

Orofacial cleft patterns and risk factors among patients at Mbeya Zonal Referral Hospital, Tanzania: A retrospective descriptive study

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Abstract

Background: Despite the global impact of orofacial clefts, there is limited information on their patterns and risk factors in Tanzania. This study aims to examine the patterns and risk factors of orofacial clefts among patients in the Southern Highlands of Tanzania treated at Mbeya Zonal Referral Hospital in collaboration with the Smile Train program.

Methods: A retrospective descriptive study was conducted at Mbeya Zonal Referral Hospital, analyzing data from November 2021 to June 2022 obtained from the Smile Train database. Descriptive and analytical methods were used with Stata 14.0, and a p-value of <0.05 was considered statistically significant.

Results: A total of 65 patients were treated during the study period, with 64.6% being male. The median age at the procedure was six months (IQR 3-10 months). The majority (33.8%) of patients were from Chunya district. The most common type of cleft was cleft lip (83.1%), followed by clefts of the hard and soft palate. Nine patients had clefts involving the lip, hard palate, and soft palate. Age at the procedure was associated with the severity of the cleft (OR 5.45, Cl: 1.09-27.28, p-value: 0.039). No significant associations were found between cleft severity and gender, family history, antenatal clinic attendance, or other malformations.

Conclusions: Understanding the patterns and types of orofacial clefts through routine clinical data is crucial for assessing the disease burden and allocating resources effectively. This study underscores the importance of routine data collection in identifying trends and potential areas for further research. Additional comprehensive studies are needed to investigate the causal factors and broader implications of these findings.

Keywords: Orofacial Clefts, Smile Train Program, Mbeya Zonal Referral Hospital, Epidemiological Patterns, Congenital Anomalies.

Background

Orofacial clefts are considered the most common congenital malformations of the head and neck region, compromising about 65% of all head and neck anomalies identified among live births (Antoszewski & Fijałkowska, 2016). The global prevalence of cleft lip and palate has shown a decline in recent years; however, data quality remains inconsistent across regions, often lacking crucial information on variables such as gender and the severity of morphological abnormalities (Zhou et al., 2022). In Tanzania, available data, largely derived from a study conducted between October 2004 and July 2009, indicated a prevalence of 2.8% associated with congenital anomalies (Manyama et al., 2011). The etiology of orofacial clefts is complex, involving multiple genetic and epigenetic (environmental) factors (Brender & Weyer, 2016). Notably, some environmental factors, such as temperature variation,

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exhibit seasonal patterns that have been correlated with an increased incidence of orofacial clefts (Wehrung & Hay, 1970).

Several environmental and lifestyle factors contribute to the incidence of orofacial clefts, with maternal smoking during early pregnancy associated with a 1.5- to 2-fold increased risk of these congenital anomalies (Shaw et al., 1996). Additionally, gender disparities exist, with males generally presenting milder forms of cleft lip, cleft lip and palate, or cleft palate, while females tend to experience more severe cases (Nagase et al., 2010). Moreover, individuals with orofacial clefts often exhibit poorer work ability and a lower quality of life compared to those without congenital deformities (Reis et al., 2018). Early rehabilitation and intervention have shown substantial improvements in health-related quality of life and work ability (Phua & de Chalain, 2008). In Tanzania, limited research has been conducted on the prevalence and risk factors of orofacial clefts, highlighting the need for further studies to understand these factors better and to develop effective treatment and prevention strategies.

The management and monitoring of orofacial clefts are crucial for the affected individuals' speech, hearing, feeding, and maxillary growth (Phua & de Chalain, 2008). The Mbeya Zonal Referral Hospital, in collaboration with the Smile Train program, has been at the forefront of managing orofacial clefts in the Southern Highlands of Tanzania. This study aims to describe the patterns of orofacial clefts among patients managed by the hospital, focusing on patients from November 2021 to June 2022. By using data from the SMILE TRAIN database, this research seeks to fill the knowledge gap, provide a better understanding of the burden, and aid in the appropriate allocation of healthcare resources for affected populations. This study also presents an opportunity to identify potential areas for further research, ensuring improved outcomes for patients with orofacial clefts.

Methods

Study settings and location

This study was conducted at the Mbeya Zonal Referral Hospital, the largest government health facility in the Southern Highlands zone of Tanzania. The hospital receives referral patients from seven regions: Njombe, Ruvuma, Songwe, Rukwa, Iringa, Katavi, and Mbeya.

