Recurrent oral angioleiomyoma

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Abstract

Angioleiomyomas are vascular variant of leiomyomas which are benign tumors of smooth muscle. They are exceedingly rare in the oral cavity. Malignant transformation of these tumors has also been reported occasionally which warrants knowledge of this soft tissue tumor. A 57 year old male patient reported with a 15 day history of an asymptomatic growth that had started insidiously in his lower left back tooth region. Clinical examination revealed a solitary, oval, sessile growth in the mandibular left retro molar region. Excisional biopsy was suggestive of Angioleiomyoma. A recurrence of the same was noted two months later which was also histopathologically reported as Angioleiomyoma. The same was confirmed using special stains. This case reports an unusual presentation of Angioleiomyoma with regards to both recurrence as well as rapid growth. It is important to be well aware of this uncommon entity as these tumors often can mimic or transform into malignancy. Precise clinicopathological examinations are therefore invaluable in establishing an accurate diagnosis and delivering suitable treatment.

Keywords: Angioleiomyoma, leiomyoma, smooth muscle tumor

Introduction

Angioleiomyomas are benign tumors of the vascular smooth muscle origin. Also termed as vascular leiomyoma and angiomyoma, they are rarely found in the oral cavity and when found, most commonly occur on the lower lip and the palate. Clinically they exhibit slow growth and seldom recur. The case being reported is atypical as it showed rapid growth and recurrence following excision.

Case Report

A 57-year-old man presented with a chief complaint of a growth in his lower left back tooth region of 15 days duration. The growth had started insidiously and gradually increased to reach the present size. It was asymptomatic, and without any discharge and was not associated with loss of weight or appetite.

On examination, a solitary, oval, sessile growth measuring 3 cm anteroposteriorly 1.5 cm mediolaterally and 1 cm superoinferiorly with distinct borders was evident in the mandibular left retro molar region distal to tooth 37. The growth showed superficial ulcerations with indentations of opposing teeth. Tooth 38 was missing. There was no tenderness or discharge on palpation and it was firm in consistency [Figure 1]. No cervicofacial lymph nodes were palpable. Hematological investigations revealed normal values with nonreactivity for HIV. Also, no radiographic changes were evident in the area of interest.

An excisional biopsy was performed of the lesion along with extraction of teeth 37, 36. The H and E stained sections showed numerous dilated vascular spaces with a single layer of endothelial lining. Smooth muscles cells were found to be arranged concentrically around the blood vessels. The smooth muscle cells had well defined, “cigar shaped” nuclei. The lesion was unencapsulated. Various amount of collagen was found to be interspersed between vascular spaces [Figures 2 and 3]. Special staining was performed using Masson's trichrome which stained the smooth muscle cells red and the collagen fibers blue [Figure 4].

A final diagnosis of angioleiomyoma of the left retromolar area was thus considered.

Two months following the excision of the tumor, the patient reported with similar growth in the same location that had started 10 days prior to his reporting and had rapidly increased in size. The clinical presentation was very much similar to the previous episode [Figure 5]. Excision of the lesion along with a 2 cm margin was performed and histopathology confirmed the lesion to be an angioleiomyoma. There has been no further recurrence, 32 months following treatment.

Discussion

Leiomyomas are benign tumors of the smooth muscle origin. They are found in areas abundant in smooth muscles...
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**Definition**

WHO defines angioleiomyoma as “a frequently painful, benign subcutaneous or deep dermal tumor composed of mature smooth muscle bundles which are surrounded and interlaced by vascular channels”.[4]

Angioleiomyomas are believed to be originating from the tunica media of blood vessels, mainly the veins.[4] Oral angioleiomyomas originate from the vascular smooth muscles, excretory duct of salivary glands[5] and also from the circumvallate papillae.[3,6]

**Etiology**

The exact etiology of angioleiomyomas is unknown. However, local infection, trauma and artero-venous malformation and hormonal influences have been proposed as possible factors.[7]

**Epidemiology**

Oral angioleiomyomas occur in the age range of 3.5–85 years with a mean of 50–55 years.[2,7] The gender predilection has been controversial as a few report it to be common in males with a male-to-female ratio of 1.43:1[2,8] whereas others have observed a female predilection with the ratio being 2:1.[3] Angioleiomyomas are common among Caucasians such as GI tract, uterus, and skin.[1] Oral leiomyomas are rare tumors. The overall prevalence ranges from 0.016% to 0.06%[2] Leiomyomas are classified into three groups based on histopathologic features as (a) solid, (b) epitheloid, and (c) angioleiomyoma.[2,3] Of the three types, angioleiomyomas are the most common.[3]
(66%) and in African-Americans (16.1%). Within the oral cavity, angioleiomyomas are virtually found everywhere, with the common locations being lower lip and palate and also centrally within the mandible. A review of oral angioleiomyomas revealed only about 20 cases involving the gingiva. The present case was seen in a 57-year-old man, in the mandibular left retro molar area.

