Sialosis: Cytomorphological significance in the diagnosis of an uncommon entity

ABSTRACT
A diffuse, chronic, usually bilateral, noninflammatory, nonneoplastic enlargement of major salivary glands is termed as Sialosis or Sialadenosis. It is an extremely uncommon cause for enlargement of the parotid gland. We hereby present a case of a 45-year-old female patient having a swelling at the left preauricular region. The swelling was gradually increasing in size since 6 months. On clinical examination, the swelling was 3 cm × 3 cm, mobile, and nontender. On ultrasonography, it was suggestive of benign parotid lesion or parotitis with cervical lymphadenopathy. On fine needle aspiration cytology, it was suggestive of sialadenosis. This is an extremely rare salivary gland lesion with specific cellular features. It is very important to distinguish sialadenosis from other causes of enlargement of the parotid gland as treatment modality differs.

Key words: Cytology; parotid gland swelling; sialadenosis

Introduction
The first important discussion regarding sialosis in dental reports was mentioned 35 years ago and since then little has been published. Sialadenosis refers to noninflammatory, often recurrent enlargement of the salivary glands, most frequently, the parotids. It is usually associated with various underlying disorders that include diabetes, alcoholism, malnutrition, anorexia nervosa, bulimia, etc. The management of sialadenosis depends upon identification of the underlying cause that must then be corrected. Thus, the unnecessary surgical intervention in such diagnosed cases can be prevented.

Case Report
A 45-year-old female came with a swelling at the left preauricular region measuring 3 cm × 3 cm. The swelling was solitary, mobile, and nontender. It was gradually enlarging in size over a period of 6 months. The overlying skin was normal. Clinically it was suggestive of parotid inflammation or parotid neoplasm. The right parotid gland was unremarkable. On investigation, the patient had hyperglycemia and showed features of mild megaloblastic anemia. All other investigations were within normal limits. Ultrasonography showed single, well-circumscribed swelling measuring 3 cm × 3 cm in the left preauricular region which was suggestive of benign salivary gland lesion. Fine needle aspiration cytology showed moderately cellular smears having acinar epithelial cells arranged in clusters, papillae, and glandular pattern as well as scattered singly [Figure 1a]. Individual cells were round, having round uniform nuclei and a moderate amount of cytoplasm [Figure 1b]. In areas mild cellular enlargement, nucleomegaly, and hyperchromasia were noted. Cells adherent to fibrovascular stroma were noted as well [Figure 1c].

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Characteristically, inflammatory cells were absent in the smears studied. The background showed numerous naked epithelial cell nuclei and red blood cells. Thus, cytological diagnosis was given as sialosis of the left parotid gland.

Discussion

Sialadenosis is a recurrent, noninflammatory, nonneoplastic enlargement of salivary glands usually associated with an underlying systemic disorder.\(^1\) It mainly occurs in the parotid gland.\(^2\) It is often bilateral and recurrent. But few cases with unilateral sialadenosis were reported as well.

Pape et al.\(^3\) reported in his series of cases, four cases of unilateral sialadenosis. Sialadenosis usually occurs in association with a variety of conditions including diabetes mellitus, alcoholism,\(^4\) endocrine disorders, pregnancy, drugs, bulimia,\(^5\) eating disorders, idiopathic, etc. Most patients present were between 40 and 70 years of age.\(^6\) Clinically, it presents as soft, often bilateral, usually painless, and recurrent swelling of the parotid gland.

Fine needle aspiration yields cellular smears having acinar epithelial cells adherent to thin fibrovascular stroma. Mainly large numbers of naked nuclei of epithelial cell origin were seen in the background. Atypical cells or inflammatory cells were characteristically absent. A similar condition that can mimic sialadenosis is low grade acinic cell tumor particularly on cytology. Acinic cell tumors generally yield abundant cellular material with poorly formed microacinar groupings.\(^7\) Naked nuclei are absent in acinic cell tumors that are numerous in sialadenosis. Atypical nuclear features were more prominent in acinic cell tumors as compared to sialadenosis. In our case, nuclear atypia was absent. The distinguishing cellular feature for acinic cell carcinoma includes large nuclei, grainy eosinophilic cytoplasm, and neoplastic cells are arranged singly or in small clusters. In addition, usually there is an absence of other normal salivary gland structures such as duct epithelium and interstitial adipose tissue. These are important features useful for differentiating acinic cell carcinoma from sialadenosis and normal salivary gland enlargement.

The diagnosis of sialadenosis must exclude inflammatory causes of salivary gland swelling, particularly Sjogren’s syndrome, human immunodeficiency virus (HIV) infection, sarcoidosis, and lymphoepithelial diseases by relevant investigations. Other condition of sialadenitis will be excluded as it contains inflammatory cells on the background.

So an unnecessary surgical intervention can be avoided by excluding other pathological conditions. The treatment of sialadenosis is unsatisfactory but it should be aimed at the correction of the underlying disorder. We are presenting this case for its rarity and its important differentiation on cytology smears from other parotid lesions.

Conclusion

In the work-up of salivary gland swelling, it is important to recognize on cytological evaluation of these underestimated entities which do not necessarily require surgical treatment and can be treated with an underlying systemic cause.

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Conflicts of interest
There are no conflicts of interest.

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