

Original article

Epidemiology of Orofacial Clefts: A Spatio-Temporal Descriptive Analysis in Tripoli, Libya, 2017-2021

Yusra Elfaidy*, Eman Naas, Arij Arebi, Hend Alfowers, Hadil Albourawi, Surur Milad, Nooralhuda Aljhem

Department of Dental Technology, Faculty of Medical Technology, University of Tripoli, Tripoli, Libya

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Corresponding Email. y.gumma@uot.edu.ly

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ABSTRACT

Orofacial clefts are the most common congenital anomalies affecting the orofacial region. An abnormal facial development causes this congenital deformity during gestation; its aetiology is multifactorial. It is crucial to understand the prevalence of craniofacial anomalies in each community to determine the size of the problem, enhance the life standards for the patients, and assess the efficacy of interventions. In this study, we aimed to provide a picture of the prevalence of orofacial clefts in Tripoli, Libya. Also, it provides a useful reference for cleft-type distribution with their etiological risk factors. The data was collected from the archives of cleft babies referred to major specialized hospitals in Tripoli, Libya, from 2017 to 2021. Variables were collected and analyzed, including data related to cleft cases and their parents. During the study period, the incidence of orofacial clefts was 1.06 per 1,000 live births. The prevalence of cleft lip was 38.3%, cleft palate was 42.2%, and cleft lip with palate was 19.6%. The results showed that 2017 had the highest incidence rate, reaching 32.2%, while it was 8.3% in 2021. The prevalence was generally higher among males than females, at a difference rate of 3.47%. A history of folic acid deficiency and unsupervised drug intake during pregnancy was noted (58.3% and 61.7%, respectively). In addition, the higher prevalence was more common among mothers older than 34. Moreover, it has observed that 46.8% of patients' fathers were smokers. The study concluded that the incidence of cleft defects in Tripoli, Libya, was relatively low, and the role of the predisposing factors in increasing the incidence of cleft deformity remains uncertain. Further studies are recommended to find out the root cause.

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INTRODUCTION

Orofacial clefts are congenital abnormal spaces or gaps in the tissues of the upper lip and palate, and they are the most common congenital anomalies affecting the orofacial region. Orofacial clefts are classified as the cleft lip, cleft palate, and cleft lip with palate. This congenital deformity is caused by abnormal facial development during gestation (failure in the union of palatal, median, and lateral nasal processes) [1].

Cleft lip and palate commonly influence the lip, alveolar edge, and hard and soft palate; issues related to these oddities are dental problems, malocclusion, nasal deformation, gross facial deformity, and feeding, ear, and speech troubles [2]. In addition, this deformity can occur isolated or along with other congenital deformities, particularly congenital heart disease [3].

The aetiology of cleft lip and palate is multifactorial, present in various cultures and races as well as in countries at different frequencies. These factors can be both genetic and environmental, including malnutrition, drugs, alcohol, smoking, infections, traumatic stress, and pollution [3]. It is crucial to understand the prevalence of craniofacial anomalies in each community to determine the size of the problem and to enhance the life standards for the patients and the efficacy of interventions. In spite of the fact that orofacial clefts occur in all races -according to the World Health Organization- the prevalence of specific cleft conditions varies greatly geographically and among ethnic groups.

Globally, the rate of cleft lip and palate is 1 in 600 to 800 live births (1.42 in 1000); therefore they are the most common congenital abnormalities of the craniofacial structure [4]. Its incidence appears high among Asians (0.82-4.04 per 1000 live births), intermediate in Caucasians (0.9-2.69 per 1000 live births), and low in Africans (0.18-1.67 per 1000 live births) [5]. In the UK, approximately 1 in 700 babies is affected by cleft lip and palate [6]. Chinese showed 1.76 per 1000 live births, while the Japanese reported 0.85 to 2.68 per 1000 live births of orofacial cleft [5].

