

## CLINICAL STUDY

## Kuttner tumor

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**Abstract:** *Objective:* Kuttner tumor or chronic sclerosing sialadenitis is a benign inflammatory condition of the salivary gland. Clinically, it produces a firm swelling of the gland and may be difficult to distinguish from neoplasia. This is an under-recognized entity in the surgical pathology and cytology literature. We describe our experience with Kuttner tumor.

*Methods:* Retrospectively, we analyzed a group of 7 patients with chronic sclerosing sialadenitis treated from January 1999 to March 2010 at the Department of Otorhinolaryngology, FD Roosevelt Faculty Hospital in Banska Bystrica, Slovakia. The authors evaluated age and sex distribution, patient's history, diagnostic procedures, extent and success of surgical treatment and postoperative complications.

*Results:* Four patients with chronic sclerosing sialadenitis were male and 3 were female. The mean age of patients was 55 years (ranging from 43 to 70 years). Submandibular gland was affected in 6 cases (85.7 %), involvement of both parotid glands was found in one patient (14.3 %). All patients in our study group were treated surgically. In 6 cases submandibular gland extirpation was performed via standard transcervical approach. Total parotidectomy with facial nerve preservation was realized in one patient with bilateral involvement of parotid glands.

*Conclusion:* Chronic sclerosing sialadenitis is a condition that is perhaps more common than thought but usually mis-recognized as it is only after excision of the gland that the correct diagnosis is made (Tab. 1, Ref. 17).

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Key words: Kuttner tumor, chronic sclerosing sialadenitis, salivary gland.

Kuttner tumor, also known as chronic sclerosing sialadenitis or cirrhosis of the salivary gland is a fibroinflammatory disease of the salivary glands, characteristically of the submandibular gland. Chronic sclerosing sialadenitis belongs to the spectrum of IgG4-related diseases (1, 11, 16).

Clinically, it produces a firm swelling of the glands and may be difficult to distinguish from neoplasia (10). It is characterized histologically by periductal fibrosis, dense lymphocytic infiltration with lymphoid follicle formation, loss of the acini, and eventually, marked sclerosis of the salivary gland (2). Excision of the mass, usually carried out diagnostically, is an adequate treatment (10).

We report our experience of Kuttner tumor. Etiopathogenesis, clinical symptoms, diagnostic procedures and treatment are discussed.

## Methods

This retrospective study analyzed a group of 152 patients with salivary gland disorder treated surgically at the Department of Otorhinolaryngology, Faculty Hospital of FD Roosevelt in Banska Bystrica, Slovakia, within the period from January 1999

to March 2010. Among these, 7 patients (4.6 %) were confirmed to be Kuttner tumor, based on the clinicopathological testing, and represented our study group. The authors evaluated age and sex distribution, patient's history, diagnostic procedures, extent and success of surgical treatment and postoperative complications.

## Results

Seven cases of chronic sclerosing sialadenitis were identified. There were 4 males (57.1 %) and 3 females (42.9 %). The mean age of patients was 55 years (ranging from 43 to 70 years).

Submandibular gland was affected in 6 cases (85.7 %), involvement of both parotid glands was found in one patient (14.3 %). Two patients (28.6 %) complained of painful swelling in the salivary gland region, while the other 5 (71.4 %) had painless masses. The median duration of the presenting masses was 20.7 months (range 3 months – 3 years) (Tab. 1).

Preoperative ultrasonography (USG) was performed in all patients. Two (28.6 %) patients underwent computed tomography imaging (CT). Stone in the Warthon's duct was detected in 2 cases (28.6 %). 5 patients (71.4 %) had preoperative fine-needle aspiration biopsy (FNAB), the other 2 were spared this investigation as stones were detected during imaging. In the cytological findings of 3 of these patients, FNAB revealed chronic inflammation of salivary gland. In one patient there was suspicion of lymphoproliferative process due to the presence of a heavy lymphoid infiltration. Carcinoma was not suspected in any of them. FNAB was not valuable due to the lack of the cells in the aspirate in one case.

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**Tab. 1. Demographic and clinical features of the patients with Kuttner's tumor.**

Case No.	Sex/age (years)	Pain (Yes/No)	Duration (months)	Gland involvement	Stone (Yes/No)
1	M/70	No	4	gl. submandibularis	No
2	M/49	No	12	gl. submandibularis	No
3	M/64	Yes	36	gl. submandibularis	Yes
4	F/67	No	36	gl. submandibularis	No
5	F/46	No	3	gl. parotis – bilateral	No
6	M/46	No	24	gl. submandibularis	Yes
7	F/43	Yes	30	gl. submandibularis	No

All patients in our study group were treated surgically. In 6 cases submandibular gland extirpation was performed via standard transcervical approach. Total parotidectomy with facial nerve preservation was realized in one patient with bilateral involvement of parotid glands. Regarding surgical complications, only one patient after total parotidectomy suffered a transient facial nerve palsy, which recovered spontaneously.

## Discussion

Kuttner tumor, known descriptively as chronic sclerosing sialadenitis, is a benign chronic fibroinflammatory disease of the salivary gland. Chronic sclerosing sialadenitis is a part of the spectrum of IgG4-associated diseases (10, 16).

