

## Original Research Article

# Kuttner's tumour: chronic sclerosing sialadenitis- a mimicker of malignancy

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### ABSTRACT

**Background:** Kuttner's tumour is a condition of the submandibular gland which is underreported as a distinct entity. Also called as the chronic sclerosing sialadenitis it resembles a plasmocytic and lymphocytic inflammatory process and presents as a hard and painful mass which clinically mimics a malignancy and raises significant concerns. The objective of the present study was to evaluate cases of Kuttner's tumor and discuss its clinical and pathological aspects to distinguish it from a definite malignancy and create a grown acceptance of the presence of such an entity in our setting.

**Methods:** We collected 170 cases of submandibular swellings and evaluated in detail the clinical and pathological aspects of eight cases out of them which were later diagnosed to as Kuttner's tumour.

**Results:** The age of the patients varied between 23 to 61 years (mean age 42.5 years) with 3 males and 5 females. 6 patients reported with a firm to hard painless submandibular mass (5 left sided; 3 right sided) while 2 patients experienced intermittent discomfort. The mean duration of presentation of symptoms was 5.3 months. Fine needle aspiration cytology was done in 6 cases preoperatively. Following submandibular sialadenectomy histopathology showed salivary gland tissue with preserved lobular architecture, but with marked fibrosis, acinar atrophy, and a dense lymphoplasmacytic infiltrates.

**Conclusions:** Kuttner's tumor may be pre operatively distinguished from a malignancy with improved imaging and a good image guided FNA Cytology saving the clinician and the patient from a lot of dilemma. However, histopathology and immunohistochemistry would be the key in establishing the diagnosis.

**Keywords:** Kuttner's, Chronic sclerosing sialadenitis, Submandibular tumour

### INTRODUCTION

Kuttner first defined chronic sclerosing sialadenitis (CSS) of submandibular gland when he described 4 of such cases of chronic inflammatory salivary gland disease in 1896.<sup>1</sup>

Difficult to distinguish these lesions from malignancies of salivary glands clinically and dreaded, such masses have been now been described and proven to be as

inflammatory lesions of the salivary glands more than anything else. This lesion has been recognised as a distinct clinicopathological entity in the latest edition of World Health Organisation classification of salivary gland tumors.<sup>2</sup> CSS is clinically characterised by a firm, occasionally painful mass of the submandibular glands. Fine needle aspiration cytology (FNAC) being a simple and effective modality to work up salivary gland masses, its efficacy in diagnosing this particular entity is still questionable.<sup>3-5</sup> Plasmocytic and lymphocytic periductal

infiltrate which eventually lead to the encasement of ducts with thick fibrous tissue of the salivary glands are the characteristic features of Kuttner's tumor.

Although described more than a century ago, this clinical entity is still under-reported and preoperatively it remains unrecognized by many surgeons. The submandibular masses are often removed following a preliminary diagnosis of carcinoma, thus inciting substantial worry to both patients and clinicians. This state of affairs is attributed to the under-reporting of Kuttner's tumor in literature. We report here a series of cases of CSS and discuss its clinical and pathological aspects to create a clearer picture and a grown acceptance of the presence of such an entity in our setting.

