP) are the most common craniofacial birth defects with a worldwide birth prevalence of approximately 1/700 (2). It varies from 1/2500 to 1/500 births depending on the geographic origin, racial and ethnic backgrounds, and socioeconomic status (3,4). Das et al (1995) stated that Asians have the highest risk (14/10,000 births) followed by Whites (10 /10,000 births) and African Americans (4 /10,000 births) (5). There has been varying reports and rates from Africa. Khan reported a birth prevalence of 1.65/1,000 births in Kenya (6). Odihambo et al (2012) in a descriptive cross-sectional study at the Kenyatta National Hospital and Pumwani Maternity Hospital done from November 2006 to March 2007 found an incidence of preauricular tags and cleft lip and palate 1.5/1000 births (7). Suleiman et al (2005) reported a prevalence of 0.9 per 1,000 live births of OFC among a group of Sudanese hospital newborns in Khartoum (8). Teopista Kesande et al in (2014) in a retrospective analysis of births at two Ugandan hospitals found a prevalence of 0.77/1000 live births (9). The reported rates of Orofacial clefts in Nigeria are low. Iregbulem (1982) reported a prevalence of 0.3/1000 in the Eastern part of Nigeria (10). Butali et al (2014) reported a countrywide prevalence of 0.5/1000 (11). The incidence and prevalence of these anomalies in Ethiopia are not known and there are only two published reports about these anomalies. The first is the study among surgical patients less than 14 years of age admitted to the Ethio-Swedish children's hospital in Addis Ababa from 1984-1988. Among 2281 surgical patients treated, 183 (8%) were cleft cases (12). The second is a study conducted at Addis Ababa health institutions by Eshete et al (2011) that reported an incidence of 1.49/1000 live births (13). OFC represent significant public health problems because their treatment requires

comprehensive surgical, orthodontic, speech, and psychological management. As Christensen et al noted in 2004, in spite of these comprehensive management efforts, patients with OFC can experience lifelong psychosocial effects from the malformation (14). They also noted that the incidence of mental health problems is higher in individuals born with OFC. These complications are more severe in the developing world where medical care is limited. In the majority of the cases, affected individuals receive only a single surgical treatment.

In Ethiopia there is only one center, which provides multidisciplinary cleft care in the entire country. This center was established in 2003 in collaboration with the Cleft Lip and Palate Team in Bergen, Norway supported by Norwegian Agency for Development Cooperation (Norad) and later strengthened by Smile Train (American based charity organization, which organizes and supports free cleft surgical treatment in Ethiopia and other countries) and Transforming Faces (a Canadian based charity organization which supports holistic cleft care in Ethiopia and other countries). There are also other hospitals, which provide surgical treatment to cleft patients in collaboration with Smile Train. Patient population to these hospitals is a clear mix of urban and rural. The publicity regarding the care of individuals with clefts in Ethiopia is optimal and widespread to all areas at the moment. Our center and other hospitals, which provide cleft surgical treatment, are involved in surgical missions to rural areas to ensure that no cleft case is left untreated

Methods

After obtaining ethical clearance from Institutional Review Board College of Health Sciences Addis Ababa University 3.10/027/2015, and permission from Smile Train .We retrieved data from Smile Train database. This included data of all cleft patients who received single surgical treatment with the support of Smile Train at 31 hospitals. The hospitals are distributed throughout the country. Children and adults identified with clefts and provided with surgical repair are included in the database. All individuals with undiagnosed or unoperated cleft were not included. The dataset analyses is based on all Ethiopian cleft patients surgically treated at the above-mentioned institutions from June 2007 to December 2013 (N=18073). The cleft types were classified as bilateral cleft lip and palate (BCLP), unilateral cleft lip and palate (UCLP) further broken down into right and left CLP, Bilateral Cleft Lip only (BCLO) and Unilateral Cleft lip only (UCLO) - again broken down into right and left CLO and Cleft Palate only (CPO). This classification does not include the syndromic & atypical clefts. The cleft types were also divided into bilateral and unilateral in order to examine the cleft laterality. This was done by merging the left/right categories into a unilateral category. That is, instead of having LCLP and RCLP, we have now Unilateral Cleft Lip and Palate UCLP. Frequency Tables were constructed for the overall sample and stratified by gender. Exact binomial tests for differences in proportions were used for the whole population, and for each gender to assess whether there was a significant difference in the proportion of bilateral cleft lip and palate, left/right cleft lip and palate, and unilateral cleft lip and palate. An analogous procedure was followed for the cleft lip only cases. Information about immediate and distant relatives with clefts was also collected and frequency tables were constructed based on this information.

