Original Article

Benign Orofacial Vascular Anomalies: Review of 47 Cases in Enugu, Nigeria

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Received: 27-Apr-2023;
Revision: 10-Jul-2023;
Accepted: 01-Aug-2023;
Published: 04-Dec-2023

Abstract

Background: A study of oral vascular anomalies has not been conducted in Nigeria to provide baseline data for comparison with reports in the literature. **Aims:** To study the prevalence and distribution of benign orofacial vascular anomalies at a tertiary hospital in Enugu. **Materials and Methods:** This is a 10-year retrospective observational study of consecutive patients with orofacial vascular anomalies, diagnosed by histology. The clinic-pathologic information was obtained from records archived in the department, and descriptive analysis was used to determine the frequency, tables for categorical variables, and a Chi-square test to determine the statistical significance. **Result:** There were 47 cases of benign vascular anomalies out of 897 orofacial lesions giving a prevalence of 5.2%. There were 35.4% (17) male and 64.6% (31) female patients. The mean age in this series was 37.4 ± 19.8 (range: 1 to 76 years). Pyogenic granuloma was the most common vascular lesion 78.7% (37), followed by hemangioma 14.9% (7) and lymphangioma 6.4% (3). The gingiva was the most frequent site of oral occurrence 65.9% (31), especially maxillary gingivae 48.9% (23). The type of orofacial vascular anomalies was significantly associated with the anatomical site of occurrence, $P = 0.00$. The mean ages for the occurrence of pyogenic granuloma, hemangioma, and lymphangioma were 37.7 ± 18.3, 50.7 ± 16.9 years, and 3.3 ± 3.2 years, respectively. Pain was a frequent occurrence in 36.2% (17) of anomalies. **Conclusion:** Oral vascular anomalies predominantly presented as pyogenic granuloma on the gingivae, while oral hemangioma was observed in adults, and lymphangioma was infrequent.

Keywords: *Hemangioma, lymphangioma, oral vascular anomaly, pyogenic granuloma, vascular malformations*

Introduction

Benign vascular anomalies refer to neoplasms and malformations of the blood and lymphatic channels and are traditional divided into tumors and vascular malformations (VM).[1,2] Some of the tumors include hemangioma (HEM) and pyogenic granuloma (lobular capillary hemangioma).[3] The vascular malformations (VM) include capillary, macrocystic lymphatic, microcystic lymphatic, venous, venolymphatic, arteriovenous malformations, and arteriovenous fistulas.[4]

Vascular anomalies occur in various parts of the body, with the head and neck region having a prevalence range of 39% to 70% in Nigeria.[5-8] Similar or higher prevalence results are reported in other countries.[9,10] Orofacial vascular anomalies constitute only 1.4% to 4.3% of all surgical biopsies[5,6] and 16% to 37.7% of soft tissue tumors in Nigerian literature.[5,6] Studies from Northern and Western Nigeria of total body vascular anomalies reported a predominance of hemangioma (capillary and cavernous) with a prevalence range of 50% to 95%.[7,8,11]

Similarly, hemangioma was reported (57.6%), with lymphangioma (36.4%) in Tanzania as the most prevalent orofacial vascular anomalies.[12] In contrast,

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Access this article online

Quick Response Code:

Website: www.njcponline.com

DOI: 10.4103/njcp.njcp_332_23

How to cite this article: Nwoga MC. Benign orofacial vascular anomalies: Review of 47 cases in Enugu, Nigeria. Niger J Clin Pract 2023;26:1723-7.

oral pyogenic granuloma (70.5%) was reported as the dominant lesion in the Iranian population,[2] while oral varix (65.6%) was most frequent in Brazil.[13]

There is currently no primary prevalence study of benign orofacial vascular anomalies in Nigeria known to the author. Previous Nigerian literature had focused only on oral pyogenic granuloma,[14,15] maxillofacial tumors, and tumor-like lesions.[15-17] This paper is a study of the prevalence, distribution, and epidemiologic features of benign orofacial vascular anomalies in a tertiary hospital in Enugu, Southeast Nigeria.