A study conducted in Sudan proved that cases of cleft lip and palate, demonstrating a prevalence of 0.9 per 1000 [7]. Egyptian researchers investigated the average prevalence value of cleft lip and palate with a result of 40/1000. In addition, they found that cleft lip and palate had the highest percentage, followed by isolated cleft palate [8]. A study conducted in Benghazi, Libya; in 2020 revealed that the proportion of cleft lip and palate was 0.875 per 1000 live births [9]. Unfortunately, the incidence data of cleft cases in Tripoli, Libya, were limited. So our study aimed to know the approximate prevalence of cleft lip and palate cases in the region and to screen the predisposing factors in newborns suffering from such defects.

METHODS

Study design

A cross-sectional descriptive and retrospective study was conducted that was limited to cleft newborns admitted to three major specialized hospitals in Tripoli, Libya: Tripoli Medical Centre, Burns and Plastic Surgery Hospital, and Aljalaa Hospital, on an interval from 2017 to 2021. A questionnaire was prepared based on previous studies [9,10,11] and validated through pretesting with parents, resulting in modifications until the questions effectively addressed their intended purpose [12].

The final form of the questionnaire included a set of self-administered questions consisting of fourteen questions that included aspects related to the newborn data: gender, date of birth, birth weight, and type of defect. The second section focused on the parents' information, addressing their age, medical conditions, medications taken by mothers during pregnancy, and family history.

Data collection

The data for this research were gathered by specialized professionals from the archives of cleft babies. Out of all referred cases, only 230 files were accessible. We obtained access to their data, and additional information was collected through direct phone contact with parents of the cases, who voluntarily participated in the study. Some detailed information was collected by direct phone contact with the parents of the cases who entirely volunteered to participate in the study, and finally, 180 parents only responded and gave detailed answers to the questionnaire. The incidence of cleft cases was measured by calculating the total number of cleft cases registered in that hospital in relation to the total birth rate in Tripoli, Libya.

Data analysis

Results were evaluated by IBM SPSS version No. 26 software and then analyzed using a chi-square test at a 0.05 level of significance. The correlation between variables was evaluated by Spearman coefficient analysis.

RESULTS

The current study showed 230 cleft newborns from 2017 to 2021, out of 216278 live births in Tripoli, Libya, resulting in a prevalence rate of 1.06 per 1000 births.

Characteristics of study sample

As illustrated in Table (1), males (51.7%) were more influenced by this deformity than females (48.3%), with a difference of (3.47%). Regarding the birth year, the annual distribution was the highest (35.2%) in 2017, while it was lowest in 2021.

Table 1. Cleft patients' distribution according to Gender and Birth year

Birth year	Gender				Total		P-value
	Male		Female		N	%	
	N	%	N	%			
2017	41	17.8%	40	17.4%	81	35.2%	0.679
2018	23	10.0%	31	13.5%	54	23.5%	
2019	30	13.0%	17	7.4%	47	20.4%	
2020	8	7.8%	11	4.8%	29	12.6%	
2021	7	3.0%	12	5.2%	19	8.3%	
Total (N)	119	51.7%	111	48.3%	230	100%	

Independent Samples T-test was done as the test of significance

Table 2 showed that in total (183, Missing=47) of cleft newborns, the birth weight ranged from 1.2 kg to 5.5 kg with a mean weight of 3.083 kg (SD=0.773). The majority of the study sample were males (53.6%) with a mean weight of 3.129 kg, and (46.4%) of the sample were females with a mean weight of 3.029 kg. However, according to the weight groups, we noted that the majority of cleft newborns (68.3%) had a weight ranging between 2.5 and 4 kg.

Table 2. Distribution of Birth weight for cleft patients according to Gender

Birth weight (Kg)	Gender				Total		P-value
	Male		Female		N	%	
	N	%	N	%			
< 2.5	16	8.7%	13	7.1%	29	15.8%	0.389
2.5 – 4	65	35.5%	60	32.8%	125	68.3%	
> 4	17	9.3%	12	6.6%	29	15.8%	
Total (N)	98	53.6%	85	46.4%	183	100.0%	
weight (kg) (Mean ±SD)	3.129 ± 0.781		3.029 ± 0.766		3.083 ± 0.773		
CI (95%)	(2.972 - 3.285)		(2.864 - 3.195)				

SD= Std. Deviation, CI (95%) = 95% Confidence Interval for mean and Missing value = 47. Independent Samples T-test was done as the test of significance. This test used to compare means between males and females.

