

## Original Article

# Prevalence and Associated Factors of Congenital Heart Disease in Cleft Lip and Palate Patients at the University of Benin Teaching Hospital

**\*Ekaniyere Benlance Edetanlen<sup>1</sup>, Ufadime Lawani<sup>2</sup>, Wilson Ehidiemen Sadoh<sup>3</sup>, Esezobor Peter Egbor<sup>3</sup>**

<sup>1</sup>Department of Oral and Maxillofacial Surgery, University of Benin Teaching Hospital, Benin City, Nigeria

<sup>2</sup>Department of Family Dentistry, University of Benin Teaching Hospital, Benin-city, Nigeria

<sup>3</sup>Department of Pediatrics, University of Benin Teaching Hospital, Benin-city, Nigeria

## Abstract

**Background:** The prevalence of congenital heart disease in patients with cleft lip and palate varies from place to place. There is a paucity of such studies in our region. This study aimed to determine the prevalence and risk factors for congenital heart defects (CHD) in children with oral clefts at the University of Benin Teaching Hospital

**Methodology:** This was a retrospective study of patients with cleft lip and palate who had echocardiography in the Department of Oral and Maxillofacial Surgery of the University of Benin Teaching Hospital (UBTH) from April 2012 to April 2022. Data were collected from the patient's case notes and the echocardiography registry in the pediatric cardiology unit of the same hospital. Both descriptive and inferential statistics were performed. In inferential statistics, the association between the prevalence of CHD and the studied variables was performed with the Chi-square test. Data were analyzed using Statistical Package for the Social Sciences, version 21.

**Results:** A total of 225 patients comprising 131 females and 94 males were recruited. The age range was 3 days to 10 years with a median age of 0.6 years. Of the 225 patients, 24(10.7%) had CHD giving a prevalence of 10.7%. The most common CHDs were atrial septal defect 12(5.33%) and ventricular septal defect 4(1.78%). The prevalence of CHD was significantly ( $p= 0.02$ ) higher in the patients living outside the study environment compared to those within the study location, and in those with isolated cleft palate ( $p=0.03$ ) compared to other types of orofacial cleft.

**Conclusions:** The prevalence of CHD is relatively high among cleft lip and palate patients. The residential location and the type of orofacial cleft were associated with higher prevalence in this study.

**Keywords:** Prevalence, Congenital Heart Disease, Cleft Lip, Cleft Palate.

**Corresponding Author:** \*Edetanlen Benlance Ekaniyere, Department of Oral and Maxillofacial Surgery, University of Benin Teaching Hospital, Benin City, Nigeria. ehiben2002@yahoo.com

This is an open-access journal, and articles are distributed under the terms of the Creative Commons Attribution-Non-Commercial-Share Alike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

**How to cite this article:** Edetanlen EB, Lawani U, Sadoh WE, Egbor EP. Prevalence and Associated Factors of Congenital Heart Disease in Cleft Lip and Palate Patients at the University of Benin Teaching Hospital. Niger Med J 2023; 64(1):54-60.

Quick Response Code:



## Introduction

Cleft of the lip and palate are congenital structural defects capable of causing significant lifelong morbidity and social embarrassment to the patients if not professionally managed. They result from the interplay of factors such as environmental, genetic, maternal intake of certain medications, and idiopathic<sup>1</sup>. Several studies have described cleft lip (CL) and/or palate (CLP/CP) as the most common craniofacial congenital facial defect, and its global incidence is said to be one out of every 500-1000 live births.<sup>2-4</sup> The condition occurs worldwide, the Asian population has been reported to have the highest prevalence, while African population has the least prevalence. Butali et al<sup>2</sup> in their study pooled from six geopolitical zones in Nigeria gave an estimated prevalence of 0.5 per 1000 births<sup>5-9</sup>.

The prevalence of congenital heart defects in children with CL and CP is reportedly higher than in the general population<sup>10-14</sup>. The incidence of cardiovascular anomalies in children with CL and CP is estimated to be 16-fold higher than that seen in the general population<sup>10</sup>. The congenital heart diseases that could be associated with cleft lip and palate include, patent ductus arteriosus (PDA), patent foramen ovale (PFO), atrial septal defect (ASD), ventricular septal defect (VSD), Tetralogy of Fallot (TOF), truncus arteriosus (TA) and mitral valve prolapsed (MVP)<sup>11,13</sup>.