Materials and Methods

This is a 10-year (April 2012 to March 2022) retrospective cross-sectional study of consecutive patients diagnosed with oral vascular anomalies by histology in a tertiary health center in Enugu. Their biodata and clinic-pathologic information from the biopsy forms, histopathology reports, and case files were assessed from the departmental archived records. Information such as gender, anatomic site, age at presentation, duration, complaint of pain, ulceration, diagnosis, treatment, and recurrences was obtained. The corresponding histologic slides were retrieved from the oral pathology departmental archives and reviewed.

Statistics: The data were analyzed with IBM SPSS Statistics for Windows, version 24.0. (Armonk, New York: IBM Corp., United States). The frequencies and percentages were calculated for the descriptive variables. The association of categorical variables using Chi-square tests was determined, and the test of significance was set at $P \leq 0.05$. This study followed the Declaration of Helsinki on medical protocol and ethics and received the institutional approval of the Research Ethics Committee of the College of Medicine of the University of Nigeria, with the Protocol Number**:** 0148/04/2022.

Inclusion Criteria: All orofacial vascular anomalies diagnosed based on histopathology were included in the study. **Exclusion Criteria**: Orofacial vascular anomalies that were conservatively managed or kept under observation without biopsy were not included in this study.

Result

A total of 55 benign and malignant oral vascular anomalies were identified among 897 orofacial lesions. There were 47 benign oral vascular anomalies with a prevalence of 5.2% of orofacial lesions. Tables 1 and 2 show the clinical features of 37 cases of pyogenic granuloma, while Table 3 shows the clinical features of ten cases diagnosed as hemangioma and lymphangioma. Three benign vascular anomalies were identified: pyogenic granuloma 78.7% (37), hemangioma 14.9% (7), and lymphangioma 6.4% (3).

Table 1: Features of pyogenic granuloma in maxillary gingivae

Sex	Age (years)	Duration (months)	Site	Pain	Recurrence
*F	45	36	Maxillary gingiva	Nil	NIL
†M	29	6	Maxillary gingiva	Nil	Yes
F	23	12	Maxillary gingiva	Nil	Nil
F	45	2	Palatal gingiva	Pain	Nil
F	16	24	Gingiva	Nil	Nil
F	28	1.5	Maxillary gingiva	Yes	Nil
F	19	-	Palatal gingivae	Nil	Yes
M	10	1	Palatal gingiva	Yes	Nil
F	11	1	Maxilla gingiva	Nil	Nil
F	65	7	Maxilla gingiva	Nil	Nil
M	40	-	Maxillary gingiva	Nil	Nil
F	44	2	Maxillary gingivae	Nil	Nil
M	35	24	Maxillary gingiva	Nil	Nil
M	51	12	Maxillary gingiva	Nil	Nil
F	17	4	Palatal gingiva	Yes	Nil
M	64	36	Maxillary gingiva	Yes	Nil
F	69	18	Maxillary gingiva	Yes	Nil
F	35	36	Maxillary gingiva	Nil	Nil
M	63	24	Maxillary gingiva	Nil	Nil
F	45	12	Maxillary gingiva	Yes	Nil

*F=Female, †M=Male N

Table 2: Orofacial pyogenic granuloma in mandibular gingivae and other sites

Sex	Age (years)	Duration (months)	Site	Pain	Recurrence
*F	24	19	Mandibular gingiva	Nil	Nil
F	12	29	Mandibular gingiva	Yes	Nil
‡M	18	27	Mandibular gingiva	Yes	Nil
F	24	76	Mandibular Gingiva	Yes	Nil
M	12	53	Mandibular gingiva	Nil	Nil
F	1	20	Mandibular gingiva	Nil	Nil
F	4	54	Mandibular gingiva	Nil	Nil
F	-	49	Mandibular gingiva	Nil	Nil
F	48	38	Mandibular gingiva	Nil	Nil
M	9	22	Mandibular gingiva	Nil	Nil
M	3	23	Lower lip, mucosal part	Nil	Nil
F	1	52	Left posterior cheek	Nil	Nil
M	1	25	Mandibular region	Nil	Nil
M	12	69	Premaxilla-Nasolabial Expansion	Nil	Nil
F	12	16	Anterior palate	Nil	Yes
F	24	25	Zygomatico-maxillary region	Yes	Nil
F	-	44	Palatal junction	Nil	Nil

*F=Female, †M=Male N

The gender distribution for vascular anomalies was 36.2% (17) and 63.8% (30) for the male and female patients, respectively. A Chi-square statistic of types of benign vascular tumors and gender shows no significant ($P > 0.05$) association, with $P = 0.87$ at 95% confidence interval.