The etiology of Kuttner tumor is still unknown. The effects of microliths (sialoliths), infectious agents, secretory dysfunctions, duct abnormalities, and immune processes have been considered (5, 6, 7, 10).

## Diagnosis

Patients presenting with Kuttner tumor are usually adults. The reported ages of patients range from 13 to 81 years with a mean age at surgery of 44 years. There is usually a slight male predominance (1).

In most cases of chronic sclerosing sialadenitis, the main complaint of the patients is intermittent and/or persistent pain or swelling in the salivary gland region, which worsens during mealtimes. However, some lesions are asymptomatic except for a firm swelling. Duration of symptoms is highly variable, ranging from less than 1 year to several decades (14). In our series the duration of symptoms ranged from 3 months to 3 years.

Most commonly there is unilateral involvement of submandibular gland, but less frequently both submandibular glands and parotid gland can be involved. Unusual cases affecting both parotid glands, minor salivary glands, lacrimal glands as well as multi-glandular involvement were also reported (1, 3, 4, 6, 7, 13, 14).

Because the disease cannot be distinguished clinically from a true neoplasm, ultrasonography is often used for the initial investigation to evaluate the nature of an enlarged major salivary gland. For the detection of focal salivary masses, ultrasonography has a sensitivity of 100 % and an accuracy of nearly 100 % compared with 92 % and 87 % by palpation. The most common ultrasonographic appearances are the diffuse cirrhotic-like patterns: diffuse involvement with multiple hypoechoic lesions against a heterogeneous background with duct dilatation, resembling a cirrhotic

liver. The involved gland shows prominent vascularity, with no mass effect or displacement of the vessels as they course through the parenchyma (4, 11).

Fine-needle aspiration biopsy (FNAB) is a simple, cost-effective, and safe technique with a high sensitivity and specificity in the diagnosis of salivary gland masses (1, 2).

Chronic sclerosing sialadenitis has some characteristic features on fine-needle aspiration cytologic specimens: (1) relatively low cellularity and thus relative difficulty in obtaining the cellular elements; (2) scattered ductal structures with paucity or absence of acini; (3) ducts intimately surrounded by collagen sheaths or lymphoid cells; (4) small isolated fragments of fibrous stroma; (5) moderate to large numbers of lymphoid cells that lack definitive atypia (5, 11).

Histopathology shows preservation of the lobular architecture, dense lymphoplasmatic infiltration, periductal fibrosis, and loss of acini (5, 14).

According to Seifert et al, lesion may evolve through four histologic stages.

Stage 1: Focal chronic inflammation with nests of lymphocytes around salivary ducts, which are moderately dilated and contain inspissated secretions.

Stage 2: more pronounced diffuse lymphocytic infiltration and severe periductal fibrosis. Ductal system still shows inspissated secretions and focal metaplasia with proliferation of ductal epithelium. Periductal lymphoid follicles are well developed. There is fibrosis in the centers of the lobules, accompanied by atrophy of the acini.

Stage 3: Shows even more prominent lymphocytic infiltration with lymphoid follicle formation, parenchymal atrophy, periductal hyalinization, and sclerosis. Squamous and goblet cell metaplasia may be seen in the ductal system.

Stage 4 (end stage): Shows cirrhosis-like stroma with marked parenchymal loss and sclerosis (7, 13).

Possible differential diagnosis of this entity includes other benign inflammatory lesions of salivary glands, benign lympho-epithelial lesions, extranodal marginal zone B-cell lymphoma of mucosa-associated lymphoid tissue, sclerosing lymphoma, neoplasms of the salivary glands and radiation effects (4, 6, 7, 8, 10).

## Treatment

Management can be conservative by adopting a “watch and wait” approach in the case where the mass is otherwise asymptomatic and the patient is comfortable, and there is adequate evidence provided that the lesion is benign. Most commonly the mass is

surgically excised, as it is difficult to differentiate it from a malignancy. However, based on immunological findings recently implicated in its development, administration of steroids has been shown to be effective in shrinking such salivary swellings (3, 4, 6).

From our point of view, surgery is the standard therapy for Kuttner tumor. We think, that excision of the affected gland should be performed for definitive diagnosis and treatment of chronic sclerosing sialadenitis. All patients in our report were treated surgically. Our results show that operative morbidity is minimal, only 1 patient suffered a temporary facial nerve palsy. No recurrence of Kuttner tumor was observed after surgical treatment in our study group.

### Follow-up

For the benign lesions of salivary glands, follow-up is usually scheduled for every 2–3 months, then every 6–12 months, with an annual ultrasonography; the length of the follow-up period was not specified (17). In our opinion, a follow-up every 3 months, for the first year, and then every 6–12 months, thereafter, seems reasonable.

### Conclusion

Kuttner tumor is a benign inflammatory condition of the salivary gland with a characteristic ultrasonographic and clinical manifestations. Its morphologic appearance, in conjunction with the elevated IgG4 expression, distinguishes chronic sclerosing sialadenitis from other inflammatory diseases of the salivary glands. Surgical excision of the affected gland is a safe operation and seems to be a definitive treatment of chronic sclerosing sialadenitis.

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Received May 25, 2010.  
Accepted September 20, 2012.