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*Case Report*

# Bilateral Kuttner Tumor of Submandibular Glands: A Case Report

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***Abstract***

***Introduction:***

Chronic sclerosing sialadenitis is a relatively uncommon disorder of the salivary gland. Because of its clinical similarity to a salivary gland neoplasm, this condition has been known as Kuttner tumor and is classified as a tumorlike lesion.

***Case Report:***

This is the first reported case of bilateral Kuttner tumor of the submandibular glands in Iran. It was initially diagnosed as a primary submandibular gland neoplasm whereas histological findings showed chronic sialadenitis. Excision of such masses is the treatment of choice.

***Keywords:***

Kuttner tumor, Lesion, Submandibular gland.

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***Introduction:***

Kuttner in 1896 reported chronic sclerosing sialadenitis for the first time (1). This tumor-like lesion is a chronic inflammatory disease of the salivary gland that causes a firm swelling of the glands with the clinical impression of a neoplasm (2,3). It seems that Kuttner tumor is reported less than the real prevalence (i.e. It is underdiagnosed). Hence, it might be due to its low prevalence which makes the histological diagnosis a little difficult for the pathologist who is not fully aware of this condition (4).

We present a case of bilateral submandibular mass initially diagnosed as a primary salivary gland neoplasm but its final histological evaluation revealed a chronic sclerosing sialadenitis bilaterally.

***Case Report:***

A 14-year-old boy with a bilateral submandibular gland mass which had been present for about 5 months was referred to the ear, nose and throat clinic. He did not complain of drying eyes or mouth but felt some discomfort on palpation of the masses and also postprandial. His past medical history record was insignificant.

Physical examination showed a 4×3 cm left submandibular mass and a 3×2.5 cm right submandibular mass (Fig. 1).

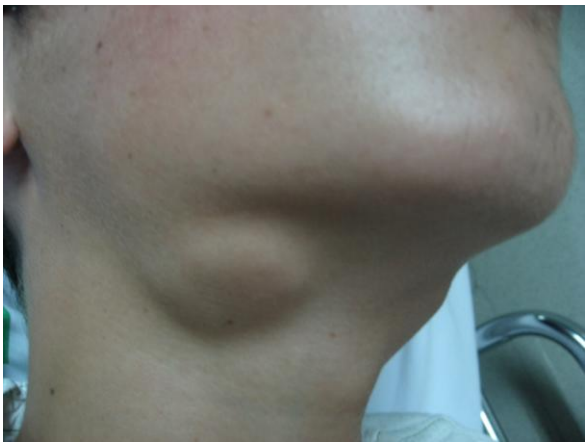

**Fig 1:** Bilateral submandibular masses

They were firm and mobile with mild tenderness but without any erythema or constitutional signs. There was no lymphadenopathy or any other salivary gland involvement.

CT scan with contrast showed a bilateral submandibular homogenous mass which enhanced mildly. There was no evidence of sialolithiasis. FNA was done to rule out a neoplastic condition and showed a nonspecific inflammation. Routine laboratory tests (CBC, ESR, CRP, and FBS) were in the normal range.

The patient underwent operation on both submandibular glands and they were removed at the same time (Fig. 2).

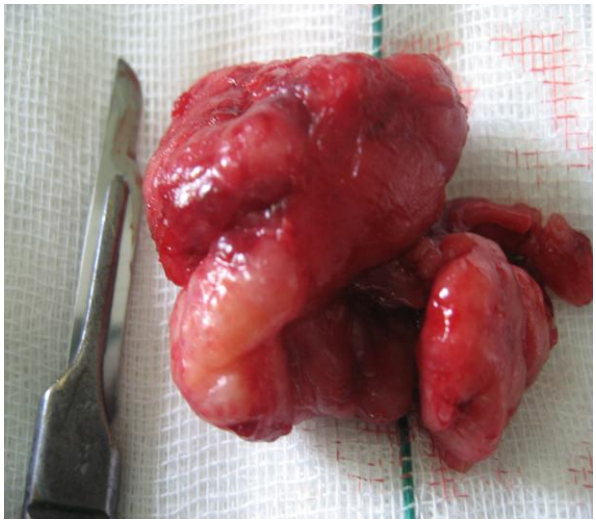

**Fig 2:** The left submandibular gland specimen which was removed surgically

The permanent pathological sections confirmed the presence of chronic sclerosing sialadenitis called Kuttner tumor with no evidence of malignancy. Histologically, the submandibular mass showed inflammatory infiltration of lymphocytes, plasma cells, and histiocytes, growth of fibroblasts and marked atrophy of acinis. The normal lobular architecture was preserved while destruction of the duct like lymphoepithelial lesion was present. There was fibrosis in the stroma and ductal hyperplasia with periductal fibrosis. Some of them were surrounded by collars of fibrous tissue often formed in an onion-skin arrangement (Fig. 3)

Progressive periductal sclerosis, dense lymphocytic infiltration, reduction of the secretory gland parenchyma, and fibrosis were also diagnosed.
No further therapy was prescribed and after one year no evidence of the disease was observed on follow up.

