CASE REPORT

Interstitial lipomatosis of minor salivary gland: Fight for a place with sialolipoma

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Abstract

The aim of this article is to report an uncommon lipomatous minor salivary gland (MSG) lesion of the upper lip that was diagnosed with pathological insight and clinical correlation. Differential diagnosis of fat containing salivary gland (SG) tumors will also be discussed. MSG lesions constitute a diverse group encompassing non-neoplastic lesions, benign neoplasms, and malignant neoplasms of varying grades. A rare subset among these includes the lipomatous SG lesions. Interstitial lipomatosis (IL) is an overgrowth of adipose tissue throughout the SG, that should be histologically differentiated from sialolipoma and infiltrating lipoma, which is neoplasms. The former is associated with systemic conditions while the latter is not. This case is of IL in a 55-year-old woman with hypothyroidism. A search in English literature did not reveal any cases of IL of the upper lip. The fact that SG neoplasms are more common in the upper lip than non-neoplastic lesions adds to the rarity of this case. Thus, this report adds another entity in the differential diagnosis of upper lip lesions and underlines the need to differentiate it from lipomatous neoplasms.

Keywords

Hypothyroidism, interstitial lipomatosis, minor salivary gland, sialolipoma

Introduction

Minor salivary glands (MSG) are small, chiefly mucous-secreting glands located in nearly all areas of the oral cavity. Diseases of the MSG encompass a diverse set of non-neoplastic lesions, as well as benign and malignant neoplasms of varying grades. MSG neoplasms constitute 10-15% of all salivary gland (SG) neoplasms, with MSG being second most commonly involved after the parotid gland. Palate is the preferred site followed by the lip, with a higher incidence in the upper lip than the lower. However, literature regarding the incidence of non-neoplastic MSG lesions is scarce.

Both neoplastic and tumor-like lesions of SG (including MSG) may contain adipose tissue. The latter may be a minor component of the lesion or may be entirely composed of adipose tissue. Among these, interstitial lipomatosis (IL) is an overgrowth of adipose tissue throughout the SG and often associated with systemic factors and conditions, that should be histologically differentiated from sialolipoma, a neoplasm.

A rare case of IL of the upper lip in a hypothyroid patient is presented here, that required pathological insight and clinical correlation for diagnosis. An internet search in English literature did not reveal any cases reported of IL of the upper lip, thereby adding to the rarity of the case.

Case Report

A 55-year-old woman reported with a complaint of swelling in inner part of her upper lip since 10 days, which caused her 'discomfort during eating and speaking'. On clinical examination, a diffuse, solitary “pea-sized” swelling in greatest dimension was observed, but a discrete mass could not be felt on palpation. Overlying mucosa appeared normal. There was no pain or discharge from the swelling. There was no history of trauma recalled, however a fractured 21 was observed adjacent to the swelling. Medical history, revealed a history of hypothyroidism since 4 years, for which she was on regular medication. Dryness of eyes and mouth were absent. A provisional diagnosis of mucocele of upper lip was considered, and an excisional biopsy was performed. Intraoperatively, no discrete mass could be found, and hence the lesion was difficult to excise. According to the surgeons, the mass appeared more localized after injection of the local anesthetic [Figure 1a]. We received multiple, irregular whitish pink bits of soft tissue that lacked a capsule [Figure 1b].

Microscopic examination revealed clusters of SG tissue interposed between mature adipose tissue. The SGs were normal-appearing and included mucous acini and ducts. Lobules were
intact in many of the clusters and were distributed throughout the sections. The ducts were dilated and some of them showed oncocytic change. An inflammatory infiltrate of lymphocytes admixed with few plasma cells and mast cells were noted in relation to the glands. Mature adipose tissue constituted about 40% of the lesion and was present in between the clusters of SG elements, and in few places, it appeared to be replacing the SG parenchyma. There was no atypical in the adipocytes. Numerous dilated and some engorged blood vessels were scattered throughout the specimens. Focal areas of fibrosis were seen [Figures 2-5]. A diagnosis of IL (fatty infiltration) of MSG of the upper labial mucosa was given on clinical correlation.

**Discussion**

The labial mucosa contains adipose tissue and MSG along with other general connective tissue components. Hence, both lipomatous proliferations and MSG diseases are expected to occur in this region. Many lipomas have been reported in the lips. Nonetheless, lipomatous conditions of MSG of the labial mucosa are extremely rare.

Fat-comprising SG tumors and tumor-like conditions have been classified and reviewed by Agaimy. They include (1) SG neoplasms with lipometaplasia (pleomorphic adenoma), (2) pure lipomatous neoplasms (lipoma and its variants), (3) mixed SG and lipomatous neoplasms (lipoadenoma, sialolipoma, and polycystic lipoadenomatous lesions), and (4) non-neoplastic lesions (traumatic pseudolipoma, IL, and lipomatous atrophy). Our case was not a distinct mass clinically and histologically, thus excluding most entities in the first three categories.