Study design

This research was a retrospective descriptive study aimed at understanding the pattern of orofacial clefts among patients from the Southern Highlands of Tanzania. Data were collected from November 2021 to June 2022 from the SMILE TRAIN database and hospital records.:

Sample size

The study included 65 patients who were attended to for orofacial clefts during the specified period. The sample comprised children with cleft lip alone, cleft palate alone, and those with both cleft lip and palate (soft and hard).

Data collection

Data were obtained from documented patient records at the hospital, the electronic medical system of the Mbeya Zonal Referral Hospital, and the SMILE TRAIN database. Key variables included patient demographics, clinical details of the orofacial cleft, family history, and maternal history, such as antenatal clinic attendance and alcohol use during pregnancy.

Study instruments

Data collection was facilitated through standardized forms used in the hospital and SMILE TRAIN programs. Information was systematically reviewed and recorded by trained data collectors to ensure accuracy and completeness.

Study analysis



Data were cleaned and analyzed using Stata 14.0. Descriptive statistical methods such as frequencies and percentages were used for categorical variables, while means, standard deviations, medians, and interquartile ranges were used for numerical data. The chi-square test or Fisher exact test measured associations between variables. Multivariable logistic regression models were applied to adjust for potential confounding effects. A p-value of <0.05 was considered statistically significant. Data was presented in tables and charts to visualize findings comprehensively.

Results

From November 2021 to June 2022, a total of 65 patients with orofacial clefts were treated at the Mbeya Zonal Referral Hospital with support from the Smile Train program. The majority of the patients were male (64.6%), and the median age at the time of the procedure was six months (IQR 3-10 months). Most of the procedures were performed between the ages of 4 and 12 months. A significant proportion of the patients (33.8%) were residents of the Chunya district, while others came from various regions within the Southern Highlands (Table 1).

Characteristic		Frequency (%)
Gender	Female	23 (35.4)
	Male	42 (64.6)
Age at procedure*	1-3 months	18 (27.7)
	4-12 months	37 (56.9)
	>1 year	10 (15.4)
Location	Kyela	1 (1.5)
	Chunya	22 (33.8)
	Mbarali	8 (12.3)
	Mbeya	10 (15.4)
	Mbozi	8 (12.3)
	Rungwe	2 (3.1)
	Songwe	10 (15.4)
	Other	4 (6.1)
Severity of the cleft	Less severe	43 (66.1)
	Severe	22 (33.9)
Procedure done	Cleft palate	12 (18.5)
	Lip bilateral	5 (7.7)
	Lip unilateral	48 (73.8)
Family history of any cleft	No	53 (81.5)
	Yes	12 (18.5)
Maternal alcohol history	No	55 (84.6)
-	Yes	10 (15.4)
Antenatal clinic booking**	Early	52 (81.2)
C C	Late	12 (18.8)
Other congenital malformations	None	57 (87.7)
Procedure done	Cardiac	2 (3.1)
	Club feet	2 (3.1)
	Genital	3 (4.6)
	Talipes equinovarus	1 (1.5)

Table 1: Characteristics of patients attended (n=65)

* It is recommended that clefts are repaired for at least three months unless the patient is not physically fit for the procedure.

** As per WHO recommendation, one should have started antenatal visits at 17 weeks of gestation



Table 2 shows the frequency of the cleft lip and palate by anatomical distribution. Thirty-three patients had a left cleft lip, while 27 had a right-side cleft lip. The majority (65%) of those with a cleft lip had a complete cleft. A total of 19 patients had a left-side cleft of the hard palate, while 6 had a right-sided cleft hard palate. The majority (21/25) of these patients had a complete cleft of the hard palate. Twenty-three patients presented with a cleft lip, which involved the left alveolar, while 15 presented with a cleft lip with right alveolar involvement. Twenty patients had a cleft of the soft palate and among these patients, 65% had complete cleft soft palate (Table 2).

Characteristics (n)		Frequency (%)
Left cleft lip (33)	Complete	23 (69.7)
	Incomplete	10 (30.3)
Right cleft lip (27)	Complete	16 (59.3)
	Incomplete	11 (40.7)
Left cleft hard palate (19)	Complete	15 (78.9)
	Incomplete	4 (21.1)
Right cleft hard palate (6)	Complete	6 (100)
	Incomplete	o (o)
Cleft lip with left alveolar involvement (23)	Complete	13 (56.5)
	Incomplete	10 (43.5)
Cleft lip with right alveolar involvement (15)	Complete	9 (60.0)
	Incomplete	6 (40.0)
Cleft soft palate (20)	Complete	13 (65.0)
	Incomplete	7 (35.0)

Table 2: Showing frequencies of cleft lip and palate by anatomical distribution

When assessing the anatomical overlap of the clefts, nine patients had a cleft involving both the lip, hard and soft palate. Seven patients had both a hard and soft cleft palate. Only four patients had cleft of the soft palate (Figure 1).

