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Case Report

Angiomyoma of the Hard Palate

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ABSTRACT

Leiomyoma is a rare benign neoplasm deriving from smooth muscle. Vascular leiomyoma is its most common subtype in the oral cavity. It may appear at any age with the greatest incidence in the 4th and 5th decade of life. The common manifestation is a slow-growing, asymptomatic, submucousal mass. The diagnosis is only through microscopic examination requiring special staining. Treatment of choice is surgical excision and no recurrence is usually seen. In this report, we present a case of angiomyoma in the midline of hard palate with description of its clinical, histological and immunohistochemical characteristics.

Keywords: Angiomyomas, Hard Palate, Leiomyoma

Introduction

Angiomyoma -also known as vascular leiomyoma- is a benign tumor and a variant of leiomyoma. This lesion is composed of multiple vessels surrounding with thickened smooth muscle layer (1).

The most frequent site of the appearance of angiomyoma is the skin of lower extremities and it is rarely found in the oral cavity (2,3). Lips, palate, buccal mucosa and tongue are the reported sites of its occurrence in the oral region (1,2). This tumor is usually a slow-growing painless submucosal mass (2-5) which can have several clinical diagnosis and just can be diagnosed histologically (4).

Diversity of the differential diagnosis list especially because of the color of the lesion, make it a distracting case among the oral lesions. As the treatment and prognosis of the other lesions of the differential diagnosis list can be widely different, it is very important to consider it as a probable diagnosis.

In this report, we present a case of angiomyoma in the midline of hard palate with description of its clinical, histological and

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immunohistochemical characteristics. We also show the result of its treatment and follow-up. It is among the first reports of oral angiomyomas in national literature.

Case report

A 75-year-old male who looked like fit and without any remarkable medical or dental problem referred to our clinic with a complaint of a mass in the midpalatal area. The patient was first noticed about the existence of the lesion 2 years ago and it was completely asymptomatic during this period. Based on his expression, there was not any pain, bleeding and also change of size.

In the clinical examination, a purple red dome shape mass without ulceration was found in the midline of hard palate. The size of the lesion was approximately 0.7×0.7 cm and its periphery was well-defined. It had a rubbery to firm consistency in palpation and moved easily without any attachment to the peripheral tissues. The aspiration was negative (Fig. 1). It had not caused any radiographic changes in conventional occlusal view and also computed tomography.

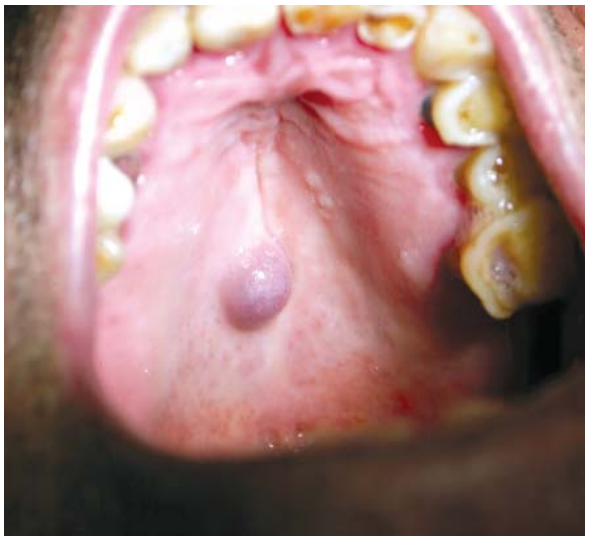

Fig. 1. Clinical appearance of the lesion in the midpalatal area before surgery

The lesion excised under local anesthesia and it easily got detached from surrounding connective tissue without considerable bleeding during surgery. It was completely above the periosteum and it had not any effect on underlying bone.

In gross examination, the size was 0.7×0.6×0.6 cm and it had gray to white color with firm consistency. Dissected surface was smooth and there was not any space on it.

Varix, benign mesenchymal tumors (probably with vascular origin), benign minor salivary gland tumor and mucocele were considered in differential diagnosis list. Based on its place, the probability of being a minor salivary gland tumor was low.

In histopathology, the lesion consisted of spindle cells arranged in small fascicular patterns and formed around a central lumen which lined with endothelial cell. Evidences of malignancy such as mitosis, necrosis, cellular atypia and pleomorphism were not seen. (Fig. 2). Masson Trichrome staining revealed the muscular nature of the lesion (Fig. 3).

