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Case Report

Angiomyoma vs solitary fibrous tumour – A histopathological dilemma

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ABSTRACT

Benign neoplasms associated with blood vessels make up a wide spectrum of lesions ranging from reactive lesions to high grade malignant tumours in the oral cavity. Angioleiomyoma is one such uncommon benign soft tissue tumor rarely observed in oral tissues. Microscopically characterized by proliferation of smooth muscle cells intermingled with abundant blood vessels. It rarely affects the upper lip and only few cases have been reported. This is a rare case of a swelling involving the upper lip in a 65-year-old man present for 4 years. History of trauma to the same region was reported. The medical, personal and dental history of the patient were unremarkable. Histopathological evaluation using routine H&E stain followed by special stains and immunohistochemical stains were done for ruling out various differential diagnosis and arriving at the diagnosis of Angiomyoma. For the year-long follow-up period after the surgical removal, no recurrence was noted. The presented case emphasizes the importance of histological and immunohistochemical examinations to arrive at the definitive diagnosis of rare tumours such as angiomyoma.

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1. Introduction

Angiomyoma also known as vascular leiomyoma or Angioleiomyoma is one of the rare types of leiomyomas among its three histological groups- (i) vascular (angioleiomyoma), (ii) solid and (iii) epithelioid (leiomyoblastomas).[1] It was first described in 1854 by Rudolf Virchow.[2] Angioleiomyomas (AL) are the most common variants affecting the oral cavity, representing 64.0%–92.3% of all variants of oral leiomyoma.[3] This may be due to the rich blood supply of the oral cavity. Clinically they are small, painless, slowly growing tumours located subcutaneously without superficial changes.[4] Lips, palate, buccal mucosa and tongue are the commonly involved sites in the oral cavity.[5] Commonly seen in the age group of 40 to 50 years with a slight male predominance and characterized histologically, as a well circumscribed lesion composed of a tortuous mass of thick walled blood vessels among mesenchymal spindle cells with eosinophilic cytoplasm and elongated basophilic cigar shaped nuclei.[6] Due to the pain producing nature of cutaneous and vascular leiomyomas they were collectively termed 'tubercular dolorosum'.[7]

The clinical features of AL overlap with many other lesions of the oral cavity like mucocele, pleomorphic adenoma, mesenchymal tumors, lymphangioma, pyogenic granuloma, and schwannoma, solitary fibrous tumor etc. These lesions need to be ruled out before giving the diagnosis of AL. It requires a histopathological confirmation not only using regular H&E but special stains and IHC markers such as α-SMA, desmin, CD34 and HHF-35 are

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also required.[8] Here we present a case of a long-standing swelling involving the upper lip of an adult male.

2. Case Presentation

A 65-year-old male presented to our department, with a chief complaint of swelling in his upper lip for 4 years. His medical history was unremarkable, and had no history of tobacco smoking or alcohol consumption. The patient was apprehensive about the labial growth. Upon eliciting the history, an oval-shaped peanut-sized swelling initially appeared along the midline of the upper lip, following blunt trauma to upper front teeth and lip which grew gradually over 4 years to reach the present size measuring approximately 1.5x2cms in size. The swelling was not associated with pain or any discomfort. On extraoral examination (Figure 1 a) a solitary dome shaped swelling was seen extending superior-inferiorly from the vermillion border of the upper lip to 2cms downwards. The colour of the overlying mucosa was normal. Intraoral examination was unremarkable with generalised attrition, abrasion, gingival recession and root stumps. On palpation, the inspe ctory findings were confirmed. The mass was superficial, non-tender, smooth, sessile, movable, and firm. No fluctuation or bruit was present. The patient was afebrile, and there was no cervical lymph nodes enlargement. Based on the clinical findings a provisional diagnosis of traumatic fibroma and a differential diagnosis of minor salivary gland tumour was given.

The patient was advised oral prophylaxis after which he underwent an excisional biopsy under local anaesthesia under antibiotic prophylaxis (Figure 1 b,c). The specimen was sent in formalin for histopathological analysis (Figure 1 d). Whole of the tissue bit was taken for processing.