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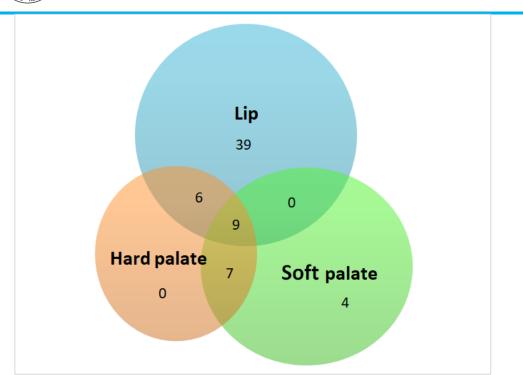


Figure 1: A Venn diagram showing the frequencies and anatomical overlap of clefts n=65

A total of 22 patients were classified as having a severe cleft, depending on the anatomical position and overlap of different clefts. Age at procedure was associated with severity of the cleft, where those aged 4-12 months had 5.45 times the odds of having a severe cleft compared to those aged 1-3 months (Cl; 1.09-27.28, p-value; 0.039) and those aged more than one year had eight times the odds of a severe cleft compared to those aged 1-3 months (Cl; 1.17-54.72, p-value; 0.034). Patients whose mothers had a positive maternal alcohol history had about three times the odds of a severe cleft compared to those who did not have an alcohol history (OR; 3.66, Cl; 0.91-14.72), though there was no statistical evidence for this association. Other factors such as gender, family history of cleft, time of antenatal clinic booking and presence of other malformations did not show any evidence of an association with severity of cleft (Table 3).

Characteristic	Total (%)	n (%)	OR (CI)	P-value†	
Gender					
Female	23 (35.4)	8 (34.8)	1		
Male	42 (64.6)	14 (33.3)	0.94 (0.32, 2.74)	0.906	
Age at procedure*					
1-3 months	18 (27.7)	2 (11.1)	1		
4-12 months	37 (56.9)	15 (40.5)	5.45 (1.09, 27.28)	0.039	
>1 year	10 (15.4)	5 (50.0)	8 (1.17, 54.72)	0.034	
Family history of any cleft					
No	53 (81.5)	17 (32.1)	1		
Yes	12 (18.5)	5 (41.7)	1.51 (0.42, 5.46)	0.531	
Maternal alcohol history					
No	55 (84.6)	16 (29.1)	1		
Yes	10 (15.4)	6 (60.0)	3.66 (0.91, 14.72)	0.064	



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Antenatal clinic booking**				
Early	52 (81.2)	19 (36.5)	1	
Late	12 (18.8)	3 (25.0)	0.58 (0.14, 2.40)	0.439
Other malformations				
No	57 (87.7)	20 (35.1)	1	
Yes	8 (12.3)	2 (25.0)	0.62 (0.11, 3.34)	0.564

Severity was defined as any co-occurrence of a cleft lip (be it unilateral or bilateral) and a cleft palate or just a cleft palate itself. Two categories were created "severe" and "less severe"

* It is recommended that clefts are repaired at least at three months of age unless the patient is not physically fit for the procedure.

** As per WHO recommendation, one should have started antenatal visits at 17 weeks of gestation. † LR test p-value

Table 4 shows the association between patient characteristics and different cleft types. There was strong evidence of an association between gender and cleft of the alveolar, where male patients had 0.22 times the odds of cleft of the alveolar compared to the females (CI; 0.07-0.67, p-value 0.005). However, other anatomical clefts did not show an association with gender. Cleft lip and cleft palate were strongly associated with age at the procedure, where older ages had higher odds of both lip and palate clefts. However, no association was observed between cleft of the alveolar and age at the procedure (Table 4). Other factors such as family history of any cleft, maternal alcohol history and time of antenatal booking of the mothers of patients and presence of other malformations did not show any association with any of the anatomical types of clefts.



