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CASE REPORT

Intramuscular Lipoma of the Tongue: An Unusual Intraoral Entity

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ABSTRACT

Lipomas are benign soft tissue neoplasms of mature adipose tissue. It is most commonly found in the torso or limbs. However, their incidence in the oral cavity is relatively rare. They are slow-growing, painless mass and usually asymptomatic in nature. As they grow, they can interfere with the physiological processes in the surrounding area. We present a case of tongue lipoma in a 55-year-old man who had the painless mass for seven years. The mass was surgically excised. Histopathological examination confirmed the diagnosis.

KEYWORDS: Intraoral lipoma, intramuscular lipoma, tongue lipoma, infiltrating lipoma

INTRODUCTION

Lipoma is a benign tumour composed of adipose tissue. The majority of cases occur in the trunk and proximal extremities, making it the most frequent mesenchymal neoplasm. The incidence of oral lipoma is estimated to be 1% to 5% of all benign oral lesions [1]. The buccal mucosa is the most commonly affected site, followed by the tongue, lips, and floor of the mouth [2,3]. Lipoma is classified histologically as classic lipoma, intramuscular lipoma, fibrolipoma, angiolipoma, spindle cell lipoma, and salivary gland lipoma [3,4]. Studies on oral lipomas have found either a similar gender distribution or slightly higher occurrence among females [2,5]. Lipomas are mostly found in the subcutaneous tissues. However, there are also rare deep-seated subtypes known as intramuscular lipoma or infiltrating lipoma that are located deep within the muscle. They are extremely rare, accounting for less than 1% of all lipomas, with only a few cases reported in literature worldwide [6].

CASE PRESENTATION

A 55-year-old Malay man presented to the clinic with a painless tongue mass. The mass had been present for an approximate duration of seven years and had been gradually increasing in size for 7 months prior to the patient's presentation. The patient finally decided to

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seek treatment because the mass was interfering with his chewing. However, there was no weight loss, loss of appetite, family history of cancer, history of contact with tuberculosis patients or neck swelling. The patient denied trauma or injury to the tongue.

On examination, there was a solid yellowish, non-tender mass measuring about 1.5 cm x 1.5 cm on the left side of the tongue, firm in consistency with a smooth surface (Figure 1). Other aspects of the intraoral examination were unremarkable. An ultrasound revealed a solid nodule of the tongue. He underwent an excisional biopsy of the mass under general anaesthesia. A well-defined mass of approximately 1.5 cm x 1.5 cm was completely removed (Figure 2). Both the intraoperative and postoperative periods were uneventful. Histopathological examination (HPE) revealed an intramuscular lipoma. The HPE showed sheets of mature adipocytes separated by fine fibrous septa. There were no lipoblasts seen. Skeletal muscle bundles were seen within the lesion. Postoperative recovery was uneventful with no residual mass (Figure 3).

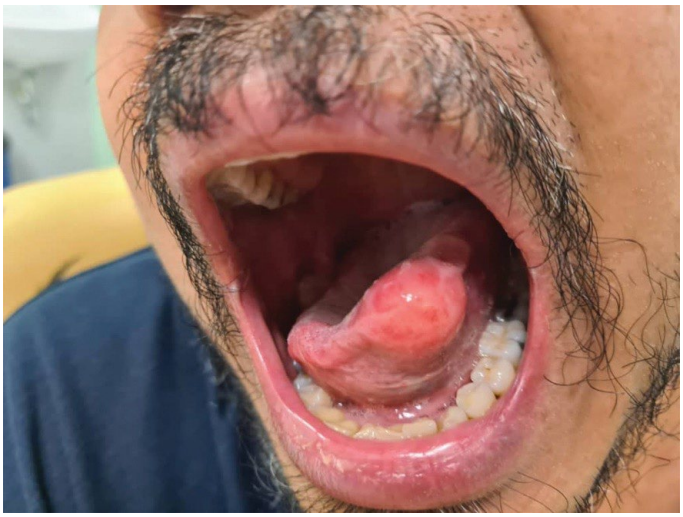

Figure 1 A painless smooth-surfaced mass on the left lateral border of the tongue

Figure 2 Floating yellowish mass consistent with lipomatous lesion

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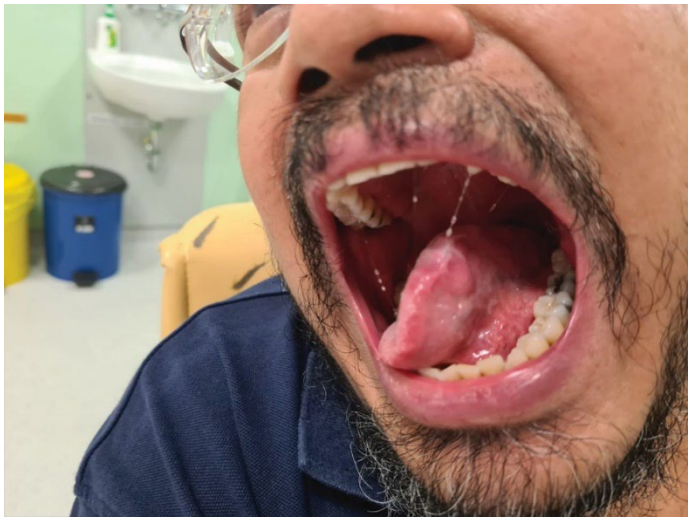

Figure 3 Post-operative tongue showed clinically no residual lesion

DISCUSSION

Oral lipomas are relatively uncommon, accounting for only 1% to 5% of all benign neoplasms of the oral cavity [1,2,4]. Egido-Moreno et al reported a 37.9% incidence of lipoma in the intraoral buccal mucosa, followed by the tongue (24%), lip (10%), palate (7%), floor of the mouth (7%), vestibule (6%), retromolar area (4%), and gingiva (2%) [5]. Other authors have reported nearly the same results [1,2,3]. A clear gender preference has not yet been established [1,2,3,4,6,8].

