Case Report

Fibrolipoma of floor of the mouth of 20 years of duration

R. Rajeev, V. T. Beena, G. Indu, Kanaram Choudhary[1], Aloka Devu

Department of Oral and Maxillofacial Pathology, Government Dental College, Trivandrum, Kerala, India, [1]320 Field Hospital, 99 APO

ABSTRACT

Intraoral solitary lipomas are very rare. In spite of under reporting, lipomas outnumber any other benign or malignant soft-tissue tumors by a considerable margin and represent the most common soft-tissue tumor. Fibrolipoma is a rare histologic variant of classic lipoma. In this paper, we have reported a rare case of solitary fibrolipoma of the oral cavity of 20 years of duration, with atypical histopathological features. In histopathology of case Lochkern cells were noted, these are atypical for usual fibroliopmas. Many lipomas remain unrecorded or are brought to the attention of a physician only if they reach a large size or cause cosmetic problems or complications due to proximity to vital structures. To the best of our knowledge, it is the first case of such long presentation of 20 years.

Key words: Adipocytes, fibrolipoma, Lochkern cells

INTRODUCTION

Solitary lipomas, consisting entirely of mature fat, have been largely ignored in the literature, as they grow insidiously and cause few problems than any other localized mass. Many lipomas remain unrecorded or are brought to the attention of a physician only if they reach a large size or cause cosmetic problems or complications due to proximity to vital structures. Therefore, the reported incidence of lipoma is undoubtedly much lower than the actual incidence. In spite of under reporting, lipomas outnumber any other benign or malignant soft-tissue tumors by a considerable margin and represent the most common soft-tissue tumor.

Lipoma is rare during the first 2 decades of life and usually makes its appearance when fat begins to accumulate in inactive individuals. Most cases become apparent in 40-60 years of age. When not excised, they persist for the remainder of life, although they hardly increase in size after the initial growth period. Statistics as to gender predilection may vary, but most reports show a higher incidence in men.[1,2]

Access this article online	
Quick Response Code:	Website: www.ccij-online.org
	DOI: 10.4103/2278-0513.138058

CASE REPORT

An 85-year-old male patient reported to Government Dental College, Trivandrum, Kerala, South India, in July 2013 with the chief complaint of swelling underneath the tongue for the past 20 years. The swelling was very small to begin with and increased to the current size during the past 1 year. Patient did not seek any treatment in between. The patient also had associated symptoms of difficulty in mastication due to the unwarranted increase in size of the lesion. Intra-oral examination revealed pedunculated soft-tissue swelling of size 5 cm × 5 cm × 5 cm in the floor of the mouth arising in relation to lingual aspect of mandibular alveolar ridge on the left side. The swelling was nonfluctuant and nontender on palpation. The lesion displaced the tongue medially and posteriorly. The color of the swelling was similar to surrounding mucosa. Oral hygiene status of the patient was poor.

Clinical diagnosis of lipoma arising from the floor of mouth was made. Further, computed tomography (CT) examination of neck showed well-defined hypo dense lesion with density ranging from − 50 to − 20 HU suggestive of fat in the sublingual region on the left side. The lesion had lobulated margins. A few soft-tissue strands could

Address for correspondence: Dr. R. Rajeev, Department of Oral and Maxillofacial Pathology, Government Dental College, Trivandrum - 695 011, Kerala, India. E-mail: drrrajeevmds@gmail.com

be seen within the lesions. No areas of calcifications were seen. The lesion of size 4.2 cm × 3.1 cm × 1.6 cm was limited anteriorly and laterally by the alveolar process of mandible [Figure 1]. A thin soft-tissue layer separated the lesion from the oral cavity proper, probably the capsule and mucosa. Inferiorly the lesion was limited by mylohyoid muscle. CT feature was also suggestive of lipoma. Excision biopsy was carried out. Gross specimen was of 5 cm × 5 cm × 3 cm in size [Figure 2]. Hematoxyline and eosin stained section showed densely collagenous connective tissue stroma infiltrated with proliferating adipocytes without regular lobular architecture and were interspersed by fibrous connective tissue septae. Lochkern cells, cells with vacuolated nuclei, were noticed [Figure 3]. No atypical features were observed. The stroma showed moderate vascularity and minimal inflammatory infiltrate. In immunohistochemical staining of section by vimentin, diffuse pattern of staining was observed, but was negative for fat cells [Figure 4]. Definitive diagnosis of fibrolipoma was made.

