

## Incidence of Cleft Deformities among Neonates in Mulago National Referral hospital, Uganda

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**Background:** Cleft deformities (lip and palate) have been reported to be the most common congenital craniofacial anomaly in several settings. In Uganda, though two previous studies were conducted to determine the incidence of cleft lip and palate, the estimates obtained from those studies may not be precise given the study settings. This study was undertaken to establish the incidence of cleft deformities and provide data to plan for better management of these deformities. The Setting was the labour wards at Mulago National Referral Hospital, Kampala Uganda. The main objective of this study was to determine the incidence of cleft deformities (lip and palate) among neonates born between February 2008 and February 2009

**Methods:** Cross-sectional study of all neonates who were born in Mulago Hospital. We examined all new born children and determined the presence or absence of cleft lip and/palate. Socio-demographic data and risk factors such as smoking, alcohol consumption, infections and exposure to drugs such as anti-convulsants and steroids were collected.

**Results:** Among twelve thousands seven hundred and thirteen neonates born in Mulago hospital between February 2008 and February 2009, seventeen neonates presented with cleft lip with or without cleft palate: this gives an incidence of 1.34 per 1000 newborns or 134 in 100,000 newborns.

**Conclusion:** The incidence of neonatal cleft deformities seems to lie between what was previously reported in 1961 and 1996 in Uganda.

### Introduction

Cleft deformities (lip and palate) have been reported to be the most common congenital craniofacial anomaly in several settings. In Uganda, though two previous studies were conducted to determine the incidence of cleft lip and palate, the estimates obtained from those studies may not be precise given the study settings. The first study by Simpkins and Lowe<sup>1</sup> in 1961 was a retrospective study from hospital records which were often poor or incomplete; the study gave the incidence of clefts to be 1.45 per 1000. The second study was done in 1996 by Byarugaba and Mirembe<sup>2</sup>, they reported on newborns admitted in the special care unit, and found the incidence to be 0.5 per 1000. Special care Unit admits only infants with complications within twenty four hours after birth, so many infants with clefts but in stable condition may have been missed during the study due to a selection bias. Our study looked at all neonates born in Mulago hospital labour wards and also those in Special care unit.

This study was undertaken to establish the incidence of cleft deformities and provide data to plan for better management of these deformities. Further more data from this study would help set up better counseling and prevention programmes with data specific to Uganda. This study would help the design of early warning system to detect

for example; new teratogens in the environment or changes in the incidence of the cleft lip and palate

The study sites comprised of Mulago Hospital labour wards and special care unit. Mulago Hospital is the National Referral hospital of Uganda, with 1500 beds; it's the largest hospital in Uganda. Uganda has a population of 34 million people, however only 41% of deliveries occur in hospital according to the Uganda Bureau of Statistics. Mulago hospital is a University teaching hospital, and serves a widely varied socio economic and ethnic population. Because of its location in Kampala which houses the government, some major industries, and is the centre of commerce, it conducts deliveries for women from all over the country. This study was approved by Mulago Research and Ethics Committee.

### **Patients and method**

A cross-sectional descriptive study of all neonates who were born in Mulago Hospital during a period of one year, February 2008 to February 2009, was undertaken. Informed consent was sought from all the mothers of the neonates. We examined all new born children and determined the presence or absence of cleft lip and/palate. The clefts were described according to Smith's modification of Kernahan's "y" classification<sup>3</sup>. This classification system was used for all cleft cases. A stamped symbolic representation appropriately filled out for the type of cleft anomaly was included in the questionnaire. Related congenital abnormalities if present were also recorded. Socio-demographic data and risk factors such as smoking, alcohol consumption, infections and exposure to known teratogenic drugs e.g., anti-convulsants and steroids were collected. Participating midwives were especially trained to look out for isolated cleft palates using a tongue depressor and examination light. Neonates with clefts in the study were followed up in the plastic outpatient clinic and operated on at the appropriate time. Frequencies and proportions were used to examine the distribution of categorical variables, whereas for continuous variables, means and their standard deviations were used.

Contingency tables and chi-square analysis tested for the association between categorical variables and for continuous variables. The student-t tests were used to test for significant differences.

### **Results**

Among twelve thousands seven hundred and thirteen neonates born in Mulago hospital between February 2008 and February 2009, seventeen neonates were born with cleft lip with or without cleft palate: this gives an incidence of 1.34 per 1000 newborns or 134 in 100,000 new borns. There were more cleft lips (CL) than cleft lip and palate (CLP); bilateral cleft lips and or palate were only 3/17 cases seen, (Table 1).

Out of the total number of neonates (12713) seen during the study period, complete data regarding risk factors for 8733 neonates could be accessed (Tables 2 and 3).

Only one mother had a relative with clefts.

