Content available at: https://www.ipinnovative.com/open-access-journals

Indian Journal of Pathology and Oncology

Journal homepage: www.ijpo.co.in

Case Report

Angiolymphoid hyperplasia with eosinophilia of the tongue – A case report

Sujata Panda[1], Ajit Surya Mohapatra[2], Sindhuja Dash[2], Shushruta Mohanty[2,*]

[1]*NIDAN Diagnostic Centre, Brahmapur, Odisha, India*
[2]*Dept of Pathology, M.K.C.G Medical College and Hospital, Berhampur, Odisha, India*

ARTICLE INFO

Article history:
Received 25-08-2023
Accepted 15-09-2023
Available online 27-09-2023

Keywords:
Angiolymphoid hyperplasia
Epithelioid hemangioma
Vascular lesion

ABSTRACT

Angiolymphoid lymphoid hyperplasia with eosinophilia (ALHE) represents a heterogenous group of benign vascular lesions that is characterized by proliferation of small to medium-sized vascular structures lined by plump epitheloid (histiocytoid) endothelial cells surrounded by mixed inflammatory infiltrate predominantly eosinophils. Intraoral lesions of ALHE are uncommon and tongue involvement is extremely rare. Here we report a case of ALHE of tongue in a 60 yr old female diagnosed on histopathology and confirmed by IHC.

1. Introduction

ALHE is an uncommon vascular lesion, characterized by florid proliferation of small to medium sized vascular structures lined by plump epitheloid (histiocytoid)endothelial cells surrounded by mixed inflammatory infiltrate of lymphocytes, eosinophils and mast cells.[1,2] Clinically it presents as intradermal papules to subcutaneous nodules that are solitary to multiple, red to brown in colour usually occurring in young adults.[1] Most common sites of predilection is subcutaneous tissues of Head and neck particularily in the preauricular area.[3] Other unusual sites reported in literature includes muscles, bone and salivary gland[4] Intraoral lesions of ALHE are uncommon, and most important site being lip, followed by tongue and buccal mucosa.[5] Its etiology and pathogenesis still remains unclear and correct diagnosis is based on distinct histopathological findings.

2. Case Report

A 60-year-old female attended Outpatient dental dept with complaints of ulcer in the right lateral border of the tongue for 3 months. Physical examination revealed a whitish, non tender ulcerated mass measuring 2.5 cm in greatest dimension opposite to 2^{nd} and 3^{rd} molar (Figure 1).

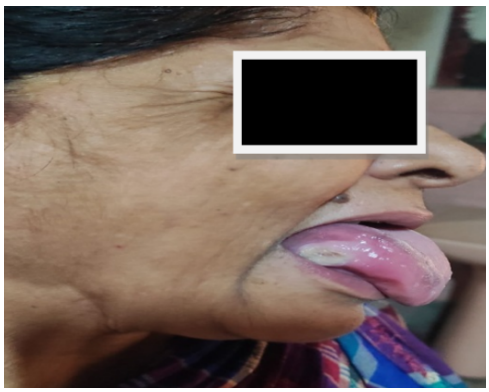

Fig. 1: Whitish non-tender ulcerated mass in the lateral border of the tongue

* *Corresponding author*.
E-mail address: sushruta.mohanty@gmail.com (S. Mohanty).