Type of Cleft Cases

Among 230 cleft newborns in the study sample, as illustrated in Figure (1), the majority of cases (42.2%) had cleft palate, followed by those with cleft lip (38.3%), and the least common were cleft lip with palate (19.6%).

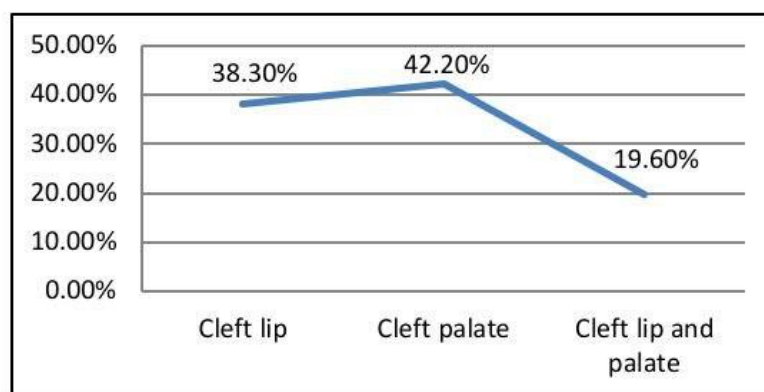


Figure 1. Relative distribution of Type of Cleft Cases

Family Background

Table (3) summarizes the results of maternal history regarding cleft deformities. It was found that cleft deformities were more common (54.7%) in newborns to mothers over the age of 34. The findings also indicated that a majority of mothers (87.2%) were in good health during pregnancy. However, 61.7% reported a history of drug intake during this period. Conversely 58.3% of mothers did not take the recommended folic acid during their pregnancies. Additionally, most of the mothers (92.6%) in the study sample did not have a family history of cleft deformity.

Table 3. Mothers' history and some factors that may have impact with the occurrence of a cleft

Factor		Number (N)	Percentage (%)
Maternal Age (years)	< 21	3	1.6%
	21 – 34	83	43.7%
	> 34	104	54.7%
Total (N, Missing (40))		190	100.0%
Medical Condition	Not good	22	12.8%
	Good	150	87.2%
Total (N, Missing (58))		172	100.0%
Drug Consumption (During Pregnancy)	NO	67	38.3%
	Yes	108	61.7%
Total (N, Missing (55))		175	100.0%
Folic Acid Consumption	NO	102	58.3%
	Yes	73	41.7%
Total (N, Missing (55))		175	100.0%
Family History of Cleft	NO	174	92.6%
	Yes	14	7.4%
Total (N, Missing (42))		188	100.0%

In considering to father's history, the results in table (4) referred to that half of the cleft cases (50%) in the sample had their fathers aged from 30 to 45. In terms of smoking, the findings showed that (46.8%) of the patients' fathers were smokers. Additionally, there was a father's family history of cleft in (6.9%) of cases.

Table 4. Fathers' history and some factors that may have impact with the occurrence of a cleft

Factor		Number (N)	Percentage (%)
Father Age (years)	< 30	12	6.3%
	30 – 45	95	50.0%
	> 45	83	43.7%
Total (N, Missing (40))		190	100.0%
Smoking Habit	NO	101	53.2%
	Yes	89	46.8%
Total (N, Missing (40))		190	100.0%
Family History of Cleft	NO	175	93.1%
	Yes	13	6.9%
Total (N, Missing (42))		188	100.0%

The Pearson Chi-Square test was used for association (independence) between the type of cleft and some factors from parental data. Table (5) showed that the P-values were more than 0.05, indicating that cleft type and factors from parental data were independent, and there were non-significant associations at the 5% level. Moreover, there were no correlations between these variables.