The most common associated anomalies were poly dactyl, accessory auricles, hypospadias and spine bifida

**Table 1** Sex distribution of cleft deformities among neonates born in Mulago hospital, (n=12713) February 2008 to February 2009

Variable	Cleft Deformity (n=17)	No Cleft Deformity (n=12696)	Total	Incidence/ 100000 population
Male	5	6345	6350	79
Female	12	6351	6363	189
Total	17	12696	12713	134

**Table 2.** Distribution of cleft deformities according to site among 17 neonates born in Mulago hospital, February 2008 to 2009

Cleft Lip		Isolated Cleft Palate	Bilateral Cleft Lip	Total
Left	Right			
8 (47.0%)	4 (23.6%)	2 (11.8%)	3 (17.6%)	17

**Table 3.** Comparison of parental risk factors between neonates with cleft deformities versus those without (n=8746) in Mulago hospital, 2009

Variable	Cleft Deformity (n=13)	No Cleft Deformity (n=8733)	p-value
Took Mumbwa during Pregnancy Yes n (%)	10 (76.9)	7816 (89.5)	0.14
Ever Used Contraception Yes n (%)	8 (61.5)	3855 (44.1)	0.207
Suffered from Syphilis during pregnancy Yes n (%)	3 (23.1)	1533 (17.6)	0.601
Mother's Parity Prime Gravida n (%)	2 (15.4)	2964 (33.9)	0.158
Alcohol intake during Pregnancy Yes n (%)	2 (15.4)	757 (8.7)	0.39
Mother's age Mean (SD)	24.5 (5.3)	24.0 (5.1)	0.733
Husband's age Mean (SD)	31.4 (4.8)	30.1 (6.6)	0.442

## Discussion

The incidence of cleft lip and palate varies very much among different countries and ethnic groups in the same region. Globally it is been noted to be highest among North American Indians (1:500), Orientals (1:600) and Caucasians (1:750)<sup>4,5,6,7</sup>. The stated figure for Africans

(0.4/1000), is grossly underestimated as evidenced by some studies from different African countries: Angola(1.35/1000),Zaire(10.46/1000),Tunisia(1.49/1000),Ethiopia(1.49/1000),and Kenya(1.92/1000).

From our study, the incidence of cleft lip and palate in Mulago hospital is 1.34/1000. This incidence lies between that from the two previous studies done in this hospital. Simpkin and Lowe found it to be 1.46/1000 in 1961 and Byarugaba and Mirembe found it to be 0.5/1000 in 1996 from a total of 12391 neonates. The differences may arise because of the different study designs: Simpkins and Lowe used a retrospective study from hospital records which often have been found to be poor or incomplete since they are not meant for research purposes. Whereas Byarugaba and Mirembe dealt with newborns from Special Care Unit which normally admits neonates with complications got within the first 24 hours of birth; so some newborns with clefts but without other life threatening complications were left out. The incidence of cleft lip and palate got from our study should be reflective of the national incidence because Mulago hospital has the largest number of hospital births in the country. Also being in Kampala the capital city, it conducts deliveries of women from diverse ethnic backgrounds.

The high incidence of clefts in African studies reported of recent (1:746 in this study) as compared to previously reported African Average of 0.4/1000 may be explained by the improved diagnosis and registration seen today in most African hospitals. Fogh- Andersen explained the lower incidence found in previous studies by saying that in earlier studies, registration of isolated cleft palates may have been missed and minor cleft lips may not have been registered if operation was not deemed imperative. From our results, despite few clefts seen (1:746), the findings are comparable with those from other African studies.

#### **Severity of deformity**

58.3% of newborns presented with CL alone against 41.7% who presented with CLP. This compares with the 58% CL reported from Nigeria by Oluwasanmi and Adekunle (1970), 59% by Adekeye and Lavery (1985) and 50% by Orrett and Ogle from Zaire (1993). Isolated cleft palate constituted 11.8% of the total clefts while, the reported incidence of isolated cleft palate from African populations is between 15 to 20% in sharp contrast with the Caucasian population of between 30 to 40%. In our sample population, 17.6% had bilateral cleft lip/palate. Simpkin and Lowe in 1961 only noted the low occurrence of bilateral clefts in their study; whereas Byarugaba and Mirembe did not comment on the laterality of cleft cases seen in their study.

#### **Sex**

For CLP, there were more females born with clefts 189/100000 compared to males 79/100000. This trend has been noted from other studies elsewhere<sup>6, 7, 8, 9</sup>. Furthermore there more left sided clefts than right sided in keeping with other studies. The isolated cleft palates were all females also as reported else where<sup>8, 9, 10, 11</sup>. The exact reasons for this are not yet clear.

#### **Risk factors**

Most mothers (80%) had taken "emumbwa" during pregnancy. This is a common clay spindle which is sun baked with herbal medicine whose exact content is unknown. It is widely used in the central parts of Uganda. Association of usage of emumbwa and development of cleft deformity was not statistically significant. The gestational period in which these drugs were taken was not stated so we could not ascertain their influence on the embryogenesis of the neonates born with clefts. 5/17 mothers reported to have suffered from syphilis in the past; although it is considered to be one of the teratogenic infections, the association was not

statistically significant. Most of the mothers were multiparous (76.5%) which contrasts from the study by Byarugaba and Mirembe who found the majority of mothers with malformed babies to be primigravidi. From our study, parity as a cause of clefts was not statistically significant. Other studies<sup>18,19</sup> reported the same result.

