

CHRONIC SCLEROSING SIALADENITIS MASQUERADING AS SALIVARY GLAND TUMOUR

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Abstract:

Küttner described 4 cases of chronic sclerosing sialadenitis (CSS) of submandibular gland in 1896 and defined it as a chronic inflammatory salivary gland disease. Although chronic sclerosing sialadenitis is an inflammatory lesion of the salivary glands but sometime mimics malignant masses of salivary glands. We reported a 35-year-old male with a neck swelling of chronic sclerosing sialadenitis which was initially diagnosed as malignancy.

Keywords: chronic sclerosing sialadenitis, sialoliths, submandibular gland, salivary glands.

INTRODUCTION:

The mass of salivary glands may result from a benign inflammatory process, which is known as Chronic sclerosing sialadenitis (CSS) or Kuttner's tumour (KT). Sometime these masses may present as stony hard tumour and masquerade as malignant lesion. It affects mainly the submandibular gland but some cases of parotid glands are also reported. (1, 2)

The histological characteristics of chronic sclerosing sialadenitis are ductal squamous metaplasia, periductal fibrosis, dense lymphoplasmocytic infiltration, loss of the acini, sclerosis of the salivary gland and sialoliths in salivary ducts. (3, 4) Because CSS appear as a hard mass, it usually assumes an immense clinical doubt of a malignant neoplasm. In recent years, fine-needle aspiration cytological (FNAC)

examination and needle biopsy have been used. It is more safer, comparatively trouble-free with quick result and economical techniques to confirm salivary gland lesions.(5,6) The assistance of CT scan in FNAC is also very valuable in correct diagnosis of salivary gland masses. We report the CT scan appearance and CT scan guided FNAC features of an adult with CSS of the unilateral submandibular gland.

CASE REPORT:

A 35 –year-old male was referred to oncology outpatient department. He observed the neck mass 2 years ago. On examination there was hard, bimanual palpable mass at level Ib on right upper neck and it seemed to be attached to underlying structures. Small Lymph node were palpable at right level II and III and also at left level Ib and II in neck. Small nodule was noted at right Floor of Mouth (FOM). Any other related events were not found in the patient’s medical history. He reported no other symptoms or complaints. His facial nerve function was intact. Patient came with FNAC report which revealed an impression of malignant lesion. Malignancy of Unknown Origin was predicted as probable diagnosis because report of malignant lesion. Identification of primary and secondary and further management of both were planned. CT scan showed mildly enlarged right submandibular gland with heterogeneous enhancement and architecture, suggestive of sialadenitis along with calculi in the submandibular duct.



Figure 1:CT scan image (transverse view) shows that Right submandibular duct was mildly dilated and there were two calcified calculi measuring 8 mm and 9 mm in the proximal part of duct and at the distal end. (Black arrows).

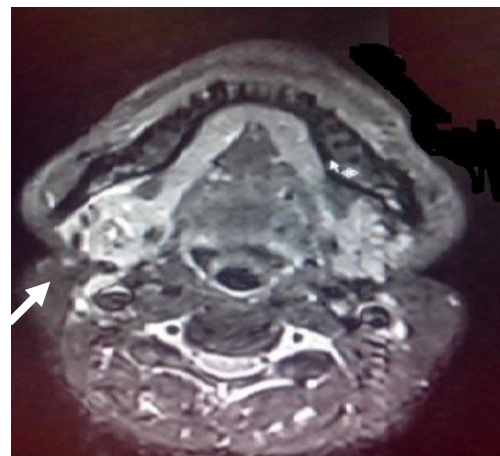


Figure 2:CT scan image (transverse view) of the head shows Right submandibular gland mass located within the neck. (white arrows).

