Case Report

Leiomyoma of Hard palate — case report and review

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We report a rare case of Leiomyoma of The hard palate : Case Report and Review. A 42 year old male presented with a small 1 cm swelling over hard palate which was painless slow growing, non tender, smooth surface, overlying mucosa was reddish. Total excision was done with safe margins. The histopathological examination shown leiomyoma. This is rare tumour of hard palate which arise from unstriated muscle presented with slow growing swelling over hard palate. Treatment is total excision with safe margins followed by regular follow up.

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***Key* words : Leiomyoma, hard palate, smooth muscle.**

Leiomyoma is a benign smooth muscle tumor that may appear in any location, Though it is more common in the uterus, gastrointestinal tract and skin. It is rarely found in the oral cavity (0.065%), due to the scarcity of smooth muscle in this territory.

Oral leiomyoma is usually seen in adults and shows no gender predilection. The most frequent locations are the lips, tongue, hard and soft palate, and the cheeks. The tumor generally manifests as a slow-growing painless lesion, often of a purplish color.

The diagnosis is exclusively based on the histological findings. Clinically, a differential diagnosis must be established with lesions of the oral mucosa or connective tissue, such as fibromas, lipomas, salivary gland neoplasms, vascular tumors such as lymphangioma or haemangioma, etc. The differential diagnosis moreover must also include the malignant form of leiomyoma, ie, leiomyosarcoma. Treatment consists of complete resection, with dew safety margins and periodic controls to ensure early identification of possible tumor relapse. The present article describes a new case of oral leiomyoma, located in the palatal region, and evaluates the clinical and histological characteristics of the lesion, with a view to including the latter in the routine differential diagnosis of oral mucosal lesion.

Case Report

A 42-year-old male presented with a history of painless small swelling over hard palate on left side (Fig 1). The physical examination revealed a lesion measuring about 1 cm in diameter, located adjacent to the palatal surface of the upper premolars in the second quadrant. The lesion was a pale color with redness of adjacent mucosa, it was painless and non-hemorrhagic in response to palpation.

Treatment done with total excision of the lesion with a safe margin under local anesthesia and was sent for histopathology. The surgical wound was allowed to heal by second intention.

The histopathological study diagnosed oral leiomyoma (angioleiomyoma type). Hematoxylin-eosin staining (Fig 2, H&E x 100) revealed the presence of intermingling smooth muscle bands separated by cellular fibrous connective tissue.

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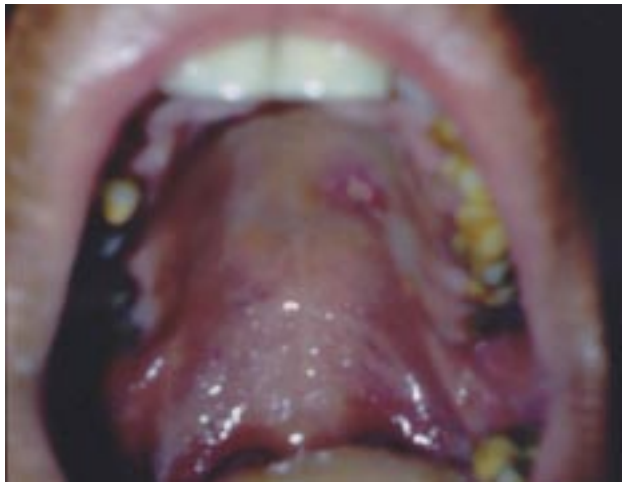

Fig 1 — Showing swelling over hrad palate

Discussion

Leiomyoma is infrequent in the oral cavity, due to the scarcity of smooth muscle in this territory. Stout suggested the smooth muscle of the tunica media of the arteries to be the probable origin of oral leiomyomas. Other authors consider leiomyomas to derive from the remains of embryonic tissue such as the lingual duct or circumvallate papilla of the tongue (Praal *et al*).