The vascular tumors had no predilection for a particular age group but were mostly well distributed over the decades, though most occurred in the third decade at 21.3% (10).

Table 4 shows a summary of the clinical information on oral vascular anomalies. The maxillary gingiva was the most frequent site of oral occurrence with 48.9% (23), followed by the mandibular gingivae with 17.0% (8). Table 5 shows that buccal mucosa 12.8% (6) was the third most common site of occurrence, while the tongue, lip, and floor of the mouth were least affected. The association of the type of orofacial vascular lesion with the anatomical site of occurrence was significant, with $P = 0.00$.

Pyogenic granuloma [Figure 1] was the most prevalent vascular lesion found mostly on the gingiva in 81.1% (30) of cases. The gender distribution of 35.1% (13) and 64.9% (24) in male and female patients, respectively, gave a ratio of 1:1.8. Mean ages for male and female patients were 39.3 ± 18.9 and 36.8 ± 18.3 years, respectively. There were only 21.6% (8) pyogenic granuloma cases in children and adolescents, while most of the cases 62.1% (23) occurred in the third to sixth decades.

Oral hemangioma was observed only in adults and exhibited no site predilection since they were widely distributed especially on the buccal mucosa 28.6% (2) and tongue 28.6% (2). Other sites with single occurrence included lip, mandible, and labial mucosa anterior to mandible. Oral lymphangioma was the only vascular

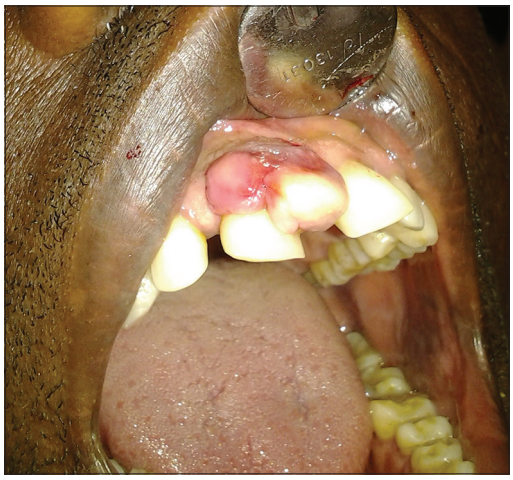

Figure 1: Pyogenic granuloma presenting as a nodular outgrowth on the maxillary labial gingivae, causing displacement and mild rotation of the central incisors

Table 3: Features of orofacial hemangioma and lymphangioma (n=10)

Sex	Age (years)	Duration (months)	Site	Pain	Ulceration	Histologic Diagnosis
*M	31	24	Lip	Nil	Yes	Cavenous hemangioma
M	48	3	Buccal mucosa	Nil	Nil	Hemangioma
†F	51	3	Tongue	Yes	Nil	Capillary hemangioma
F	60	1	Buccal mucosa	Nil	Nil	Capillary hemangioma
F	67	1.5	Tongue	Yes	Nil	Capillary hemangioma
F	27	7	Mandible	Yes	Nil	Hemangioma
M	71	‡N/A	Lower labial mucosa	Yes	Nil	Hemangioma
F	2	24	Floor of mouth	Yes	Nil	Cystic lymphangioma
M	1	0.75	Floor of mouth	Nil	Nil	Lymphangioma
F	7	84	Tongue	Yes	Nil	Congenital cavenous lymphangioma

*M=Male, †F=Female, ‡N/A=Not available

Table 4: Summary of the clinical information on benign orofacial vascular anomalies

