

Clinical features of oral angioliipoma – A review

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Abstract: *The aim of this paper was to analyze the data obtained from case reports of oral angioliipoma in terms of age and gender distribution, site of occurrence, nature (intraosseous/extraosseus) and infiltration. an internet search using Google scholar and Pubmed engine was carried out using search terms 'angioliipoma' and 'oral'/'mouth'. English literature full text articles and abstracts of oral angioliipoma obtained from 1976-2016 were analyzed for clinical data and presented in this article.*

Keywords: *angioliipoma, oral, mouth*

INTRODUCTION

Angioliipoma (AL) is rare variant of lipoma which consists of both fatty and vascular elements. [1] ALs were first describe by Bowen in the year 1912. [2] The first published report of an oral angioliipoma was by Davis et al in 1976. [3] Angioliipomas account for 5 to 17% of all the lipomas occurring in the oral cavity, lipomas themselves account for 1-5% of all oral benign tumors [1]. Since oral angioliipoma is rare benign tumor, published reports are mainly restricted to case reports. [4,5]

CLASSIFICATION OF ANGIOLIIPOMAS

Based on their studies Gonzalez-Crussi et al. classified angioliipomas into infiltrating and non-infiltrating. angioliipomas.[6] The non-infiltrating angioliipomas are encapsulated, whereas infiltrating angioliipomas lack this capsule.[6] Among the two, infiltrating angioliipoma (IAL) is a rare lesion typically characterized by infiltration of the surrounding structures, especially skeletal muscle.[7] Based on

whether the angioliipoma occur in bone or soft tissues they can be classified as intra-osseous or extra-osseous types.[8] The intra-osseous variety is extremely rare in comparison to the extraosseus variety. [8,9]

METHODOLOGY FOR ANALYSIS OF THE CASE REPORTS

An analysis of the data obtained from case reports of oral angioliipoma in terms of age and gender distribution, site of occurrence, nature (intraosseous/extraosseus) and infiltration. An internet search using Google scholar and pubmed engine was carried out using search terms 'angioliipoma' and 'oral'/'mouth'. Full text and abstracts of oral angioliipoma cases obtained from 1976-2016 were analyzed. (Table 1)

GENDER PREDICTION OF ORAL ANGIOLIIPOMA

Fregnani ER et al (2003) stated that male to female

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ratio from these intraoral angioliipomas was 1.4:1, in contrast to cutaneous angioliipomas and intraoral lipomas as a group, which either shows no sex predilection or a slight female predilection.[10]

Table 1: Clinical data obtained from full texts and abstract of oral angioliipoma case reports published from the year 1976-2016

Researcher	Year of publication	Type infiltrating/ non-infiltrating	Gender	Age group	Site	Extrasosseous/ intraosseous
Davis et al [3]	1976	-	-	-	hard palate	
Polte HW et al [30]	1976	-	-	-	mandible	intraosseous
Weitzner et al [35]	1978	-	male	4-year-old	cheek	
Campos et al [34]	1980	-	-	-	cheek	
Lewis DM et al [31]	1980	-	-	-	mandible	intraosseous
Brahney CP et al [21]	1981	-	-	-	tongue	
Flaggert et al [19]	1986	-	female	8-year-old	palate	
Reilly et al [15]	1988	-	female	infant	parotid	
Lin SC et al [17]	1989	-	male	49-year-old	tongue	
Manganaro AM et al [32]	1994	-	-	-	mandible	intraosseous
Ali MH et al [36]	1996	-	-	-	cheek	
Sugiura J et al [16]	1999	infiltrating	-	-	muco-buccal fold	
Ida-Yonemochi H et al [12]	2005		male	69-year-old	buccal mucosa	
Altug HA et al [11]	2009	non-infiltrating	male	22-year-old	cheek	
Gerard N et al [20]	2009	-	-	-	upper lip	
Thakur B et al [37]	2010	non-infiltrating	female		cheek	
Palaia G et al [38]	2011	non-infiltrating	female	66-year-old	cheek	
Yanase S et al [13]	2011	non infiltrating	male	76-year-old	cheek	
Sah K et al [4]	2012	non infiltrating	female	9-year-old	upper lip	
Hemavathy S et al [9]	2012	-	female	21-year-old	mandible	intraosseous
Silva-Junior GO et al [5]	2013	non-infiltrating	male	57-year-old	cheek	
2 cases	2013	non-infiltrating	female	29-year-old	masseteric region	
Shahi AK et al [14]	2014	infiltrating	female	9-month-old	cheek	
Mohiuddin SA et al [39]	2014	non-infiltrating	male	46 year old	mandibular buccal vestibule	
Ohnishi Y et al [7]	2015	infiltrating	female	74 year old	lower lip	
Chandrasekaran D et al [1]	2016	non infiltrating	female	55-year-old	palate	