Leiomyoma of palate is rare tumour as reported. In the review published by Farman in 1975, involving 7748 smooth muscle tumors located throughout the body, only 5 cases (0.065%) corresponded to the oral cavity. The most common location was the female genitourinary tract. Hachisuga *et al* recorded 15 cases (2.7%) of oral angioleiomyomas in a series of 562 angioleiomyomas registered in a General Pathology Department. Brooks *et al* published a retrospective study of 12 angioleiomyomas, one solid leiomyoma and one leiomyosarcoma, out of a total of 76,412 biopsies/oral lesions registered in a Department of Oral Medicine in the period between 1963 and 2001. The incidence of angioleiomyoma was 0.016%, and represented 92.3% of all benign smooth muscle tumors located in the oral cavity.

Leiomyoma is usually seen in adults with the greatest incidence

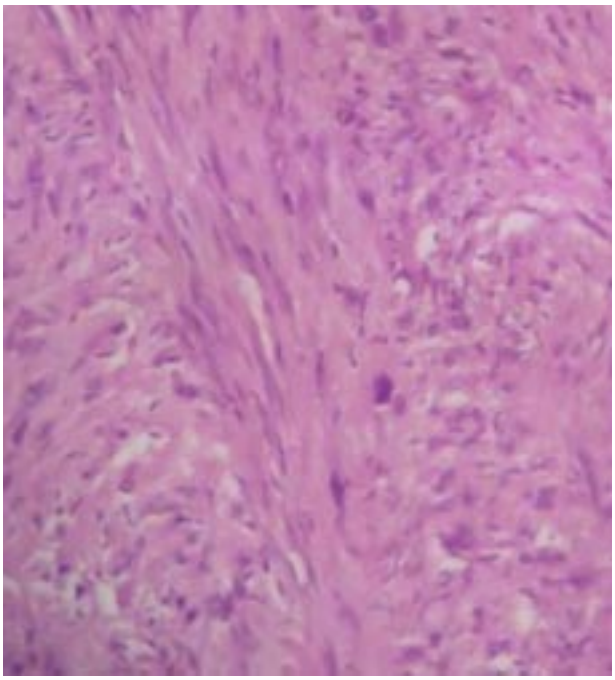

Fig 2 — Hematoxylin-eosin staining revealed the presence of intermingling smooth muscle bands separated by cellular fibrous connective tissue

corresponding to the 40-59 years age interval. Regarding gender distribution, some authors consider both males and females to be affected in equal proportion, though considerable controversy exists on this point. The present case was 45 years male, coincides with the typical presentations reported in the literature in relation to age distribution of leiomyoma.

The most frequent locations of oral leiomyoma are the lips, tongue, hard and soft palate, and the cheeks. Leung KW, *et al* reported a case of oral leiomyoma of buccal sulcus. In the series described by Svane *et al*, 21% of the leiomyomas were located in the palate. Out of these, angioleiomyoma represents 74%, solid leiomyoma 25%, and only one case of leiomyoblastoma has been documented. Brooks et al. recorded two angioleiomyomas at palatal level (16.7%) one in the hard palate and the other in the soft palate. The World Health Organization classifies leiomyomas into three histological types: leiomyoma (solid), angiomyoma (vascular leiomyoma) and epithelial leiomyoma (leiomyoblastoma).

According to Natiella *et al* the most frequent histological presentation of oral leiomyoma is angioleiomyoma (derived from smooth muscle of blood vessels). According to other authors, angioleiomyomas represent 64-66.2% of all types of oral leiomyomas. This is probably because the main source of smooth muscle tissue in the oral cavity is tunica media of the arteries. Duhig and Ayer suggested that vascular leiomyoma represents only a stage within a continuous process of smooth muscle maturation. The maturation sequence would be as follows: hemangioma, angioma, vascular leiomyoma, leiomyoma and solid leiomyoma. According to Damm and Neville, solid leiomyoma is histologically very different from angioleiomyoma, and the two entities therefore should be regarded as separate tumors. Leiomyoma tends to manifest as a smooth-surfaced submucosal nodule.