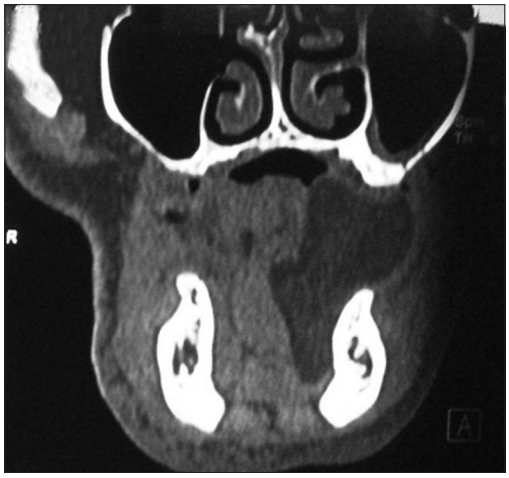

Figure 1: Computed tomography image showing well-defined hypodense lesion

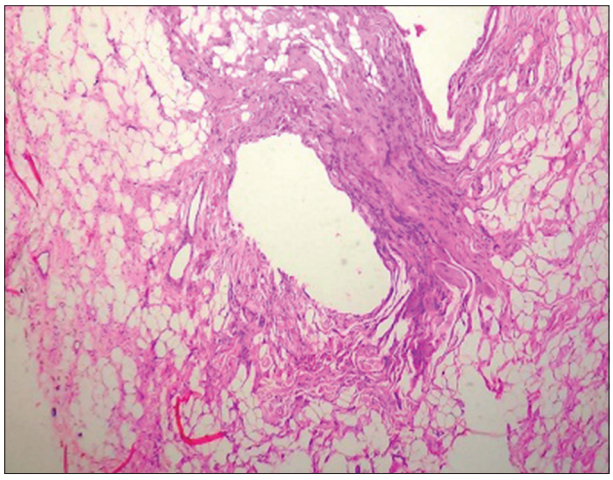

Figure 3: Histopathological section showing mature adipocytes in fibrous tissue (×10)

DISCUSSION

Lipomas are rare tumors of the oral cavity. A study by Juliasse *et al*.[3] in 2010 in Brazilian population showed that there was the predominance in females (2.4:1), with a peak incidence between the 6th and 7th decade. The buccal mucosa was the most affected site (53.7%), followed by the buccal sulcus (14.6%) and tongue (9.8%). Tumor size ranged from 0.5 to 10 cm and the mean reported duration was 48 months. Histologically, the following variants were identified: Lipoma (41.5%), fibrolipoma (34.1%), spindle cell lipoma (9.8%), sialolipoma (9.8%), osteolipoma (2.4%), and chondrolipoma (2.4%). Most tumors were well-delimited, irrespective of the variant. A review by Fregnani *et al*.[4] revealed the buccal mucosa as the most common site of occurrence and the tongue as the second most common site. Fibrolipolma rising from the floor of mouth has rarely been reported. In 2010, Manjunatha *et al*.[5] summarized 33 cases of oral fibrolipoma with bucaal mucosa as the most common site followed by tongue. Maximum and minimum duration was 10 years and 8 month respectively. Follow-up data showed no recurrence in 5 years duration. Duration of our