	Oral vascular tumors (47)	Pyogenic granuloma	Hemangioma	Lymphangioma
Prevalence	5.2%	78.7% (37),	14.9% (7)	6.4% (3)
Gender ratio (male:female)	1:1.8	1:1.8	1:1.3	1:2
Mean age (range, years)	37.4±19.8 (1-76)	37.7±18.3 (10-76)	50.7±16.9 (27-71)	3.3±3.2 (1-7)
Mean duration (range, months)	14.8±16.2 (0.8-84)	14.1±12.5 (1-48)	6.6±8.8 (1-24)	36.3±43 (0.75-84.0)
Site predilection % of cases (no.)	Gingiva 65.9% (31)	Gingiva 81.1% (30)	Buccal mucosa 28.6% (2), tongue 28.6% (2)	Floor of mouth 66.7% (2)
Pain	36.2% (17/47)	29.7% (11/37)	57.1% (4/7)	66.7% (2/3)
Recurrences	6.4% (3/47)	6.4% (3/47)	Nil	Nil
Ulceration	4.3% (2/47)	Nil	28.6% (2/7)	Nil

Table 5: Site distribution of orofacial vascular anomalies

Site	*n* (%)
Maxillary gingivae	48.9 (23)
Mandible gingivae	17 (8)
Buccal/labial/mucosa	12.8 (6)
Tongue	6.4 (3)
Lip	4.3 (2)
Floor of mouth	4.3 (2)
Palate	4.3 (2)
Mandibular bone	2.1 (1)
Total	100 (47)

malformation identified. The floor of the mouth and tongue were the sites affected.

There were no recurrences noted in the cases of hemangioma and lymphangioma. In addition, there was no significant ($P > 0.05$) association between the type of benign vascular tumor and history of pain in the participants as shown by the P value, 0.308. The cases of benign oral vascular anomalies were submitted as incisional or excisional specimens. Surgery was the treatment of choice.

Discussion

The prevalence of 5.2% for vascular anomalies among orofacial lesions was low when compared to Nigerian reported values for other groups of orofacial lesions,[18] especially odontogenic cysts (23.3%)[15] and tumors (49%).[19] The absence of primary studies on oral vascular anomalies as a subject in Nigerian could be because of their low incidence, under-reporting of cases, and greater research interest in odontogenic lesions,[15,16] salivary tumors, and oral epithelial malignancies.[20] However, oral pyogenic granuloma is the only vascular lesion that has attracted much research interest and literature because of its high prevalence in Nigeria.[14,21] This study corroborated the high frequency of pyogenic granuloma in Nigeria with an observed prevalence of 78.8% and similar to that reported in Iran (70.5%).[2] In contrast, authors in other countries reported oral hemangioma (57.6%) in Tanzania,[12] oral varix (65.6%) in Brazil,[13] and venous malformations (25.4%) in Belgium as more frequent.[22]

There were no cases of arteriovenous, capillary (pot wine stains), and venous vascular malformations in this series probably due to reduced awareness, under-reporting, and conservative management. Similar reasons could explain the low prevalence of oral lymphangioma and oral hemangioma in this study and as reported in Iran.[2]

The female predilection of 63.8% in oral vascular anomalies in this study is due to the same gender predilection in pyogenic granuloma, a common vascular lesion in Calabar South-South Nigeria where it was reported with 100% female occurrence[15] and in Iran.[2] A similar female gender preponderance in the range of 56%–66.7% was attributed to oral varix in a Brazilian study[2] and in other studies with high prevalence of hemangioma and vascular malformations.[13,23,24] The reasons for the female gender prevalence of orofacial vascular lesions are unknown to the author.

The mean ages for oral vascular anomalies depended on the ages of the most prevalent lesion; low if congenital oral hemangiomas or lymphangiomas, but high if oral pyogenic granulomas. The orofacial vascular study in Tanzania with the predominant occurrence of lymphangiomas mostly at birth and hemangioma after birth reported a low mean age of 9.4 years.[12] In contrast, the overall high mean age of 37 years for oral vascular anomalies in this study was similar to the 36 years reported in other studies where pyogenic granuloma was most prevalent.[2,14,21]