**Objectives**

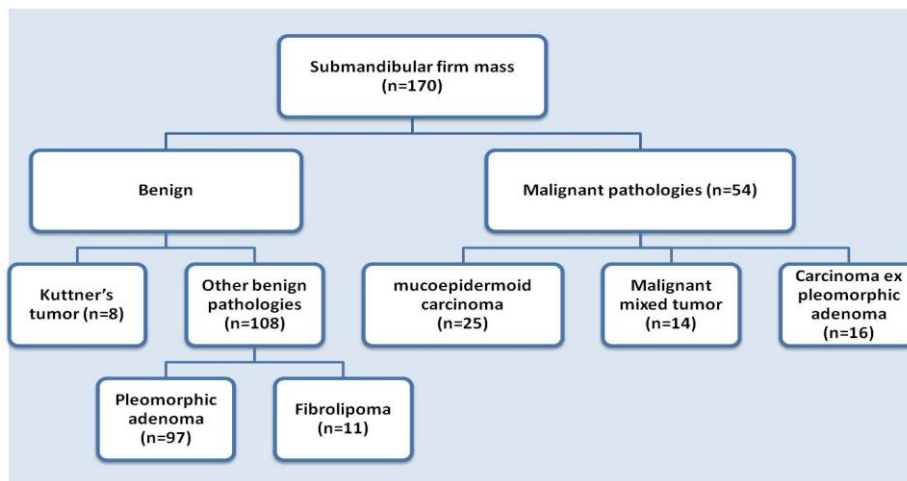
The objectives of the present study were to evaluate cases of Kuttner's tumor and discuss its clinical and pathological aspects to distinguish it from a definite malignancy and a grown acceptance of the presence of such an entity in our setting.

**METHODS**

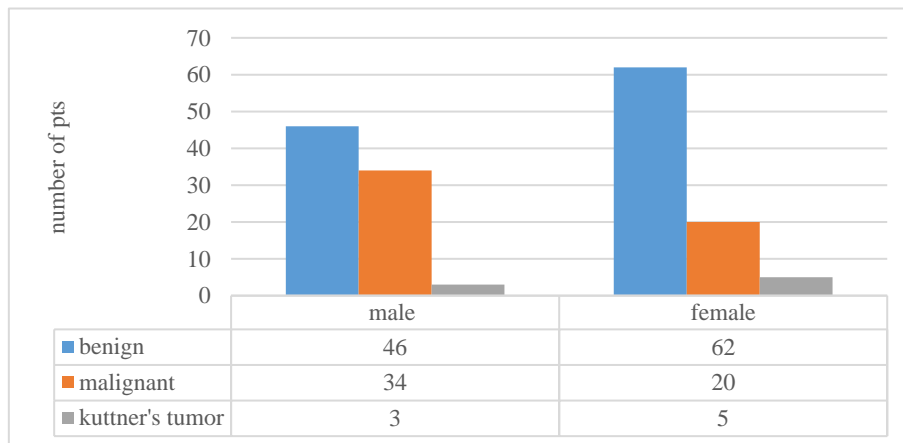
This clinicopathological study entails 8 cases of Kuttner's tumor, which were treated at Institute of Medical Sciences and SUM Hospital, Bhubaneswar, Odisha, India, a tertiary care hospital, during the period from July 2014 to January 2019. We evaluated and followed up 170 cases which presented with submandibular masses with or without pain. All the cases were clinically assessed, preliminary cytology done and subjected to surgery which was then followed by a histopathology. Out of these, 8 patients were diagnosed as CSS (Kuttner's tumor) histopathologically and thus were included in the present study (Table 1). Due consent was taken from each of the above patient before any investigations, surgery and inclusion in the above study. The statistical analysis was done using SPSS.

**RESULTS**

The incidence of Kuttner's tumor in our setting was <0.05% of all submandibular masses encountered during the above time period (i.e., exactly 0.04%; 8 out of 170) (Figure 1 and 2).



**Figure 1: Epidemiology of firm painless, submandibular masses.**



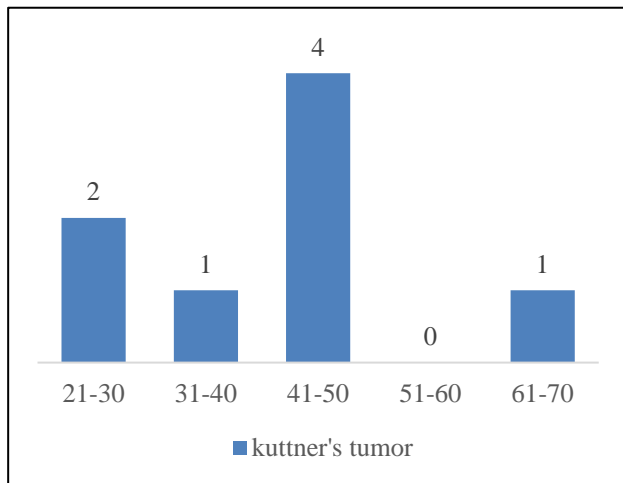
**Figure 2: Incidence of various submandibular pathologies.**

