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Gingival angioleiomyoma - A rare case report

Twinkle Thakker [a], Venkatesh Anehosur [a,*], Dinesh U.S [b], Jawahar Anand [a], Niranjan Kumar [c]

[a] Oral and Maxillofacial Surgery, SDM Craniofacial and Research Centre, SDM College of Dental Sciences and Hospital, Sattur, Dharwad, 580009, Karnataka, India
[b] Department of Pathology, SDM College of Medical Sciences and Hospital, Sattur, Dharwad, 580009, Karnataka, India
[c] Professor Plastic Surgery Department, SDM Craniofacial and Research Centre, SDM College of Dental Sciences and Hospital, Sattur, Dharwad, 580009, Karnataka, India

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ABSTRACT

Angioleiomyoma which was initially classified as vascular leiomyoma has been reclassified as a tumor of perivascular origin with its etiology still being uncertain. It is a rare entity to be observed in head and neck region with an incidence of less than 0.06%. In the oral cavity its origin from the gingiva is rare. Clinical presentation of pathologies may vary and oral cavity being a constituent of mixed tissues there may be pathologies found which are seen in other parts of body. Thus, histopatholgy plays an important role in diagnosis of these rare lesions. Present a rare case of large angioleiomyoma arising from gingiva.

1. Introduction

Angioleiomyoma which was initially classified as a vascular benign tumor of smooth muscle (leiomyoma) has been reclassified as a tumor of perivascular origin by WHO in 2013 [1,2,5]. The common sites for it's occurrence include uterus, gastrointestinal tract, skin and subcutaneous tissue whereas one of the unusual site involved may be oral cavity [4]. Of all the angioleiomyomas, less than 0.06% is found to be associated with head and neck region [6]. Middle-aged males have a higher prevalence of angioleiomyoma [2].

Clinically, angioleiomyoma appear as a well-defined, painless, gradual growing swelling which is a feature similar to many other lesions, hence the diagnosis can be derived on only basis of histolopathologic findings which includes characteristics features of proliferation of mature smooth muscle cells and numerous blood vessels [1]. In cases of unclear histologic features, further specific immunohistochemistry is used [7].

The present report describes an unusual case of large angioleiomyoma arising from the gingiva.

2. Case report

A 17yrs old male presented with a growth in the left posterior mandible since 3 months which had gradually increased to a size of 4 × 3cm. On examination patient appeared normal with no extra-oral abnormality. On palpation there was a single, firm and tender

* Corresponding author. Oral and Maxillofacial Surgery, SDM Craniofacial and Research Centre, SDM College of Dental Sciences and Hospital, Sattur, Dharwad, 580009, Karnataka, India.
E-mail addresses: twinks.ht@gmail.com (T. Thakker), venkysdm@gmail.com (V. Anehosur), drusdinesh@sdmmedicalcollege.org (D. U.S), jawanand@gmail.com (J. Anand), drniranjankumar@sdmmedical.org (N. Kumar).

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left submandibular lymph node. Intra-orally, a well defined pedunculated, oval soft tissue mass with smooth surface noted arising from the lingual interdental gingiva between mandibular left 1st and 2nd molars *(*Fig. 1*)*. The boundaries between the growth and normal tissue were relatively clear. It was non-tender, firm in consistency with presence of bleeding on probing. It was freely movable with no adhesion to the surrounding tissue. No restriction in tongue movements noted. Grade 2 mobility observed with respect to mandibular left 1st and 2nd molars. Fine needle aspiration cytology (FNAC) of the lesion was performed which was suggestive of a granulation tissue growth. On the basis of clinical evaluation, diagnostic hypothesis of peripheral giant cell granuloma was made. Clinical differential diagnosis focused on ruling out pyogenic granuloma, central giant cell granuloma, fibroma, peripheral ossifying fibroma and mesenchymal tumor.

Further investigations included radiographic evaluation with mandibular occlusal view x-ray *(*Fig. 2*)* and orthopantamogram (OPG) which revealed periodontal ligament widening with respect to 36 to mandibular left 1st and 2nd molars and increase interdental gap between them.