The predominant site of oral vascular anomalies varied in countries of study and sometimes depended on the prevalent lesion. The gingiva was the most common site observed in this study due to the prevalence of pyogenic granuloma, which were more on the maxillary than the mandibular gum. In studies focused only on pyogenic granuloma such as at Ibadan Western Nigeria and in Yobe Northern Nigeria, gingival predilection was characteristic and preferentially on the maxillary gum.[2,14,21] Owobu *et al*.[21] reported that anterior (labial) surface of both upper and lower gingivae was the site for 78% (98/137) of all pyogenic granulomas, out of which 70% (69/98) were located on the anterior (labial) surface of the upper (maxillary) gingivae. This anterior (labial) surface often shows poor oral hygiene which is one of the several aetiologic factors[21,25,26] and was corroborated by the reporting of poor oral hygiene in 95% of patients with pyogenic granuloma.[21] It is probable that the preference for anterior labial gingivae by pyogenic granuloma may be due to reduced exposure to outlets of the major salivary glands and reduced saliva presence with lowered cleaning function especially on the maxillary labial gingivae resulting in poor oral hygiene. Other prevalent sites have been reported for oral vascular anomalies in Tanzania: the lip (45.5%) for hemangiomas and the cheek (33.3%) for vascular malformations.[12] The ventral surface of the tongue was the most favored site for oral varix (65.6%) in a Brazilian study.[13]

Pain was not commonly reported in benign oral vascular anomalies,[2,12,21] but this study observed it in 36.2% of anomalies in contrast to the 10% reported by Fernandes *et al*.[23] Majority of the complaints of pain were attributed to cases of pyogenic granuloma. It is probable that these may be due to the inflammatory process that could be associated with pyogenic granuloma lesions. Besides pain, ulceration could be observed in benign

orofacial vascular anomalies without implying any sinister diagnosis. The one case of ulceration observed in this study was in oral hemangioma, and this could be attributed to trauma. Ulceration was also reported in a large Nigerian study of pyogenic granuloma.[21]

Many treatment options are available for benign oral vascular anomalies,[12,13,23] and some authors have recommended sclerotherapy for some vascular malformations, and oral hemangioma,[12,13,23] or a combination therapy.[13] However, surgery was the treatment of choice in this series because all the anomalies were mostly tumors and in accessible sites. Though recurrence was observed in three cases of pyogenic granuloma, they were thought to be residual lesions after inadequate excision.[21]

In conclusion, this study has established the prevalence of benign oral vascular anomalies in Enugu as 5.2% and observed that these occurred predominantly as pyogenic granuloma in the gingivae. Acquired oral hemangioma and oral lymphangioma were infrequent in contrast to reports from other countries.