The age of the patients varied between 23 to 61 years (mean age 41.5 years) with 3 males and 5 females. The age distribution is shown in Figure 3 with a 50% incidence in 5<sup>th</sup> decade of life.

Six patients reported with a firm to hard painless submandibular mass (5 left sided; 3 right sided) while 2 patients experienced intermittent discomfort. The mean duration of presentation of symptoms was 5.3 months.

**Table 1: Clinical details of patients with Kuttner’s tumor (CSS).**

Case no.	Age /sex	Clinical manifestations	Side affected	FNAC findings	Calculi
1	45/M	Painless, firm, non-tender, Left sided submandibular mass for 6 months	Left	Scanty acini and normal-looking ductal cells, with a variable degree of chronic inflammatory cell infiltration	Absent
2	23/F	Recurrent occurring firm, slightly tender with intermittent discomfort left sided submandibular mass for 8 months	Left	Epithelial proliferation; chronic inflammation	Present
3	36/F	Painless, firm nontender right sided submandibular mass for 2 months	Right	Variable degree of chronic inflammatory cell infiltration	Absent
4	50/F	Painless, firm to hard, nontender left submandibular mass; intermittent discomfort for 6 months	Left	Not done	Present
5	43/M	Recurrent occurring soft to firm, non-tender left submandibular mass for 4 months	Left	Epithelial proliferation with chronic inflammation	Absent
6	44/M	Painless, firm to hard non tender right submandibular mass for 1 year	Right	Lymphoproliferative lesion	Absent
7	30/F	Painless, firm non tender left submandibular mass for 3 months	Left	scanty acini and normal-looking ductal cells, with a variable degree of chronic inflammatory cell infiltration	Absent
8	61/F	Painless, firm nontender right sided submandibular mass for 2 months	Right	Epithelial proliferation; chronic inflammation	Absent

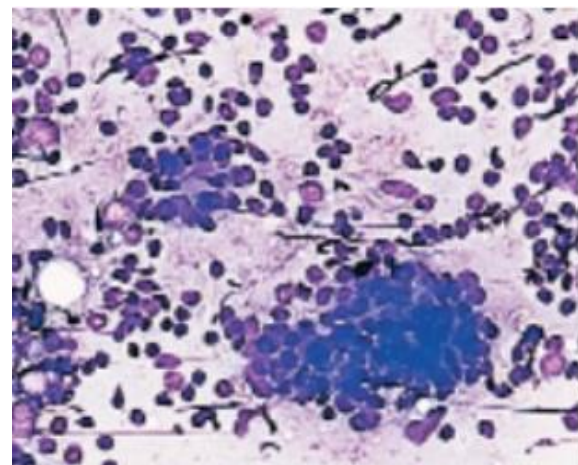


**Figure 3: Age distribution of patients with Kuttner’s tumor.**

All the patients were subjected to ultrasonography of the submandibular region of both the sides. Calculi were detected in 2 cases along with dilated duct which were confirmed by submandibular sialography and computed tomography imaging (Figure 5).

FNAC was done in all 6 cases pre operatively (Figure 4 and 5).