Results

During the study period 18,073 patients with cleft lip and palate were operated, Out of the total operated cleft patients, 8,232 are under seven years old. In these seven years (the study period) the total number of live births was 18,811,316. This gives an incidence of 0.44/1000 live births of orofacial clefts in Ethiopia, although this is likely to be an underestimate. We also estimated the prevalence to be 0.20/1000 using the total number of clefts (N=18,073) and number of total population in 2013 (N= 88,703,914). Individuals with no diagnosis, no cleft but prior unspecified surgeries are under the N/A category. The syndromic & atypical clefts are not included in the tables. Table 1 presents the distributions of these categories by gender based on the individuals that presented for surgery. We noted that these do not include termination of pregnancy, stillbirths, neonatal deaths with clefts and untreated cases or misdiagnosed cases. Overall, most of the individuals examined had LCLO. The category with the least individuals examined was CPO. When stratified by gender, these patterns remained the same.

The distributions of the cleft types by laterality and by gender are presented in Table 2. Most of the individuals had unilateral cleft lip only, and the category with the least individuals examined is cleft palate only.

Tests for differences in proportions were performed to see if there was a significant difference in the proportion of bilateral cleft lip and palate, left/right cleft lip and palate, and unilateral cleft lip and palate. An analogous procedure was followed for the cleft lip only cases. These assessments were done for all individuals (Table3).

Table 3 shows that the proportion of bilateral cleft lip and palate is smaller than the proportion of unilateral, right and left cleft lip and palate (all p-values <0.0001). It also shows that the proportion of left cleft lip and palate is smaller than the proportion of right cleft lip and palate (p-value 0.00367). Likewise, the proportion of bilateral cleft lip only is smaller than the proportion of right, left and unilateral cleft lip only (all p-values <0.0001). However, the proportion of left cleft lip only is bigger than the proportion of right cleft lip only (p-value <0.0001).

An analogous procedure was done but now stratifying by gender. The results were similar to the ones without the stratification. The only exception happens when we compare the proportion of left cleft lip and palate versus right cleft lip and palate among females. In this case, the difference is not significant at the $\alpha = 0.05$ level (Table 4).

Information about immediate and distant relatives with clefts was also collected. The frequencies and percent are shown in Table 5. The majority of the individuals reported that they did not have an immediate relative with cleft or a distant relative with cleft. Less than 2% of them had immediate or distant relative with cleft, and less than one percent reported that they did not know if they had either of those.

Discussion

There is no relevant information about orofacial clefts in Ethiopia. The multidisciplinary cleft care, which was started in 2003 in collaboration with the Cleft Lip and Palate Team in Bergen supported by Norwegian Agency for Development Cooperation (Norad) and strengthened by Transforming Faces and Smile Train created an opportunity for teaching and research. This research was done based on the database of Smile Train, which is the largest and most representative database available at the moment. It revealed an incidence of 0.44/1000 live births and prevalence of 0.20/1000. The distribution of the cleft types which is done for all operated cleft patients (18073) during the study period is: cleft lip only (CLO = 12831, cleft lip and palate (CLP=4632), and cleft palate only (CPO = 541). The number of isolated cleft palate in this study (3%) is low similar to other African studies (15, 16,11), It is also more common in males similar to the study done by Conway et al (16).

