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Case Report

A rare case of sialolipoma of submandibular gland

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ABSTRACT

Lipomas in head and neck region are common but prevalence in salivary glands is rare with majority in parotid. Sialolipoma is a rare tumour of the salivary gland that is composed of mature adipocytes and normal salivary gland tissue. We report an extremely rare case of sialolipoma of left submandibular gland in a 39-year-old male who presented with complaints of insidious onset gradually progressive swelling in left submandibular region with no pain, discolouration or discharge. On examination, a 5×4 cm soft, non-tender swelling was noticed in (L) submandibular region. Ultrasonography of neck revealed enlarged (L) submandibular gland with hypoechoic area of 5 mm diameter and small areas of echogenic foci with no cervical lymphadenopathy. Contrast enhanced computed tomography (CECT) neck showed a 5.6×2.7×4 cm well defined fat attenuated lesion in (L) submandibular space, suggestive of lipoma with a well-defined enhancing soft tissue component within the lesion. He underwent excision of (L) submandibular gland along with fibrofatty tissue under general anaesthesia. Histopathological examination revealed submandibular gland with normal histomorphological features along with mature adipocytes in fibrofatty tissue. The patient is on regular follow-up with no recurrence.

Keywords: Sialolipoma, Submandibular gland, Salivary gland, Lipoma

INTRODUCTION

Of all the benign mesenchymal origin neoplasms, lipomas are the commonest. They usually arise in locations where fat is normally present. Only 13% of lipomas are present in head and neck region of which the posterior neck space is the most common site.[1] Fat-containing tumours of the salivary glands are uncommon with less than 0.5% of all parotid tumours.[2] These have a wide histological spectrum varying from pure lipomatous neoplasms to mixed lipo-epithelial lesions.[3] Nagao et al became the first one to propose the term "sialolipoma" in 2001, for a variety of lipoma of salivary gland consisting of mature adipocytes and normal salivary gland tissue.[4]

Here we report an extremely rare case of sialolipoma of left submandibular gland in a 39-year-old male who underwent surgical excision for the tumour.

CASE REPORT

A 39 years old male with no comorbidities presented with complaints of swelling in left submandibular region for past 3 years which was insidious in onset and gradually progressed in size. There was no pain, redness or discharge associated with the swelling. There was no change in size of swelling in relation to blowing air, exertion or eating food. There was no other neck or facial swelling noticed by the patient.

On clinical examination, 5×4 cm soft, non-tender swelling was noticed in (L) submandibular region which was non-ballotable and skin over the swelling was normal (Figure 1). Ultrasonography of neck revealed enlarged (L) submandibular gland (20.9 mm in largest diameter) with hypoechoic area of 5 mm diameter and small areas of echogenic foci. There was no cervical lymphadenopathy or swelling noticed. Fine needle aspiration cytology from

the swelling showed occasional cluster of acinar cells with haemmorhagic background. Contrast enhanced computed tomography (CECT) neck showed a 5.6×2.7×4 cm (AP×TR×CC) well defined fatty attenuated lesion epicentred in (L) submandibular space at the level of angle of mandible, suggestive of lipoma (Figures 2a and b). There was a well-defined enhancing soft tissue component measuring 17×7×16 mm (AP×TR×CC) noted within the lesion along the platysma.

Figure 1: Submandibular region swelling (arrow).

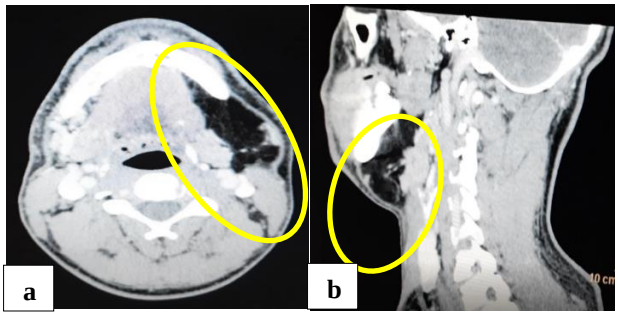

Figure 2: (a) and (b) CECT neck reveals lesion in (L) submandibular region as described (marked in yellow).

The patient underwent excision of (L) submandibular gland along with fibrofatty tissue under general anaesthesia on 26 February 2021 (Figures 3 and 4). Intraoperatively, fibrofatty tissue along with submandibular gland was seen at level Ia and Ib, which were excised and sent for histopathological examination.

