CASE REPORT

Chondrolipoma: Report of a rare intra oral variant with review of histiogenetic concepts

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ABSTRACT

Chondrolipomas are benign mesenchymal tumors characterized by the proliferation of mature adipocytes associated with variable amounts of mature cartilaginous tissue. Herein, we describe a case of chondrolipoma of the tongue in a 35-year-old Indian male. The lesion presented as a nodular, sessile, pink mass on posterio-dorsal surface of left side of the tongue since approximately 20 years. Histopathologically, the mass revealed a well circumscribed, encapsulated proliferation of mature adipocytes with islands of well formed mature cartilaginous tissue. Chondrolipomas are uncommon in the oral cavity, with only 14 cases being reported in the English literature.

Key words: Chondrolipoma, lipoma, tongue

INTRODUCTION

Lipomas are benign, well circumscribed, expansile connective tissue neoplasms predominantly composed of mature white fat cells. They may occur anywhere in the body and usually present as slow-growing, solitary and asymptomatic subcutaneous or superficial lesions. About 20% of cases of lipoma affect the head and neck region with only 1%-5% of these neoplasms involving the oral cavity.[1-3] Histologically, lipomas are composed of mature adipocytes arranged in lobules that are separated by fibrous connective tissue septa and are occasionally associated with one or more secondary mesenchymal elements. Different variants of lipoma have been described, such as fibrolipoma, angiolipoma, myolipoma, spindle cell lipoma, chondroid lipoma, chondrolipoma and osteolipoma.[4] The buccal mucosa is the most affected intra oral site; and lipomas and fibrolipomas are the most frequently observed histological types in the oral cavity.[1-3] Among the histopathological variants, lipomas with cartilaginous or osseous metaplasia, called chondrolipomas or osteolipomas respectively, have been described in the subcutaneous and deep soft tissues, particularly in the parosteal localization but are rare in the oral cavity.[5,6] As per Pubmed database, till date only 14 cases of chondrolipoma in oral cavity have been reported in English literature.[7-19] Herein, we report a case of this rare intraoral variant in a 35-year-old male patient.

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CASE REPORT

A 35-year-old Indian male patient presented with a painless mass on the postero-dorsal region of tongue since last 20 years. The lesion showed slow growth and the patient reported that there was no increase in size for past many years. Clinically, the lesion was an oval, well-circumscribed nodule measuring approximately 1.0 × 1.0 cm. The overlying mucosa was pale and the surface was smooth. On palpation, the mass was firm in consistency with no tenderness or discharge [Figure 1]. A clinical diagnosis of fibroma was made and based on the long standing history and benign appearance, an excisional biopsy was performed. The surgical area healed uneventfully and six-month follow-up of the patient revealed no evidence of any recurrence.

Gross examination of the excised mass revealed a pale yellowish cut surface with foci of shiny white areas [Figure 2]. Microscopically, the tumor was composed of well circumscribed, encapsulated lobular mass of mature adipose tissue separated by fibrous septae. Foci of metaplastic hyaline cartilage were evident within the mass along with delicate capillaries [Figure 3]. The cartilage cells as well as the fat cells did not show any mitosis, pleomorphism or any other histological evidence of malignancy. No evidence of lipoblasts or hibernoma-like cells was seen [Figure 4]. Based on the histopathological features a diagnosis of chondrolipoma was made.