The incidence and prevalence rates reported in this study are less than what has been reported in Addis Ababa, Ethiopia by Eshete et al in 2011(13). They are lower than the Nigerian prevalence reported by Butali et al in 2014(11) and the prevalence report in African American by Gundlach (17). However, these rates are similar in the sense that they are lower than other population and consistent with what has been reported for clefts in Africa. Kesande et al (2014) in a retrospective analysis of births at two Ugandan hospitals found a prevalence of 0.77/1000 live births (9). This also is higher than our study. In our current study, most of the patients with orofacial clefts (55.6%) were males. This is similar to the Ugandan study (9) and the study conducted in Tanzania by Manyama et al (18). The study done by Martelli Junior et al (19) in a Brazilian population reported similar findings. They found 54.5 % males and 45.6% females. In the previous Ethiopian study done by Eshete et al (13) cleft lip alone and isolated cleft palate were more common in females, whereas cleft lip and palate were more common in males, in contradiction to the current study. In the current study all types of clefts including isolated cleft palate are more common in males than in females. This can be explained by the fact that this study captured only those patients who came to get surgical treatment, and the previous one captured all hospital deliveries at specified institutions. This might also be the reflection of the attitude of the community to give priority for males for everything including treatment. Isolated cleft lip constituted the most common type of cleft (70%), cleft lip and palate (26%), isolated cleft palate (3%). This is the same finding with a study done in Tanzania by Manyama et al (18). In their study isolated cleft lip constituted 49.2% of all cleft deformities, while clefts of both lip and palate and isolated cleft palate constituted 39.2% and 11.7% of cleft deformities respectively. In our study isolated cleft palate is low as it is in Manyama et al (18) study and other studies in Africa. One of the reasons for this could be lack of proper examination of the neonate before discharge from the delivery ward and unattended deliveries. Congenital anomalies like isolated cleft palate are not evident to everybody including parents and physicians unless a proper physical examination is done. It is very common to find patients with an isolated cleft palate who do not exactly know the pathology they have until adulthood at our set-ups. We think it is not different in other institutions in developing world. The other reason could be the higher mortality rate in these patients because of difficulties in feeding neonates and

infants in the absence of supportive feeding devices (10, 20). This raises several concerns that can be addressed by surveillance, community participation and education.

There is no established system of birth defect registry including orofacial clefts in Ethiopia (recent unpublished review). We think this has contributed to the non-existence of relevant information on the incidence of congenital anomalies including orofacial clefts. The main reason for planning and conducting this research is to obtain relevant information on the incidence and prevalence of orofacial clefts. We retrieved data from Smile Train database. During the past seven years more than 18,073 patients were operated at different hospitals. Of the total operated patients, 8232 were born and received surgical cleft repair during the study period. This data contains the information of all the patients operated during this period. It is limited by the use of data only from the hospitals and may not be representative of the true estimate of the prevalence. A population-based study is preferred but there is lack of resources human and capital to undertake such an exercise at this moment. However, the current study provides a baseline data on the prevalence and incidence that will serve as reference for future population based studies.

Limitation

This study is hospital based and captures only patients who presented for surgical treatment. This is bias and may not give a true reflection of the prevalence and incidence of clefting. However, it gives a baseline upon which future population studies can be conducted. Our rates are smaller than other African countries but the methodology reported in these other studies are different. Most of them reported single center data for the prevalence estimate, which is largely an under-ascertainment. This observation was reported by Butali and Mosey, 2009. Our approach is similar to other standardized studies in Africa conducted in Nigeria (11) and South Africa (Kromberg and Jenkins, 1982). Furthermore, our rates are comparable to rates reported by Kromberg and Jenkins, 1982 in South Africa and Butali et al., 2014 in Nigeria.

Conclusion and recommendation

The incidence rate (0.44/1000 live births) and prevalence rate (0.20/1000 population) found in this study are lower than previously reported in Ethiopia and other African countries. The reason for this lower rate could be that in the numerator we included only individuals who presented for surgery through the Smile Train outreach, considering the available surgical set-up. This finding could not be representative therefore we highly recommend establishing a system of birth defect registry to know the burden of birth defects including cleft lip and palate.