Incisional biopsy was performed under local anesthesia and the histopathological report with the use of immunohistochemical (IHC) markers was suggestive of angioleiomyoma. IHC study was positive for vimentin and smooth muscle actin (SMA) and negative for cytokeratin, epithelial membrane antigen (EMA) and S100.

The tumor was excised with 1cm clear margin under general anesthesia along with extraction of 36, 37 and 38 and closure of the defect done using buccal fat pad *(*Fig. 3,4*)*. The specimen was sent for histopathological analysis.

Histological sections of the specimen with hematoxyline and eosin staining showed nodule with surface covered with fibrinous exudates, neutrophils and granulation tissue with proliferating blood vessels. Underlying were noted plump spindle shaped cells in fascicles and bundles (Fig. 5a and b). Chondroid metaplasia was noted throughout the specimen which was characterized by multiple circumscribed lobules of chondroid material without evidence of hyaline cartilage. IHC analysis confirmed presence of smooth muscles with positive SMA marker (Fig. 6a and b), vimentin marker (Fig. 7a and b) and CD 34 marker (presence of vascular channels) was found positive. Based on these features the final diagnosis of angioleiomyoma of gingiva was established.

Patient was followed up for a period of 1 year with uneventful healing and no signs of recurrence.

3. Discussion

Angioleiomyoma of the oral cavity is a rare entity to be observed due to limited presence of smooth muscles which is restricted to blood vessels (tunica media), circumvallate papillae (ductus lingualis) and arteriovenous anastomoses [1,2,4]. In the review of 562 cases of angioleiomyoma done by Hachisuga et al., the incidence of oral cavity lesions accounted for only 2.7% [4].

Ishikawa et al. reported the incidence of angioleiomyoma at various oral subsites of which lip (48.6%) being most common followed by palate (21.1%), buccal mucosa (9.2%), tongue (9.2%), mandible (8.3%), buccal sulcus (0.9%), labial sulcus(0.9%), mouth floor (0.9%), and gingiva (0.9%) [6].

Though the actual etiology of the tumor is still unresolved, the postulated hypothesis include minor traumas, decreased venous flow, hormonal impairments, and genetic variations in which mutations to the BRAF, NF1, NOTCH2 and NOTCH3 genes are under investigation [2,6,8]. Morimoto et al. reported an increased prevalence of angioleiomyoma of the extremities in women (male: female ratio, 2:3) whereas that of the head and neck region in men (male: female ratio, 3:1) [6]. Usually, 4th to 6th decades of life is the peak

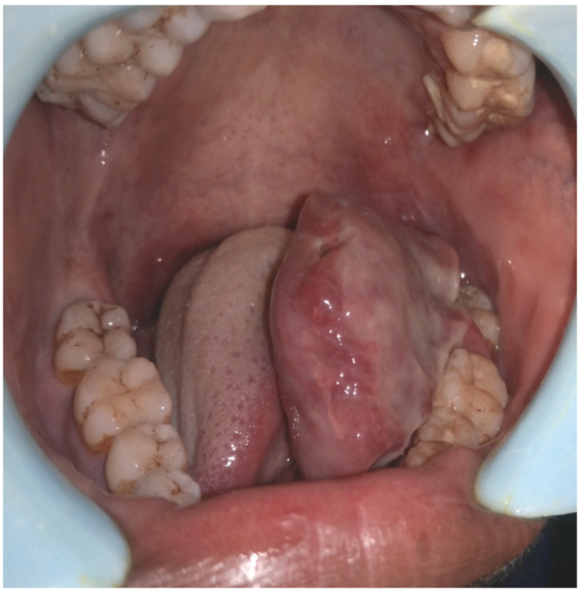

Fig. 1. Intraoral soft tissue mass.