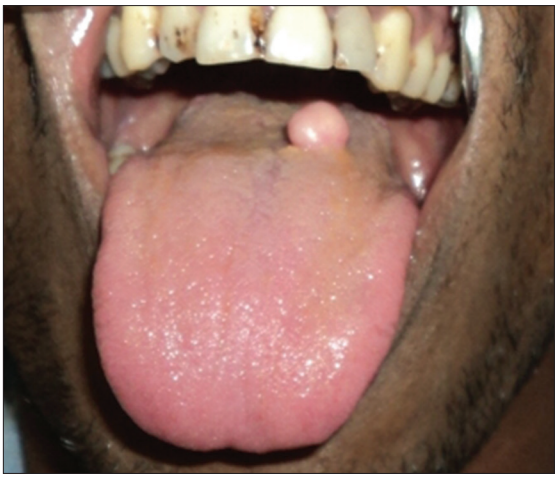

Figure 1: Figure shows nodular growth on the posterior dorsal surface of tongue

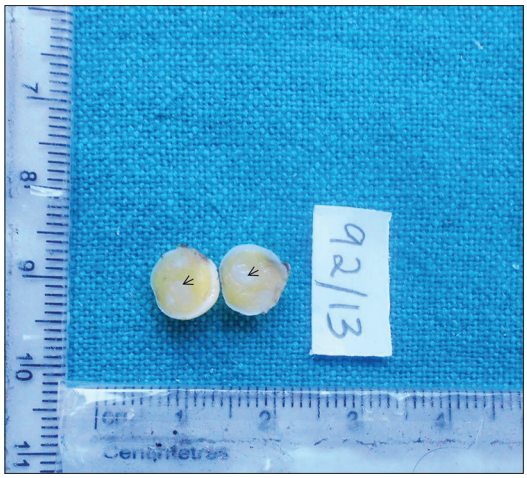

Figure 2: Figure shows cut surface of gross specimen. Note the yellowish color of the cut surface of the lesion with foci of white hyalinized areas *(arrows)*

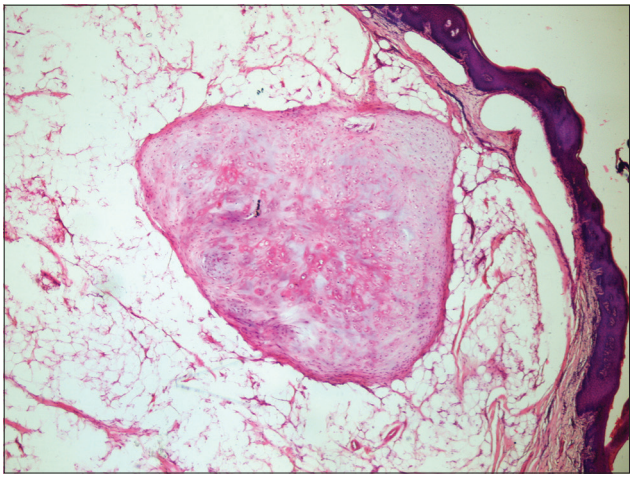

Figure 3: Low power photomicrograph showing stratified squamous epithelium overlying a well-circumscribed mass of adipose tissue, surrounded by a fibrous capsule and containing a focus of hyaline cartilage. (H&E stain, ×40)

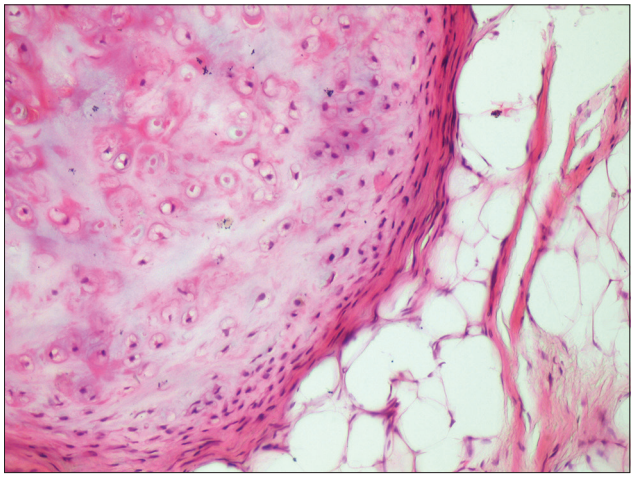

Figure 4: High-power photomicrograph showing cartilaginous foci made up of mature normal appearing chondroid tissue with peripheral rim of spindle shaped lining cells. (H&E stain, ×400)

DISCUSSION

Chondrolipoma is a rare tumor, usually occurring in the parosteal region. The first case of oral chondrolipoma was reported by McAndrew and Greenspan[7] in 1976 in a 72-year-old male in lower lip and since then only a handful of cases been reported in oral cavity. Many clinical and histopathological characteristics of chondrolipomas of the oral cavity continue to be poorly understood because of the rarity of this tumor, together with the lack of data in the literature. A review of the PUBMED database revealed 14 such cases in English literature.[7-19] We have included only those cases which presented strictly within the oral cavity 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 summarized in Table 1.