However most of the other researchers have stated that no gender predilection. [1,7,11,12] According to the data obtained from the analysis of the reports, seven cases were in males, ten cases occurred in females. However we could not obtain enough data regarding gender in 9 case reports.

AGE PREDILECTION OF ORAL ANGIOLIIPOMA

Fregnani ER et al (2003) stated that the mean age of the affected patients was 37 years (range, 4 to 81

years), which was in contrast to other variants of lipomas occurring in the oral cavity that occur in the 50-60 year age group.[10] On analysis of the available case reports we found that occurrence of oral angioliipoma ranged from a 4-month-old to 76-year-old individual.[13,14,15] Infiltrating angioliipomas are usually diagnosed in older individuals.[7, 16, 17, 18]

SITE PREDILECTION OF ORAL ANGIOLIIPOMA

Angioliipoma have a prediction of occurrence in the

buccal mucosa, followed by lip.[5] Occurrence in palate has been reported in few cases.[1,3,19]. Upper lip involvement is more common than lower lip.[4, 20] Few cases have also been reported in tongue.[17, 21] However many of the cases of angiolipoma of tongue have the muscle component along with fat and vasculature and thus are termed as angiomylipoma.[22,23, 24, 25]. The occurrence of angiolipoma in the parotid has also been reported.[15] The benign angiolipomas occurring the salivary gland have glandular component apart from adipose and vascular components and are called as sialoangiolipoma.[26] Such cases have been reported to have been observed in parotid and submandibular gland.[27, 28] A rare case of a lipoma with vascular and salivary gland components has been reported in palate also.[29]

INTRAOSSEOUS ANGIOLIPOMA

The first case of intraosseous angiolipoma was by Polte in the year 1976.[30] Mandible is the most common site of occurrence of intraosseous angiolipoma.[9,31,32] Intraosseous lipoma may present a range of clinical manifestations such as swelling, or presence of neurologic signs (hypesthesia or paresthesia) or can be totally asymptomatic.[9] On radiographic examination the lesion usually presents

as well-circumscribed radiolucency.[9] Extraction related trauma is believed to be cause for intraosseous angiolipoma of the jaws. Fatty metamorphosis of a central hemangioma and possible variation osteoporotic bone marrow defect are other suspected etiologies. [33]

INFILTRATING ANGIOLIPOMA

Infiltrating angiolipomas lack encapsulation and are typically characterized by infiltration of the surrounding structures.[6,7,14,16] In contrast to the non-infiltrating angiolipomas are well encapsulated. [34,35,36,37,38]. Infiltrating angiolipoma usually occur in older individuals but a report of 9-month-old diagnosed with infiltrating angiolipoma has recently been published.[14] Infiltrating angiolipomas are also associated with symptoms muscular pain and neural deficits.[39,40]

CONCLUSION

In this review we have made an attempt to analyze the clinical features of oral angiolipoma in terms of its gender predilection, site predilection, infiltrative nature, intra/extraosseous types. The information obtained from this review could aid the clinicians in diagnosis this extremely rare benign lesion.