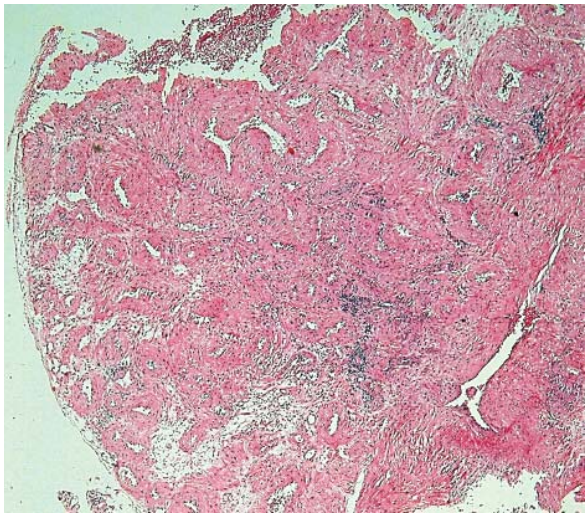

Fig. 2. Photomicrograph of the lesion, fascicular pattern of the spindle-shaped smooth muscle cells around blood vessels of varying sizes (Hematoxylin and eosin, original magnification ×40)

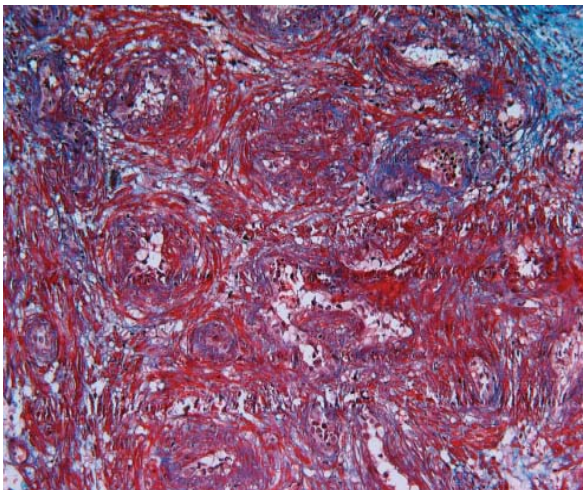

Fig. 3. Masson's trichrome staining; exhibiting collagen fibers in blue and smooth muscle cells in red, forming the main part of the lesion (Original magnification ×200)

Immunohistochemical stainings were also applied for SMA, CD31 and S100. Positivity was seen with SMA and the vascular compartment was positive with CD31 (Fig. 4 &5).

The final diagnosis was angioleiomyoma and no recurrence has been occurred after six months follow up.

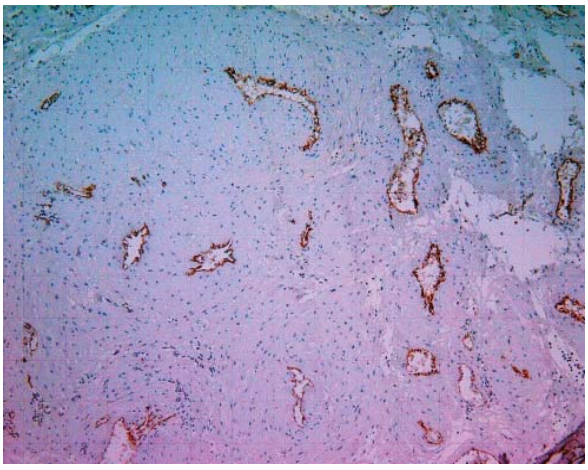

Fig. 4. CD31 immunostain. A positive reaction is seen in the endothelial cells of vessels (Original magnification ×100)

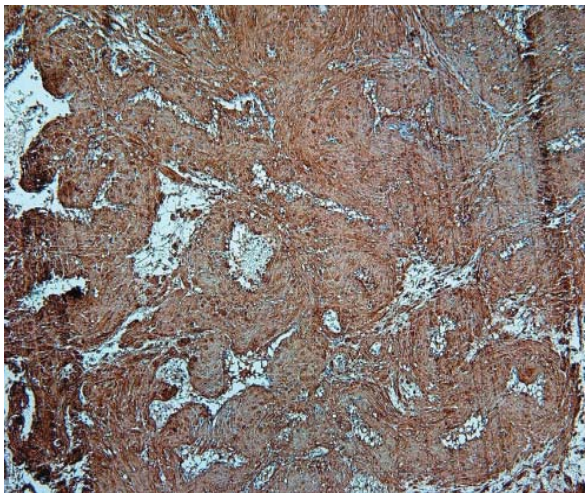

Fig. 5. Smooth Muscle Actin immunostain. Positivity is seen in perivascular spindle cells (Original magnification ×100)

Discussion

According to Brooks *et al.* the overall incidence rate of angiomyoma in the oral cavity is around 0.016% (2) and its incidence in the hard palate is much lower. Brooks *et al.* investigated 76412 biopsies registered in a Department of Oral Medicine in the period between 1963 and 2001 in a retrospective study. Only 12 cases of angiomyoma with 1 in hard palate identified (2). Farmen in 1975 found 5 oral angiomyomas out of a total 7748 smooth muscle tumors located throughout the body (0.065%) (4). In a survey done by Hachisuga *et al.* on 48 cases of angiomyoma in the head region, they found only 13 cases in the oral cavity with one on the hard Palate (1). Blanc presented the first case of oral leiomyoma in 1884(4).