Cases of oral chondrolipoma have been diagnosed in patients aged from 14 to 72 years, but it seems to be a tumor of older individuals with majority of cases being diagnosed after the age of 50 years. Only two cases have been reported before the age of 30.[15,19] While these lesions are usually diagnosed in older adults, they may originate at a younger age as some of these tumors including the present case have shown long duration of presentation.[14,15,17] There may be an actual bimodal age predilection as substantial number of cases (almost one-third including the present case) seem to originate in the first two decades of life[13-15,19] but a larger number of cases will be required to accept or refute this observation. This feature of chondrolipoma slightly differs from a conventional lipoma, which are rarely seen in the first two decades of life and are thought to make their appearance at the age when fat starts accumulating in inactive individuals.[4] Hence, most lipomas including those of oral cavity become apparent between 5th to 7th decades of life.[2] In fact all of the cases of chondrolipoma involving young individuals have been reported in recent literature probably highlighting the fact that awareness about this variant is increasing among histopathologists and possibly

Table 1: Clinical features of 14 cases of oral chondrolipoma described in English literature

Author	Age at diagnosis (years)	Duration at the time of diagnosis	Probable decade of onset	Sex	Site	Size	Treatment
McAndrew, Greenspan 1976	72	6 months	8th	Male	Lower lip	2.5×1.5 cm	Surgical excision
Allard *et al.* 1982	69	2 years	7th	Female	Lower lip	1.0×1.0 cm	Surgical excision
Maes, Eulderink 1989	47	Some months	5th	Male	Tongue	Less than 1cm	Surgical excision
Fujimura, Enomoto 1992	56	2 months	6th	Male	Tongue	1.5×1 cm	Surgical excision
Szudrowicz, Jakobi-Roz 1995	52	Some months	6th	Male	Lower lip	1.7×1.7×1.3 cm	Surgical excision
Hietanen, Makinen 1997	68	NA	NA	Female	Tongue	1.4×1.0 cm	Surgical excision
Goel G *et al.* 2008	36	Since childhood	1st	Female	Tongue	3.0×2.0×1.0 cm	Surgical excision
Nonaka *et al.* 2009	30	10 years	2nd	Male	Tongue	1.4×1.0 cm	Surgical excision
Kuyama *et al.* 2009 (2 cases)	28 and 59	27 years, 2 months	1st and 6th	Female, Male	Tongue, Lower labial vestibule	1.6×1.5×1.2 cm, 0.9×0.5×0.5 cm	NA
Bezerre *et al.* 2010	68	20 years	5th	Female	Tongue	NA	Surgical excision
Berg, Gorsky 2010	69	1 month	7th	Male	Tongue	1.0 cm	Surgical excision
Shabbir, Greenwood 2011	71	NA	NA	Male	Tongue	NA	Surgical excision
Batchvarova *et al.* 2012	14	NA	NA	Male	Tongue	NA	Surgical excision
Present case	35	20 years	2nd	Male	Tongue	1.0×1.0 cm	Surgical excision

some of these lesions may have earlier been overlooked. Similar to other lipomas at other sites, oral lipomas are believed to be more common in men[2] and this seems to be true even for chondrolipomas in the oral cavity. Oral chondrolipomas seem to have a strong predilection for occurrence on tongue but interestingly none of the lesions have been described in buccal mucosa which is the preferential site for conventional lipomas.[1,2]

Histologically, variants of lipoma differ from ordinary lipoma by characteristic microscopic picture and specific clinical setting. This group is chiefly represented by 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.[4,14,15] Some authors consider these lesions as one of the subsets of mesenchymomas.[13] Though this term has been used in past,[20] it was first defined by Stout[21] for tumors that are composed of two or more mesenchymal elements. On the other hand, Jones *et al*.,[22] described mesenchymomas as an unencapsulated soft tissue neoplasms composed of two or more mature mesenchymal tissues not normally associated with each other and no single mesenchymal tissue should predominate with respect to the other mesenchymal elements. Usually the cartilage in lipoma represents only a small part of the tumor and hence would not strictly fit to the above criteria. Mature cartilaginous areas in a chondrolipoma should be distinguished from chondroid lipoma, a lipoma variant that is uncommon in the oral cavity and consists of mature adipocytes admixed with multivacuolated lipoblast like cells in a myxohyaline and chondroid matrix.[4,23] This newly described lesion, having an immature aspect may give a pseudosarcomatous appearance and may be mistaken for lipoblastic or chondroblastic malignancies.[24] On the other hand, in true chondrolipoma the adipose component is entirely composed of mature tissue with lack of any lipoblastic cells. Due to similarity in nomenclature, the two entities are often confused with each other and it is possible they may have been diagnosed interchangeably in past. In this review, we have included only those cases which show typical features of a chondrolipoma and all cases showing immature lipoblast like component or hibernoma-like areas have been excluded. In addition, chondrolipoma may sometimes need to be differentiated from extra-skeletal chondroma specially those occurring in deep submucosal areas and hence surrounded by adipose tissue. Histologically, chondromas are characterized by greater proportion of cartilaginous tissue arranged usually in distinct lobular pattern.[25] whereas the chondroid component of chondrolipoma is focal and lacks any lobular arrangement.