References:

1. Chandrasekaran D, Chinnaswami R, Narasimhan M, Nithia Kumar AE, Natarajan P. Non Infiltrating Angiolipoma of the Palate in Geriatric Patient: A Case Report with Review of Literature. *Journal of Clinical and Diagnostic Research*. 2016 Jan, Vol-10(1): ZD01-ZD02.
2. Bowen JT. Multiple subcutaneous hemangiomas, together with multiple lipomas, occurring in enormous numbers in an otherwise healthy, muscular subject. *Am J Med Sci*. 1912;144:189–192
3. Davis GB, Stoelting P, Tideman H. Angiolipoma of the hard palate: A case report and review of literature. *J Maxillofac Surg*. 1976;4:242-44.
4. Sah K, Kadam A, Sunita J, Chandra S (2012) Non-infiltrating angiolipoma of the upper lip: a rare entity. *J Oral Maxillofac Pathol* 16, 103-106.
5. Silva-Junior GO, Picciani BL, Raphael C. Costa RC, Barbosa SM, Silveiras MG, Souza RB
6. Cantisano MH, Pires FR. Oral soft-tissue angiolipoma: report of two cases of rare oral
7. lipomatous lesion with emphasis on morphological and immunohistochemical features. *Journal of Oral Science*, Vol. 55, No. 1, 85-88, 2013.
8. Gonzalez-Crussi F, Enneking WF, Aream VM. Infiltrating angio- lipoma. *J Bone. Joint Surg* 1966;48:1111-24.
9. Ohnishi Y, Watanabe M, Fujii T, Yasui H, Kubo H, Kakudo K. Infiltrating angiolipoma of the lower lip: A case report and literature review. *Oncology letters* 2015; 9: 833-836.
10. Nguyen L, Zwagerman N, Grandhi R, McFadden K, Richardson M. Intraosseous Angiolipoma of the Cranium: Case report and review of the literature. *Surg Neurol Int*.

2014; 5: 79.

