

Pediatric leiomyoma of the oral cavity: A rare entity

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Abstract

Leiomyoma is a benign smooth muscle tumor that frequently occurs in the uterine myometrium of the uterus and gastrointestinal tract rarely but seldom encountered in the oral cavity, particularly in children. Its occurrence in the oral cavity is considered rare probably because of the scarcity of smooth muscle tissue in the oral cavity. The frequently affected anatomical areas intraorally are lips, palate and tongue. The purpose of this case report is to put forth a rare case of oral leiomyoma in a 10-year-old girl child involving the retromolar region.

Keywords: Leiomyoma, retromolar trigone, spindle cell neoplasm

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INTRODUCTION

Neoplasms originating from the smooth muscle are seldom encountered in the oral cavity due to the paucity of smooth muscle in this particular anatomical region of the body.^[1,2] The frequency of smooth muscle tumors in the oral cavity accounts to 0.092%.^[1,3,4] Among the smooth muscle tumors of the oral cavity, leiomyomas are more common than leiomyosarcomas.^[1,5]

Leiomyomas are generally encountered in the myometrium of the uterus followed by the cutaneous region and the gastrointestinal tract. The prevalence of leiomyoma in the oral cavity accounts up to 0.065%.^[1,2,6-8] In the oral cavity, it is generally encountered in the fourth and fifth decades of life.^[1,2,9] However, it is very rarely encountered in children.^[10,11] The lips followed by the tongue are the commonly affected, while the mandibular retromolar trigone is rarely affected.^[2,10,12]

CASE REPORT

A 10-year-old female child reported to our unit complaining of a swelling in the left lower jaw region behind the teeth for the past 4 months. Swelling started as a peanut size and increased to the present size. Facial symmetry was noted extraorally, but cervical lymph nodes were not palpable. Intraoral examination revealed a solitary, pedunculated, well-circumscribed, nonfluctuant, nonsuppurative swelling, distal to the lower left first molar tooth extending into the retromolar region measuring 3 cm × 3 cm approximately in the greatest dimensions. Overlying surface on the superior aspect revealed indentations of upper teeth [Figure 1]. Swelling was nontender without localized rise in temperature. Fine needle aspiration was negative. The permanent mandibular left first molar was slightly mobile. No anesthesia/paresthesia was noticed. No relevant medical history was noticed.

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An orthopantomogram revealed that the swelling was not odontogenic in origin. Radiolucent shadow of the soft tissue growth with erosion of bone in distal to first molar and retromolar region was observed [Figure 2]. Based on the clinical and radiographic examination, a provisional diagnosis of fibroma was made. Considering the age and location of the swelling, an excisional biopsy was planned under general anesthesia (GA). Patient written consents were obtained. Following complete systemic evaluation, surgical excision was done under GA. Macroscopically, it is pinkish in color, elastic in consistency, oval in shape and well encapsulated [Figure 3]. No teeth were extracted.

Microscopic findings revealed a stratified squamous epithelium with irregular acanthosis with ulceration covered by neutrophilic exudate and underlying granulation tissue. Subepithelial stroma shows an ill-defined lesion composed of spindle cells arranged in interlacing fascicles and bundles with ill-defined cytoplasm and mildly pleomorphic vesicular nuclei with inconspicuous nucleoli. Interspersed capillaries are seen suggestive of low-grade spindle cell neoplasm [Figure 4]. Immunohistochemistry was done which revealed immunoreactivity for SMA and negativity

for the desmin and S-100 protein [Figure 5]. The patient was kept on periodic follow-up without any signs of recurrence.

DISCUSSION

The occurrence of oral leiomyoma is extremely rare due to the scantiness of smooth muscles in the oral cavity.^[1,2,13,14] However, it is advocated that the tunica media in the walls of the blood vessels, the smooth muscle of ductus lingualis, the circumvallate papillae and heterotopic smooth muscle could be the potential source for the origin.^[11,13] Majority of the leiomyomas occurring in the head and neck region are slow growing and asymptomatic.^[1,2,15,16] They manifest as a smooth-surfaced submucosal nodule and the overlying epithelium seldom ulcerates. The color of the lesions generally is similar to that of the normal mucosa.^[17-19] The lesion usually grows up to a few centimeters.^[19,20] Our case is in accordance with previous studies. Intraosseous leiomyomas are extremely rare.^[11,21] The differential diagnosis based on the clinical presentation includes fibroma, lipoma, neurofibroma, mucocele, lymphangioma and hemangioma.^[2,17]