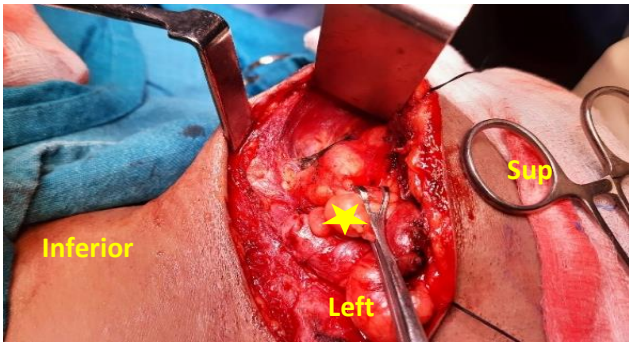

Figure 3: Intraoperative image of (L) submandibular gland with fibrofatty tissue (star marked).

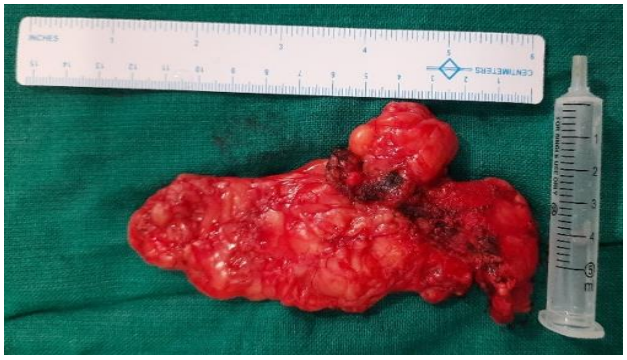

Figure 4: Surgically excised specimen.

Histopathological examination revealed submandibular gland with normal histomorphological features along with mature adipocytes and fibrofatty tissue suggestive of sialolipoma (Figure 5). The patient was discharged after an uneventful hospital stay and is on regular follow-up with no recurrent swelling.

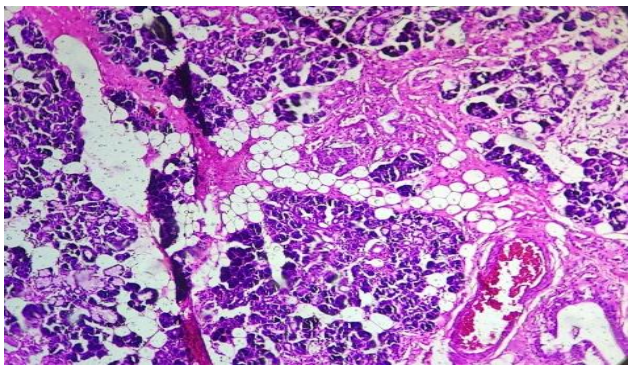

Figure 5: HPE – mature adipocytes interspersed with glandular morphology.

DISCUSSION

Lipoma is the most common benign soft tissue neoplasm of mesenchymal origin in adults. These tumours commonly occur in the head and neck area. Although uncommon, lipomas are found in the oral cavity, with a reported incidence of only 1% to 4.4% of all benign oral lesions.[5,6] Fat-containing tumors of the salivary glands are uncommon. They comprise of less than 0.5% of all parotid tumours.[2] Their wide histological spectrum varies from pure lipomatous neoplasms similar to their cutaneous and soft tissue counterparts to mixed lipoepithelial lesions specific to the salivary glands. With few exceptions, these uncommon lesions affect mainly the elderly, with a mean age at presentation of >50 years and show a predilection for males.[3] Sialolipoma was firstly described in 2001 by Nagao et al. It was included in the 2005 World Health Organization (WHO) classification of head and neck tumors. Sialolipomas have been reported under diverse names in the English literature. This neoplasm is characterized by a well-defined and encapsulated lipomatous tumor with entrapped normal salivary gland tissue morphologically similar to normal salivary gland

parenchyma. Sialolipoma can arise in either major or minor salivary glands, and the parotid gland is the most common location to occur.[4]

Clinical symptoms of sialolipoma depend on the size, location and rate of growth of the lesion. Lipoma usually presents as a painless, well-circumscribed mass with gradually increase in size over time. Possible aetiological factors include heredity, obesity, diabetes, trauma, radiation, endocrine disorder, insulin injection and corticosteroid therapy. Some associated syndromes include hereditary multiple lipomatosis, adiposis dolorosa, Gardner's syndrome and Madelung's disease.[1] Trauma is one of the causes of lipoma with several cases of lipoma in salivary glands following trauma been reported in various studies. The protection of submandibular gland by mandible may be a possible explanation to account for less incidence in submandibular gland as compared to parotid gland.[7] In our case, the patient was a 39-year-old soldier with no comorbidity but had taken part in many sport activities involving trauma to face and neck like boxing and karate, so trauma as the cause of sialolipoma cannot be ruled out. The swelling had progressed gradually over 3 years without any pain which corresponds to the above description of lipoma.