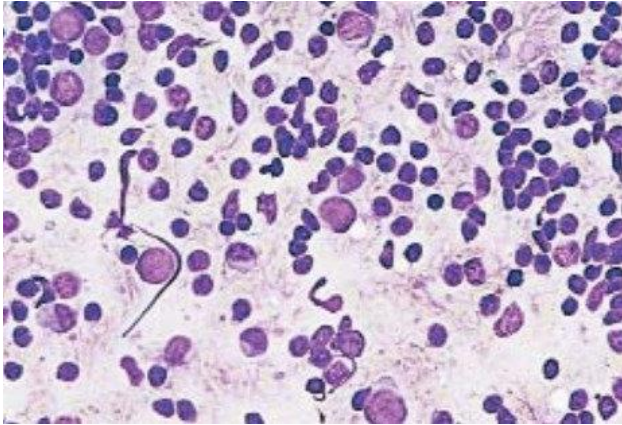
Only 1 patient on FNAC had features suggestive of a lympho-proliferative disease though malignancy was not ruled out.



**Figure 4: FNAC the aspirate is moderately cellular and shows ducts scattered in a background of inflammatory cells.**

**Table 2: Fine needle aspiration findings.**

Variable degree of chronic inflammation with scanty acini (Figure 4).
Normal looking ductal cells (Figure 4).
Tubular ductal elements surrounding collagen fibres around Plasmolymphocytic cells (Figure 5).



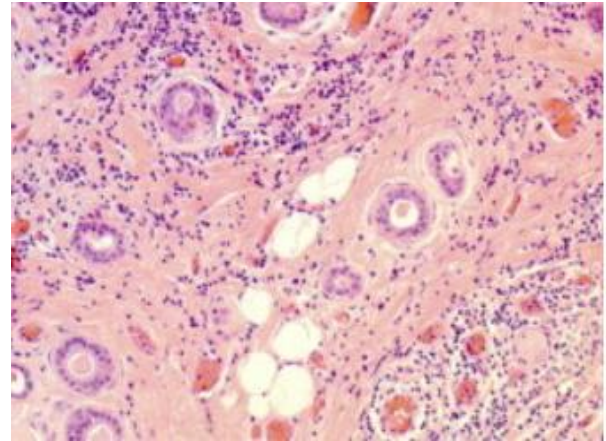
**Figure 5: FNAC numerous lymphoid cells admixed with plasma cells and histiocytes.**



**Figure 6: CT SCAN Showing submandibular sialolith.**

All the 8 patients were subjected to surgery as the submandibular lesion could not be ascertained to be benign on any of the pre-operative investigations. Submandibular sialadenectomy of the affected side under general anaesthesia was performed on each of the above patients. On gross examination, the lesions of the submandibular gland measured between 6-8 cms approx. Intra operative findings included a firm submandibular mass with dense surrounding adhesions and difficulty in delineation of regional anatomy, though in each case the

duct was duly identified and secured. The gross samples were firm, indurated, lobulate yellowish white. Histopathology showed salivary gland tissue with preserved lobular architecture including the lobular septa. Marked periductal fibrosis was noted in all cases though the extent varied in each case. Dense lymphoplasmocytic infiltrate (Figure 7) was noted both within and in between the lobules. There was acinar atrophy with residual ducts being encased by thick collagen bundles.



**Figure 7: Histopathology of the gland showing chronic plasmolymphocytic infiltrate with acinar atrophy and ductal sclerosis.**

## DISCUSSION

CSS is an inflammatory process that primarily affects the submandibular gland of middle aged adults and presents clinically as a painful swelling. The inflammatory lesions in Küttner's tumor may occur on one side (unilateral) or both sides (bilateral), predominantly involving the submandibular gland, but is also known to occur in other major and minor salivary glands including the parotid gland.<sup>6-9</sup> Salivary tumors especially those of submandibular glands show a great deal of morphological diversity, as well as variations in the nature of the lesion (malignant vs benign) 35-40% of submandibular tumors. This situation underscores the diagnostic challenges in respect of Küttner's tumor; despite being benign, this condition mimics the clinical appearance of malignancy in the salivary gland.