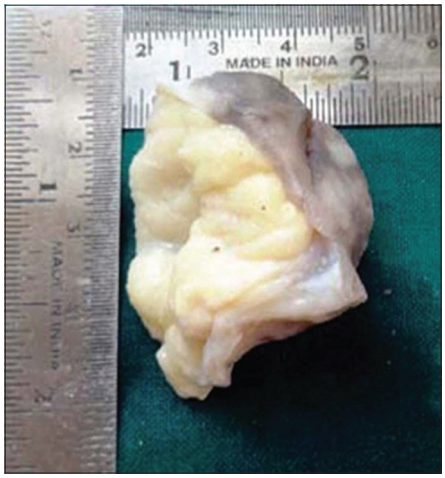

Figure 2: Gross specimen

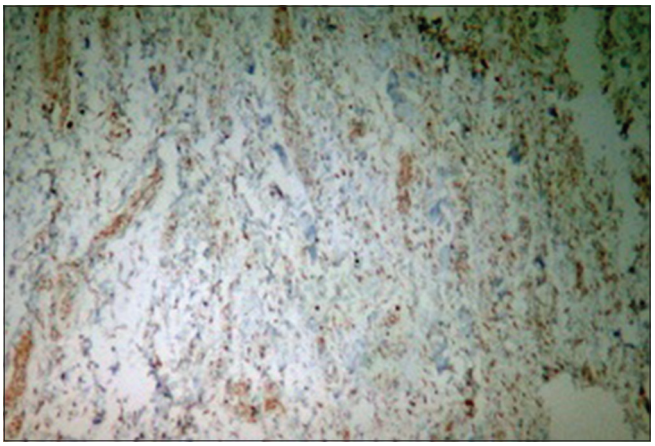

Figure 4: Scattered positivity for vimentin shown by collagenous stroma (×10)

case was 20 years, which is the longest duration reported till date. In 2010, Manjunatha *et al*.[5] have reported three cases of oral fibrolipoma with duration of 10 years. In this case, series no recurrence was noted in 6 months follow-up.

Data from the literature review showed no evidence of recurrence in any reports and the duration of follow-up ranged from minimum 10 months to maximum 5 years. The average follow-up period in the literature review was approximately 3 years.

Magnetic resonance imaging (MRI) and CT can aid in diagnosis and to determine the accurate extension of all kind of lipomas, especially for intramuscular and infiltrative giant lipomas. In MRI, these tumors have high signal intensity on T1-weighted images, with relative decreasing signal on T2-weighted images. A fat-suppressed MRI is particularly beneficial for diagnosis. On the other hand, fibrolipomas are more heterogeneous than lipomas on MRI images. On CT images, lipomas can be seen as an uncontrasted hypodense mass, as demonstrated in the image of the subject.[6] In the present case, accurate extension of mass was delineated by CT and no features of infiltration were found.

Histologically, classic lipomas are composed of mature adipose tissue with true lipoblasts showing no cellular atypia. Adipose tissues can be admixed with other mature benign mesenchymal tissues, thus necessitating sub-classification. Several variants of lipoma have been described, such as fibrolipoma, spindle cell lipoma, intramuscular or infiltrating lipoma, angiolipoma, salivary gland lipoma (sialolipoma), pleomorphic lipoma, myxoid and atypical lipoma.[7,8] Though adipocytes and benign and malignant fatty tumors stain positively for vimentin and S-100 protein. In the present case, immunohistochemical marker used was vimentin, which turn out to be negative. However, definitive fat cells were observed, that was indicative of adipocytic origin of the tumor. Fibrolipoma should be differentiated from spindle cell lipomas which are composed of mature lipocytes and uniform spindle cells in a mucinous and fibrous background. Normal white fat consists of spherical cells containing one large lipid vacuole that displaces the thin oval nucleus to one side. On routine sections, the nucleus of most fat cells is barely perceptible. From time to time, a section grazes an adipocyte nucleus such that it is viewed en face, displaying its characteristic central vacuole, termed Lochkern (German: Hole in the nucleus) nuclei. Lochkern are viewed more frequently in thick sections and therefore, are sometimes misinterpreted as evidence of lipoblastic differentiation and hence a liposarcoma. In the present case, we observed Lochkern cells [Figure 5].[1] In our case, the diagnosis of fibrolipoma was given based on the presence of mature adipocytes interspersed with dense collagen fiber bundles.

