

An Intramuscular Lingual Lipoma: A Case Report and Review

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ABSTRACT

Context: About 15 to 20% of lipomas occur in the head and neck region and only 1 to 4% of them affect the oral cavity. The review of an intramuscular lipoma (IML) of the tongue has been presented here.

Aims: Intramuscular oral lipomas are rare. They commonly affect the buccal mucosa followed by the tongue.

Settings and design: The representative tissue received was 10% formalin fixed, was 1 × 1 cm in diameter, and was floated on the surface of the fixative.

Materials and methods: Thick sections (0.3 μ) were obtained from paraffin-embedded tissues that had been processed and stained with routine hematoxylin and eosin (H&E) stain. Stained sections were then reviewed.

Conclusion: Examination of paraffin-embedded sections showed mature adipocytes traversing between the bundles of skeletal muscles. No cellular atypia, necrosis, or mitotic activity was seen thereby indicating absence of malignancy. Intramuscular lipomas infiltrate subjacent muscles and recur locally.

Keywords: Infiltrating lipoma, Intramuscular lipoma, Skeletal muscles, Tongue.

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INTRODUCTION

Lipoma is a common mesenchymal neoplasm affecting the human body.¹ Benign lipomatous tumors have been classified by the World Health Organization (WHO) into fibrolipoma, angiolipoma, spindle cell/pleomorphic lipoma, myxolipoma, chondroid lipoma, osteolipoma, myolipoma, lipomatosis of nerve, lipoblastoma/lipoblastomatosis, and hibernoma.^{2,3} The lipomas are rare in the head and neck region and still rarer in the oral cavity.^{1,4}

The buccal mucosa is the predominant intraoral site affected by the lipoma followed by the tongue.³ Most lipomas occur sporadically, while some of them may be associated with inherited disorders, such as multiple lipomatosis, Gardner syndrome, Cowden syndrome, and Madelung disease. Lipomas are morphologically indistinguishable from normal fat, yet differ from it, as its lipid is not available for metabolism. Unlike normal fat, it is usually surrounded by a thin fibrous capsule.⁵

Intramuscular lipoma (IML) is a slow-growing and painless lesion affecting the large muscles of the extremities of adult males exhibiting a characteristic infiltration by the adipose tissue, followed by muscular atrophy. It comprises <2% of all lipomas and is very rare in oral and maxillofacial regions.⁶

CASE REPORT

A 60-year-old healthy female patient presented to the outpatient department with a 3-month history of a swelling at the base of the tongue. Clinical examination revealed a diffuse, non-tender, soft tissue mass on the ventral surface of the anterior two-thirds of the tongue located deep within the lingual mucosa (Fig. 1). The lesion was pale in color, mobile, and diffuse and the overlying mucosa was intact. The patient had no tongue dysmotility and dysarthria, and did not have dysgeusia and sensory changes. Following strict aseptic precautions, an excision

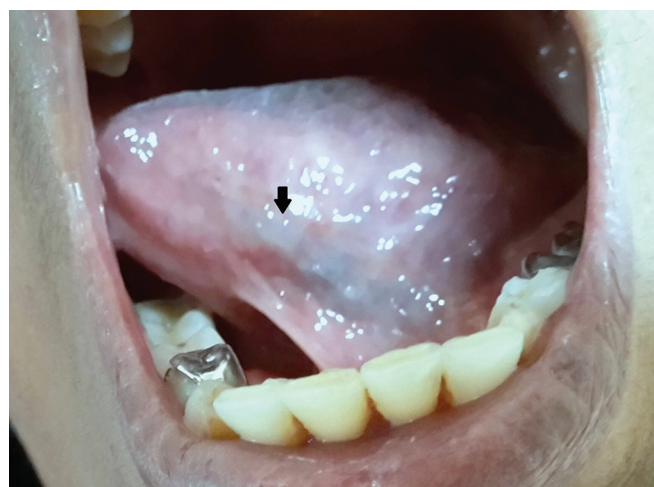


Fig. 1: Ill-defined, deep-seated lesion on the ventral surface of the tongue

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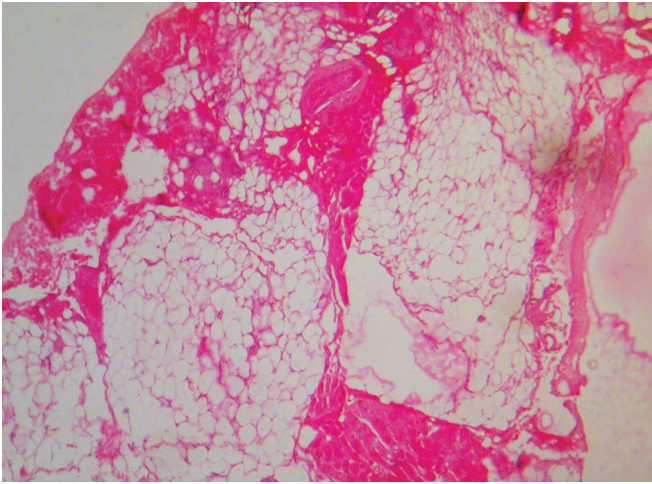