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Table 1**The distribution into the various cleft types by gender**

	BCLP	BCLO	CPO	LCLO	LCLP	N/A	RCLO	RCLP	Total
Female	344	764	248	2809	559	24	1086	626	6460
Male	902	1214	293	5002	1049	45	1956	1152	11613
Total	1246	1978	541	7811	1608	69	3042	1778	18073

BCLP- bilateral cleft lip and palate, BCLO- bilateral cleft lip only, CPO- cleft palate only, LCLO- left cleft lip only, LCLP- left cleft lip and palate, N/A- cleft type unknown, RCLO- right cleft lip only, RCLP- right cleft lip and palate.

Table 2

Laterality distribution of the cleft types by gender

	BCLP	UCLP	CPO	BCLO	UCLO	Total
Female	344	1185	248	764	3895	6436
Male	902	2201	293	1214	6958	11568
Total	1246	3386	541	1978	10853	18004

BCLP- bilateral cleft lip and palate, BCLO- bilateral cleft lip only, CPO- cleft palate only, UCLO- unilateral cleft lip only, UCLP- unilateral cleft lip and palate.

Table 3
Tests for difference in proportions

Test	Percent of first category	Confidence Interval	Exact p-value*
BCLP vs UCLP	26.90	(25.63, 28.20)	<0.0001
BCLP vs LCLP	43.66	(41.83, 45.50)	<0.0001
BCLP vs RCLP	41.20	(39.44, 42.98)	<0.0001
LCLP vs RCLP	47.49	(45.80, 49.19)	0.00367
BCLO vs UCLO	15.42	(14.79, 16.05)	<0.0001
BCLO vs LCLO	20.21	(19.41, 21.02)	<0.0001
BCLO vs RCLO	39.40	(38.05, 40.77)	<0.0001
LCLO vs RCLO	71.97	(71.12, 72.81)	<0.0001

BCLP- bilateral cleft lip and palate, UCLP- unilateral cleft lip and palate, BCLO- bilateral cleft lip only, UCLO- unilateral cleft lip only, LCLO- left cleft lip only, LCLP- left cleft lip and palate, RCLO- right cleft lip only, RCLP- right cleft lip and palate.

* Significance probability (p-value) associated with the test of the null hypothesis that equal proportions (50%) of subjects were found in the two cleft subcategories specified, assessed by the exact binomial test.

Table 4
Tests for difference in proportions by gender

Test	Female		Male	
	Percent of first category (95% CI)	Exact p-value *	Percent of first category (95% CI)	Exact p-value *
BCLP vs UCLP	22.50 (20.43, 24.68)	<0.0001	29.07 (27.48, 30.70)	<0.0001
BCLP vs LCLP	38.10 (34.92, 41.35)	<0.0001	46.23 (44, 48.47)	0.00094
BCLP vs RCLP	35.46 (32.45, 38.57)	<0.0001	43.91 (41.75, 46.09)	<0.0001
LCLP vs RCLP	47.17 (44.30, 50.06)	0.05516	47.66 (45.56, 49.77)	0.02967
BCLO vs UCLO	16.40 (15.35, 17.49)	<0.0001	14.86 (14.09, 15.65)	<0.0001
BCLO vs LCLO	21.38 (20.05, 22.76)	<0.0001	19.53 (18.55, 20.54)	<0.0001
BCLO vs RCLO	41.30 (39.04, 43.58)	<0.0001	38.30 (36.60, 40.01)	<0.0001
LCLO vs RCLO	72.12 (70.68, 73.52)	<0.0001	71.89 (70.82, 72.94)	<0.0001

BCLP- bilateral cleft lip and palate, BCLO- bilateral cleft lip only, LCLO- left cleft lip only, LCLP- left cleft lip and palate, RCLO- right cleft lip only, RCLP- right cleft lip and palate.

* Significance probability (p-value) associated with the test of the null hypothesis that equal proportions (50%) of subjects were found in the two cleft subcategories specified, assessed by the exact binomial test.

Table 5
Relatives with a diagnosis of cleft

Test	Immediate Relative with Clefts		Distant Relative with clefts	
	Frequency	Percent	Frequency	Percent
Yes	351	1.95	272	1.51
No	17609	97.81	17688	98.24
Do not know	44	0.24	44	0.24