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Oncopathology CHONDROLIPOMA OF TONGUE: A RARE CASE REPORT	
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ABSTRACT Condition of mature carrilaginous tissue. Herein, we describe a case of chondrolipoma of tongue in a 37 year old Indian lady. The lesion presented as a single, sessile, pinkish nodule at junction of anterior two-third and posterior one-third of dorsal aspect of tongue since 6 months. Histopathologically, the mass revealed a well circumscribed, nodule lined by stratified squamous epithelium with proliferation of mature adipocytes and islands of well-formed mature cartilaginous tissue. Chondrolipomas are uncommon in the oral cavity, with only 12 cases being reported in tongue in the English literature.

INTRODUCTION

KEYWORDS : Chondrolipoma, tongue, oral lipoma

Lipomas are benign tumours of mesenchymal origin. They are well circumscribed, expansile connective tissue neoplasm, predominantly composed of mature adipose cells. They usually presents as slow-growing, solitary and asymptomatic subcutaneous or superficial lesions. They may occur anywhere in the body. About 20% of lipoma affect head and neck region with only 1-5% of neoplasm involving oral cavity. The buccal mucosa is most affected intra-oral site. Oral lipomas are more common in males than females, and most common age group affected is between 40-60 years of age.⁽¹⁻³⁾

Histologically lipomas are composed of mature adipocytes arranged in lobules separated by fibrous connective tissue septae and are ocassionally associated with one or more secondary mesenchymal elements. Various histopathological variants of lipoma have been recognized, such as fibrolipomas, anigiolipomas, myolipomas, spindle cell lipomas, chondroid lipoma osteolipomas, chondrolipomas. Lipomas and fibrolipomas are the most frequently observed histological types in oral cavity.⁽⁴⁾ Chondrolipomas are histological variants of lipomas with cartilaginous metaplasia. They have been described in subcutaneous and deep soft tissues, particularly in parosteal location, but are rare in oral cavity particularly tongue.^(5,6) As per Pubmed database, till date only 12 cases of chondrolipoma of tongue have been reported in English literature among these only two cases are from India.⁽⁷⁻¹⁸⁾ As per literature review, this is a third case reported in India, we wish to present a case of 37-year-old lady diagnosed with chondrolipoma of tongue.

Case Study

A 37 year old lady presented with painless swelling over dorsum of the tongue since 6 months. Intraoral clinical examination revealed a single, sessile, pinkish nodule at junction of anterior two-third and posterior one-third of dorsal aspect of tongue. No other alterations were observed in the oral cavity.

The lesion was sharply demarcated from the surrounding area, hence, an excisional biopsy was done and sample was sent for histopathological examination. Gross examination revealed a single, oval, well circumscribed, pinkish, polypoidal nodule measuring $1.5 \times 1.5 \times 0.8$ cm. The cut surface was yellowish greasy and soft. The nodule was submitted entirely. Microscopic examination revealed a polypoidal lesion lined by stratified squamous epithelium with underlying sheets of mature adipocytes arranged in lobules separated by fibrous septae, admixed with mild mononuclear inflammatory infiltrate and few small blood vessels. Few islands of benign chondroid tissue were noted. No granulomas were seen. No necrosis was observed. There was no evidence of invasive malignancy seen in sections examined. Histopathological diagnosis of Chondrolipoma of tongue was made.



Fig 1 : Gross specimen of Chondrolipoma of tongue showing yellowish greasy cut surface

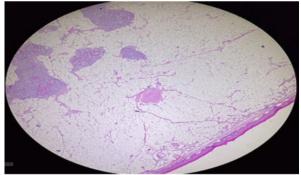


Fig 2: Hematoxylin and eosin stained slide demonstrate stratified squamous epithelium overlying a well circumscribed mass of adipose tissue (100X)

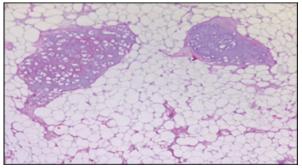


Fig 3: Hematoxylin and eosin stained slide demonstrate islands of mature cartilage surrounded by mature fat cells (400X)

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DISCUSSION

Chondrolipoma is an infrequent tumor, preferably presenting in the parosteal region. The first case of oral chondrolipoma was described in 1976 in a 72-year-old male in lower lip by McAndrew and Greenspan (19), whereas, first case of chondrolipoma of tongue was reported in 1989 in a 47-year-old male by Maes and Eulderink⁽⁷⁾. Since then only a few cases have been reported in oral cavity. A review of the PUBMED database revealed only 12 cases of Chondrolipoma of tongue in English literature $^{(7-18)}.$ We have included only those cases which presented over tongue and histologically showed presence of only cartilaginous tissue in a predominantly mature adipocyte proliferation. The clinicopathological features of these cases including the present case have been epitomized in Table 1.