11. Hemavathy S, Roy S, Asif Kiresur A. Intraosseous angioliipoma of the mandible. *J Oral Maxillofac Pathol* 2012;16:283-7.
12. Fregnani ER, Pires FR, Falzoni R, Lopes MA, Vargas PA. Lipomas of the oral cavity: clinical findings, histological classification and proliferative activity of 46 cases. *Int J Oral Maxillofac Surg* 2003; 32:49-53.
13. Altug HA, Sahin S, Sencimen M, Dogan N, Erdogan O. Non-infiltrating angioliipoma of the cheek: a case report and review of the literature. *J Oral Sci.* 2009 Mar;51(1):137-9
14. Ida-Yonemochi H1, Swelam W, Saito C, Saku T. Angioliipoma of the buccal mucosa: a possible role of mast cell-derived VEGF in its enhanced vascularity. *J Oral Pathol Med.* 2005; 34(1):59-61.
15. Yanasea S, Nomurab J, Matsumurab Y, Katob H, Takeokab T, Imurab H, Matsuurab R, Ko Nakanishib K, Tagawab T. Angioliipoma of the cheek: A case report with a literature review *Asian Journal of Oral and Maxillofacial Surgery* 2011;23: 35–37
16. Shahi AK, Hiralal Ash H, Chatterji K, Singh R. Cellular infiltrative angioliipoma of cheek in an infant. *Natl J Maxillofac Surg.* 2014; 5(2): 202–205.
17. Reilly JS, Kelly DR, Royal SA. Angioliipoma of the parotid: case report and review. *Laryngoscope.* 1988;98(8 Pt 1):818-21.
18. Sugiura J, Fujiwara K, Kurahashi I and Kimura Y: Infiltrating angioliipoma of the mucolabial fold: A case report and review of the literature. *J Oral Maxillofac Surg* 57: 446-448, 1999
19. Lin SC, Wang TY and Hahn LJ: Angioliipoma of the tongue: Report of a case. *Ann Dent* 48: 37 38, 1989.
20. Dalambiras S, Tilaveridis I, Iordanidis S, Zaraboukas T and Epivatianos A: Infiltrating angioliipoma of the oral cavity: report of a case and literature review. *J Oral Maxillofac Surg* 68: 681-683, 2010.
21. Flaggert JJ, Heldt LV, Keaton WM. Angioliipoma of the palate *Oral Surg Oral Med Oral Pathol Oral Radiol* 1986; 61(4):333–336.
22. Gerard N, Schultz DA, Angioliipoma of the Upper Lip: Report of a Case *J Oral Maxfac Surg* 2009; 67(6):1340–1341.
23. Brahney CP, Aria AA, Koval MH, Najjar TA. Angioliipoma of the tongue: report of case and review of literature. *J Oral Surg.* 1981;39(6):451-3.
24. Yura S, Terahata S, Sugiguchi S. A Case of Angiomyoliipoma Arising in the Tongue. *Case Reports in Pathology* Vol 2011, Article ID 698139doi:10.1155/2011/698139
25. Bauer V, Aleric Z, Bujas T. Huge angioliipoma of the tongue. *Otolaryngol Head Neck Surg* 2012; 146(3): 512-513
26. Koizumi H, Ishihama K, Enomoto A, Kogo M. Angiomyoliipoma of the tongue. *Br J Oral Maxillofac Surg.* 2008;46(1):e3-4. Epub 2007 Jul 12.
27. Ide F, Shimoyama T, Horie N. Angiomyoliipomatous hamartoma of the tongue. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1998;85(5):581-4.
28. Maiorano E, Capodiferro S, Fanelli B, Calabrese L, Napoli A, Favia G. Hamartomatous Angioliipoma of the Parotid Gland (Sialoangioliipoma) *Head and Neck Pathol* 2008; 2:36–40.
29. Ingle SB, Patle YG, Jatal SN, Ingle CS, Bhosle SS. Unusual Case of Sialoangioliipoma *Indian Medical Gazette* 2012;146(2): 78-80.
30. Gulati HK, Deshmukh SD, Bhayekar PD. Submandibular sialoangioliipoma: A rare hamartomatous lesion causing diagnostic dilemma. *Natl J Maxillofac Surg.* 2012 Jan-Jun; 3(1): 98–99.
31. Handra-Luca A. Vascular changes in hard palate sialoliipoma: Sialoangioliipoma or vascular malformation? *J Oral Maxfac Pathol* 2015;19(2) 269
32. Polte HW, Kolodny SC, Hooker SP. Intraosseous angioliipoma of the mandible. *Oral Surg Oral Med Oral Pathol.* 1976 May;41(5):637-43.
33. Lewis DM, Brannon RB, Isaksson B, Larsson A. Intraosseous angioliipoma of the mandible. *Oral Surg Oral Med Oral Pathol.* 1980;50(2):156-9.
34. Manganaro AM, Hammond HL, Williams TP. Intraosseous angioliipoma of the mandible: a case report and review of the literature. *J Oral Maxillofac Surg.* 1994;52(7):767-9.
35. Greer RO, Richardson JF. The nature of lipomas and their significance in the oral cavity: A review and report of cases. *Oral Surg Oral Med Oral Pathol* 1973;36:551-7.
36. Campos GA, Grandini SA, Lopes RA. Angioliipoma of the cheek. *Oral Maxfac surg* December 1980;9(6): 486–490.
37. Weitzner S, Moynihan PC. Angioliipoma of the cheek in a child. *Oral Surg Oral Med Oral Pathol Oral Radiol* 1978; 45(1)95–97.
38. Ali MH, el-Zuebi F. Angioliipoma of the cheek: report of a case. *J Oral Maxillofac Surg.* 1996 ;54(2):213-5.
39. Thakur B, Bhalerao S, Sethna K, Gundre N. Giant, recurrent angioliipoma of the cheek – a case report. *Oral Surgery* 2010; 3(3) : 96–99.
40. Palaia G, Gaimari G, Giudice R, Galanakis A, Tenore G, Romeo U, Excision of an oral angioliipoma by KTP laser: a case report. *Annali di Stomatologia* 2011; II (1-2): 28-31

41. Alvi A, Garner C, Thomas W. Angiolipoma of the head and neck. *J Otolaryngol.* 1998;27:100–3.

42. Howard WR, Helwig EB. Angiolipoma. *Arch Dermatol.* 1960;82:924–31.