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Research Paper

Intramuscular Lipoma of the Maxillofacial Region: A Case Report and Review of Literature

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ABSTRACT
Lipomas are benign tumours originating from mature adipocytes that rarely occur in the oral and maxillofacial region. It constitutes of about 1 to 4% of all benign neoplasms and 50% of all soft tissue tumours.They commonly occur in the buccal mucosa and cheek in the oral and maxillofacial region respectively.
A 60-year-old man reported with a chief complaint of swelling on the left side of the face since one year. The swelling was initially small in size, which exhibited a slow gradual continuous enlargement to its present size. On clinical examination, the swelling was non-tender with no rise in temperature of the overlying skin. The swelling was soft in consistency, compressible and non-fluctuant with well-defined margins. On the basis of clinical presentation and location of the lesion, differential diagnosis oflipoma, epidermoid cyst and minor salivary gland tumour were considered. The tumour was excised and histopathological examination showed it to be a classic soft tissue lipoma. This case of facial lipoma occurring in the lower third of the face is reported for its rarity and can be considered as a possibility in the differential diagnosis of various swellings of the maxillofacial region.

KEYWORDS: ***Facial lipoma, Soft tissue neoplasm***

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I. INTRODUCTION

Lipomas are benign, slow growing, mesenchymal tumours that develop from mature adipocytes.[1]Due to its wide distribution in the human body, it is known as Universal Tumour.[2]These lesions are the most common soft tissue tumour, making up to 50% of all soft tissue tumours.[3]They usually affect the region of the trunk, shoulders, neck and axilla[2] with rate of occurrence of 15-20% in head and neck region[4] and infrequent rate of occurrence of 1%-4%in the oral and maxillofacial region.Oral lipomas are rare comprising only 2.2% of all lipomas and 2.4% of all benign tumours of oral cavity.[5]Most commonly Lipomas occur in 4th and 5th decade of life[2]with no gender predilection and are less commonly seen in children and teens.[6]

Most lipomasclinically present asa slow- growing, painless, soft and well-circumscribed lesion usually associated with submucosal nodules with either a sessile or a pedunculated base.[7] Most cases are clinically diagnosable but in cases where imaging is required,ultrasonography (USG),Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) are the preferred diagnostic modalities.[3] Despite the availability of all these techniques, histopathology remains the gold standard for diagnosis of lipomas.[7]

II. CASE REPORT

A 60-year-old man reported with a chief complaint of swelling on the left side of the face since one year. It was a small asymptomatic swelling which exhibited a slow gradual continuous enlargement to its present size. Patient gave a medical history of increase in serum cholesterol level since 5years and was under medication for the same. His dental and personal history were non-contributory. No associatedsecondary changes like discharge, bleeding or ulcerations werenoted during the course of progression. Patient also did not exhibit any sensory changes associated with the swelling.

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On extra-oral examination, a single, localized, elliptical, lobulated, subdermal nodule of approximately 4*3cm was noted in the lowerone-third of the face extending from the level of left commissure of the mouth to the inferior border of mandible superio-inferiorly.No change in color of the skin over the swelling was noted. [Figure:1]

On palpation, there was no rise in temperature of the skin over the swelling and the swelling was non-tender.It had a smooth surface with normal overlying mucosa. The swelling was soft in consistency, compressible and non-fluctuant with well-defined margins.Slip-sign was positive indicating the swelling was mobile in all planes.[Figure 2] Submandibular, submental and cervical chain of lymph node were non palpable.On intraoral examination, no swelling was noted.No change in colour of the oral mucosa was noted either.

On the basis of clinical presentation and location of the lesion, the differential diagnosis oflipoma, epidermoid cyst and minor salivary gland tumour were considered.

Patient's blood investigation revealed no abnormalities.Fine needle aspiration was attempted twice which yielded negative aspiration. Orthopantomography revealed no bony involvement. Ultrasonography did not reveal any muscle involvement and bony cortices appeared normal. A well-defined echogenic lesion was noted in the intramuscular plane at the site of swelling measuring 2.2×0.8cm in the region of left side of mandible There was no evidence of posterior acoustic shadowing or enhancement. No calcification or vascularity noted within.

Thus, upon clinico-radiographic evaluation, a most probable diagnosis of Lipoma was considered. However, confirmation was possible upon histological examination.