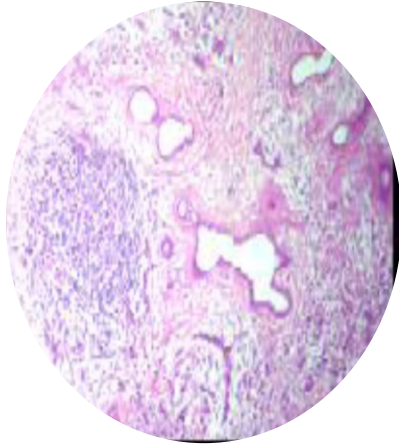


Figure 3

FNAC: A CT scan guided fine-needle aspiration cytological (FNAC) examination was performed. Cytology revealed occasional acinar clusters with inflammatory cells against a dense necrotic background. No pathologic nodes were identified (Figures 3).

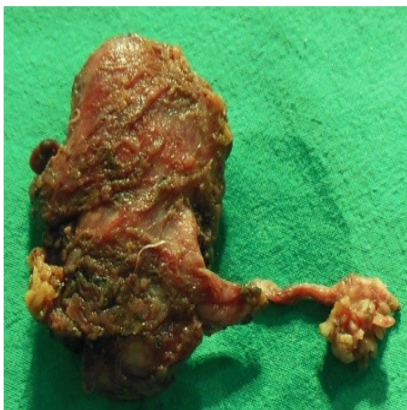


Figure 4

Histopathological examination:

Gross features: The specimen collected for pathological examination measured 5x4x1.8 cm. External surface was unremarkable, and its cut surface revealed lobulated pale tan

tissue. The whole tissue sample was submitted for pathological examination. Salivary duct also received that measured 3cm in length, dilated and one end with a calculus. (Figure 4)

Microscopy: For microscopic examination, multiple sections studied from salivary gland reveal preserved lobular architecture. There was dense lymphoplasmocytic infiltrate, surrounding the duct and acini with accompanying periductal fibrosis. The salivary acini proximal to obstructed and dilated ducts were atrophic. Reactive lymphoid follicles were seen. There was varying degree of fibrosis surrounding the lobules. Dilated larger duct revealed squamous metaplasia. Two adjunct lymph node revealed reactive hyperplasia. (Figure 5). (Figure 6). Malignant cells were not found, and the chronic sclerosing sialadenitis was diagnosed.

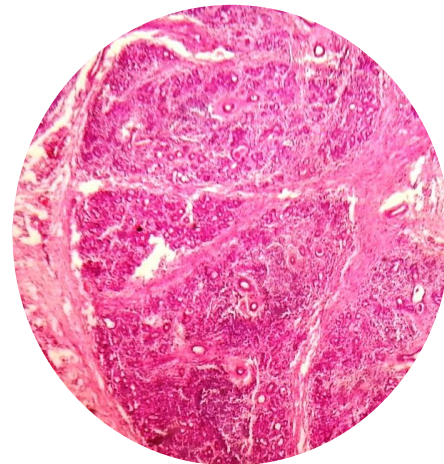


Figure 5: Histology of the salivary gland tissue showing chronic sclerosing sialadenitis (Küttner's tumour) fibrosis and few residual ducts and foci of lymphocytic

infiltrate often with germinal centres. (Haematoxylin and Eosin).

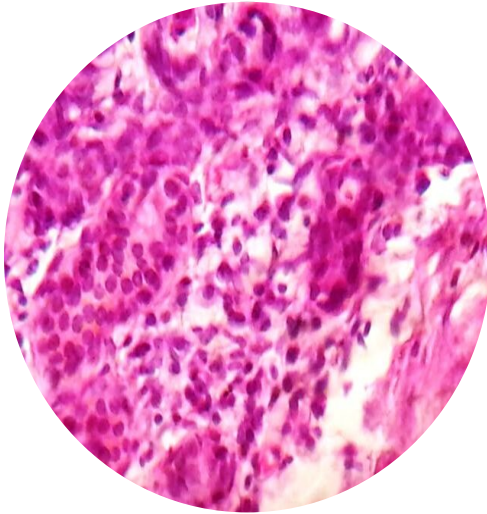


Figure 6: Histology showing the salivary gland residue embedded in collagenised fibrous tissue and dense lymphoplasmacytic infiltration. (Haematoxylin and eosin, 100 x magnifications)