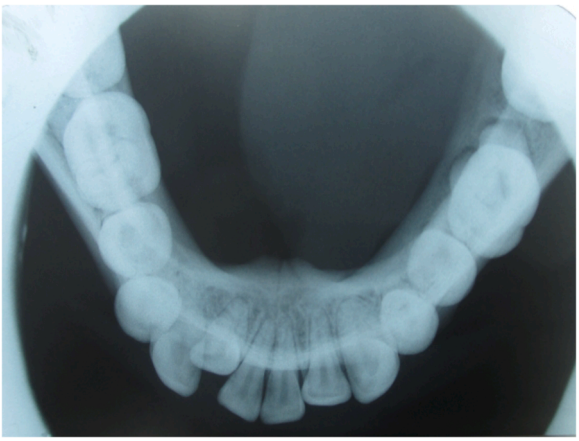

Fig. 2. Mandibular occlusal view showing radiolucent soft tissue shadow on the lingual aspect.

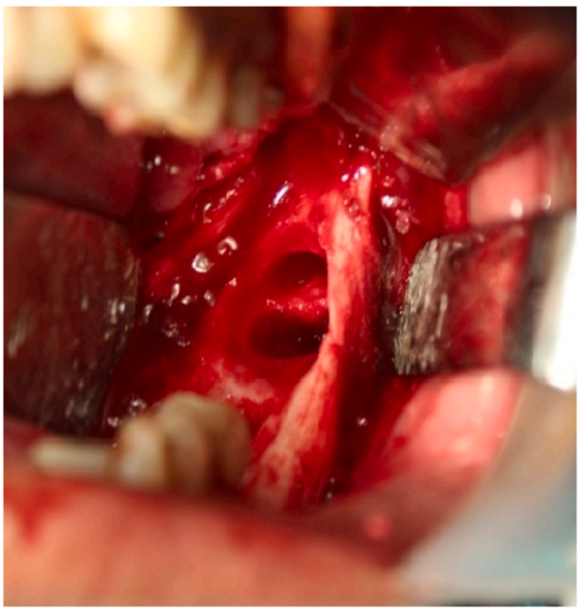

Fig. 3. Surgical excision with extraction of teeth.

age for presentation of angioleiomyomas; but there have been cases reported in pediatric age group as well [2]. The patient in the present report was a 17 year-old male.

Based on the clinical characteristics, the differential diagnosis pertinent to angioleiomyoma usually includes variety of other benign mesenchymal tumors (e.g. lipoma, fibroma and neurofibroma), benign salivary gland lesions like deep-seated mucocele and pleomorphic adenoma, vascular lesions such as lymphangioma and hemangioma, pyogenic granuloma, and soft tissue cysts, such as dermoid cyst. Whereas, based on the histomorphologic features of proliferation of smooth muscle cells, the differentials such as neurofibroma, neurilemoma, nodular fasciitis, and fibrous histiocytoma which are primarily composed of spindle-shaped cells [9].

Morimoto in 1973 put forth 3 histologic subtypes of angioleiomyoma which included solid, venous, and cavernous [10]; later this categorization was adopted by the World Health Organization and was found that the venous type is more frequently associated with oral cavity.

In histologic sections, the identification of the smooth muscle nature of the cells in angioleiomyoma can be done by using either routine histochemical stains, such as Mallory's phosphotungstic acid, Masson trichrome or immunohistochemical stains, such as smooth muscle actin, when needed [9]. Morphological findings along with immunohistochemical results are helpful for better differentiation between various lesions [4,9,11,12]. In present case, the muscular origin of the neoplastic cells in this tumor was confirmed based on strong and diffuse immunostaining observed for myogenic proteins, such as α-SMA and vimentin.