Table 5. Pearson Chi- square tests to check association (independence) between variables

Comparison between variables	P-value
Cleft type with medical condition	0.598
Cleft type with drug consumption	0.397
Cleft type with necessary folic acid consumption	0.408
Cleft type with mother family history	0.079
Cleft type with father smoking	0.561
Cleft type with father family history	0.598

DISCUSSION

This study showed that the incidence of cleft lip and/or palate in Tripoli was 1.06 per 1000 live births. This is considered a low incidence of cleft deformity when it compared to other parts of the world [13]. This result could be related to the fact that Libyan women in general live in a very conservative society where smoking and alcohol intake among females is almost zero, and there is a lot of research suggesting alcohol and smoking to have a teratogenic effect on fetus development during pregnancy [14]

Analysis of our results showed that there was a difference in the gender distribution of the study sample. As 51.7 % of

patients were boys. This means males are more likely to be affected by the deformity. However, another study in Nigeria reported an equal male to female ratio [15]. On the other hand, higher female incidence was the result of another study made in Sudan [16].

The type and extent of cleft defects vary according to race. In a study published on a Caucasian population, the prevalence of Cleft lip was 25%, Cleft lip and palate was 50%, and Cleft palate only was 25% [17]. Another study on an African population showed 49% prevalence of Cleft lip, 32% Cleft lip and palate, and 19% Cleft palate only [15].

Our study showed that the higher prevalence of cleft cases was cleft palate only (42.2%), while cleft lip cases was 38.3% and cleft lip and palate was 19.6 %, and this was in accordance with Trigos et al. [18] in 1994. As well as, the mother's medical condition could increase the incidence of congenital malformations. Viruses and bacteria, to somewhat, may traverse the placenta and reach embryonic tissue. Also, poor oxygen, poor blood supply, and malnutrition, which are associated with some systemic diseases may have a negative impact on the newborn's development at different stage [18]. Our study's statistical analysis revealed a relation between the deformity incidence and the mother's general medical condition. 12.8% of the patient's mothers have been diagnosed with at least one poor medical condition (hypertension, diabetes, heart disease, etc.). This finding could not be overlooked. However, a detailed investigation should be conducted to indicate which and how these systemic diseases may affect embryonic development. In addition, Saver et al. [19] in 1980 believe there is a connection between mother intakes of some types of medical drugs during pregnancy and the pathogenesis of cleft deformity. They claim that the exposure to certain chemical agents during the first trimester may interfere with the normal development of the lip and palate. Our results revealed that 61.70% of the patients' mothers had a history of drug intake during pregnancy. A relation could be found here; nevertheless, further research should be done on the exact type of chemical agents and when and how they disturb normal growth. On the other hand, we also revealed that only 41.7% of patients' mothers received the necessary folic acid during pregnancy, and more than half of them did not.

According to the World Health Organization's (WHO) report on 2016, the tobacco smoking epidemic is one of the largest public health problems globally, and the number of non-smokers exposed to secondhand smoke (SHS) has been steadily increasing [20]. In addition, concern is that maternal (SHS) exposure is also associated with adverse birth outcomes such as low birth weight, spontaneous abortion [21], and birth defects [22]. Our investigation studied the relationship between the incidence of orofacial clefts and smoking habits, and we concluded that more than one-third (46.8%) of patients' fathers were smokers. There could be a relation between secondary smoking and the aetiology of the cleft lip and palate; again, wider studies on bigger samples need to be done.

Our understanding of the aetiology and the pathogenesis of orofacial cleft remains relatively poor. Jones et al. [23] in 1995 believed that both genetic and environmental factors play an important role at the molecular level during embryogenesis. Primary evidence for a genetic role has been available; the sibling risk for cleft lip and palate is 30 times higher than that of the normal population prevalence as reported by Mitchell et al. [24] in 1992.

A higher prevalence of cleft lip and/or palate in perinatal infants for older mothers had been observed. As the parents get older, their offspring with cleft lip and palate were at a higher risk of more severe disease [25]. Our research results agreed, as it revealed that clefts were more common in children born to mothers over the age of 34, where they were 54.7%.

Our research found only 14 of the patients' mothers (7.40%) and only 13 of the patients' fathers (6.90%) had a history of cleft lip and/or palate running in their families. This proves that environmental factors play an important role, and an important area of future research will be needed to unravel interactions that occur between candidate genes and environmental factors during the early development of embryos.