The histogenesis of this tumor is still not completely understood. Several hypotheses have been tried to explain the occurrence of cartilage within the mass of adipose tissue. One of the possible explanations could be that this tumor represents a true mesenchymoma i.e. both the chondroid as well as the adipose components being neoplastic in nature. The initial neoplastic change may take place in the pleuripotent mesenchymal stem cells, present in oral mucosa and these neoplastic stem cells may then differentiate into adipogenic and chondrogenic cells. Presence of pluripotent mesenchymal stem cells, capable of such differentiation, has been demonstrated in the connective tissue of skeletal muscles and dermis[26] and it may be prudent to assume that similar cells may also be present in the lamina propria and submucosal tissues of oral cavity. Another possibility is the development of cartilage within neoplastic proliferation of adipocytes. Ability for multipotential differentiation including chondrogenic potential has been demonstrated in adipose

tissue derived stem cells.[27,28] Hence, it could be possible that some of the neoplastic adipocytes may differentiate into chondroblastic cells, the stimulus being either spontaneous or a metaplastic reaction triggered by long-term irritation. Similar metaplastic chondrogenesis has been described in peripheral fibromas of gingiva.[29] and may be attributed to chronic mechanical stress. An interesting, albeit a little farfetched possibility is that the neoplastic adipose tissue may develop in a pre-existing cartilaginous choristoma. Such choristomas, though rare, are known to occur in the oral cavity with tongue being the most common location.[30] This may also explain the rarity of this lesion and preferential occurrence in tongue as compared to the higher propensity of involvement of buccal mucosa for conventional lipoma. Finally, the lesion may not be neoplastic at all and may result from a combination of hamartomatous and choristomatous growth. Such combined "lipomaotus" hamartomas and choristomas made up of mature adipose tissue containing cartilage have been described in other sites of the body[31] as well as in inbred laboratory mice.[32] It was suggested that those cases containing additional tissue elements in these chiefly lipomatous growths should be considered choristomas. Even in the murine lipomatous growths additional tissue elements were found more frequently in long standing cases and it was considered that these additional elements like cartilage could be present at an earlier stage but may constitute only a minor volume of the lesion, hence could be easily overlooked and thus require careful complete serial sectioning of the entire growth. Recently, role of factors like Sox-9, RUNX-2, TGF-β, bone morphogenic protein (BMP) has been discussed in chondrogenic potential of lipomas.[15,33] We believe that oral chondrolipomas constitute a heterogenous group of lesions, some of which may represent true neoplasms while others, especially those appearing at a younger age, may be hamartomatous/choristomatous proliferations. The pluripotentiality of adipose derived stem cells and preferential expression and interplay of prochondrogenic molecules either through some genetic alteration or environmental factors are responsible for chondrogenesis in these lesions.

Whatever the pathogenesis, lesion is essentially benign and surgical excision is the treatment of choice. No recurrences have been reported in the literature.

CONCLUSION

Chondrolipomas are rare benign variants of lipoma characterized by slow growth. The cartilage found in the tumor most likely represents a metaplastic change or harmartomatous/choristomatous proliferation and could be attributed to multipotentiality of mesenchymal stem cells. The characteristics of oral lesions tend to show few deviations from the classical oral lipoma, chiefly being the marked predilection for involvement of tongue and possibly bimodal age of onset and warrants reporting of such cases with detailed clinicopathological evaluation in future.

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