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Case Report

Angiomyoma – A requisite in differential diagnosis of palatal growths

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ABSTRACT

Vascular variant of leiomyomas termed angioleiomyomas rarely present in the oral cavity. They present themselves as well-circumscribed, slow-growing, superficial lesions covered by normal mucosa and sometimes bluish or purple in color. Clinicians often tend to overlook this entity due to the rarity and asymptomatic nature of the lesion. Scrupulous knowledge of this entity and a thorough examination is of value, as the lesion can impersonate or transform to malignancy, as a malignant counterpart exists. We present a case of angiomyoma of hard palate in a 46-year-old patient with a detailed review.

Keywords: Angioleiomyoma, angiomyoma, oral vascular leiomyoma

Introduction

Leiomyoma is defined as a benign neoplasm of smooth muscle, classified into solid leiomyomas, angioleiomyomas (vascular leiomyomas), epithelioid leiomyomas (leiomyoblastomas), and myxoid leiomyomas.[1-3] The most common variant to occur in the oral cavity is the vascular leiomyomas or angioliomyomas,[4,5] defined by the WHO as a frequently painful, benign subcutaneous, or deep dermal tumor composed of mature smooth muscle bundles which are surrounded and interlaced by vascular channels.[1,6]

Case Report

A patient aged 46 years reported to the Department of Oral Medicine and Radiology, College of Dental Sciences, Davangere, India, with the chief complaint of growth on the palate for 12 years. Pain was present for 10 days and difficulty in eating was reported by the patient. Patient's medical, dental, personal, and family history was non-contributory. He chewed quid with tobacco once per day for 4 years. On intraoral examination, a solitary, dome-shaped, well-defined, exophytic growth of 1.5 cm × 1.5 cm × 0.5 cm in diameter was present on the hard palate, extending 1.5 cm posterior to the rugae area and mesiodistally extending from mid-palatine raphae to 1.5 cm lateral to midline, roughly spheroidal in shape. Lateral 1/3 of swelling was bluish-pink in color with smooth contours and a glossy appearance, and the remaining surface was ulcerated having sloping edges, well-defined border, and floor covered with a yellowish necrotic slough, which was surrounded by an erythematous halo. The growth was tender on palpation, soft to firm in consistency, sessile, and non-indurated [Figure 1]. Therefore, a provisional diagnosis of infected adenoma was given, with a differential diagnosis of cavernous hemangioma, adenoid hyperplasia, inflammatory hyperplasia, low-grade mucoepidermoid carcinoma, neurofibroma, angiomyoma, and lipoma. Maxillary true occlusal radiographs showed no bony changes in the site of the lesion [Figure 2]. Blood investigations showed normal parameters except increased erythrocyte sedimentation rate. The lesion was excised with a scalpel, under local anesthesia with minimal bleeding. It did not affect the underlying bone. The gross specimen was gray-white in color and firm in consistency, unencapsulated. Histopathologically, parakeratinized stratified squamous surface epithelium was seen, with numerous thick-walled blood vessels in the connective tissue formed of hyperplastic smooth muscle fibers arranged concentrically around the lumen with spindled cells having ovoid to blunt-ended nucleus [Figure 3]. Myxoid and fatty changes were evident in the stroma and the immunohistochemical study showed that the tumor cells were positive for smooth muscle actin (SMA) [Figure 4]. Based on history, clinical features, and histopathology, a final diagnosis of angiomyoma of hard palate was given. The patient was also counseled regarding his habit and was under frequent follow-up and showed no recurrence in 6 months.

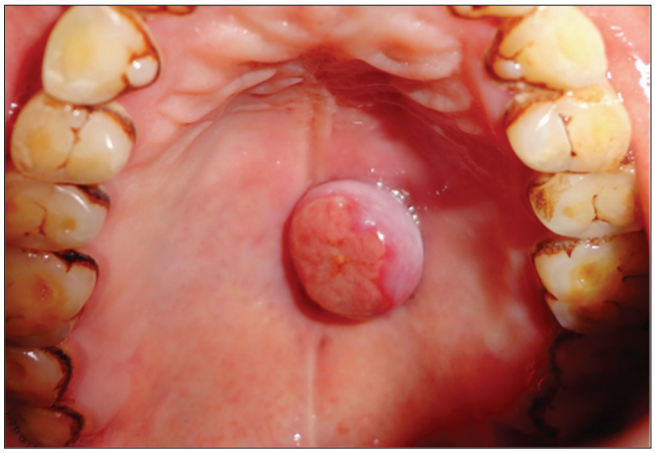

Figure 1: Intraoral presentation of the lesion on the palate showing well-defined, exophytic lesion, with ulceration on surface and sloping edges