Clinical examination is often insufficient for identifying exact origin, nature and extent of the mass, and usually an imaging modality is essential, especially if tumour is deep seated. In ultrasonography, they appear as an elliptical mass parallel to skin surface with echogenicity as hyperechoic in relation to adjacent muscle.[8] Our patient's ultrasound neck had showed enlarged (L) submandibular gland with hypoechoic area of 5 mm diameter and small areas of echogenic foci. There was no increased vascularity or calcification within the lesion and no cervical lymphadenopathy was present. Lipomas have a consistent appearance on computed tomography (CT) images, with specific CT Hounsefield Units (HU) (134 to 83 HU) values and displacement of the surrounding soft tissues. With MRI, the lesions demonstrated a high signal intensity similar to that of subcutaneous fat.[9] In the present case CECT was done which showed 5.6×2.7×4 cm (AP×TR×CC) well defined fatty (average HU 98) attenuated lesion epicentred in (L) submandibular space at the level of angle of mandible, suggestive of lipoma.

Imaging studies do not distinguish sialolipoma from conventional lipoma and other variants. Although fine needle aspiration cytology may be useful for diagnosis of lipomatous tumours of salivary gland, the definitive diagnosis is based on histopathological features. Sialolipoma is a non-oncocytic tumour that displays prominent lobulation and is predominantly composed of mature adipose tissue admixed with evenly distributed normal salivary tissue elements. The fatty component predominates over the epithelial elements (20–90%).[3,10] In our case, histopathological examination showed mature adipocytes along with serous and mucinous acini with normal lobular and ductal architecture and mild patchy lymphoplasmacytic infiltrate around few ducts, features suggestive of sialolipoma.

The treatment of lipomatous masses involves complete surgical resection of the tumour. Submandibular gland resection can be performed considering the location of the tumor and the needs of the patient, who did not want to undergo a possible reoperation. There are no reported cases of recurrent sialolipoma in previous studies.[7] In the present case, complete surgical removal of the submandibular gland along with fibrofatty tissue was performed. Patient who is on regular follow-up for past 1 year hasn't shown any symptoms or signs of recurrence.

CONCLUSION

Sialolipomas are one of the rarest types of salivary gland neoplasms with multiple variants. Imaging modalities like ultrasonography along with fine needle aspiration cytology, CECT can lead to suspicion but definitive diagnosis of these type of tumours can only be done by histopathological examination due to presence of different variants. Surgical excision remains the main modality for treatment and there are no reported recurrences.

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REFERENCES

1. Kim KS, Yang HS. Unusual locations of lipoma: Differential diagnosis of head and neck mass. Aust Fam Physician. 2014;43(12):867-70.
2. Ruangritchankul K, Connor S, Oakley R. Oncocytic Sialolipoma of Parotid Gland: Case Report and Literature Review. Head Neck Pathol. 2019;13(4):548-53.
3. Agaimy A. Fat-Containing Salivary Gland Tumors: A Review. Head Neck Pathol. 2013;7(1):90-6.
4. Nagao T, Sugano I, Ishida Y, Asoh A, Munakata S, Yamazaki K, et al. Sialolipoma: A report of seven cases of a new variant of salivary gland lipoma. Histopathology. 2001;38(1):30-6.
5. Sato K, Gotoh C, Uchida H, Kawashima H, Yoshida M, Kitano Y, et al. Sialolipoma of the submandibular gland in a child. J Pediatr Surg. 2011;46(2):408-10.
6. 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(1):49-53.
7. Lee YW, Chung J. Intraglandular Ordinary Lipoma of the Submandibular Gland. Ear, Nose Throat J. 2021;100(5):587S-9S.
8. Ahuja AT, King AD, Kew J, King W, Metreweli C. Head and neck lipomas: sonographic appearance. AJNR Am J Neuroradiol. 1998;19(3):505-8.
9. Chikui T, Yonetsu K, Yoshiura K, Miwa K, Kanda S, Ozeki S, et al. Imaging findings of lipomas in the

orofacial region with CT, US, and MRI. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1997;84(1):88-95.
10. Ahn D, Park TI, Park J, Heo SJ. Oncocytic sialolipoma of the submandibular gland. Clin Exp Otorhinolaryngol. 2014;7(2):149-52.

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