The overlying epithelium rarely ulcerates, though in some cases there is histological evidence of ulceration. The color of the lesions depends on their vascularization and depth. However, although their origin is related to the blood vessels, only 55.9% are red, blue or purple in color. The rest show the appearance of normal mucosa, or have a grayish tone. At palpation, the tumors appear firm and are generally well delimited, with free displacement within the lax tissues of the lip and oral mucosa. The lesions tend to grow slowly, with a size ranging from a few millimeters to 3 cm. In the cases described by Brooks *et al*, all the lesions were between 2-10 mm in size. Hemani DD *et al* presented a large leiomyoma of palate. The tumor in our patient was 1 cm in diameter, and presented a pink-purple color, as commented above. Most oral leiomyomas present as asymptomatic lesions, same in present case, though different authors have described clinical symptoms like pain on palpation, chewing and swallowing difficulties, and abnormal tooth mobility. The diagnosis of leiomyoma is relatively difficult to establish, due to the similarity with other fusiform cell tumors. The differential diagnosis must include other mesenchymal tumors (fibroma, neurofibroma, lipoma, etc), salivary gland neoplasms (mucocele, pleomorphic adenoma, etc), vascular tumors (lymphangioma, hemangioma, pyogenic granuloma, etc), and soft tissue cysts such as dermoid cysts. When located in the region of the hard palate, adjacent to teeth the tumor can be confused with a periodontal lesion.

The definitive diagnosis of leiomyoma is therefore based on the histological study of the lesion. Leiomyomas are composed of fusiform smooth muscle cells with elongated nuclei, similar to fibroblasts. The cells are distributed in parallel bundles, and the lesions are encapsulated or well delimited within the surrounding tissue. No fibrous stroma is noted only small capillaries among the tumor cells. In order to differentiate leiomyoma from the rest of fusiform cell tumors, specific stains are used to identify collagen and muscle cells, such as the Van Gieson, Masson trichromic and Mallory phosphotungstic acid-hematoxylin (PTAH) stains. Van Gieson staining is recommended for muscle. The Masson trichromic stain differentiates the cytoplasmic elements of the smooth muscle cells, which stain red, from collagen and fibroblasts, which stain blue or green. However, both the Van Gieson and Masson trichromic stains can give rise to false positive results for muscle and collagen fibers; it is therefore advisable to confirm the presence of myofibrils by using the Mallory PTAH stain. Immunohistochemical techniques can also be used. In this context, specific monoclonal antibodies for actin (a smooth muscle marker) are useful for confirming the diagnosis of leiomyoma (Gonzalez sanchez MA *et al*). Six cases diagnosed as leiomyoma were retrieved from the files of two oral biopsy were done with suitable controls. The haematoxylin and eosin and Masson's trichrome stains supported a diagnosis of leiomyoma in all 6 cases but PTAH was positive in only 3 of them. The immunohistochemical study confirmed the diagnosis of leiomyoma in 3 cases. The other 3 were identified as granular cell tumour, myofibroma and neurofibroma, respectively. Immunohistochemistry is a precise and reliable method for definitive diagnosis of oral leiomyoma (Baden E *et al*).

Leiomyoma must be carefully differentiated from leiomyosarcoma, particularly low-grade leiomyosarcoma. To this effect, a determinant factor is the presence of mitotic figures. In the presence of over 10 mitoses per high-magnification field (x40), the lesion is considered to have a malignant behavior, while fewer than two mitotic figures per 10 high-magnification fields is indicative of a good

prognosis. The presence of ulceration is also considered to be indicative of malignancy. Immunohistochemical techniques and molecular markers such as PCNA, bcl-2, CDK4, p53 and MDM2 are correlated to malignant lesions; the diagnostic procedure for differentiating muscle tumors is therefore based on these methods.

The treatment of choice is local resection, including an adequate safety margin of normal-appearing tissue. Despite the vascular origin of these lesions, important bleeding after excision is rare. Likewise, these benign smooth muscle tumors rarely relapse. Nevertheless, Brooks et al. documented relapse two weeks and 9 months after resecting two hard palate leiomyomas.

Conclusion

The leiomyoma is a rare benign tumor of the oral cavity, and with a good prognosis, though it must be included in the differential diagnosis of oral mucosal lesions. The treatment of choice is surgical resection with adequate safety margins in all cases, due to the high incidence of malignancy of this tumor within the oral cavity, when compared with the rest of anatomical locations. The possibility of relapse moreover requires periodic patient controls after resection. Histopathological examination should also include the immunohistochemical study.

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