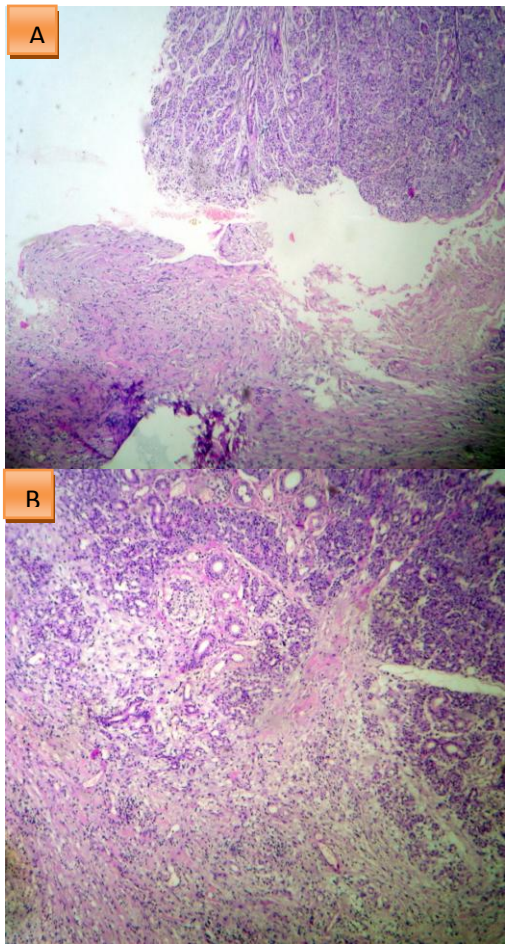


**Fig 3:** Chronic sclerosing sialadenitis :
A) Foci of residual salivary ducts embedded in extensive amounts of fibrous connective tissue. .(H&E staining,X 10 ) .
B) acinar atrophy, lymphoid infiltrate and marked fibrosis.(H&E staining,X 40 )

***Discussion:***

Chronic sclerosing sialadenitis of the submandibular gland was reported by Küttner in 1896 for the first time[1].The clinical presentation varies from asymptomatic swelling to a recurrent postprandial pain (2,3,5). The lesion is usually firm and it grows gradually. The submandibular gland is the most common salivary gland that is involved in KT (2,3). KT is seen in all ages, from 12 to 83-year-old cases, however the mean age of the affected patients is 39 to 45 years (2,3,5,6,7). Some reports have shown a slightly higher incidence in males (2,6) whereas this has been vice versa in others (5).
It is often presented as an enlarging firm mass for which the clinical diagnosis of a salivary gland neoplasm, especially carcinoma might be suggested (4). Kuttner tumor should be distinguished from benign lymphoepithelial lesions, Kimura's disease, lymphoma of mucosa-associated lymphoid tissue, and inflammatory pseudotumors (7).

Duration of the disorder is highly variable and ranges from 2 weeks to 28 years (5). In our case the duration of symptoms before the patient's referral was 5 months.

The cause of inflammation seen in KT is still unknown (7). The origin may be attributable to many etiologic factors. Sialoliths are found in 29–83% of cases (2,8,9) but the cause and effect relationship is not yet clear. In this case, there was no sialolith in either gland. But anything that obstructs the salivary flow or causes stasis of secretions can lead to acinar cell swelling, necrosis, ductal dilation and retention of salivary secretions resulting in edema and inflammatory cell infiltration consequences (10).

For diagnosis of KT open or excisional biopsy is necessary. The diagnosis of chronic sclerosing sialadenitis in the submandibular glands is based on histology; however, it is difficult to distinguish KT from other forms of sialadenitis hiostologically in advanced stages. Periductal lymphocytic infiltration and irregular ductal ectasia is seen from the morphologic aspect. In the final stage, fibrosis of the periductal and periacinar parenchymal gland is the prominent feature (2,10).

KT is absolutely benign, and up to now there have been no reports of malignancy. Complete removal of the affected gland is the entire procedure that should be done as a treatment and no additional measures are warranted.

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