Microscopic examination of the H&E- stained section revealed a well-encapsulated lesion with hyper cellular and hypo cellular areas in connective tissue (Figure 2 a). The hyper cellular areas show round to ovoid cells with vesicular nuclei and prominent nucleoli surrounding the blood vessels. Congested blood vessels are exhibiting slit like and stag horn pattern (Figure 2 b). The hypo cellular areas show a dense hyalinised stroma interspersed with a few spindle-shaped cells. Histological differential diagnoses of Solitary fibrous tumour (SFT), Heamangipericytoma (HPC) and Angiomyoma were considered. However, numerous thick-walled, congested blood vessels were seen within these bundles of smooth muscle (Figure 2 c). Individual smooth muscle cells possess abundant eosinophilic cytoplasm and elongated cigar-shaped nuclei (Figure 2 d). These cells lack both nuclear hyperchromasia and significant numbers of mitosis. Mild inflammatory infiltrate composed of lymphocytes and few plasma cells are also seen. These features pointed towards Angiomyoma. To rule out confusion between SFT and Angiomyoma, Masson Trichrome staining was done which distinguishes smooth

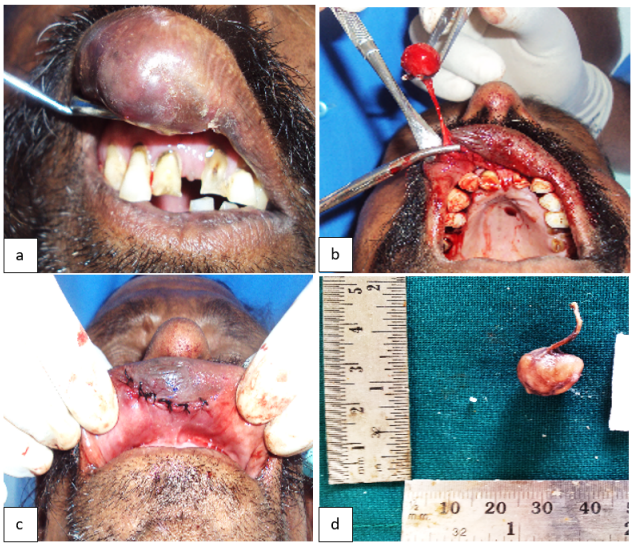

Fig. 1: a): Well defined dome shaped swelling; **b**): Surgical excision under LA; **c**): Post-operative suture placement; **d**): Excised specimen received

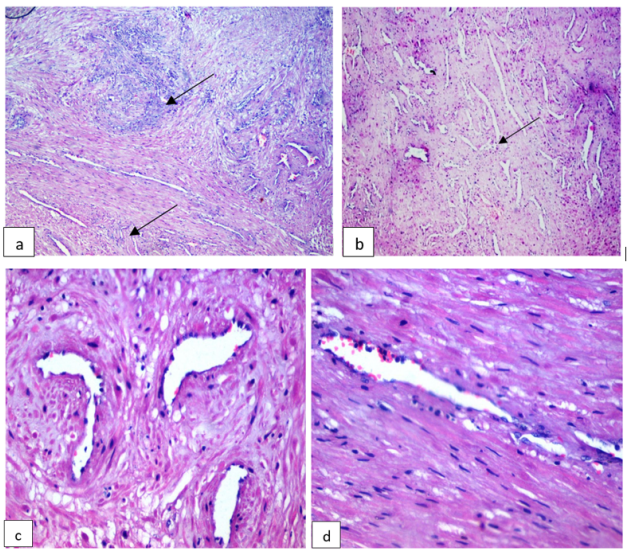

Fig. 2: a): Connective tissue exhibiting hypercellular and hypocellular areas; **b**): Slit like and stag horn pattern of blood vessels; **c**): Thick-walled blood vessels surrounded by smooth muscle cells; **d**): Thick-walled blood vessels surrounded by smooth muscle cells

muscle and collagen by staining smooth muscle cells as red and collagen fibers as blue. As our case stained with this pattern, we could recognize the smooth muscle as the main part of the lesion. To further confirm our diagnosis, immunohistochemical staining was performed using SMA and H-caldesmon that showed a diffuse positivity which is characteristic of Angimyoma (Figure 4 a, b). S100 was negative (Figure 5 a) ruling out any neural tumours. On the other hand, CD34, CD99 were negative which are distinct markers in diagnosing SFT (Figure 5 b, c).