Table 4: She	<u> </u>			patient			ut differe			5)
	Total	Cleft Lip			Cleft Palate ††			Cleft Alveolar		
Characteristic	N (%)	n (%)	OR (CI)	Pvalue†	n (%)	OR (CI)	Pvalue†	n (%)	OR (CI)	Pvalue
Gender										
Female	23	21	1		9	1		17	1	
	(35.4)	(91.3)			(39.1)			(73.9)		
Male	42	33	0.35	0.171	17	1.06 (0.37,	0.916	16	0.22 (0.07,	0.005
	(64.6)	(78.6)	(0.07,		(40.5)	2.99)		(38.1)	0.67)	
			1.78)							
Age at										
procedure*										
1-3 months	18	18	1		2 (11.1)	1		11	1	
	(27.7)	(100)						(61.1)		
4-12 months	37	32	9.6	0.005	18	7.58(1.52,	0.013	19	0.67 (0.21,	0.496
	(56.9)	(86.5)	(1.98,		(48.7)	37.73)		(51.4)	2.11)	
			46.5)							
>1 year	10	4	1		6	12 (1.73,	0.012	3	0.27 (0.05,	0.123
	(15.4)	(40.0)			(60.0)	83.45)		(30.0)	1.42)	
Family history of a	any cleft									
No	53	44	1		19	1		25	1	
	(81.5)	(83.0)			(35.9)			(47.2)		
Yes	12	10	1.02	0.979	7	2.50 (0.69,	0.155	8	2.24 (0.60,	0.218
	(18.5)	(83.3)	(0.19,		(58.3)	8.98)		(66.7)	8.35)	
			5.48)							
Maternal alcohol										
history										
No	55	46	1		20	1		26	1	
	(84.6)	(83.6)			(36.4)			(47.3)		
Yes	10	8	0.78	0.782	6	2.63(0.66,	0.165	7	2.60(0.61,	0.180
	(15.4)	(80.0)	(0.14,		(60.0)	10.42)		(70.0)	11.12)	
			4.31)							
Antenatal clinic be	-									
Early	52	44	1		20	1		29	1	
	(81.2)	(84.6)			(38.5)			(55.8)		
Late	12	10	0.91	0.912	5	1.14 (0.32,	0.837	4	0.39 (0.11,	0.158
	(18.8)	(83.3)	(0.17,		(41.7)	4.09)		(33.3)	1.48)	
			4.95)							
Other										
malformations										
No	57	47	1		23	1		30	1	
	(87.7)	(82.3)			(40.4)			(52.6)		
Yes	8	7	1.49	0.713	3	0.89 (0.19,	0.877	3	0.54 (0.12,	0.421
	(12.3)	(87.5)	(0.16,		(37.5)	4.08)		(37.5)	2.48)	
			13.4)							

* It is recommended that clefts are repaired at least at three months of age unless the patient is not physically fit for the procedure.

** As per WHO recommendation, one should have started antenatal visits at 17 weeks of gestation.

† LR-test p-value except for age at procedure

*††*This included both soft and hard palates.

Discussion

This study is one of the few in Tanzania and the first in the Southern Highlands to describe the patterns and explore the factors associated with orofacial clefts. The study's findings indicate a higher prevalence of orofacial clefts among males, accounting for 64.6% of the total cases. This male predominance aligns with previous research conducted in Mwanza, Tanzania, and Tokyo, Japan,



where similar patterns were observed (Brender & Weyer, 2016; Manyama et al., 2011). However, contrasting results were reported in South Africa, underscoring potential geographic and demographic variations in orofacial cleft incidence (Hlongwa et al., 2019). The predominance of cleft lip over other types of orofacial clefts was another key finding, with 83.1% of cases presenting cleft lips. This result is consistent with studies conducted in Mwanza, Nigeria, and Norway, where cleft lips were also more prevalent (Hlongwa et al., 2019; Kurita et al., 2021; Manyama et al., 2011). These findings highlight the need for region-specific studies to better understand the epidemiology of orofacial clefts.

Notably, 33.8% of the patients in this study were residents of the Chunya district, an area known primarily for mining activities and agricultural undertakings. This high prevalence rate calls for epidemiological studies to explore the potential causal link between environmental factors, such as mining activities, and the incidence of orofacial clefts. Environmental factors, such as exposure to harmful chemicals and poor maternal nutrition associated with such occupational environments, could be contributing factors (Candotto et al., 2019; Kurita et al., 2021; Wilcox et al., 2007). The large influx of patients from a specific geographic location emphasizes the need for targeted public health interventions and awareness campaigns in high-risk areas to mitigate the prevalence of orofacial clefts.