CONCLUSION

The overall incidence of orofacial clefts in Tripoli, Libya, was relatively low. Most of them had cleft palate only, followed by cleft lip, while cleft lip with palate cases had the lowest incidence rate. The older the mother, the more likely the baby will have a cleft. However, males were more vulnerable than females. In addition, more than half of cleft patients' mothers didn't receive the necessary folic acid during pregnancy. On the other hand, again, more than half of the patients' mothers had a history of drug intake during pregnancy. Additionally, more than one-third of cleft patients' fathers were smokers. However, the role of these predisposing factors in increasing the incidence of cleft deformity cannot be conclusively confirmed or ruled out. Further studies are recommended to find out the root cause.

Conflicts of Interest

The authors declare no conflicts of interest.

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معدل حدوث الشفة الأرنبية مع أو بدون الحنك المشقوق: تحليل وصفي مكاني وزماني في طرابلس، ليبيا، 2017-2021

يسرا الفاندي*، إيمان النعاس، أريج عريبي، هند الفوارس، هديل البوراوي، سرور اليونسي، نورالهدى الجهيمي
قسم تقنية الأسنان، كلية التقنية الطبية، جامعة طرابلس، طرابلس، ليبيا

المستخلص

تعد الشقوق الفموية الوجهية من أكثر التشوهات الخلقية شيوعاً التي تصيب منطقة الوجه والفم. يحدث هذا التشوه الخلقي بسبب نمو غير طبيعي للوجه أثناء الحمل، وأسبابه متعددة العوامل. و من الأهمية فهم مدى انتشار الشذوذ القحفي الوجهي في كل مجتمع لتحديد حجم المشكلة، وتحسين معايير الحياة للمرضى، وتقييم فعالية التدخلات العلاجية. في هذه الدراسة، هدفنا إلى تقديم صورة عن مدى انتشار الشفة الأرنبية مع أو بدون الحنك المشقوق في طرابلس، ليبيا. وهدفنا أيضاً لتوفير مرجع مفيد لتوزيع أنواع تشوهات الشقوق الفموية الوجهية وعوامل الخطر المسببة لها. تم جمع بيانات هذا البحث من ملفات الأرشيف للأطفال المصابين الذين تم إحالتهم إلى المستشفيات المتخصصة الرئيسية في طرابلس، ليبيا، للفترة من 2017 إلى 2021م. تم جمع المتغيرات، بما في ذلك البيانات المتعلقة بتلك الحالات وأبائهم، وتحليلها. خلال فترة الدراسة، بلغ معدل حدوث الشفة الأرنبية مع أو بدون الحنك المشقوق 1.06 لكل 1000 ولادة حية. ومن إجمالي عدد الحالات، كان معدل انتشار الشفة الأرنبية 38.3%، والحنك المشقوق 42.2%، والشفة الأرنبية مع الحنك المشقوق 19.6%. وأظهرت النتائج أن عام 2017 كان أعلى معدل انتشار، حيث بلغ 32.2%، بينما كان 8.3% في عام 2021. وبالإضافة إلى ذلك كان معدل الانتشار أعلى عموماً بين الذكور منه عن الإناث، بمعدل فرق 3.47%. كما سجلت حالات عدم تناول حمض الفوليك اللازم بنسبة 58.3%، وحالات تناول لأدوية بدون إشراف طبي أثناء فترة الحمل بنسبة 61.7%. ولوحظ أن معدل الانتشار الأعلى كان أكثر شيوعاً بين الأمهات فوق سن 34 عاماً. علاوة على ذلك، وجد أن 46.8% من آباء الأطفال المصابين هم من المدخنين. وخلصت الدراسة إلى أن معدل حدوث تشوهات الشقوق الفموية الوجهية في طرابلس، ليبيا، كان منخفضاً نسبياً، ولا يمكن التأكيد على دور العوامل المذكورة في زيادة الإصابة بهذه التشوهات، كما لا يمكن استبعادها. ويوصى بإجراء المزيد من الدراسات لمعرفة العوامل الرئيسية المسببة.

الكلمات المفتاحية: الشفة الأرنبية، الحنك المشقوق، معدل الانتشار، عوامل الخطر، طرابلس-ليبيا