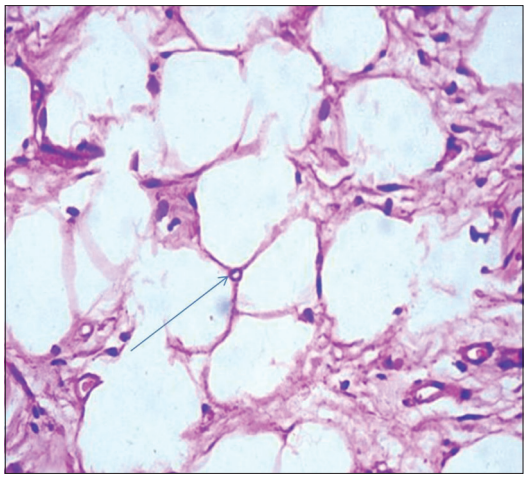

Figure 5: Histopathological section showing Lochkern cells (cells with vacuolated nucleus) (×40)

Fibrolipoma have been reported to show more proliferative activity than classic lipomas. Occasionally, fibrolipoma can be confused with herniated buccal pad of fat, but the characteristic well-circumscribed nature and lack of history of trauma will help in differentiating it.[7] The treatment of lipomas including fibrolipoma is usually surgical excision and the recurrence is rare. Capodiferro *et al*. in 2008,[9] treated a case of lip fibrolipoma by diode laser surgery and concluded that such treatment modality for benign lesions of the oral mucosa appears to be a convenient alternative to conventional blade surgery. In the present case, simple surgical excision of the tumor was done without any reported complication. A 6 month follow-up period was uneventful.

CONCLUSION

Fibrolipoma is a rare histologic variant of classic lipoma. It shows more proliferative activity than a classic lipoma. Though, lipomas not associated with any syndromes and have low recurrence rate, unwarranted increase in size of oral lipomas can lead to unexpected complications. Large oral lipomas of the floor of the mouth, as in the present case, can displace tongue posteriorly causing airway obstruction. Systematic documentation and early diagnosis is much essential for implementing correct treatment and to prevent further complications in the future.

REFERENCES

1. Weiss SW, Goldblum JR, editors. Benign lipomatous tumors. In: Enzinger and Weiss's Soft Tissue Tumors. 6th ed. St. Louis: Mosby; 2013. p. 445-96.
2. Rydholm A, Berg NO. Size, site and clinical incidence of lipoma. Factors in the differential diagnosis of lipoma and sarcoma. Acta Orthop Scand 1983;54:929-34.
3. Juliasse LE, Nonaka CF, Pinto LP, Freitas Rde A, Miguel MC. Lipomas of the oral cavity: Clinical and histopathologic study

of 41 cases in a Brazilian population. Eur Arch Otorhinolaryngol 2010;267:459-65.
4. Fregnani ER, Pires FR, Falzoni R, Lopes MA, Vargas PA. Lipomas of the oral cavity: Clinical findings, histological classification and proliferative activity of 46 cases. Int J Oral Maxillofac Surg 2003;32:49-53.
5. Manjunatha BS, Pateel GS, Shah V. Oral fibrolipoma-A rare histological entity: Report of 3 cases and review of literature. J Dent (Tehran) 2010;7:226-31.
6. Cappabianca S, Colella G, Pezzullo MG, Russo A, Iaselli F, Brunese L, *et al*. Lipomatous lesions of the head and neck region: Imaging findings in comparison with histological type. Radiol Med 2008;113:758-70.
7. Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 2nd ed. USA, Elsevier Publication; 2004.
8. Gujjari SK, Shah M, Hegde U, Doddawad VG. Fibrolipoma: Report of two intraoral cases. J Clin Diagn Res 2012;6:524-6.
9. Capodiferro S, Maiorano E, Scarpelli F, Favia G. Fibrolipoma of the lip treated by diode laser surgery: A case report. J Med Case Rep 2008;2:301.

Cite this article as: Rajeev R, Beena VT, Indu G, Choudhary K, Devu A. Fibrolipoma of floor of the mouth of 20 years of duration. Clin Cancer Investig J 2014;3:394-7.

Source of Support: Nil, **Conflict of Interest:** None declared.