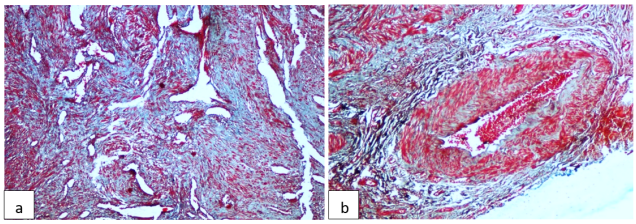

Fig. 3: Masson's trichrome staining exhibiting collagen fibers in blue and smooth muscle cells in red (**a**) 4x (**b**) 10x

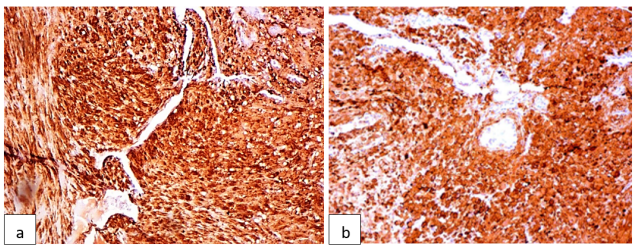

Fig. 4: a): Positive immunohistochemical reaction of smooth muscle actin; **b):** Positive immunohistochemical reaction of H-caldesmon

A diagnosis of Angiomyoma was given based on the clinical features, histologic appearance under routine H&E stain, Masson Trichrome stain and IHC markers. The postoperative period was uneventful. For the year-long follow-up period after the surgical removal, no recurrence was noted.

3. Discussion

Angioleiomyoma (AML) are benign tumors of the vascular smooth muscle origin. They are rarely encountered in the oral cavity and when found, most commonly seen on the palate and the lower lip.[9] WHO defines AML as "A frequently painful, benign subcutaneous or deep dermal tumor composed of mature smooth muscle bundles which are surrounded and interlaced by vascular channels".[5] The exact etiology of AML is unspecified. However, trauma, local infection, hormonal influences and arterio-venous malformations are some of the possible causes.[10] In our case, trauma was considered as an etiological factor.

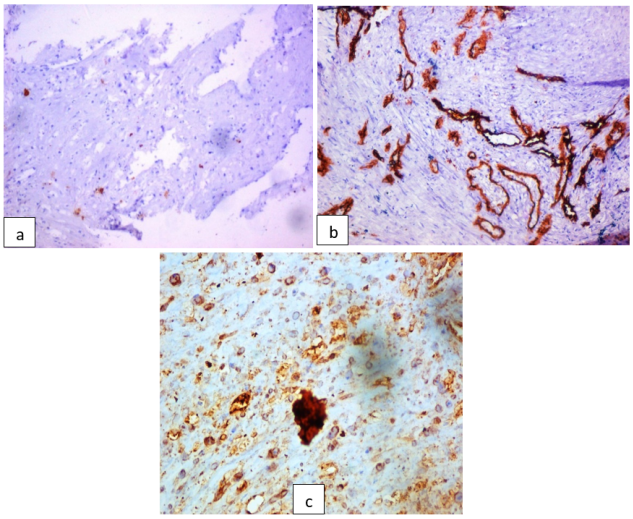

Fig. 5: a): Negative immunohistochemical reaction of S100; **b):** Negative immunohistochemical reaction of CD34; **c):** Negative immunohistochemical reaction of CD99

Clinically it is very difficult to diagnose AML as it has no distinct features. One must rely on the histopathology which in itself is strenuous because of similarity with various other entities. It is believed that AML starts as an hemangioma, proceeds to form vascular leiomyoma and then ends up becoming a solid leiomyoma.[11]

AMLs being spindle cell neoplasms, careful differentiation is critical from other spindle cell neoplasms such as neurofibroma, neurilemmoma, fibrous histiocytoma, solitary fibrous tumour. Neurofibroma has spindle cells with wavy nuclei and interlacing bundles, neurilemmoma shows Antoni A and Antoni B cell pattern. Apart from the histological differences, a negative S100 reaction ruled out the neural tumours. Furthermore, Fibrous histiocytoma has spindle cell proliferation with vesicular nuclei and the cells are arranged in a storiform pattern which is absent in AML. Solitary myofibroma can cause confusion where numerous proliferating fibroblasts and myofibroblasts are seen positive for SMA marker but the cells have tapered nuclei whereas in AML cigar shaped nuclei is present and desmin is positive in AML whereas it is negative in the former.[4,5]

Myopericytoma can also be confusing because AML and the latter share similar histologic features. Nonetheless, H-caldesmon is strongly positive in AML hence differentiating between the two lesions. H-caldesmon is a more specific immunohistochemical indicator for smooth muscle fibers than SMA.[12]