**Clinical features**

Clinically they appear as sessile growths ranging from 2 mm to 2 cm in size. They are firm in consistency and color varies from red to pink to gray. Although the lesions have a vascular origin, only about half of them exhibit red, purple, or blue color. They grow slowly and are mostly symptom free. When present, pain is the common symptom, followed by difficulty in swallowing and chewing food. The cause of pain in angioleiomyomas has been predicted to be either due to local ischemia or neural irritation in the tumor. The surface of the lesions is usually smooth and rarely can get ulcerated as a result of trauma from occlusive forces. The present case is atypical as it exhibited rapid growth during recurrence contrary to the previous reports.

**Clinical differential diagnosis**

The clinical appearance of this entity may mimic many lesions such as pyogenic granuloma, benign, and malignant salivary gland tumors namely pleomorphic and monomorphic adenomas, mucoepidermoid carcinoma and adenoid cystic carcinoma, mesenchymal tumors such as fibrous histiocytoma and schwannoma as well as non-Hodgkin's lymphoma.

**Histopathology**

Angioleiomyomas contain dilated vascular spaces with a single layer of endothelial lining. Smooth muscles are found to be arranged concentrically around the blood vessels. The smooth muscle cells have a well defined, “cigar shaped” nuclei. The lesions may be encapsulated or unencapsulated. Various amounts of collagen fibers can be found interspersed between vascular spaces. Based on the type of blood vessel involved, angioleiomyomas have been further classified as (a) cavernous, (b) solid, and (c) venous types. The venous type is the most common type. The case being reported showed consistently all the features of a typical venous angioleiomyoma.

Few authors have pointed out that angioleiomyomas represent only a stage in the formation of solid leiomyomas. They all start as hemangiomas, proceed to form vascular leiomyomas and then end up becoming solid leiomyomas. Others have proposed that angioleiomyomas and solid leiomyomas are entirely different entities.

**Special stains and immunohistochemistry**

Special stains such as Masson’s trichrome, Van-Gieson, and Mallory’s phosphotungstic acid haematoxylin have been variably used to confirm this entity. In Masson’s trichrome, the cytoplasmatic elements of the muscle cells are stained red and fibroblasts and collagen fibers are stained blue. It is also suggested that in situations of histopathologic controversy, immunohistochemistry can be sought. Markers toward muscle actin, vimentin, desmin, and S-100 can be utilized. Angioleiomyomas are positive for all the above markers except for S-100. In the present case, Masson’s trichrome was used that showed positive staining in the muscle cells.

**Histopathologic differential diagnosis**

Since angioleiomyomas are spindle cell neoplasms, careful differentiation is necessary from other spindle cell neoplasms such as neurofibroma, neurilemmoma, fibrous histiocytoma as well as solitary myofibroma. Neurofibroma consistently shows spindle cells arranged in interlacing bundles with cells having wavy nuclei, neurilemmoma having cells arranged in Antoni A and Antoni B areas, fibrous histiocytoma having spindle cell proliferation with vesicular nuclei and the cells arranged in a storiform pattern and lastly solitary myofibroma shows numerous proliferating fibro and myofibroblasts having tapered nuclei.

**Management**

Angioleiomyomas are managed by surgical excision. Although the lesion has vascular components, it seldom bleeds massively.

**Recurrence**

Albeit rare, recurrence has been reported. Irrespective of the type, about 5% of leiomyomas show local recurrence. This has been attributed to incomplete excision or deeply situated lesions. The present case recurred 2 months following excision, probably due to incomplete excision. The patient is under follow-up, and no further recurrence has been noted 32 months after the last recurrence.

**Malignant transformation**

Angioleiomyomas are benign lesions but malignant transformation is a possibility. So far no case of malignant transformation has been reported in literature. Malignant counterpart of angioleiomyoma which is angioleiomyosarcoma exists, which histopathologically exhibits dysplastic features.

In conclusion, oral angioleiomyomas are rare. They are considered to be slowly growing and asymptomatic although occasionally they can display rapid growth and recurrence as in the case reported. Dentists and oral diagnosticians should be aware of this entity as the rapidly growing tumors can closely mimic malignancies clinically. Careful histopathologic evaluation is indispensable as they need to be differentiated from other spindle cell neoplasms as well as malignant smooth muscle tumors.
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