Fig. 2: Unencapsulated, lesional tissue showing mature adipocytes infiltrating into the skeletal muscle bundles (H&E 40×)

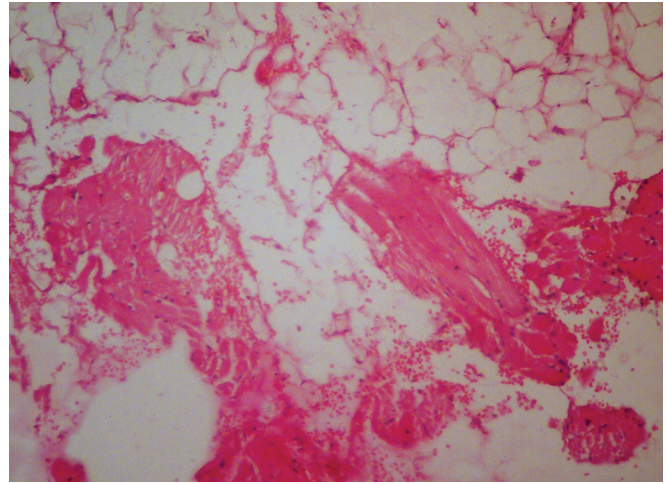


Fig. 3: Mature adipocytes infiltrating the skeletal muscle bundles without cytological atypia, mitosis, or necrosis (H&E 100×)

biopsy was performed under local anesthesia in a routine clinical set up. Routine blood examination and the vital signs, when examined prior to the surgery, were within normal limits. Gross examination revealed a pale, yellowish colored specimen measuring 1×1 cm in size and soft in consistency. The specimen was sent for routine histopathological processing. The tissue floated when placed in the fixative. Microscopic examination showed an unencapsulated lesion (Fig. 2) with mature adipocytes containing peripherally placed nucleus. The cells were interspersed between the striated muscle bundles (Fig. 3). They did not exhibit pleomorphism, mitotic figures, or necrosis; features that could rule out the diagnosis of a liposarcoma. The histopathological diagnosis of an IML was given. Special stains like the Oil Red O stain or immunohistochemistry may be done to confirm the diagnosis. Complimentary diagnostic tests, such as magnetic resonance imaging (MRI) and the computed tomography (CT) scan, may help in the exact demarcation of the tumor and may also help in diagnosing the benign nature of the lesion.

Healing was uneventful and the patient was well satisfied with the surgical results. Follow-up examination of the patient was done for up to 6 months; during which period, no recurrence of the lesion was noted.

DISCUSSION

Muscular lipoma, a rare variant of lipoma, was first described by Paget et al in 1853 as a lipoma infiltrating into the trapezius muscle. In 1946, Regan et al introduced the term "infiltrating lipoma" for lesions infiltrating into muscle or other subjacent structures. As of now, only lesions showing clear radiological, surgical, or microscopic features are considered as "infiltrating." Greenberg et al further added that infiltrating lipomas may be intermuscular or intramuscular. Moriconi based

his classification on differentiation of lipomas located between muscles or within them. Intramuscular lipoma⁷ was further classified into infiltrative, well-defined/non-infiltrative, or mixed (with areas of infiltration and well-defined areas). Fletcher et al established that 83% were of the infiltrative type and 17% were of the well-defined type.¹ Infiltrative type tends to recur and often resembled a well-differentiated liposarcoma histopathologically.⁸

Intramuscular extension of lipoma is clinically diagnosed by the change in consistency of the swelling on the contraction of the muscle involved. The most common site of occurrence of this tumor is the skeletal muscle of both the upper and lower extremities. Intramuscular lipomas almost certainly have neoplastic pathogenesis and signify a true neoplasm arising directly from the multipotent mesenchymal cells. Reactive pathogenesis has been proposed as well. Chronic irritation, trauma, endocrine and genetic factors, obesity, and developmental disorders stimulating the uncontrolled growth of lipomas have all been suggested as predisposing factors for development of IMLs.¹ Genetic study has shown MDM2 amplification in large or aggressively recurring IMLs even in the absence of atypia.⁹