Figure 1: Preoperative intraoral view of the swelling



Figure 2: Preoperative orthopantomogram of the patient

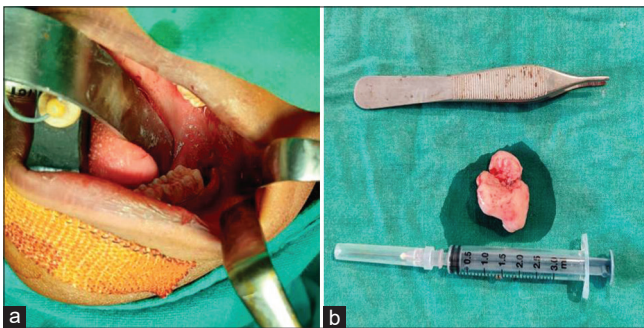


Figure 3: Intraoperative view: (a) Intraoperative view of the surgical area. (b) Macroscopic view of the excised lesion

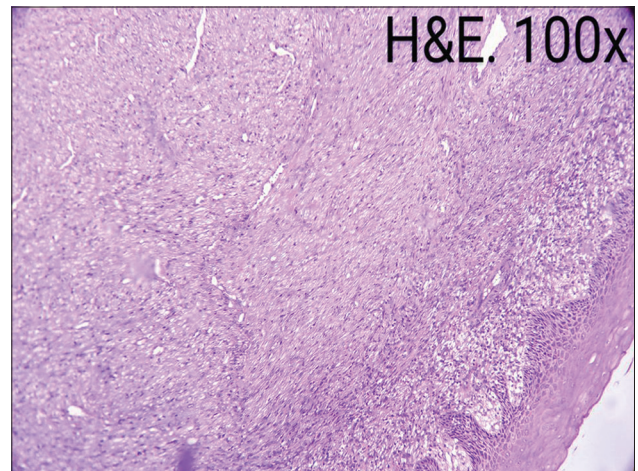


Figure 4: Histopathological picture of the excised lesion

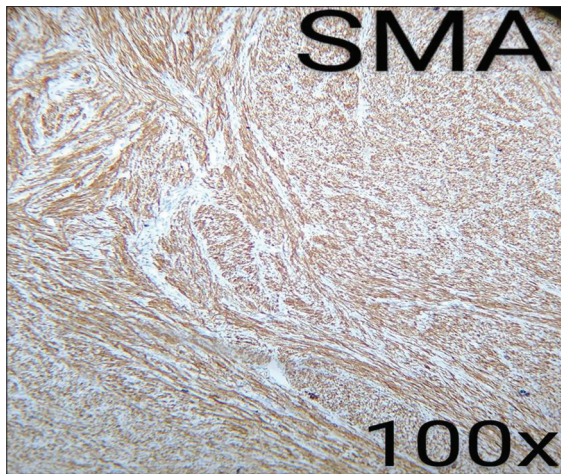


Figure 5: Immunohistochemistry of the excised lesion showing SMA positive

The etiology of leiomyoma is still unclear. It could be attributed that minor trauma, venous stasis, hormonal changes and genetic translocation could be the potential reasons.^[22] The undefined nature of clinical presentation of leiomyoma necessitates a microscopic evaluation.^[13,18] Microscopic differential diagnosis includes benign spindle cell tumors in the form of schwannoma, neurofibroma, fibrous histiocytoma, nodular fasciitis and malignant ones such as leiomyosarcoma.^[2]

Immunohistochemical studies are essential for final diagnosis.^[13,23] The key markers include SMA, vimentin, CD34, desmin and S-100.^[24-27] Leiomyomas express immunoreactivity for SMA and negativity for desmin and S-100 protein.^[23,26,27] It is advocated that each marker is representative of a particular tissue and should be correlated with clinical finding and the measurement of other markers.^[24] They can be managed by surgical excision with good prognosis exhibiting only 6% of recurrence.^[10,17,28]

CONCLUSION

Leiomyomas are rare, benign smooth muscle neoplasms but seldom encountered in the oral cavity. Considering their less incidence and nonspecific clinical presentation, making a precise diagnosis is challenging. An accurate diagnosis necessitates a histopathological evaluation and the use of stains. The authors feel that it is essential to document such rare clinical entities to enhance their understanding.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understand that

name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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