Cleft lip and palate have remained a social, economic, and psychological burden to both the parents and the children that suffer the condition's ultimate impact on the quality of life<sup>15</sup>. Those with associated congenital diseases, especially the cardiovascular system has increased morbidity and mortality following surgical procedures<sup>16</sup>. Therefore, it is not far-fetched that factors that can cause increased morbidity are identified before undergoing any surgical procedures on the group population. The information from this study will help to identify cleft and lip patients with the likelihood of congenital heart disease in a setting where echocardiography is a luxury as in most regions of developing countries.

The prevalence of congenital heart disease has been studied globally and differs among different populations and it varies widely<sup>11-24</sup>. There have been several reports on the prevalence of cardiovascular anomalies among patients with cleft lip and palate in various parts of Nigeria with prevalence ranging from 3.76% to 30.7% of such patients seen in the echocardiograph.<sup>17-24</sup> A prevalence of 16.7% was reported in a previous study in the study center that evaluated children undergoing surgery for congenital heart diseases including those with craniofacial clefts, using echocardiography. However, the study did not evaluate the factors associated with heart defects. Also, the previous study was for a shorter duration and a smaller sample size.<sup>24</sup>

This study aimed to determine the prevalence and factors associated with the presence of congenital heart disease in children with oral clefts at the University of Benin Teaching Hospital (UBTH). It is hoped that this study will assist the surgeons and anaesthetic to be aware of the likelihood of having an underlying cardiovascular disease in patients with cleft lip and palate to allow for adequate planning of patients' overall management.

## Methodology

This was a retrospective study on cleft lip and palate patients that had echocardiography and were treated in the Department of Oral and Maxillofacial Surgery of the University of Benin Teaching Hospital (UBTH) from April 2012 to April 2022. Ethical approval was obtained from the Research and Ethics Committee of the institution (ADM/E22/A/VOL.VII/14831005). All cases for repair were referred to the paediatric cardiopulmonary clinic headed by one of the authors. All were routinely sent for echocardiographic studies irrespective of the presence of symptoms. Data were collected from the patient's case notes and the record registry of the pediatric cardiology unit of the same hospital. Excluded from the study were those with incomplete information in their case notes, those without echocardiography records, and those that reside outside Edo and Delta States. Collected were age, gender, place of residence, type of cleft, laterality of cleft, presence of congenital heart disease, and type of congenital heart disease. Patients that reside in Delta state were considered as those living outside Benin City and this was deduced from a previous study that

reported that majority of patients with an orofacial cleft that sought treatment in our centre resided in Delta state compared to the other states in the Niger-Delta region<sup>9</sup>. All transthoracic echocardiography studies were done by one of the authors. M-mode and two-dimensional images with Doppler interrogation for the presence of congenital heart diseases were carried out in the standard echo windows. All studies were carried out using ALOKA SSD cardiac ultrasound system and Philips SSI-8000 Sono Scape ultrasound system (Aloka, Meerbusch, Germany). Analysis of the echocardiographic finding was in accordance with the recommendations of the American Society of Echocardiography<sup>25</sup>. Descriptive and inferential statistics were performed. In descriptive statistics, categorical data were presented in proportions and percentages while continuous data were summarized in median and range when the data was not normally distributed. In the inferential statistics, the chi-square of association was used to test the association between the prevalence of CHD and the studied variables. All Data were analyzed using the statistical package for the social sciences (SPSS), version 26 (IBM, Armonk, NY, United States of America). A *p*-value < 0.05 was considered statistically significant.

## Results

A total of 225 patients were seen during the study period comprising 131 females and 94 males giving a male-to-female ratio of 1: 1.3. The age range was 3 days to 10 years with a median age of 0.6 years. Table 1 shows the demographic characteristics of patients with cleft lip and palate in UBTH. The majority (69.3%) of the patients were within the age group of 0-20 months and this was followed by those between 21 and 40 months (13.8%). More than half (53.8%) of the patients with cleft lip and palate reside outside Benin City.

Table 2 shows the clinical characteristics of patients with cleft lip and palate. Cleft lip and palate patients constitute the highest proportion (38.2%) of patients with congenital heart disease while the least proportion (28.0%) was the cleft lip alone. The majority (96.0%) of the patients presented with unilateral cleft of the lips and palates. Of the 225 cleft and lip patients, 24(10.7%) had associated congenital heart diseases given a prevalence of 10.7 %. Out of the 24 patients with congenital heart disease, atrial septal defect had the highest proportion of 12(5.33%) which was followed by a ventricular septal defect (1.78%).