Epidermoid cysts are benign encapsulated, subepidermal nodule filled with keratin material and are the most common cutaneous cyst usually seen on face, neck and trunk region.[8] As fine needle aspiration did not reveal any positive aspirate, this diagnosis was ruled out. Whereas minor salivary gland tumors most commonly occur in the oral cavity in the head and neck region with palate being the most frequent site.[9] On USG, they usually appear as well defined, homogenous, hypoechoic mass.[10]

The lesion was excised completely from an intra-oral approach to avoid extra-oral scar [figure 3] and was sent for histopathological examination.

Haematoxylin and eosin-stained section showed a thinly encapsulated mass consisting of lobules of adipocytes divided by connective tissue septae. Mature lipocytes were present with centrally positioned large lipid vacuole and peripherally placed cytoplasm and nucleus giving a definite diagnosis of simple lipoma. [Figure4] One year of follow-up of the patient post-surgery revealed no recurrence. [Figure 5]

III. DISCUSSION

Benign lipomas are the most common soft tissue mesenchymal neoplasms, with 15% to 20% of the lesion occurring in the head and neck region, but only 1% to 4% of the patients in the maxillofacial region.[11]Lipomas are usually solitary, but can be multiple in 5%–15% of cases.[12]The first description of soft tissue lipoma was given in 1848 by Roux as a "yellowish epulis". The incidence of intraosseous lipomais very low, about 0.1% and infrequently seen in the maxillofacial region.[13]

Etiopathogenesis

The exact aetiology is still unclear however, according to a systematic review, there are two major theories responsible for the formation of lipoma. This includes, the "Hypertrophy theory", which states that obesity and inadvertent growth of adipose tissue may contribute to their formation. This theory was not well accepted as it couldn't explain the lesions occurring in areas with inadequate pre-existing adipose tissues. Also, during periods of starvation, they were not used up by the metabolic process as were the normal body fats. And the "Metaplasia theory" which suggests that the lipoma formation occurs due to atypical differentiation of mesenchymal cells in lipoblasts. [14]

Cytogenetics

Various studies have shown a genetic link wherein chromosomal abnormalities are seen in two-third of the cases.[15]Most of the ordinary lipomas (60%) have chromosomal aberrations. The most frequent site for aberration includes 12q13–15 region followed by 6p21–23 and 13q resulting in expression of truncated HMGA2 protein or a fusion protein. Chromosomal rearrangements usually occur in these regions through translocation and deletion.[16,17]

Other factors

Another theory suggests a possibility of trauma causing lipoma formation.[18]Some studies also reveal other contributing factors like, hypercholesterolemia, endocrine disorders, hormonal imbalance, inflammation, mucosal infections, radiation, for the development of oral lipoma.[19,7]

Clinical characteristics

Lipomas occur especially in areas of fat accumulation, and the most common maxillofacial site of occurrence is cheeks, followed by the tongue, the floor of the mouth, buccal sulcus and vestibule, lip, palate, and gingiva.[20]In a study of 125 cases of lipoma in the oral and maxillofacial region, it was found that the maximum number of lipomas were exhibited in the parotid region (n=30), followed by oral mucosa. (n=29).[21]They usually present as a soft non-tender mobile sessile mass which gradually enlarges in size, covered by normal mucosa within a course of less than 3yrs in 70% of the cases. The average size of the lesion ranges from 1.5-2.5cm.[21,22,23]The present case report also had a similar presentation of lipoma with an unusual site of occurrence atthe lower-third of the face close to the left corner of the lip.

Considering the age and gender of the patients involved, majority of the studies showed no sex predilection.[20,23,24] However few studies suggested a male predilection for localized cases of maxillofacial lipoma.[22,25] Usually adults of 5^{th}-7^{th} decade of life[6,23] are affected by lipomas however studies showing lipomas in young adult,[21,25-27]andpaediatric patient[21,28] as well as congenital lipoma have been reported.Ours was a male adult patient of 60years of age which is a common age of occurrence of such lesion.

Morphologically, intraoral lipomas are classified by Rajendra et al in 3types, diffuse form, superficial form and encapsulated form. Diffuse form usually affects the deeper tissues and are soft of almost fluid filled consistency and hence can be misdiagnosed as a cyst.[29] Intramuscular lipoma have been recognized with variant presentations of infiltrative type, well-defined/non-infiltrative and mixed variant.[30] The present report presented an intramuscular, encapsulated and non-infiltrative form of facial lipoma.

