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CASE REPORT

Oral leiomyoma: A case report on a rare lesion in oral cavity

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Abstract:

Introduction: Leiomyoma, a benign tumour of smooth muscles, often affects uterine myometrium, GIT, skin and lower extremities of women of middle age group. Its occurrence in oral cavity is quite rare with reported incidence of 0.065%. It is contemplated that in oral cavity leiomyoma arise from smooth muscle wall of blood vessels chiefly affecting lips, tongue, palate, buccal mucosa, and rarely gingiva and mandible. Clinically the disease exhibits as a slow growing asymptomatic mass, occasionally associated with pain. Histopathological study is mandatory to help determine the diagnosis due to deceptive clinical appearance. Surgical excision is widely accepted as the best treatment modality with rare recurrence. **Objective:** The rationale of this case report is to help improve our understanding of this rare disease (oral leiomyoma) and to report its occurrence in the literature. **Case Report:** We report a rare case of a 25 years old female patient presenting with the chief complaint of a slow growing mass since 6 months, diagnosed as leiomyoma after clinical and histopathological examination. **Conclusion:** Leiomyoma is a benign tumour of smooth muscles with rare presentation in the oral cavity with good prognosis. Due to deceptive clinical and histopathological characteristics it is important to meticulously evaluate clinically and histopathologically to arrive at conclusive diagnosis.

Keywords: Leiomyoma; Mouth Mucosa; Smooth Muscle Tumor.

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INTRODUCTION

The World Health Organization (WHO) in 2002 defined "Leiomyoma as a circumscribed benign, often cutaneous tumour composed of intersecting bundles of mature smooth muscle cells." Leiomyoma was first described by Virchow (1854) and first reported by Blanc (1884). Kloeffer et al. (1958) first illustrated a hereditary form which causes multiple leiomyoma[1]. Leiomyoma, predominantly affects the female genital tract (95%), followed by skin (3%), GIT (1.5%) and infrequently the head and neck (less than 1%)[2-4].

Its occurrence in the oral cavity is rare, with an incidence of 0.065%, due to lack of smooth muscles and accounts for only 0.42% of soft tissue tumour[5].

Oral leiomyoma may present at any age, but its peak prevalence is in the 4th-5th decade of life, with a marginal female predilection[5]. Most frequent sites in the oral cavity include lips, tongue, hard and soft palate, and less commonly the cheeks[6,7]. Although it occasionally may be painful, the lesion manifest as slow growing, asymptomatic submucosal nodule. Surgical excision of leiomyoma has proved to be the most reliable and effective treatment till date with least recurrences being reported.

The occurrence of oral leiomyoma remains extremely low and hence it is critical to report new cases to enhance our understanding of the clinical and histopathological characteristics of the disease, the present case report on oral leiomyoma is our one such attempt.

CASE REPORT

A 25 years old female patient reported to our department with the chief complaint of a growing mass inside the right side of the cheek since 6 months. On history taking, it was found that the patient had been apparently asymptomatic before 6 months, when she noticed a nodular painless growth in right side of the buccal mucosa which gradually increased in size. Furthermore, the patient revealed a habit of cheek biting since childhood. Her past medical and dental history was non-contributory.

Clinical examination revealed a sessile nodular mass on the right buccal mucosa 0.5cm above the occlusal plane, measuring around 1x1cm in size. The overlying mucosa presents with ulceration, well defined margins with irregular borders. The surrounding mucosa inferior to the growth appears to impart greyish hue and superior to growth appears erythematous (Figure 1). Hard tissue examination revealed sharp cuspal edges of right maxillary and mandibular second molar. The lesion was firm; non tender; non compressible and non fluctuant on palpation. A solitary, tender and mobile lymph node was palpated in right submandibular region measuring around 0.5cm in size.

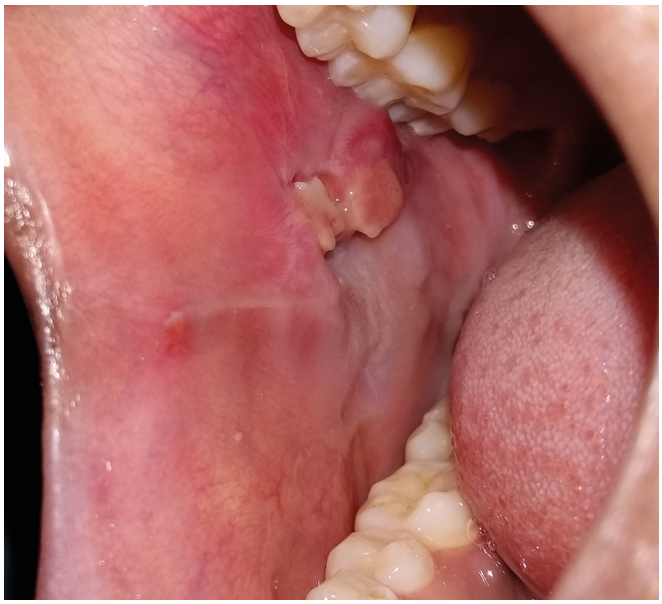

Figure 1. Presence of nodular growth in the right buccal mucosa.