The median age at the procedure was six months, with most patients undergoing surgery between four and twelve months. This relatively early timing of surgical intervention aligns with recommended practices and reflects the well-coordinated efforts of the Smile Train program in the region (Louis et al., 2018). Early surgical intervention is crucial for improving outcomes related to speech, hearing, feeding, and maxillary growth (Aycart & Caterson, 2023). However, age at the procedure was also found to be positively associated with the severity of the orofacial cleft, suggesting that more severe cases might require timely and multiple surgical interventions. This association should be interpreted cautiously, as the timing of surgery also depends on the patient's general health and readiness for surgery rather than delayed presentation to healthcare facilities (Hamdan et al., 2023). This finding underscores the importance of early diagnosis and comprehensive care protocols to address the complexities of severe orofacial clefts.

The study also explored the association between various environmental factors and the severity of orofacial clefts. While maternal alcohol consumption was associated with about three times the odds of having a severe cleft, the association did not reach statistical significance, possibly due to the small sample size and potential responder bias (Kurita et al., 2021). Although epidemiological studies have shown the role of environmental factors such as alcohol and smoking in the onset of orofacial malformations, these factors were not significantly represented in this study (Fell et al., 2022; Inchingolo et al., 2022). Environmental exposure and genetic predispositions are complex and require larger, multi-centered studies to draw definitive conclusions. Moreover, the lack of significant associations between gender, family history, time of antenatal clinic booking, and the presence of other malformations with the severity of orofacial clefts calls for further investigations into other potential risk factors.

This study's main limitation lies in its small sample size, which restricted the ability to conduct more robust statistical analyzes and multivariable regressions to adjust for potential confounding variables. The hospital-based nature of the dataset may also limit the generalizability of the findings to a larger population, as it may not represent undiagnosed or untreated cases in the community. Furthermore, the reliance on routine data collection and patient records may lead to information gaps or inaccuracies, impacting the study's comprehensiveness. Social desirability bias might have influenced the self-reporting of maternal habits, such as alcohol consumption, leading to underreporting. For future research, larger, population-based studies with detailed data collection and possibly genetic analysis are recommended to provide a more comprehensive understanding of the



patterns and risk factors associated with orofacial clefts. Despite these limitations, this study contributes valuable insights into the epidemiology of orofacial clefts in the Southern Highlands of Tanzania and underlines the importance of targeted public health interventions and further research to improve outcomes for affected individuals.

Conclusions

This study offers valuable insights into the patterns and risk factors associated with orofacial clefts among patients in the Southern Highlands of Tanzania, particularly those treated at the Mbeya Zonal Referral Hospital in collaboration with the Smile Train program. The predominance of cleft lip, especially on the left side, and the higher incidence among males align with global observations but also underscore the need for region-specific data to address unique environmental and genetic factors. A significant proportion of patients from the Chunya district point to potential environmental exposures that warrant further epidemiological investigation. Early surgical intervention, facilitated by the Smile Train initiative, has demonstrated effectiveness in managing these congenital anomalies, emphasizing the importance of timely medical care. While factors like maternal alcohol consumption showed a positive association with cleft severity, the small sample size limits definitive conclusions.

Additionally, no significant associations were found between cleft severity and gender, family history, or antenatal care timing, suggesting the need for more extensive, multi-centered studies to unravel the complex etiology of orofacial clefts. Understanding these factors is crucial for developing targeted public health interventions and allocating resources effectively. This study highlights the importance of routine data in informing healthcare practices and identifying gaps in current research. Future studies should aim for larger sample sizes and possibly incorporate genetic analyzes to provide a more comprehensive understanding. Despite these limitations, this research contributes to the foundational knowledge required to improve healthcare outcomes for individuals with orofacial clefts in Tanzania.

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Author's contributions

HM, AL, WO and OH designed the study and wrote the manuscript. HM, AL, WO, OH, FN, EM, CNM and OH reviewed and edited the manuscript. HM, AL and CNM contributed to the final manuscript. All authors reviewed the manuscript.

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None

Availability of data and materials

All data generated or analyzed during this study is included in this published article.

Declarations

Ethical approval and consent to participate

Ethical clearance to carry out the study was sought from Mbeya Research Committee. Permission was obtained from the Executive director of Mbeya Zonal Referral Hospital and the director of NIMR - MMRC and informed consent was also sought from patients and patient's parents for patients below 18 years, and their participation was voluntarily after seeking informed consent from them.

Consent for publication

Not applicable.

Competing interests

The authors declare that they have no competing interests.

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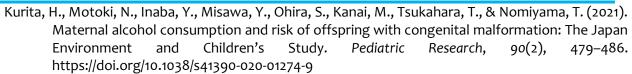
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