Table 3 shows the factors associated with the prevalence of congenital heart disease in cleft lip and palate patients in the UBTH. While the age of the patients, gender, and laterality was not related to the presence of congenital heart disease, the place of residence and type of cleft were associated with the prevalence of congenital heart disease. The prevalence of congenital heart disease was significantly (*p* =0.02) higher in the patients that reside outside the study environment than in those in Benin City. In the same vein, patients with cleft palate alone had a significantly higher prevalence compared to the other types of clefts as shown in Table 3.

**Table 1:** Demographic characteristics of patients with cleft lip and palate in UBTH (n=225)

Variables	Category	Frequency(n)	Percentages (%)
Age(months)	0-20	156	69.3
	21-40	31	13.8
	41-60	21	9.30
	61-80	4	1.80
	81-100	1	0.40
	101-120	12	5.30
Gender (n (%))	Male	94	41.8
	Female	131	58.2
Place of residence (n (%))	Within Benin	104	46.2
	Outside Benin	121	53.8

**Table 2:** Clinical characteristics of patients with cleft lip and palate in UBTH (n=225)

Variables	Category	Frequency(n)	Percentage (%)
Type of cleft	Cleft lip alone	63	28.0
	Cleft lip and palate	76	33.8
	Cleft palate alone	86	38.2
Laterality of cleft	Unilateral	216	96.0
	Bilateral	9	4.00
Presence of CHD	Yes	24	10.7
	No	201	89.3
Type of CHD	VSD	4	1.78
	ASD	12	5.33
	PDA	2	0.89
	TOF	3	1.33
	PFO	3	1.33

N.B VSD= Ventricular septal defect, A trial septal defect, Patent ductus arteriosus, Tetralogy of Fallot, Truncus arteriosus, Patent foramen ovale

**Table 3:** Factors associated with the prevalence of congenital heart disease in cleft lip and palate patients in UBTH (n = 225)

Variables	Category	Presence of CHD		P-Value
		Yes (n=24)	No (n=201)	
Age	0-20	16	140	0.89
	21-40	5	26	
	41-60	2	19	
	61-80	0	4	
	81-100	0	1	
	101-120	1	11	
Gender	Male	13	81	0.19
	Female	11	120	
Place of residence	Within Benin	6	98	0.03
	Outside of Benin	18	103	
Type of cleft	Cleft lip alone	8	55	0.02
	Cleft lip and palate	9	67	
	Cleft palate alone	7	79	
Laterality of cleft	Unilateral	22	194	0.25
	Bilateral	2	7	

NB: CHD = Congenital heart disease

## Discussion

The 10.7% prevalence of CHD in children with orofacial clefts in the present study is consistent with the 9.5% reported by Aimede et al<sup>23</sup> in Abeokuta but higher than the 3.76% and 9.50% reported by Akhiwu et al<sup>19</sup> in Kano state. The sample size and the duration of the study may be the reason for the lower prevalence in the previous study. However, the percentage in the present study is lower than the 15.0%, 17.2%, 20.0%, and 30.7% reported by Otaigbe et al<sup>18</sup> in Port-Harcourt, Sadoh et al<sup>24</sup> in Benin-city, Asani, and Aliyu<sup>20</sup> in Kano, and Erinoso et al<sup>22</sup> in Lagos state respectively. The finding of lower prevalence in the present study may likely be variations in the methodological differences. Scientific inferences could be justifiably deduced

Edetanlen EB, et al - Congenital Heart Disease in Cleft Lip and Palate Patients from the present study because of its larger sample size and the longer study duration. More so, the higher percentage of 16.7% reported by Sadoh et al<sup>24</sup> in the same centre could be attributable to the nature of the study population who were all children that had undergone non-cardiac surgery including children with orofacial clefts while the present study exclusively recruited only patients with orofacial clefts.

The prevalence of CHD was affected by the type of oral cleft and the location of the patients. Individuals with cleft palate tend to develop CHD more compared to those with isolated cleft lip or cleft lip and palate. This finding could be related to the deficiency in the migration of neural crest cells which occurred during the simultaneous fusion of the heart tubes and the palatal shelves<sup>26</sup>. As regards the effect of the residence of the patients on the prevalence of CHD, these findings may suggest that most of the patients outside the study environment are from the Niger Delta region where there have been claims of increasing risks to the child and maternal health as a result of environmental degradation and industrial pollution attributed to petroleum mining activities in the area<sup>27</sup>. It appears this study was the first to assess the factors associated with the prevalence of CHD in children with orofacial cleft thereby making a comparison of these findings with previous studies not feasible.