Solitary Fibrous Tumour (SFT) was considered in our case since this tumour also has spindle cells arranged in pattern-less pattern around slit like blood vessels and has hypocellular & hypercellular areas as seen in the case of AML. Special stains like Masson Trichome, Van-

Gieson along with immunostains such CD34, CD99 can help in clearing the dilemma between the two facsimileing tumours.[13]

Angioleiomyomas are treated by surgical excision. Around 5% of the tumours show local recurrence owing to incomplete excision.[10] There are no cases of malignant transformation reported in literature till now. Although angioleiomyosarcoma exists, hence one should be vigilant to differentiate between the benign AML and the malignant counterpart. Pathological features such as atypia, necrosis, pleomorphism, cellularity, and mitotic activity indicate a malignant tumour.[14]

4. Conclusion

The diagnosis of histopathologically similar and uncommon benign tumours is crucial to avoid confusion with morphologically similar but more aggressive and histologically bland-appearing lesions. The presented case emphasizes the importance of histological and immunohistochemical examinations to arrive at the definitive diagnosis of rare tumours such as angiomyoma.

5. Source of Funding

None.

6. Conflict of Interest

None.

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None.

References

1. Reddy BVR, Rani BS, Anuradha CH, Chandrasekhar P, Shamala R, Lingamaneni KP. Leiomyoma of the mandible in a child. *J Oral Maxillofac Pathol*. 2011;15(1):101–4.
2. Virchow R. Cavernosal expandability is an erectile tissue mechanical property which predicts trabecular histology in an animal model of vasculogenic erectile dysfunction. *Arch Pathol Histopathol*. 1854;6:553–4.
3. Hassona Y, Sawair F, Scully C. Angioleiomyoma of the upper lip. *BMJ Case Rep 2017; 2017:* . 2017;2017. doi:bcr2016219172.
4. Cepeda LAG, Rivera DQ, Rocha FT, Huerta ERL, Sánchez ERM, et al. Vascular leiomyoma of the oral cavity. Clinical, histopathological and immunohistochemical characteristics. Presentation of five cases and review of the literature. *Med Oral Patol Oral Cir Bucal*. 2008;13(8):483–8.
5. Al-Amad SH, Angel C, Grady JO, Mccullough MJ. Angioleiomyoma on the hard palate. *Oral Oncol Extra*. 2006;42:244–6.
6. Damm DD, Neville BW. Oral leiomyomas. *Oral Surg, Oral Med, Oral Pathol*. 1979;47(4):343–8.
7. Mcgowan DA, Jones JH. Angioma (vascular leiomyoma) of the oral cavity. *Oral Surg, Oral Med, Oral Pathol*. 1969;27(5):649–52.
8. Gueiros LA, Romañach MJ, Pires-Soubhia AM, Pires FR, de Almeida OP, Vargas PA. Angioleiomyoma affecting the lips: report of 3 cases and review of the literature. *Med Oral Patol Oral Cir Bucal*. 2011;16(4):e482–7.
9. Mahima VG, Patil K, Srikanth HS. Recurrent oral angioleiomyoma . *Contemp Clin Dent*. 2011;2(2):102–5.
10. Hung KF, Yan AH, Kao SY, Chang CS. Angioleiomyoma in the right lingual gingiva a case report. *Chin J Oral Maxillofac Surg*. 2005;16:179–8.
11. Sánchez MAG, Bonnin CM, Aytés LB, Escoda GC. Leiomyoma of the hard palate: a case report. *Med Oral Patol Oral Cir Bucal*. 2007;12(3):221–4.
12. Amy PB, Yaffe D, Kawar V, Akrish R, S. Abu El-Naaj I. Oral myopericytoma: a rare pediatric case report and a review of the literature. *BMC Oral Health*. 2021;21(1):1–6.
13. O'regan EM, Vanguri V, Allen CM, Eversole LR, Wright JM, Woo SB. Solitary fibrous tumor of the oral cavity: clinicopathologic and immunohistochemical study of 21 cases. *Head Neck Pathol*. 2009;3(2):106–15.
14. Rey RL, Franco FL, Oliveira GG, Seijas BP, Guitián D, Cembranos J. Oral leiomyoma in retromolar trigone: A case report. *J Coll Physicians Surg Pak*. 2007;25(Suppl 2):130–1.

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