There are some theories discussing about the origin of this lesion. It has been a theory that says the lesion may have some originations like tunica media of blood vessels, circumvalate papillae and heterotopic smooth muscle (4-6).

There is also another opinion that considers vascular leiomyoma as a stage in a process

of smooth muscle proliferation. Based on this idea, vascular leiomyoma occur in a part of the spectrum of (hemangioma→angioma →vascular leiomyoma→leiomyoma solid →leiomyoma) (4, 6).

It is also proposed that angiomyoma can appear as a result of infection, trauma, hormones and arteriovenous malformation (3, 6). A pericyte is a mesenchymal-like cell, associated with the walls of small blood vessels. This cell plays a critical role in regulating endothelial proliferation, differentiation, migration and survival. It involves in the earliest stages of capillary sprouting (7). On the other hand, pericytes can differentiate into other mesenchymal cell types such as smooth muscle cell, fibroblast and osteoblast (8). Similarity of vascular Smooth Muscle Cell (vSMC) and pericytes and their common markers like α-smooth muscle actin (SMA) and desmin show that vSMC and pericytes are phenotypic variants of a continuous population of mural cells. Pericytes have an intermediate phenotype between vSMC and fibroblasts. Irritants can make pericytes to give rise to vSMC. Thus, pericytes can be considered as progenitors for vSMC in angiomyoma (8).

Some cases of angiomyoma may appear as a result of chromosome abnormalities. Genome mapping and DNA screening has revealed this alteration in a few cases, especially those in lower extremities and uterus (3).

Angiomyoma has been reported in a range from 3.5- to 85- year-old with the highest incidence in 4th and 5th decade of life (2,4-6). In oral cases male predilection has been reported unlike the rest of the body (1, 2). Some authors believe about female predilection (2, 5) or no gender difference (4, 6).

Most of the reported cases are between 30 and 50 years old, except those occurring in the hard palate. Hard palate angiomyomas are seen in older ages than other regions. Unlike our case, these angiomyomas show a female predominance (9). Usual sites of its occurrence in the oral region are lips, hard and soft palate, buccal mucosa and tongue (1, 2).

It usually appears as a submucosal mass growing slowly without any symptom (1-5). In some cases, symptoms such as pain (4) (more frequent in solid leiomyoma) (1) and difficulties in chewing or swallowing has been reported (2, 4). Our case did not cause any symptom such as most angiomyomas.

Reported lesions consistency show different degrees of firmness and resiliency. The present case had a rubbery to firm consistency in palpation with an easy movement due to its little attachment to the peripheral tissues seen during excision; such as some reported cases. The mass color is mainly based on its depth and vascularity (2). Our case wasn't deep-seated so it appeared as purple. The lesion rarely becomes more than 2 cm in size (9) which is seen in most reports and also this case.

The diagnosis of angiomyoma is only rested on its histological features; because it does not present any special clinical characteristic (4). Clinical differential diagnosis has to include: red lesions such as hemangioma and varicosis, lymphangioma, benign mesenchymal tumors (fibroma, lipoma etc) (1-4). Because of the location of the present case – midline of palate – the probability of salivary gland origin was low.

In a histological point of view, differential diagnosis includes: neurofibroma, schwannoma, fibromatosis, fasciitis, fibrous histiocytoma, solitary myofibroma, spindle cell lipoma , peripheral nerve sheath tumor, hemangiopericytoma and leiomyosarcoma (1,2).

In its histological appearance, the lesion con-

sists of spindle-shaped smooth muscle cells with unclear cytoplasm borders and relatively hyperchromatic nucleus. These spindle cells arranged in small swirling patterns which placed packed and next to each other with just a little connective tissue stroma. A vascular component is also seated in the central part of the above structures (2-4).

For accurate histological diagnosis and to differentiate an angiomyoma from other mesenchymal tumors, special staining like Masson trichrome and immunostaining is necessary (2). Masson trichrome staining distinguishes smooth muscle and collagen by showing smooth muscle cells as red and collagen fibers as blue (3,4). As our case stained with this pattern, we could recognize the smooth muscle as the main part of the lesion. Monoclonal antibodies against Smooth Muscle Actin (SMA) also approved the result.

Positivity with CD31 staining revealed the presence of endothelial cells in the center of muscular fascicles which leads to the definitive diagnosis of angiomyoma.

The treatment of choice for angiomyoma is surgical excision and recurrence is very rare (1-6). Mostly, angiomyoma does not have the tendency to recur by nature; but recurrence was seen in 2 reported cases, both occurring in hard palate (9). Our case did not show any recurrence during 6 months follow up.

In conclusion, oral angiomyomas though rare, can be misdiagnosed as other lesions of its differential diagnosis list and should be considered as a diagnosis just with a very low probability.

Acknowledgements

The authors declare that there is no conflict of interests.

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