Similarly, parental age was not an important factor for the development of cleft lip and palate. This was found to be true in other studies<sup>18,20</sup>; whereas Habib<sup>16</sup> found an increased incidence of cleft lip and palate among older parents. There was only one positive family history of clefts(5.8%) a maternal uncle of a mother had a baby born with incomplete CL. Koguchi<sup>13</sup> reported a risk of recurrence for CLP in first degree relatives of 4.1%. Jensen reported that 7% of oral clefts cases had affected first order relatives, while Fogh-Andersen<sup>14</sup> found that 10.4% of cases had affected first order relatives.

### **Associated anomalies**

Overall, 4/17 or 23.5% of mothers had babies born with CLP associated with other anomalies. Two of these had multiple abnormalities; talipes and spina bifida, whereas the other two had polydactyl, accessory auricles and hypospadias. Byarugaba and Mirembe noted that 17.2% of neonates in Special Care unit had musculoskeletal malformations and that 23.2% had neural tube defects.

### **Conclusions**

- The incidence of clefts among neonates in Mulago hospital is 134/100,000 which is actually high compared to the African average of 40/100,000.
- The pattern of clefts seen in Mulago hospital was similar to that reported from other African countries
- No apparent risk factor could be ascertained from this study

### **Recommendations**

There is need for a national congenital malformations register to establish the occurrence and patterns of these malformations countrywide

There is a need to setup specialised multidisiplinary treatment centres in the country to manage better these malformations

There is a need to design an analytical case-control study to establish possible risk factors for clefts.

### **Study limitations**

Not all mothers in Uganda deliver in the hospital. Inheritably cleft incidences among Africans are low necessitating very large sample sizes often limited by resources. Because of financial constraints the study could only last for one year, so this study could not capture yearly fluctuations.

### **References**

1. Simpkins M, Lowe A Congenital abnormalities in the African new born. , : Arch Dis Child 1961; 36: 404-406
2. Byarugaba E W, Mirembe F: valuation of foetal and infant dysmorphisms in Mulago Hospital, Uganda. Tropical Health vol. VI no 2 august 1996

3. Smith AW, Khoo AK and Jackson I J: A modification of the Kernahan "Y" classification in cleft lip and palate deformities. *Plast. Reconstr. Surg* 102: 1842, 1998
4. Derijcke A, Eerens A and Carels C: The incidence of oral clefts: A review. *Br. J Oral maxillofac Surg.* 34: 488,1996
5. Owens JR, Jones JW, Harris F: Epidemiology facial clefting. *Arch Dis Child* 1985; 60:521-4
6. Vanderas AP: Incidence of cleft lip, cleft palate and cleft lip and palate among races: A review. *Cleft Palate J* 1987, 24: 216-225
7. Hagberg C, Larson O, Milerad J: Incidence of cleft lip and palate and risks of additional malformations *Cleft Palate Craniofac J* 1998, 35:40-5
8. Suleiman Am, Hamzah ST, Abusalab MA : Prevalence of cleft lip and palate a hospital based population in Sudan. *Int J Paediatr Dent* 2005, 15: 185-9
9. Sullivan WG: Cleft lip with and without cleft palate in blacks: Analysis of 81 patients. *Plast. Reconstr. Surg.* 84: 406, 1989
10. Adekeye EO, Lavery Km: Cleft lip and palate in Nigerian children and adults. a comparative study. *Br J Oral Maxillofac Surg* 1985, 23:398-403
11. In Orrett E Ogle: Incidence of cleft lip and palate in a new born Zairian sample. *Cleft Palate Craniofac. J* 30: 250, 1993
12. C Oluwasanmi JO, Adekunle OO: Congenital clefts of the face in Nigeria. *Plast. Reconstr. Surg.* 1970, 46:246-251
13. Irebulim LM: The incidence of cleft lip and palate in Nigeria. *Cleft Palate J* 19:201,1982
14. Koguchi A: Recurrence rate in offspring and siblings of patients with cleft lip and/or cleft palate. *Jpn J Hum Genet* 1975, 20,207-229
15. Fogh-Andersen P: Genetic and non genetic factors in the aetiology of facial clefts. *Scand J Plast Reconstr Surg* 1967, 1: 22-29
16. Habib Z: Factors determining occurrence of cleft lip and cleft palate. *Surg Gynecol Obstet* 1978, 146: 105-10
17. Rintala AE, Gyling U: Birth weight of infants with cleft lip and palate. *Scand J Plast Reconstr Surg* 1967, 1:109-12
18. Jamilian A, Nayeri F, Babayan A Incidence of cleft lip and palate in Tehran.. *J Indian Soc Pedod Prevent Dent.* December 2007: 174-6
19. Rajabian MH, Sherkat M: An epidemiologic study of oral clefts in Iran: Analysis of 1669 cases. *Cleft Palate Craniofac J* 2000, 37: 191-6
20. Bianco-Davila F: Incidence of cleft lip and palate in northeast of Mexico. A 10 year study. *J Craniofac Surg* 2003, 14: 533-7