https://doi.org/10.18231/j.ijpo.2023.069

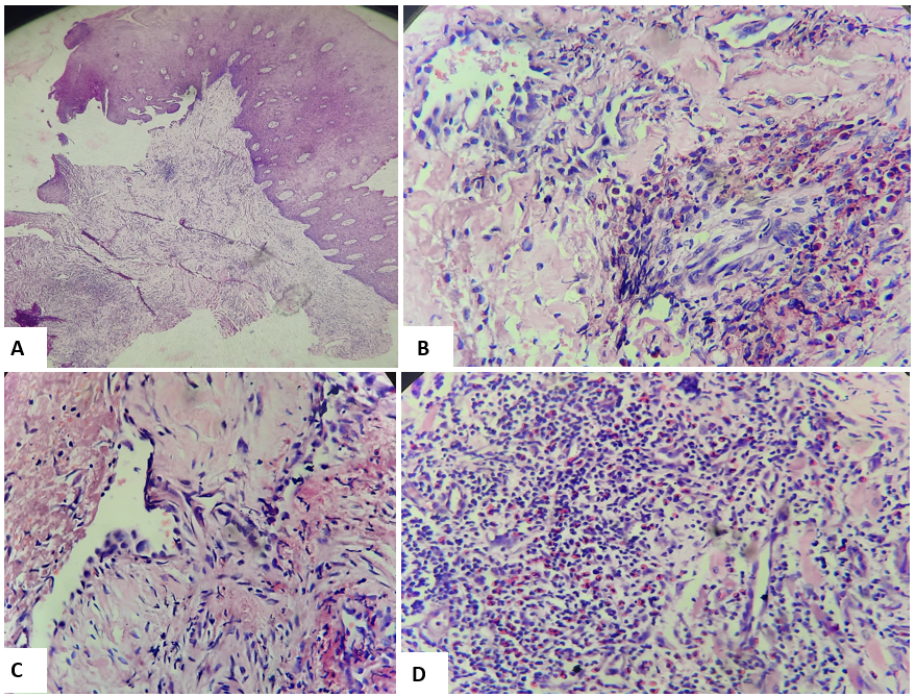

Fig. 2: A): Scanner view 40x Hyperplastic squamous epithelium with underlying inflammatory stroma showing prominent vascularity; **B**): LP 100x- blood vessels lined by epithelioid like endothelial cells with vesicular nuclei that protruded into the lumina in a background of mixed inflammatory infiltrate; **C**): LP 100X- Blood vessel lined by epithelioid endothelial cells with vesicular nuclei protruding into the lumina; **D**): LP 100x showing mixed inflammatory infiltrate with predominantly eosinophils

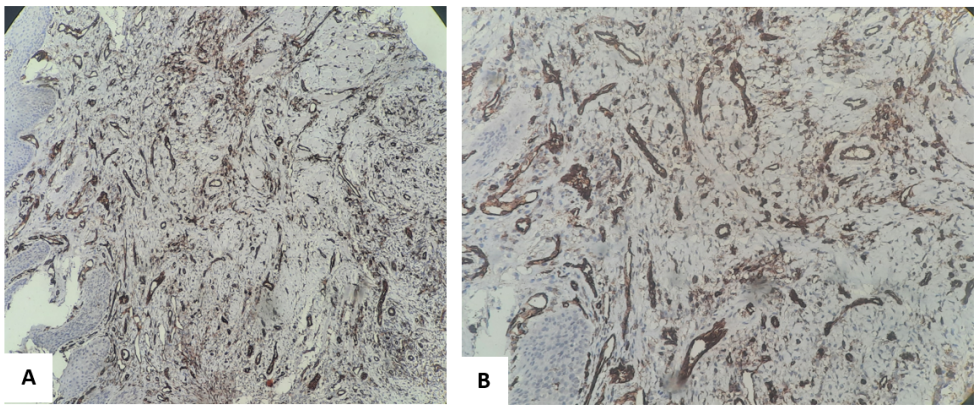

Fig. 3: A): (Scanner view 4x); **B**): (Low power view) - Showing CD31 positive in vascular lumina lined by endothelial cells

Medical history and personal history were not significant pertaining to the ulcerated lesion. No regional lymphnodes were enlarged. Incisional biopsy from the ulcerated mass was taken and sent to our dept for histopathological study. Microsection showed a hyperplastic squamous epithelium with underlying inflammatory stroma showing prominent vascularity (Figure 2 a). These blood vessels were lined by epithelioid like endothelial cells with vesicular nuclei that protruded into the lumina and large vacuolated eosinophilic cytoplasm. A dense cellular infiltrate comprising of lymphocytes and predominantly eosinophils were seen surrounding the vessels (Figure 2 b,c,d). The inflammatory process extended beyond the skeletal muscle tissue. No mitosis or nuclear atypia was seen. Immunohistochemical stain for antibodies against CD31 and CD34 showed intense positive staining for vascular structures (Figure 3 a, b). Based on histopathology and IHC, diagnosis of angiolymphoid hyperplasia with eosinophilia was signed out.

3. Discussion

ALHE was first described in the year 1969 by Wells and Whimster.[2] In 1983 Enzinger and Weiss described the same lesion as epithelioid haemangioma [EH].[6] Other terminologies used to describe the lesion included inflammatory angiomatous nodule, pseudo or atypical pyogenic granuloma and histiocytoid hemangioma.[7] Intraoral lesions of ALHE are uncommon with tongue being one of the rarest site of involvement. Extensive search in PubMed data base indicated that, till date 12 cases have been previously reported.[3,5–12] Regarding pathogenesis of ALHE there is still an ongoing debate as to whether ALHE represents a vascular neoplasm or is a reactive inflammatory condition as a result of trauma induced due to mastication bites.[3,5] The inflammatory cells play an important role in proliferation of endothelial cells. A diagnosis of ALHE was not suspected preoperatively in our case as tongue is one of the rarer sites of involvement and moreover clinically the lesion mimicked that of malignancy. Histopathology played a vital role in clinching our diagnosis. Histologically ALHE is characterized by proliferation of small to medium-sized vascular structures lined by epithelioid like endothelial cells with eosinophilic vacuolated cytoplasm and vesicular indented nuclei that protrude into the lumina forming tomb stone appearance. IHC was positive for CD31 and CD34 in the vascular structures thus confirming the presence of endothelial differentiation in the lesion. Most common differential diagnosis of ALHE is kimura's disease. Kimura disease is rather a inflammatory process with primary nodal involvement and occurs in younger men, in contrast to ALHE where it's usually neoplastic (benign vascular tumor) showing soft tissue involvement and occurs in middle-aged women. In ALHE the lesions are mostly dermal and there is prominent vascular proliferation with lobular arrangement however in Kimura's disease there is minor vascular proliferation, lymphoid hyperplasia and eosinophilic microabcesses.