The most indicated treatment is conservative surgical excision [2,3]. Despite of the vascular nature of the tumor, profuse bleeding during removal is rarely encountered [9]. The case was treated with surgical excision of the lesion along with extraction of the involved

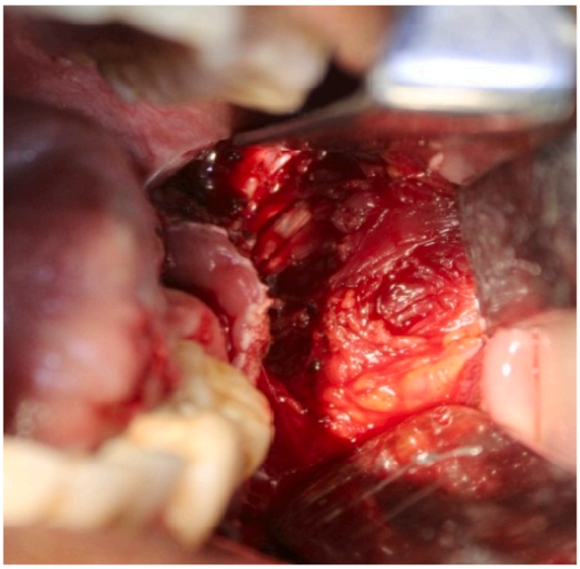

Fig. 4. Closure using buccal fat pad.

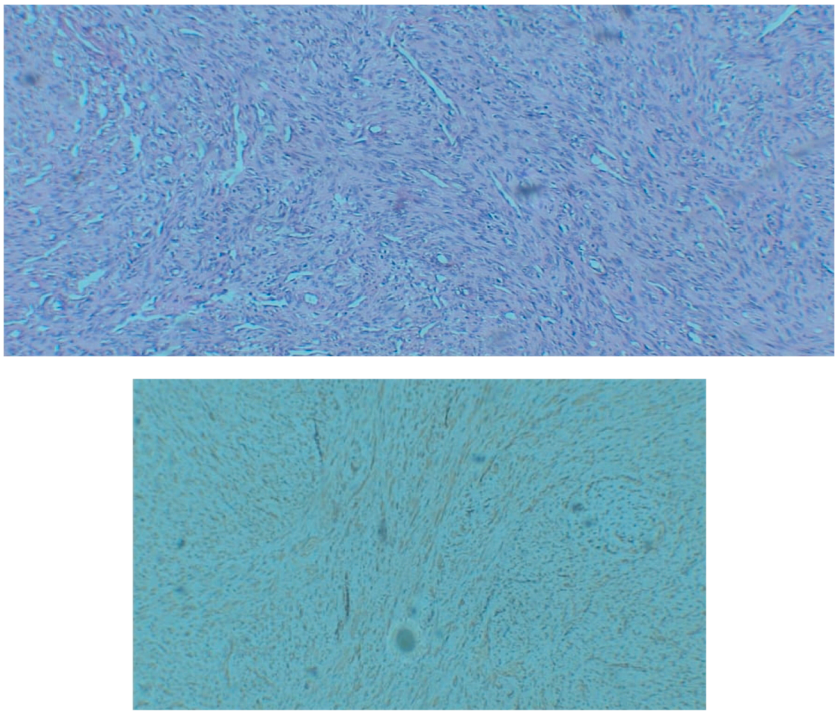

Fig. 5. a: Histological section - Hematoxyline and eosin staining (low magnification), b: Histological section - Hematoxyline and eosin staining (high magnification).

teeth and closure of defect done using buccal fat pad. Recurrence chances are quite rare and could be associated with incomplete surgical excision. No reports of malignant transformation of the lesion are noted and patient's prognosis is considered excellent.

4. Conclusion

To conclude, angioleiomyoma of gingiva is a rare pathology which needs to be differentiated from commonly occurring benign pathologies with clinical and histomorphological findings. Surgical excision remains the main modality with regular follow-up to