Intraoral lipomas are predominantly seen on the buccal mucosa and the lingual mucosa (0.3%)⁴; the vestibule, palate, lips, and the gingiva are the less commonly affected sites.^{2,4} According to some authors, the tongue is the most common site of involvement accounting for about 40% of all cases followed by the buccal mucosa. Following cases till date, including the cases from India, the age of occurrence was 17 to 81 years with no gender predilection (19 males and 10 females).⁴ Turkish researchers identified a female preponderance as compared to males and an average age of occurrence of 59 years. A Brazilian study reported the median patient age to be 53 years, similar to the Turkish study.¹⁰

Intraoral IMLs of the tongue were rare. Lipomas of the tongue usually present as soft nodular, yellowish colored asymptomatic painless swelling covered by normal mucosa. Once present, a mucosal oral lipoma may increase to a size of 5 to 6 cm over a period of years, but in most cases are <3 cm in their greatest dimension at the time of diagnosis.⁵ They are usually located submucosally and are found to affect the organ's lateral edge and its anterior two-thirds. The lesion may be either solitary or multiple.¹¹

The oral IML is usually asymptomatic (Table 1). The lesions may be widespread and may cause pressure and infiltration of the deeper structures, such as the nerve trunks and muscles, resulting in sensory change, such as dysmotility, dysarthria, or muscle dysfunction.⁷

Magnetic resonance imaging findings normally show a single mass with a homogeneous internal structure, while the well-differentiated liposarcomas (Table 2) appear as multinodular masses with an inhomogeneous internal structure and thick septa; an IML may also show features of malignancy on the MRI.⁷ Hence, for a definitive diagnosis, correlation between the histological and clinical features rather than radiological findings is important.

The lesion has to be distinguished from a myofibroma, which consists of an admixture of smooth muscle and vascular spindle cells scattered among normal and mature adipocytes.⁶

Histopathologically, IMLs are usually unencapsulated with well-defined borders and the tumor mass is composed of sheets of abundant mature adipocytes that are uniformly polygonal in shape and size, with clear cytoplasm, and eccentrically placed nucleus compressed against the cell membrane. The adipocytes diffusely infiltrate the striated muscles with varying degrees of degenerative changes.

Table 1: Difference between intermuscular lipoma and IML¹

<i>Intramuscular lipoma</i>	<i>Intermuscular lipoma</i>
Occurs within skeletal muscle	Occurs between muscles
May be infiltrative or circumscribed	Circumscribed
Recurrence rate 0–20%	Virtually no recurrences

Table 2: Differential diagnostic criteria of IML with well-differentiated liposarcoma¹⁰

<i>Features</i>	<i>Intramuscular (infiltrating) lipoma</i>	<i>Well-differentiated liposarcoma</i>
Lipoblast	No	Yes
Myxoid differentiation	No	Yes
Cellular pleomorphism	Rarely	Usually
Increased vascularity	Minimal capillary	Abundant
Mitosis	No	Yes

There is a reported recurrence rate ranging from 19 to 62.5% after 6 to 7 years following surgery.⁷ Since the infiltrative type tends to recur, a precautionary wide excision of the tumor would have to be done. Subsequent to an adequate tumor clearance of >1.5 cm, follow-up every fortnight for a year, every half yearly for 2 years, and annually for the next 7 years is suggested because of a high probability of distant metastasis. The role of adjuvant radiotherapy or chemotherapy is minimal.⁹ Suitable clinical monitoring of patient is mandatory. No malignant changes have been reported in recurrent IMLs. This high recurrence rate is normally related to the difficulty in achieving a thorough surgical excision, as IMLs are perpetually more deeply placed, not encapsulated, and have an infiltrative growth pattern.⁷

CONCLUSION

- Most common tumors of the tongue are malignant.
- Tongue is an uncommon site for lipomas.
- Though WHO has stated histological variants for lipomas, the lipomas of the tongue are benign and ordinary.
- Intramuscular lipomas of the tongue are very unusual and are likely to recur if not excised adequately wide.
- A well-differentiated liposarcoma can impersonate a benign lipoma histologically.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

AUTHORS' CONTRIBUTIONS

The oral surgeon excised the tumor and all authors have contributed to the analysis, reading, writing, and researching this article.

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