DISCUSSION:

CSS is a benign disease that results from different causes. In recent years some etiological processes have been suggested by authors to define underlying pathology of CSS, for instance salivary gland stones, secretory abnormality with ductal stasis of saliva, infections, ductal disorders and an autoimmune pathology. (7,8) Seifert et al showed that the findings of CSS were analogous to obstructive sialadenitis. (9) However, the obstructive sialadenitis or sialolithiasis could not explain the mechanisms of the inflammatory process clearly. Immunologic pathogenesis of CSS was explained by some researchers. There was a close connection between the T cell-lymphocyte with plasmacytic infiltrate,

surrounding the duct and acini with accompanying periductal fibrosis, equally with the persistent presence of monoclonal and oligoclonal cytotoxic T cells and their relevant histopathological features. Tiemann et al concluded that intraductal inflammatory chemo-attractant may elicit an immune process and histological changes in CSS. (10) Geyer JT et al also showed other immunological markers in CSS. Immunohistochemical staining shows abundant IgG4 and IgG positive cells. The IgG4/IgG ratio is high compared to other inflammatory diseases of the salivary glands. (11)

Mucous plugs and salivary stones are reported in 29% to 83% of cases of CSS. (12) In this case, two sialoliths were found which may be a cause of dilation of right submandibular duct. Salivary gland stones may obstruct salivary discharge or accumulation of secretions. A hypothesis of obstructive electrolyte sialadenitis, is given by Seifert and Donath. (13) They postulated that secretion abnormality makes mucous plug that obstructs the small ducts, obliteration further cause inflammatory reaction, parenchymal and ductal atrophy, periductal fibrosis, and an immune reaction towards the duct system. Benign differential diagnosis of CSS include simple chronic sialadenitis, granulomatous sialadenitis, necrotising sialometaplasia, sialolithiasis, an inflammatory pseudotumour, radiation effects and benign lymphoepithelial lesions. Another common cause of CSS of the salivary glands is associated to rheumatoid arthritis, which is also explained the immune pathogenesis. (14) The malignant differential

diagnosis includes extra nodal marginal zone B-cell lymphoma of MALT, fibrohistiocytic tumours, Kimura's disease, sclerosing lymphoma, sarcoidosis and neoplasms of the salivary glands. CSS have a very good prognosis as this disorder has benign lesions that are not liable to recurrence. No reports were found to support the view that this condition may be causative factor for malignancy. (15)

The disease mimics true neoplasm and sometimes difficult to distinguish clinically. (16) Radiological imaging is frequently used for the primary examination to assess the character of salivary gland mass. For the detection of focal salivary masses, sonography has a sensitivity of 100% and an accuracy of nearly 100% compared with 92% and 87% by palpation. (17)

MRI is also a sensitive tool for diagnosis of CSS. In MRI, signal intensity ratios for T2 weighted and STIR images, ADC values and patterns of enhancement may help to distinguish Kuttner's tumours from benign submandibular gland tumours, but not from malignant tumours. Although the intensities, ADC values and enhanced patterns of Kuttner's tumours were similar to those of malignant tumours, but there were some morphological differences. (18)

Repeat FNAC may provide a cytological diagnosis in cases where the initial diagnosis is not clear, although cytology should be used in combination with other investigations of salivary tumours, including image-guided biopsy examination where appropriate. Ideally salivary gland FNAC

should be interpreted by a specialist pathologist. (19)

CONCLUSION:

Kuttner tumour should be kept in mind during the differential diagnosis of any firm to hard swelling of salivary gland as it is rare swelling of salivary glands that clinically masquerade as malignancy. Early and correct diagnosis is essential for the planning of management. FNAC is good tool but the sensitivity will be increased if it is image guided and repeat FNAC also give a correct diagnosis in case of any confusion.

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Ethical approval: The study was approved by the institutional ethics committee

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