There was a female predilection in the present study like in previous studies<sup>22,23</sup> compared to other studies that showed a male preponderance<sup>17,19-21,24</sup>. Yet the study by Otaigbe et al<sup>18</sup> reported no gender predilection. The role of gender variation in the aetiology of the orofacial cleft is yet to be scientifically proven. The mean age of 7 months in this study is comparable with the 6 months reported by Erinoso et al<sup>22</sup> in Lagos State, Nigeria. It is, however, higher than the 1 and 4 months reported by Otaigbe et al<sup>18</sup> and Asani and Aliyu<sup>20</sup> respectively but lower than 112 and 96 months reported by Barbosa et al<sup>13</sup> and Aimede et al<sup>22</sup> respectively.

The wide variation of this finding could be attributable to the time of presentation to health facilities by the patients.

The commonest type of orofacial cleft in the present was cleft palate alone which is consistent with that of Otaigbe et al<sup>18</sup> and Asani and Aliyu<sup>20</sup> but contrast with that of Akhiwu et al<sup>19</sup>, Aimede et al<sup>23</sup>, Sadoh et al<sup>24</sup>, Erinoso et al<sup>22</sup>, James et al<sup>21</sup> and Ademiluyi et al<sup>17</sup> who reported cleft lip and palate as the commonest orofacial cleft. Though the probable reason for this contrasting finding is not readily deducible, the higher sample size and longer duration of the present study make its findings scientifically reliable.

ASD was the most common CHD in children with an orofacial cleft in the present study and this finding is like that reported by Akhiwu et al<sup>19</sup>, Asani and Aliyu<sup>20</sup>, and Sadoh et al<sup>24</sup> that reported ASD as the most common CHDs in their various studies. This finding, however, contrasts that of James et al<sup>21</sup>, and Erinoso et al<sup>22</sup> which reported PFO as the most predominant CHD in patients with orofacial clefts. It is noteworthy however that ASD and PFA are both defects in the interatrial septum. Also, the finding of a preponderance of ASD in the present study disagrees with the VSD and MVP reported by Otaigbe et al<sup>18</sup> and Barbosa et al<sup>16</sup> respectively.

This study has a few limitations. Data could have been missing due to the retrospective nature of this study and this study was a single-center study that may have omitted patients residing outside the Niger Delta regions.

## Conclusion

The prevalence of congenital heart defects is relatively high among cleft lip and palate patients which buttresses the need for routine echocardiography in all patients with orofacial cleft patients. The residential location and the type of cleft lip and palate were associated with the prevalence in this study.