Limitations: This study is limited to only patients who had biopsy for their vascular anomalies and histopathology examination, thereby excluding patients with other vascular malformations managed conservatively. This omission probably affected the prevalence values, distribution patterns, and other results.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. ISSVA Classification of Vascular Anomalies ©2018 International Society for the Study of Vascular Anomalies. Available from: issva.org/classification. [Last accessed on 2022 Oct 04].
2. Ranjbar Z, Lavaee F, Dordahan H, Shahrokhi sardo M. Prevalence of oral vascular lesions in patients referred to the oral medicine department of Shiraz Dental school during 2001-2017. J Res Dent Maxillofac Sci 2022;7:28-34.
3. Enjolras O, Mulliken JB. Vascular tumors and vascular malformations (new issues). Adv Dermatol 1997;13:375-423.
4. Brahmbhatt AN, Skalski KA, Bhatt AA. Vascular lesions of the head update on classification and imaging review. Insights Imaging 2020;11:19.
5. Abudu EK, Oyebadejo TO, Akinde OR, Efunshile AM, Musa OA, Awolola NA, *et al*. Vasoformative neoplasms in a teaching hospital, Sagamu, Ogun State: A Histopathological review. Niger J Health Biomed Sci 2009;8:32-6.
6. Yusuf I, Mohammed AZ, Iliyasu Y. Histopathological study of soft tissue sarcoma seen in a teaching hospital in Kano, Nigeria. Niger J Basic Clin Sci 2013;10:70-5.
7. Malami SA, Banjo AF. Pathologic features of vascular tumours in infants and children in Lagos, Nigeria. Ann Afr Med 2002;1:92-8.
8. Ngadda HA, Gali BM, Tahir M. Histopathological pattern of vascular tumours in North Eastern Nigeria. High Med Res J 2003;1:52-6.
9. Baker LL, Dillon WP, Hieshima GB, Dowd CF, Frieden IJ. Hemangiomas and vascular malformations of the head and neck: MRI characterization. Am J Neuroradiol 1993;14:307-14.
10. Donnelly LF, Adams DM, Bisset GS. Vascular malformations and hemangiomas: A practical approach in a multidisciplinary clinic. Am J Roentgenol 2000;174:597-608.
11. Obaseki DE, Akhiwu WO, Aligbe JU, Igbe AP, Eze GI, Forae GD. Morphologic patterns of vascular tumours in Benin City, Nigeria: A 12 year retrospective review. Niger J Surg Sci 2013;23:9-13.
12. Moshy J, Owibingire S, Shaban S. Vascular lesions seen among patients treated at Muhimbili National Hospital in Dar es Salaam, Tanzania. East Cent Afr J Surg 2011;16:94-101.
13. Corrêa PH, Nune LCC, Johann ACBR, de Aguiar MCF, Gomez RS, Mesquita RA. Prevalence of oral hemangioma, vascular malformation and varix in a Brazilian population. Braz Oral Res 2007;21:40-5.
14. Lawoyin JO, Arotiba JT, Dosumu OO. Oral pyogenic granuloma: A review of 38 cases from Ibadan, Nigeria. Br J Oral Maxillofac Surg 1997;35:185-9.
15. Bassey GO, Osunde OD, Anyanechi CE. Maxillofacial tumors and tumor-like lesions in a Nigerian teaching hospital: An eleven year retrospective analysis. Afr Health Sci 2014;14:56-63.
16. Arotiba JT, Adebola RA, Ajike SO, Adeola DS, Ladeinde A. Orofacial tumours and tumour-like lesions in Kano, Nigeria. Niger J Surg Res 2003;5:134–9.
17. Adebayo ET, Ajike SO, Abite MG. Audit of oral and maxillofacial surgical conditions seen at Port Harcourt, Nigeria. Ann Afr Med 2008;7:29-34.
18. Aregbesola SB, Ugboko VI, Akinwande JA, Arole GF, Fagade OO. Orofacial tumors in suburban Nigerian children and adolescents. Br J Oral Maxillofac Surg 2005;43:226-31.
19. Orikpete EV, Iyogun CA, Omitola OG. Clinicopathologic analysis of biopsied orofacial lesions seen in children in a tertiary health centre in Port Harcourt: An 11-year review. J Biosci Med 2020;8:1-8.
20. Adekeye EO, Asamoa E, Cohen B. Intraoral carcinoma in Nigeria: A review of 137 cases. Ann R Coll Surg Engl 1985;6:181-2.
21. Owobu T, Omeje UK, Sulaiman SA, Oba OA. Oral pyogenic granuloma: Analysis of 137 cases that presented in a Nigerian tertiary health institution. Niger J Dent Res 2021;6:152-9.
22. Leyman B, Govaerts D, Dormaar JT, Coropciuc BM, Politis C. A 16-year retrospective study of vascular anomalies in the head and neck region. PREPRINT (Version 1). Research Square 2023 Jan 24. doi: 10.21203/rs.3.rs-2456726/v1. Available from: https://doi.org/10.21203/rs.3.rs-2456726/v1.
23. Fernandes DT, Elias RA, Santos-Silva AR, Vargas PA, Lopes MA. Benign oral vascular lesions treated by sclerotherapy with ethanolamine oleate: A retrospective study of 43 patients. Med Oral Patol Oral Cir Bucal 2018;23:e180–7.
24. CobiánI OG, TrigueroII RJP, Pérez HS. Treatment of orofacial vascular malformations by endoluminal sclerosis with diodo laser. Rev Habanera de Cienc Méd 2012;11:511-8.
25. Kamal R, Dahiya P, Puri A. Oral pyogenic granuloma: Various concepts of etiopathogenesis. J Oral Maxillofac Pathol 2012;16:79-82.
26. Osuh ME, Oke GA, Lilford RJ, Owoaje E, Harris B, Taiwo OJ, *et al*. Prevalence and determinants of oral health conditions and treatment needs among slum and non-slum urban residents: Evidence from Nigeria. PLOS Glob Public Health 2022;2:e0000297. doi: 10.1371/journal.pgph.0000297.