To elucidate further, we did not see features of pleomorphic adenoma thus eliminating the first category. In the second category, classic lipomas and most lipoma variants (except infiltrating lipoma and infiltrating angiolipoma) were discounted because of their strict circumscription within the SG tissue. Few SG elements may be observed at the periphery, but evenly distributed SG that we found in our case are definitely absent in lipomas. Infiltrating lipoma occurs within (intramuscular lipoma) or in between muscles (intermuscular lipoma) and exhibits adipose tissue and blood vessels permeating between skeletal muscle fibers and other surrounding tissues.

Even though the present case had numerous blood vessels and was not circumscribed, the absence of skeletal muscle...
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Involvement excluded this diagnosis. A case of infiltrating lipoma entrapping residual SG tissue has been described by Ponniah et al.[2] Infiltrating angiolipoma is another tumor that infiltrates skeletal muscles and characterized by a network of branching blood vessels and fibrin microthrombi.[13,15] One such case involving the upper labial mucosa has been reported by Dalambiras et al.[15]

In the third category of mixed SG and adipose tissue tumors, lipoadenoma was not considered since this entity is circumscribed and acinar/myoepithelial cells are absent.[2,16] Similarly, lipomatous polycystic lesions were not included in the differential diagnosis due to the absence of microcysts in our case.[2] Evenly distributed SG elements amid mature adipose tissue similar to our case is also seen in sialolipoma. Other histologic features include encapsulation, ductal dilatation, periductal fibrosis, lymphocytic infiltration, dilated and congested blood vessels, gland atrophy, oncocytic and squamous metaplasia of ducts, myxoid changes and peripheral nerve entrapment.[2,7,12,13-19] Nagao et al., 2001, were the first to describe sialolipoma as a separate entity.[17] It mainly affects the parotid gland followed by the MSG.[7,16,17] Qayyum et al. have detailed the differences between sialolipomas of major and minor SG.[18] All the above histologic features were found in our case except for encapsulation. The characteristic feature of sialolipoma is encapsulation, with rare exceptions. Even in the cases reported without capsule, it was clinically a discrete mass and well circumscribed histologically.[16]

A non-neoplastic SG condition that imitates sialolipoma is interstitial/diffuse lipomatosis. Here, there is diffuse enlargement of the gland due to deposition of adipose tissue throughout the gland. It does not form a distinct mass clinically and does not have a capsule.[7,19] These two features differentiate IL from sialolipoma,[7,8,12,16,17,19] and hence our case was diagnosed as IL. This condition is uncommon and chiefly affects the major SG (primarily the parotid). Involvement of MSG is rare.[7] The first case of IL of MSG of nasal cavity was reported by Saleh et al. in 1998.[1] An internet search in English literature did not reveal any other case of IL in MSG till date. Causes of IL include diabetes mellitus, cirrhosis, chronic alcoholism, malnutrition, and hormonal disturbances.[1,4] Our patient had hypothyroidism since 4 years. It is difficult to establish whether hypothyroidism resulted in this fatty infiltration of MSG, due to lack of literature. Saleh et al. suggested that etiology of IL of MSG was different from that of parotid lipomatosis.[1] Other non-neoplastic SG diseases such as traumatic pseudolipoma and lipomatous atrophy were not considered due to lack of history of trauma and absence of prominent glandular atrophy, respectively.[2,7]

This case showed ill-defined deposition of fat throughout labial MSG as described in literature. Furthermore, other histologic features such as ductal dilatation, lymphocytic infiltration and many dilated blood vessels, oncocytic metaplasia of ducts were observed. The blood vessels are attributable to normal histology of labial mucosa and oncocytic metaplasia could be an age change. The numerous lymphocytes may have

Figure 5: Oncocytic metaplasia of ducts. (Haematoxylin and eosin stain, ×20)

Flowchart 1: Clues for diagnosis of lesions with mixed salivary gland and lipomatous tissue
infiltrated the glands in response to irritation of the labial mucosa by fractured 21.

The salient features of mixed SG and lipomatous lesions have been summarized in Flowchart 1.

Conclusion

To the best of our knowledge, this is the first case report of IL of labial MSG, in a hypothyroid patient. IL has to be differentiated from sialolipoma by lack of a distinct mass and encapsulation.

Clinical significance

IL is a rare entity that may be included in the differential diagnosis of diffuse enlargement of the labial mucosa and needs to be distinguished from neoplasms. Fatty infiltration of MSG may ensue hormonal disturbances, similar to major SG.

References