Diagnostic characteristics

Ultrasonography

The lesions usually appear as well-defined, echogenic mass without posterior acoustic enhancement in USGwhich was similar to the present report. Deeper lipomas may however appear isoechoic or hyperechoic to the adjacent muscle and may have posterior acoustic enhancement. [3]

To determine the accuracy of ultrasonography in the diagnosis of soft tissue lipoma, Prasuna et al (2004) found a wide range of echogenicity with most common being isoechoic (28-60%) or hyperechoic (20-52%) with wide inter-reader variability. They also tend to display other ultrasound features, such as no acoustic shadowing, no or minimal color Doppler flow.[31]

CT-scan

On a non-contrast CT scan, Lipomas appear as circumscribed, homogenous hypointense mass of -120 to -65 H.U.[3]Thin and thick soft tissue density streaks are usually appreciated. CT Scan is the preferred diagnostic tool in cases of ossification.[32]

MRI Imaging

On MRI sequences,lipomas resemble subcutaneous fat, but may also contain a few thin septa measuring less than 2 mm in thickness. Enhancement of these thin septae occurs after administration of contrast material which are better appreciated through MRI imaging than CT scan. In general, marked enhancement of these septae and nonfatty elements are seen in well-differentiated liposarcomas which could be the differentiating factor. In cases of Lipomatosis, MR imaging shows infiltration of unencapsulated fat in subcutaneous or deep soft tissue, with characteristic T1- and T2-hyperintense signal that suppresses at fat-suppressed sequences . Mineralization is a complicating feature of some lipomas which is present in up to 11% of imaged lesions. The lipomas may also undergo necrosis or atrophy. Lipomas are mostly benign without risk of malignant transformation. In cases where there is atypical appearance on imaging, biopsy should be considered in order to rule out liposarcoma. [17]

To evaluate the reliability of MRI in distinguishing lipoma from other variants the sensitivity and specificity was found to be 100% in simple cases of lipoma with lowered specificity in cases of atypical lipoma.[33]

Histopathology

Lipomas show no evidence of cellular atypia and are subdivided into lobules by septae of fibrous connective tissue, sometimes associated with other mesenchymal elements andusually circumscribed by a thin capsule. They can be classified as simple lipomas, fibrolipomas, spindle cell lipomas, intramuscular or infiltrating lipomas, angiolipomas, salivary gland lipomas, pleomorphic lipomas, myxoid lipomas, and atypical

lipomas. [11] Simple lipoma(non-infiltrating type), which is also found in the present study, is the most common diagnosis in the literature.[22,23,25]

Management

Well-encapsulated lipomas usually involve conservative surgical removal with minimum possibility of recurrence. Aesthetic approaches are incorporated in the surgical removal facial lipoma.[6]In the present patient, complete excision was made intra-orally, through a blunt dissection due to the thin capsule of the lesion and no signs of recurrence were observed.

IV. CONCLUSION

Lipoma in the maxillofacial region is quite infrequent. It should be differentiated microscopically and radiographically from its other variants. Complete surgical excision is the treatment of choice. In case of surgical excision, intra-oral approach should be considered keeping in mind the patients aesthetics. In this case the lesion was surgically excised via intra-oral approach and there has been no recurrence after a follow up of one year.

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Figures

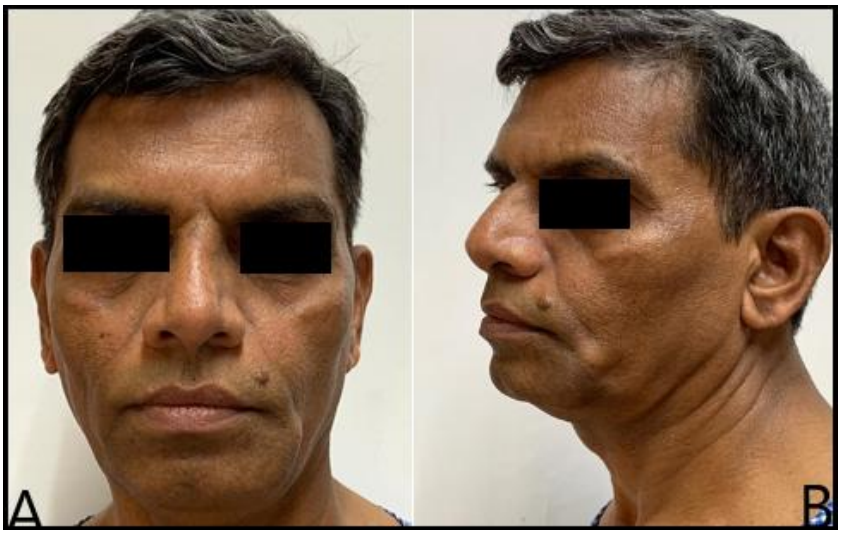

Figure 1: Frontal(A) and lateral view(B) of face showing location and extent of the lesion