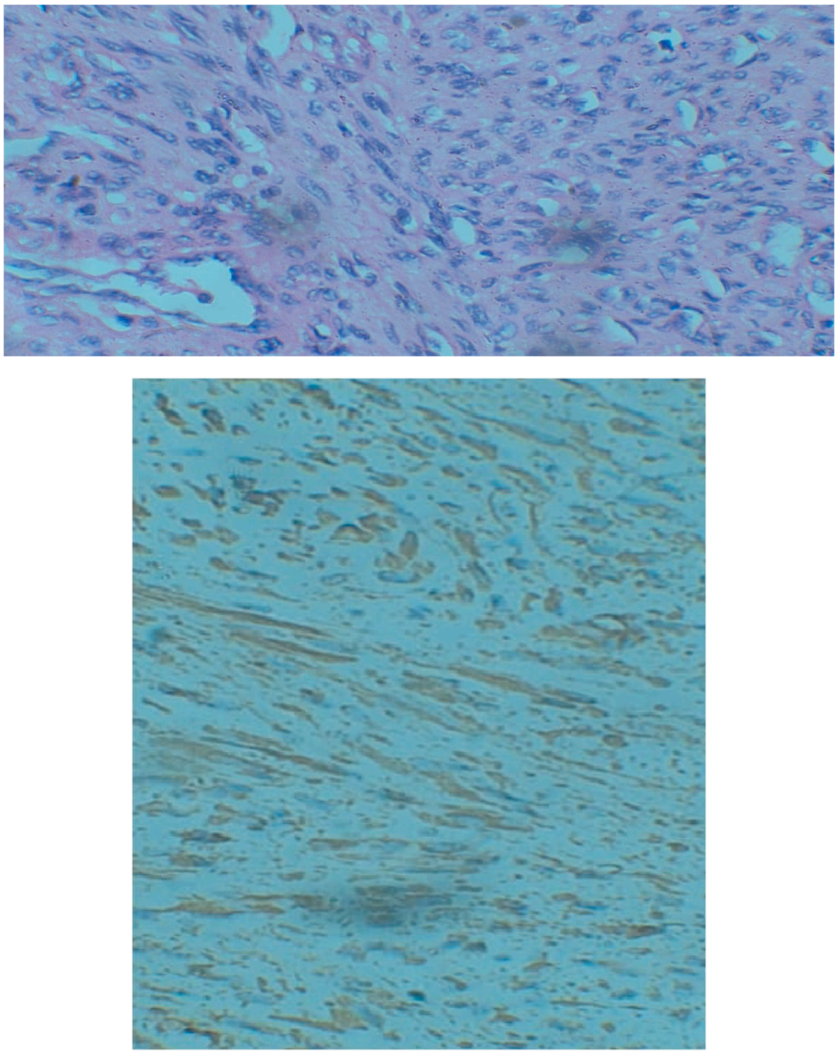

Fig. 6. a: Histological section – Immunohistochemistry staining: SMA positive ((low magnification), b: Histological section – Immunohistochemistry staining: SMA positive (high magnification).

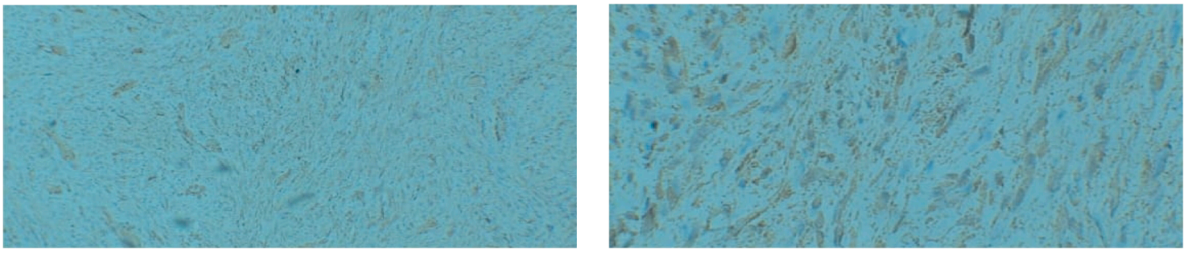

Fig. 7. a: Histological section - Immunohistochemistry staining: Vimentin positive (low magnification), b: Histological section - Immunohistochemistry staining: Vimentin positive (high magnification).

prevent recurrence which is unlikely.

Conflicts of interest

All the authors declare that there is no conflict of interest regarding the publication of this paper.

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Informed consent

Informed consent from the patient has been obtained.

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