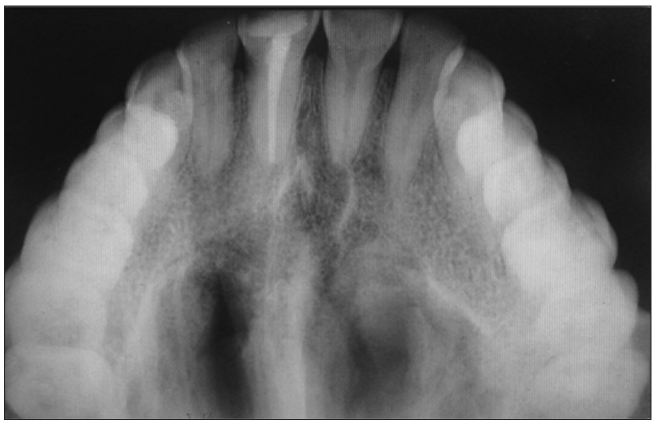

Figure 2: True occlusal radiograph showing no underlying bony changes

Discussion

The first case of oral leiomyoma was reported by Blanc in 1884 and since then, a number of additional cases have been documented.[1,3] This tumor is thought to originate from tunica media of blood vessels and heterotopic smooth muscle,[7] though some authors do suggest them to be arising from the remains of embryonic tissue such as the lingual duct or circumvallate papilla of the tongue.[5,7] However, the most accepted theory is that pericyte, a mesenchymal-like cell associated with the walls of small blood vessels is responsible for angiomyoma. These pericytes represent an intermediate phenotype between fibroblasts and vascular smooth muscle cells (VSMCs) and thereby can be considered as progenitors for VSMC in angiomyoma.[1,7] Various etiological factors such as infection, trauma, hormones, and arteriovenous malformations have been proposed to evoke the proliferation of pericytes.[7-9]

Since only a few leiomyoma of the head and neck have been reported in literature, the gender prevalence cannot be confirmed, but it may appear at any age with the greatest

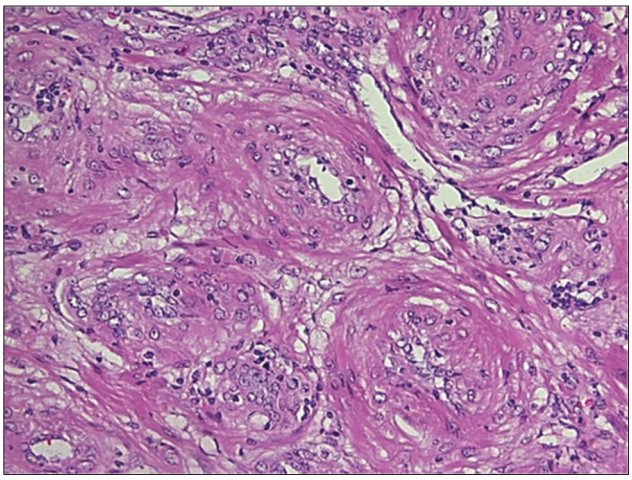

Figure 3: Pictomicrograph of the lesion showing numerous thick-walled blood vessels formed of hyperplastic smooth muscle fibers arranged concentrically around the lumen with spindled cells having ovoid to blunt-ended nucleus. Hematoxylin-eosin staining (×40)

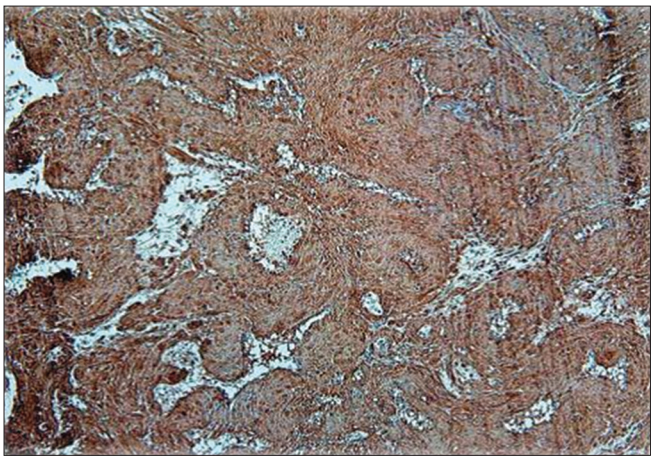

Figure 4: Smooth muscle actin immunostaining – positivity visible in the perivascular spindle cells [×20]

incidence in the 4th and 5th decades of life and is more frequently seen in men.[1,5,6,8] According to Brooks *et al*., the overall incidence rate of angiomyoma in the oral cavity is around 0.016%.[1,4,7]

Clinically, the lesion has a slow asymptomatic growth, although clinical symptoms such as pain in response to palpation, chewing and swallowing difficulties, and tooth mobility can be noted.[5] Clinical differential diagnosis has to include benign lesions of salivary gland origin such as deep-seated mucocele and pleomorphic adenoma, vascular lesions such as hemangioma, varicosis, and lymphangioma, benign mesenchymal tumors (fibroma, lipoma, neurofibroma, myofibroma, etc.), epidermoid cysts, and non-Hodgkin's lymphoma.[3,5-7,10] The final diagnosis is only possible through histopathological examination with special staining and immunohistochemistry. Various special stains such as Van Gieson, Mallory's, and Masson's trichome can be used to differentiate angiomyoma from other lesions. Immunohistochemical study on the tumor with S100, SMA,

and monoclonal antibodies against actin, vimentin, desmin, and cytokeratin will help achieve a definitive diagnosis. Vimentin is a structural protein of cytoplasmic elements of mesenchymal cells, whereas desmin is a type III intermediate filament near the Z line in sarcomere.[8,9] CD-34 is useful to define the degree of vascularization of the lesion.[2,4,7,8,10]

The treatment of choice is excision, including an adequate safety margin of normal-appearing tissue, to prevent relapse as documented by Brooks *et al*., wherein relapse was seen 2 weeks and 9 months after resecting two hard palate leiomyomas.[1,5,7,10] Furthermore, malignant changes of the lesion can happen,[3,5,7,8] and hence, the clinicians should be aware of pathological features and behavior of this lesion to ensure prompt diagnosis, appropriate management, and follow-up for these patients.

Patient's consent

Written informed consent has been taken from the studied subject.

Conflicts of interest

None.

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