## References

1. Pavri S, Forrest CR. Demographics of orofacial clefts in Canada from 2002 to 2008. *Cleft Palate Craniofac J*. 2013; **50**:224-230
2. Butali A, Adeyemo WL, Mossey P et al. The Nigerian collaboration. Prevalence of orofacial clefts in Nigeria. *Cleft Palate Craniofac J*. 2014; **51**:320-325
3. Silva HPVd, Arruda TTS, Souza KSCd, et al. Risk factors and co-morbidities in Brazilian patients with orofacial cleft. *Brazilian oral research*. 2018;**32**
4. Ferencz C, Rubin JD, McCarter RJ, Brenner JI, Neill CS, Perry LW, et al. Congenital Heart Disease: Prevalence at live births. The Baltimore Washington infant study. *Am J Epidemiol*.1985; **121**:31-36
5. Panamonta V, Pradubwong S, Panamonta M, Chowchuen B. Panamonta V, et al. Prevalence of Congenital Heart Diseases in Patients with Orofacial Clefts: A Systematic Review. *J Med Assoc Thai*. 2015; **98**:22-27.
6. Cooper ME, Ratay JS, Marazita ML. Asian oral-facial cleft birth prevalence. *Cleft Palate Craniofac J* 2006; **43**:580-9.
7. Croen LA, Shaw GM, Wasserman CR, Tolarová MM. Racial and ethnic variations in the prevalence of orofacial clefts in California, 1983-1992. *Am J Med Genet*. 1998; **79**:42-7.
8. Chan KW, Lee KH, Pang KK, Mou JW, Tam YH. Clinical characteristics of children with orofacial cleft in a tertiary centre in Hong Kong. *Hong Kong J Paediatr* 2013; **18**:147-51.
9. Obuekwe ON, Akapata O. Pattern of cleft lip and palate in Benin City, Nigeria. *The Centr Afr J Med* 2004; **50**:65-9
10. Rao G, Desai A, Kumar N. Rao G, et al. Congenital Heart Diseases Associated with Cleft Lip and Palate and Its Impact on Surgical Treatment Planning of Patients With Cleft Lip and Palate-A Cross-Sectional Study. *Cleft Palate Craniofac J*. 2021; **58**:163-169
11. Mottaghi Moghaddam Shahri H, Farzaneh M, Chamani A, Jahanbin A. Associated Congenital Heart Anomalies in Children with Cleft Lip and Palate: A Cross-Sectional Study. *Int J Pediatr* 2022; **10**:15737-15745.
12. Sun T, Tian H, Wang C, Yin P, Zhu Y, Chen X, et al. A survey of congenital heart diseases and other organic malformations associated with different types of orofacial clefts in Eastern China. *PLOS One*. 2013; **8**:e54726
13. Geis N, Seto B, Bartoshesky L, Lewis MB, Pashayan HM. The prevalence of congenital heart disease among the population of a metropolitan cleft lip and palate clinic. *Cleft Palate J* 1981; **18**:19-23.
14. Liang CD, Huang SC, Lai JP. A survey of congenital heart disease in patients with oral clefts. *Acta Paediatr Taiwan* 1999; **40**:414-7.
15. Rawashdeh MA, Jawdat Abu-Hawas B. Congenital associated malformations in a sample of Jordanian patients with cleft lip and palate. *J Oral Maxillofac Surg* 2008; **66**:2035-41.
16. Barbosa MM, Rocha CM, Katina T, Caldas M, Codorniz A, Medeiros C. Barbosa MM, et al. Prevalence of congenital heart diseases in oral cleft patients. *Pediatr Cardiol*. 2003; **24**:369-74.
17. Ademiluyi SA, Oyeneyin JO, Sowemimo GO. Associated congenital abnormalities in Nigeria children with cleft lip and palate. *West Afr J Med* 1989; **8**:135-8.
18. Otaigbe B, Akadiri O, Eigbob J. Clinical and echocardiographic findings in an African pediatric population of cleft lip and palate patients: A preliminary report. *Niger J Cardiol*.2013;**10**:6
19. Akhiwu BI, Efunkoya AA, Akhiwu HO, Adebola RA. Congenital heart disease in cleft lip and palate patients: How common is the association? *J Acad Dent Res*. 2017; **8**:53-56
20. Asani MO, Aliyu I. Pattern of congenital heart defect among children with orofacial cleft in Northern Nigeria. *J Cleft Lip Palate CranioFac Anomal*. 2014; **1**:85-87
21. James O, Erinoso OA, Adamson OO, Sokunbi OJ, Agbogidi FO, Adekunle AA, et al. Risk of congenital cardiovascular anomalies in patients with non-syndromic orofacial cleft: A preliminary case-control study. *Niger J Clin Pract*. 2020; **23**:1561-1565
22. Erinoso OA, James O , Sokunbi OJ , Adamson OO , Adekunle AA, Agbogidi OF et al. Congenital heart defects in orofacial cleft: A prospective cohort study. *Afr J Paediatr Surg* 2021; **18**:219-223.

23. Aimede OS, Olalere GO, Adedayo O, Adeshola S. Orofacial clefts: Our experience in two suburban health facilities. *Dentistry* 2013; **3**:155.
24. Sadoh WE, Ikhurionan P, Imarengianye C. Pre-anesthetic echocardiographic findings in children undergoing non-cardiac surgery at the University of Benin Teaching Hospital, Nigeria. *Cardiovasc J Afr* 2016; **27**:1- 5
25. American Society of Echocardiography recommendation for continuous quality improvement in echocardiography. *J Am Soc Echocardiogr* 1995; **8**:1-8.
26. Vega-Lopez GA, Cerrizuela S, Tribulo C, Aybar MJ. Neurocristopathies: New insights 150 years after the neural crest discovery. *Dev Biol.* 2018; **444**:110-143.
27. Niger Delta natural resources damage assessment and restoration Project-Phase I scoping report. Report from the IUCN Commission on Environmental, Economic, and Social Policy; 2006. p. 14