Chondrolipoma of tongue have been diagnosed in between 14 to 71 years, but it appears to be a tumor of older individuals. Only three cases have been testified before the age of 30 ^(12,16,18). Although these lesions are established in adults, they may initiate at a younger age since some of these cases have shown extended period of appearance ^(11,12,14). There may be a definite bimodal age preference as significant number of cases seem to originate in the first two decades of life but more study is required to make a remark. (10-12,16) Analogous to other lipomas at other sites, chondrolipomas of tongue are believed to be more common in men.

Histologically, variants of lipoma includes angiolipoma, myolipoma, angiomyolipoma, myelolipoma, chondroid lipoma, spindle cell and pleomorphic lipoma.⁽⁴⁾ Chondro lipomas are characterized by the proliferation of mature adipocytes with additional mature cartilaginous tissue formation. ^(4,11,12) Mature cartilaginous areas in a chondrolipoma should be distinguished from chondroid lipoma, which consists of mature adipocytes admixed with multivacuolated lipoblast like cells in a myxohyaline and chondroid matrix.^(4,20) Since chondroid lipoma have an immature trait, it may give a pseudosarcomatous appearance and may be misguided for lipoblastic or chondroblastic malignancies.⁽²¹⁾ Conversely true chondrolipoma have entirely mature tissue with privation of any lipoblastic cells. Due to resemblance in nomenclature, the two entities are often confused with each other and there is a likelihood of misdiagnosis. In this review, we have encompassed cases of true chondrolipoma only and all cases with immature lipoblast like component or hibernoma-like areas have been omitted. Another differential of chondrolipoma is extra-skeletal chondroma occurring in deep submucosal areas surrounded by adipose tissue. Microscopically chondromas have greater proportion of cartilaginous tissue arranged in lobular pattern whereas, the chondroid element of chondrolipoma is focal and lacks any lobular arrangement.⁽²²⁾

The pathogenesis of chondrolipoma is uncertain.^(2,12) Several hypothesis have been tried to elucidate the occurrence of cartilage within the mass of adipose tissue. The main mechanisms proposed include the distinction of pluripotent mesenchymal cells into adipose tissue and cartilage^(2,7,1) analogous to benign mesenchymomas, development from different cell lines⁽¹¹⁾ and the existence of cartilaginous metaplasia in a pre-existing lipoma^(4,8,9). Moreover, Hietanen and Mäkinen ⁽⁹⁾and Maes and Eulderink⁽⁷⁾ also propose that the tumor is essentially a lipoma or chondroma and the associated cartilage or fat is a form of metaplastic stromal reaction. Mesenchymal cells can be altered by local or systemic factors such as local trauma and prolonged ischemia. (4) According to Rau et al. (6) the derivation of these lesions from pluripotent cells, which also have been identified in adult differentiated fat tissue seems likely. In vitro and animal models discovered multidirectional differentiation capacity of adiposederived stem cells. This permits development of bone, cartilage, fat, muscle, blood vessels and fibrous tissue from the same precursor cells.⁽¹¹⁾ The synonyms for this variant of lipoma include chondrolipoma, lipoma with chondroid metaplasia, benign mesenchymoma and lipoma with cartilaginous change. According to Fujimura and Enomoto, the term lipoma with cartilaginous (osseous) change, which only defines the histological findings, would be desirable because pathogenesis of this tumor is still questionable.⁽⁸⁾ However, Furlong et al. refer to these tumors as lipoma with cartilaginous metaplasia.⁽²

Recently, Nakano et al. detected important differences in the pattern of immunohistochemical expression of transforming growth factor-beta (TGF-B), latent TGF-B binding protein-1 (LTBP-1), and bone morphogenetic protein (BMP) between chondrolipoma, normal human tracheal cartilage and osteochondroma. Therefore, Nakano et

al. recommend that the countenance of TGF-B, LTBP-1 and BMP might be important in the pathogenesis of chondrolipoma.

Whatever the pathogenesis, lesion is essentially benign and surgical excision is the treatment of choice with no cases of recurrences been reported in the literature. Chondrolipomas are well demarcated tumors that can be easily removed.

CONCLUSIONS

Chondrolipomas are rare benign deviations of lipoma. The cartilage found in the tumor most likely signifies a metaplastic change or choristomatous proliferation and could be attributable to multipotentiality of mesenchymal stem, cells. The oral chondrolipomas shows few eccentricities from classical oral liopma, predominantly being the marked preference for involvement of tongue and conceivably bimodal age of onset and merits reporting of such cases with detailed clinicopathological evaluation in future. These tumors are acquiescent to excision and do not recur.

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