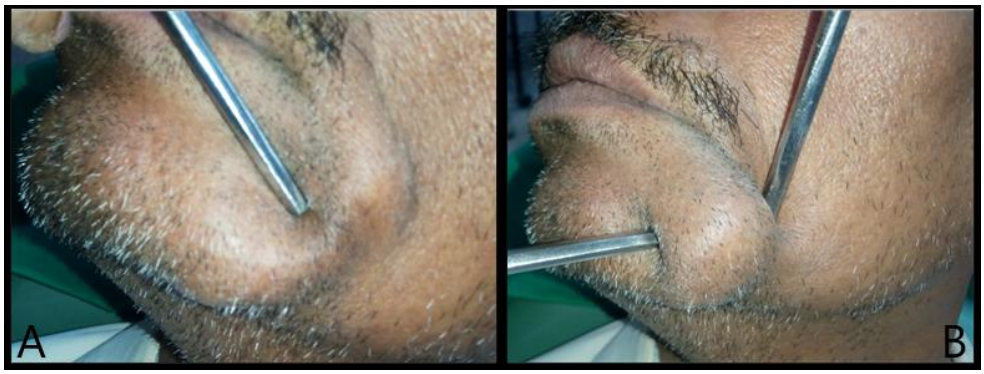

Figure 2: Palpatory finding of the lesion: showing its compressibility(A) and mobility(B)

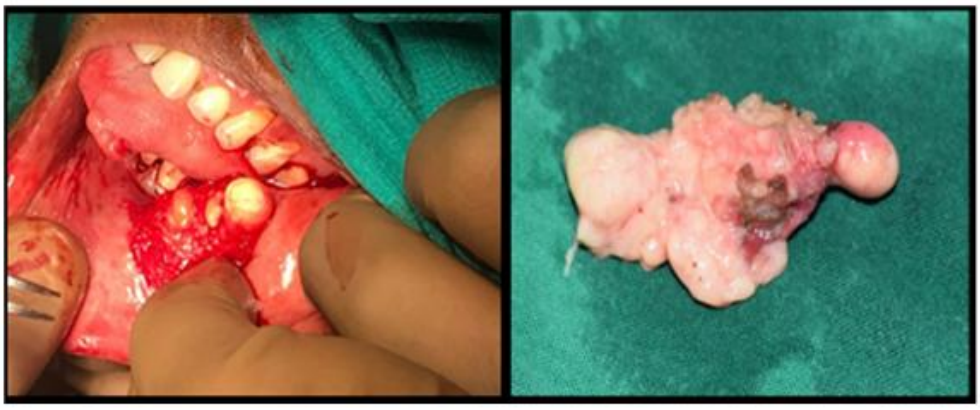

Figure 3: completely excised lesion from an intra-oral approach

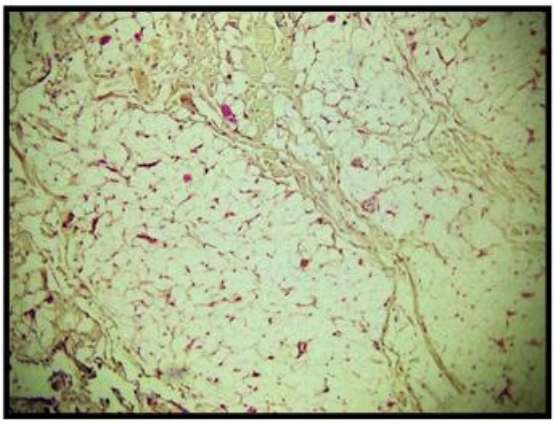

Figure 4: Microphotograph of H & E-stained section showing lobules of mature adipocytes interspersed by connective tissue septae

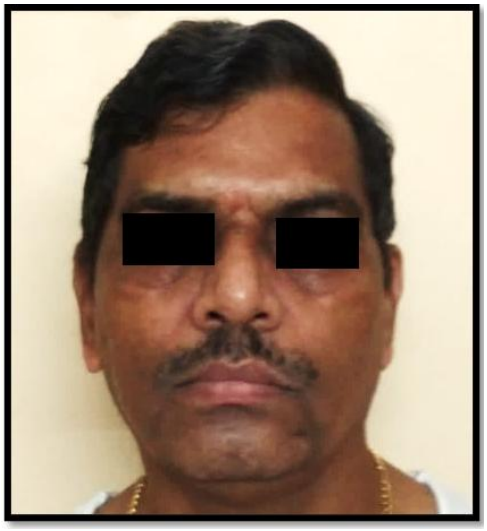

Figure 5: Follow-up frontal view of face post-surgery.