Lipomas are slow-growing, painless swelling that manifest for a long period of time. It may occasionally appear pedunculated or submerged. Clinically, intraoral lipoma presents as a well-circumscribed painless, rubbery, solitary, and submucosal swelling [7]. The differential diagnoses include dermoid cyst, fibroma, mucocele, minor salivary gland tumours, haemangioma, neuroma, rhabdomyoma or lymphangioma [5]. As the tumour grows in size, functional limitations may occur. It can cause discomfort during speech, deglutition, or mastication, especially if it is located on the tongue [2]. Our patient only sought treatment after seven years, because the swelling had begun to interfere with mastication.

Classic lipomas and its variants, that include spindle lipomas, pleomorphic lipomas, intramuscular lipomas, fibrolipomas, angiolipomas, salivary gland lipomas, myxoid lipomas, and atypical lipomas, are classified histologically. [2]. Intramuscular lipoma is a rare type of lipoma that usually develops in the trunk and extremities of the major muscles [8]. Intramuscular lipoma accounts for only 3% of intraoral lipoma, making intramuscular lipoma of the tongue extremely rare [5]. A total of 20 articles were retrieved from the PubMed database using the search term “intramuscular lipoma of the tongue”. Of these retrieved articles, only 10 were case reports [6].

The gold standard in the diagnosis of lipoma is histopathology. A crucial feature of the lipoma is that the excised lipoma specimen tends to float when placed in 10% formaldehyde solution [10]. Histologically, intramuscular lipomas are classified into three types: infiltrative (80%), well-circumscribed (17%), and mixed (with infiltrative margins and well-circumscribed or encapsulated areas) [8]. Intramuscular lipomas are found to be encased in an indistinct capsule containing skeletal muscle fibres. The stroma is composed of mature adipose cells that are wrapped around and separated by irregular bundles of skeletal muscle fibres [2].

Intramuscular lipomas can sometimes be misdiagnosed as liposarcoma. However, the absence of cellular pleomorphism and nuclear chromatin, as well as low mitotic activity, can help to confirm the diagnosis of intramuscular lipomas [3]. The presence or absence of lipoblasts also distinguishes intramuscular lipoma from liposarcoma. Furthermore, lipoma is made

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up of homogeneous mature adipocytes with low vascularization, while liposarcoma has hyperchromasia, vascularization, and cellular pleomorphism [8,9]. When a lesion is suspected to be malignant, an incisional biopsy or punch biopsy is recommended. We did not suspect malignancy in this case due to the painless, long-standing duration, and smooth-surfaced swelling, along with the ultrasound findings that were suggestive of lipoma rather than malignancy. In our case, the HPE revealed sheets of mature adipocytes separated by fine fibrous septa, a bundle of skeletal muscle bundles was seen within the lesion and no lipoblast was seen which led to the diagnosis of intramuscular lipoma.

Lipoma is surgically resected as the definitive treatment. In comparison to other lipoma subtypes, intramuscular lipoma has a high recurrence rate. These high rates are due to intramuscular lipoma's infiltrating tendencies and the difficulty in removing the entire mass [9]. The post-treatment recurrence rate ranges from 3% to 62% [1,6,8]. To prevent recurrence, the well-circumscribed area should be completely excised, and the infiltrative areas excised with a wide margin. However, complete removal of intramuscular lipomas is difficult when it has already infiltrated the muscle fibre [6]. The most important steps to achieve a clear margin and lower the local recurrence rate are a careful preoperative MRI evaluation of the margin and a combined marginal and broad excision method at the infiltrative region during the surgery [11,12]. Su et al. used this approach to treat eight patients with intramuscular lipomas surgically in varied locations, with no local recurrence in an average of 40 months [11]. Han et al. also reported that using a large excision margin, they were able to drastically reduce intramuscular lipoma recurrence rates by about 11% [12].

If the lesion is small, simple excision and primary suture can usually be used as the technique during operation. However, when the lesion is big, it is more prone to causing aesthetic and functional adverse effects due to defects after surgical removal. Thus, in order to widely excise a benign lesion in a small organ such as the tongue, the functional compromise needs to be justified. The enveloped flap design, as used by Hue et al., is advantageous in terms of surgical field of view, healing time and postoperative aesthetics when excising extensively a large lesion [6].

CONCLUSION

Intramuscular lipomas though rare can occur in the oral cavity, particularly on the tongue. Tissue biopsy is necessary to confirm the diagnosis. Surgical excision needs to be carefully designed to avoid functional and aesthetic issues. Patient follow-up is necessary due to the possibility of tumour recurrence. The clinician must be aware of the clinical characteristics and progression of oral lesions in order to differentiate between malignant and benign tumours and plan the appropriate treatment.

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Conflict of Interest

Authors declare none.

Authors' contribution

KM drafted the manuscript. AHS conceived of the study and provided the required information. IM critically revised the manuscript. All authors read and approved the final manuscript.

Informed Consent

The patient's informed consent was obtained.

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