Recurrent Glossal Leiomyoma

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SUMMARY

Oral leiomyomas are rare benign tumour of smooth muscle. The first case of oral leiomyoma was reported by Blanc in 1884 and since then more cases has been published following advancement in immunohistochemical study. This tumour has an excellent prognosis and recurrences are extremely rare. We report a case of a recurrent glossal leiomyoma in a patient with HIV infection and the lesion recurred one year after the first excision.

KEY WORDS:
Glossal, leiomyoma, recurrent

INTRODUCTION

Leiomyoma is a benign neoplasm originating from smooth muscle. It is mostly found in the uterine myometrium where smooth muscles are abundant. It is less frequently seen in the gastrointestinal tract and subcutaneous tissue. The scarcity of smooth muscle in the mouth has made leiomyoma account for less than 0.5% of all benign tumours of oral cavity [1].

The greatest incidence of oral cavity leiomyoma is in the 40 to 59 year age-group with slight predominance in men [2]. The diagnosis of oral leiomyoma is based primarily on histological examination. So far, four cases of recurrence have been reported and all were histologically confirmed as the vascular type [3]. To the best of our knowledge, there is no previously reported recurrent case of solid type found in English literature.

CASE REPORT

A 28-year-old woman, seropositivity for HIV since 10 years ago, presented with a complaint of a painful swelling at the posterior part of the tongue for 2 months duration. The swelling had not changed in size over that period of time. The swelling had caused odynophagia but the patient denied any difficulty in breathing. There was no history of trauma.

She gave a similar history about 1 year ago and the lesion was excised at the local hospital. The pathological report affirmed that the lesion was a leiomyoma. Clinical examination revealed a solitary nodule localized at base of the tongue measuring 1x1cm, firm and slightly tender on palpation. There was no induration and the overlying mucosa was intact (Figure 1). Cervical lymph nodes were not palpable. A wide excision of the tumour with cuff of surrounding tissue was performed under general anaesthesia. As the mass was more on the right side, the endotracheal tube was passed without difficulty from the opposite side of the oropharynx. A throat pack was inserted before the procedure to avoid aspiration of blood. The mass was excised using monopolar diathermy in order to minimize bleeding. After extubation, she was observed in the recovery unit for any signs of airway obstruction.

The specimen consisted of a gray white tissue measuring 10x10x7mm. Microscopic findings showed fibrocollagenous tissue covered with non keratinizing stratified squamous epithelium. There was a circumscribed non encapsulated lesion in deeper dermis composed of fairly uniform spindle cells with round to oval nuclei and wavy cytoplasm forming fasicular pattern. Mitosis was rare. These cells expressed SMA and Vimentin but were negative for CD117, CD68, Desmin and EMA. The final diagnosis was leiomyoma of the tongue.

The patient did well during follow up in the clinic for one year. She was symptom free throughout the period and endoscopically confirmed that there was no tumour recurrence. However she default the subsequent follow up.

DISCUSSION

Leiomyoma in the oral cavity is a rare. The lips are the most common site (27.46%), followed by the tongue (18.30%), cheeks and palate (15.49%), gingiva (8.45%) and mandible (5.63%) [1]. The World Health Organization (WHO) describes three types of leiomyoma: leiomyoma (solid), angiomyoma (vascular) and epithelioid (leiomyoblastoma). In this case it was the solid subtype.

Clinical feature of oral leiomyomas includes a painless slow growing mass with firm and elastic consistency. Although most of them are asymptomatic, leiomyomas arising in tongue especially at the posterior region are most likely to cause symptoms like dysphagia, foreign body sensation in the throat and dyspnea. In the first case report of leiomyoma in an AIDS patient, the complaint was also a painful tongue mass,[4] as similarly featured in this case. Recurrence does not always occur in immunosuppressed patients. The uniqueness about our case was that it was recurrent, unlike the reported first case.

From clinical appearance alone, it is difficult to differentiate the tumour from other mesenchymal tumours or its malignant counterpart, leiomyosarcoma. In view of a history of leiomyoma, leiomyosarcoma needs to be considered, especially in this case when she presented with recurrent painful mass of the tongue.

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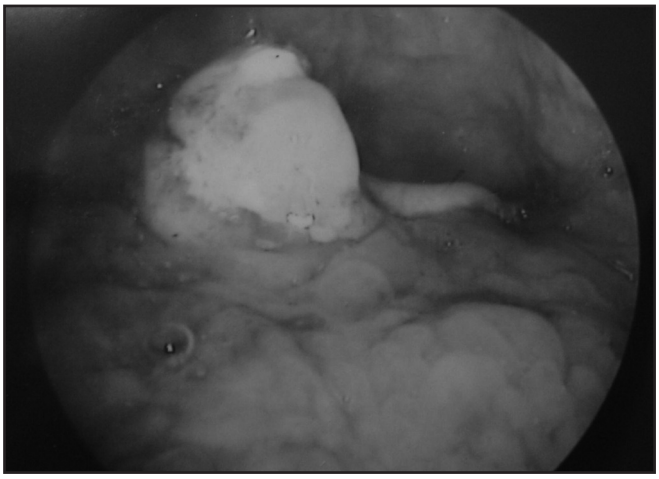

Fig. 1: A nodule-like mass mucosa seen at base of tongue.

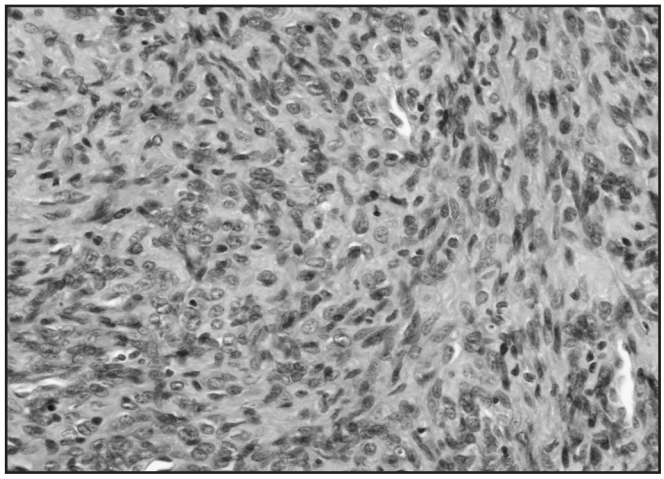

Fig. 2: The monotonous spindle cells in interlacing pattern with scanty mitosis(x20).

Histologically oral leiomyoma may resemble neurilemmoma, neurofibroma or other spindle cell tumours such as spindle cell pleomorphic adenoma or well differenciated leiomyosarcoma [5].

Immunohistochemistry is an accurate and reliable method for definitive diagnosis of oral leiomyoma. In our case, the marker studies have conclusively made a diagnosis of leiomyoma of the tongue and rare mitotic figures exclude leiomyosarcoma.

The prognosis of oral leiomyoma is excellent [1]. Complete surgical excision has been advocated as the primary treatment modality and possibility of recurrence is more likely if generous margins are not included in the specimen. Recurrences are rarely seen after a normal follow-up period of 6 years.

There were only four recurrences reported, which were of the vascular type [3]. In another study of 142 cases, 2 cases (4.2%) recurred after a few months [1]. Inadequate surgical margins could be the possible cause of recurrence in this patient. Thus a wide excision including 5mm surgical margin was contemplated and to date there has been no evidence of recurrence. However, a longer follow up period of at least 5 years has been recommended [5].

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