After clinical examination a provisional diagnosis of fibrous hyperplasia secondary to chronic cheek biting was made, and the following investigations were advised. A complete blood heamogram, which revealed significantly elevated ESR levels (68mm/1st hour, westergren method) and ultrasonography of right side of cheek with Doppler study (Figure 2), which showed elongated hypoechoic soft tissue lesion in the submucosal layer measuring about 2cm in length and 7mm in thickness, with no abnormal vascularity, suggestive of fibrous hyperplasia.

An incisional biopsy was performed and the specimen was sent for histopathological examination, which suggested the presence of short intersecting fascicles of numerous spindle shaped cells with blunt ended or cigar shaped nucleus and endothelial cells in a sparse stroma without any atypia in deep submucosal area (Figure 3). Furthermore, Masson's trichrome special stain was performed which demonstrated intercellular collagen fibres between the smooth muscle cells (Figure 4).

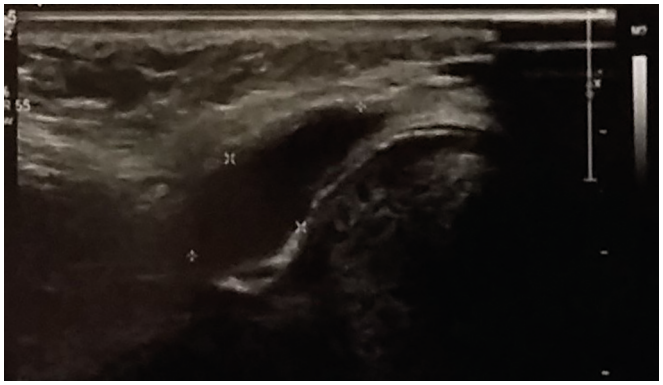

Figure 2. HRUSG of right cheek, showing elongated hypoechoic soft tissue lesion in the submucosal layer of right cheek.

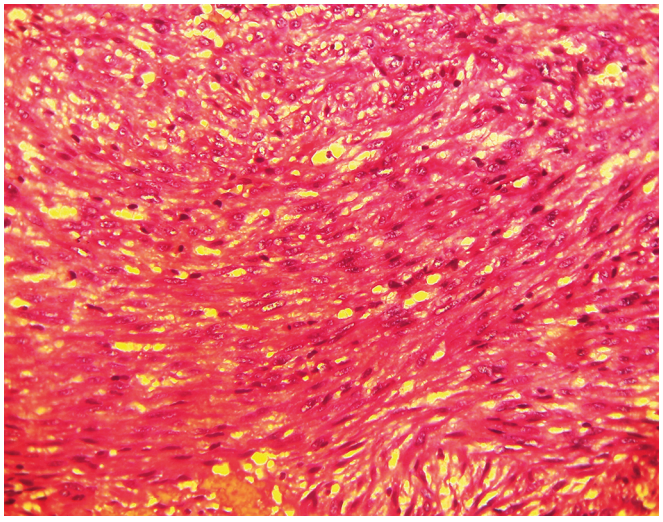

Figure 3. Photomicrograph showing intersecting fascicles of spindle shaped cells exhibiting blunt ended or cigar shaped nuclei (Hematoxylin and Eosin stain, 40x).

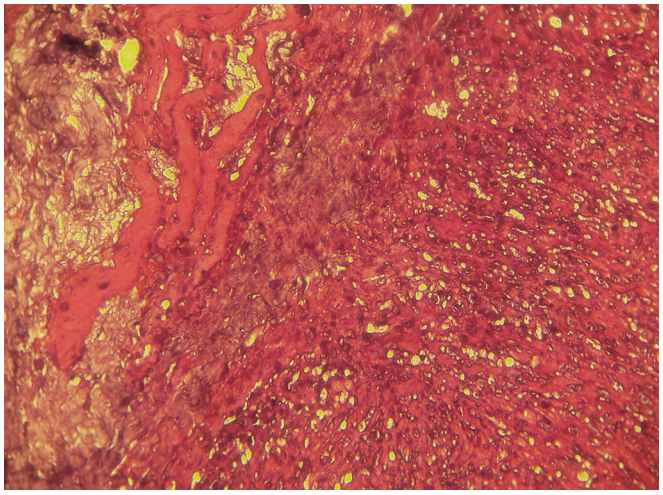

Figure 4. Photomicrograph showing smooth muscle cells (pink) in longitudinal and transverse sections with intervening collagen fibers (blue) (Masson's trichrome stain, 40x).