4. Conclusion

Angiolymphoid hyperplasia with Eosinophilia is a rare entity and should be considered in differential diagnosis when abnormal proliferation of blood vessels are seen. The most common treating modality includes surgical excision and a care must be taken to ensure complete margins of the surgical specimen so as to minimize the possibility of recurrance that often poses a therapeutic challenge. Review of literatures has depicted a recurrence rate of 44.2% in ALHE.[13] Though it's a benign lesion a prompt diagnosis and close monitoring of the patient is essential for futher intervention and better management of the patient.

5. Source of Funding

None.

6. Conflict of Interest

None.

References

1. Olsen TG, Helwig EB. Angiolymphoid hyperplasia with eosinophilia. A clinicopathologic study of 116 patients. *J Am Acad Dermatol*. 1985;12(5 Pt 1):781–96.
2. Wells GC, Whimster IW. Subcutaneous angiolymphoid hyperplasia with eosinophilia. *Br J Dermatol*. 1969;81(1):1–14.
3. Martín-Granizo R, Mu~noz E, Naval L, Martín R, Goizueta C, Diaz FJ. Epithelioid hemangiomas of the maxillofacial area. A report of three cases and a review of the literature. *Int J Oral Maxillofac Surg*. 1997;26(3):212–4.
4. O'connell JX, Kattapuram SV, Mankin HJ, Bhan AK, Rosenberg AE. Epithelioid hemangioma of bone. A tumor often mistaken for low-grade angiosarcoma or malignant hemangioendothelioma. *Am J Surg Pathol*. 1993;17(6):610–7.
5. Sun ZJ, Zhang L, Zhang WF, Alsharif MJ, Chen XM, Zhao YF. Epithelioid hemangioma in the oral mucosa: a clinicopathological study of seven cases and review of the literature. *Oral Oncol*. 2006;42(5):441–7.
6. Enzinger FM, Weiss SW. Soft tissue tumors. *Am J Dermatopathol*. 1989;11:593–4.
7. Park Y, Chung J, Cho CG. Angiolymphoid hyperplasia with eosinophilia of the tongue: report of a case and review of the literature. *Oral Oncol*. 2002;38(1):103–6.
8. Razquin S, Mayayo E, Citores MA, Alvira R. Angiolymphoid hyperplasia with eosinophilia of the tongue: report of a case and review of the literature. *Hum Pathol*. 1991;22(8):837–9.
9. Toro J, Rocher FP, Marques JBV, Galofre JD. Pathologic quiz case 3. Angiolymphoid hyperplasia with eosinophilia of the tongue. *Arch Otolaryngol Head Neck Surg*. 1992;118(2):216–8.
10. Garrido-Ríos AA, Sanz-Mu~noz C, Torrero-Anton MV, Martínez-García G, Miranda-Romero A. Angiolymphoid hyperplasia with eosinophilia on the tongue. *Clin Exp Dermatol*. 2009;34(8):729–31.
11. Iguchi Y, Inoue T, Shimono M, Yamamura T, Shigematsu T, Takahashi S. Kimura's disease and its relation to angiolymphoid hyperplasia with eosinophilia: report of three cases and review of the literature. *J Oral Pathol*. 1986;15(3):132–7.
12. Jacob J, Roy S, Dildeepa SN. Angiolymphoid hyperplasia with eosinophilia — A case report. *Indian J Otolaryngol Head Neck Surg*. 2006;58(3):285–7.
13. Lagha BI, Souissi A. Angiolymphoid hyperplasia with eosinophilia (Internet). Treasure island (FL): StatPearls Publishing; 2020.

Author biography

Sujata Panda, Consultant Pathologist

Ajit Surya Mohapatra, Assistant Professor

Sindhuja Dash, PG Student

Shushruta Mohanty, Assistant Professor https://orcid.org/0000-0002-3122-6892

Cite this article: Panda S, Mohapatra AS, Dash S, Mohanty S. Angiolymphoid hyperplasia with eosinophilia of the tongue – A case report. *Indian J Pathol Oncol* 2023;10(3):310-312.