On the basis of histopathological examination a final diagnosis of leiomyoma was established. Further the case was planned for complete excision, and grinding of 17 and 47 performed prior to surgery. Under general anesthesia a wide excision was carried out followed by reconstruction with buccal pad of fat and simultaneous extraction of 18 and 48. The patient has been kept under follow up for periodic evaluation.

DISCUSSION

Leiomyoma, a benign, soft tissue tumour affects the smooth muscles, and is frequently seen in uterine myometrium (95%), skin (3%), GIT (1.5%), lower extremities and head and neck (1%). The occurrence of smooth muscle tumours is rare in oral cavity due to the lack of smooth muscles[8]. Scout (1938) proposed the source of smooth muscle in the oral cavity to be the tunica media of the blood vessel wall. Whereas the other sources suggested for smooth muscle tumour in oral cavity are ductus lingualis (Glass), circumvallate papillae of the tongue, and smooth muscles of excretory duct of salivary gland and heterotropic embryonal muscle tissue[8]. Since Glass reported the second case of leiomyoma, only 139 cases have been described in the literature[8].

Although oral leiomyoma may occur at any age, its peak prevalence is seen in 40-49 years of age, with slight female predilection[9]. Hormonal variations may be attributed to higher incidence of leiomyoma (F:M, 3.75:1) in females. Recently, sex steroid receptors have been found in leiomyomas, which implies that the growth of these tumours could be hormone dependent[10].

WHO[11] differentiated leiomyomas into three types: solid leiomyoma, angiomyoma (vascular leiomyoma), and epitheloid leiomyoma (leioblastoma). The most common type being angiomyoma (74%), followed by solid leiomyomas (25%) and only a single case of an epitheloid leiomyoma documented in the literature[12,13]. The histopathological report in our case confirmed the lesion to be a solid variant of leiomyoma. Most leiomyomas in the head and neck region are asymptomatic, although few may be sometimes associated with pain[5].

The solid variant manifest as slow growing, small (<2cm), asymptomatic, submucosal nodule with similar colour as that of adjacent mucosa or may sometimes show a greyish tone. The surface of the lesion is usually smooth and seldom ulcerates. The vascular variant presents as blue or red discolouration, with

size measuring from few millimetres to 3cm, frequently tender on palpation. In general it is well delimited with free displacement within the tissues of the oral mucosa.

Clinically the differential diagnosis pertinent to intraoral leiomyoma include fibroma, myofibroma, neurofibroma, schwannoma, nodular fasciitis and lipoma. However, the vascular variant of leiomyoma should be differentiated from vascular lesions such as hemangioma, pyogenic granuloma, lymphangioma. Hence, these entities should be excluded before arriving at the final diagnosis. Histopathologically, one should consider the following differential diagnosis: benign spindle cell tumours like schwannoma, neurofibroma, fibrous histiocytoma, nodular fascitis and malignant tumours like leiomyosarcoma[5].

Differentiation of leiomyoma from other mesenchymal tumours is difficult due to their similar clinical appearance[12,14], consequently the final diagnosis of oral leiomyoma is primarily established histopathologically via hematoxylin and eosin stains. Muscle fiber stains like Masson´s trichrome, help in displaying intercellular collagen fibres between smooth muscles and myofilaments inside[8].

We made use of hematoxylin-eosin and Masson´s trichrome stains to reach to the conclusive diagnosis. However, in cases when H&E staining does not yield decisive information, immunohistochemical studies can be performed to arrive at final diagnosis. Various smooth muscle markers like desmin, MSA and SMA can be positive for smooth muscle tumours while other markers such as; S-100 protein, CD34, CK and epithelial membrane antigen can be used for differentiating from other spindle cell tumours.

However, in our case IHC was not carried out, partially due to financial constraints, and also because, our histopathological report yielded decisive information. At times, differentiating leiomyoma from a low grade leiomyosarcoma becomes tough, hence Robbins and Corten[15], advocated that 10 mitotic figures per field signify plausible malignant behaviour, while less than two mitotic figures per 10 high power fields, and in general denotes a good prognosis.

When malignancies of oral smooth muscle are compared with the similar lesions in the female genitourinary tract, the proportion is reported to be higher in the oral counterpart, with 20% tumours diagnosed as malignant[3]. Complete excision or en bloc resection of the lesion with wide margins remains the first line of treatment for benign smooth muscle tumours[7].

In our case complete excision with wide margins, which was confirmed histopathologically followed by reconstruction with buccal pad of fat was performed under general anaesthesia. The prognostic outcome after surgery is exceptionally favourable in cases of smooth muscle tumours and rarely any case has been reported with recurrence.

CONCLUSION

Leiomyoma is a benign tumour of smooth muscles with infrequent incidence in oral cavity. The features of leiomyoma may be similar to those of various soft tissue lesions, clinically and histopathologically. Hence arriving at accurate diagnosis is both challenging and critical. With attention to the clinical scenario, meticulous knowledge of differential diagnosis, careful evaluation of the histopathological features and judicious use of special stains and IHC can help us in deciphering these lesions.

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