Postulations are still the mainstay in the etiology of Küttner's tumor, the chief among which comprise sialoliths and intrinsic ductal abnormality with inspissated secretions which can give rise to chronic inflammatory changes. Mucous plugs and salivary stones have been reported in 29-83% of cases of CSS.<sup>10</sup> A hypothesis of obstructive electrolyte sialadenitis, is given by Seifert et al.<sup>11</sup> 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.

However, the obstructive sialadenitis or sialolithiasis was unable to explain clearly how the actual inflammatory process initiated and progressed. Some studies put forward an immunologic connection for the above pathology and described 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. An intraductal inflammatory chemo-attractant may elicit an immune process and histological changes in CSS as was concluded by Tiemann et al.<sup>12</sup> Geyer et al also showed other immunological markers in CSS.<sup>13</sup> Kamisawa et al reported four patients with Kuttner's tumor, two of whom also manifested autoimmune pancreatitis.<sup>14</sup> They suggested that the serum IgG4 level was useful in diagnosing Kuttner's tumor. Abundant IgG4 and IgG positive cells were seen on immunohistochemical staining with IgG4/IgG ratio high compared to other inflammatory diseases of the salivary glands. Our study being retrospective in nature IgG4 studies were not available.

Since Kuttner's tumor manifests as a hard mass which may be painful or sometimes even painless, it usually raises a strong clinical suspicion of a malignant neoplasm. FNAC of the mass is a simple and cost-effective technique which has been employed in the diagnosis of these tumors and has been quite handy. The FNA cytology of Kuttner's tumor is characterized by the following features: relatively low cellularity, probably attributable to the fibrosis, making it difficult to aspirate the cellular elements; scattered ductal structures with paucity or absence of acini; ducts intimately surrounded by collagen sheaths or lymphoid cells; small isolated fragments of fibrous stroma; and moderate to large numbers of lymphoid cells that lack definite atypia.<sup>15-19</sup> So the FNAC findings of scattered ducts surrounded by collagen and infiltrated by lymphoid cells, although not specific but along with the absence of malignant cells are somewhat suggestive of the diagnosis in the appropriate clinical settings.

Radiological imaging is primarily used 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.<sup>20</sup>

In magnetic resonance imaging signal intensity ratios for T2 weighted and Short Tau Inversion Recovery images, Apparent Diffusion Coefficient values and patterns of enhancement may help to distinguish Kuttner's tumours from benign submandibular gland tumours, but not from malignant tumours. There do exist some very subtle morphological differences on magnetic resonance images of Kuttner's from those of malignancies.<sup>21</sup>

Surgery is the standard therapy for Kuttner's tumor. From a surgeon's perspective, we believe submandibular sialadenectomy offers the best treatment option. It removes the mass entirely and a very few to almost no post-operative complains have been received from patients who had undergone surgery. It is only upon postsurgical histopathology of the excised mass that the diagnosis is confirmed.

Histologically, CSS demonstrates a loss in acinar tissue, dense fibrosis, which is mainly periductal, and lymphocytic infiltration with lymphoid follicle formation in some cases.<sup>22</sup> According to Seifert CSS may progress through 4 different histological stages with different degrees of inflammation.<sup>23</sup>

Immunological studies have revealed an abundance of cytotoxic T cells especially near the acini and ducts while significantly lower proportion of B-cell population lacking Bcl-2 expression were restricted to areas of lymph follicles. The histopathological nature of the populations of T cells were more suggestive of an immune process triggered by intraductal epithelial agents.<sup>12,24</sup>

## CONCLUSION

So putting it in a nutshell we may conclude that an indurated submandibular mass with paucicellularity and scattered tubular ductal structures on FNAC with lymphoplasmacytic infiltration in a background of fibrous stroma and having diffuse heterogenous echogenicity and multiple hypoechoic shadows may be highly suggestive of Kuttner's tumor or CSS of the submandibular gland. Though it has been a diagnostic nightmare but now with improved imaging, a good image guided FNAC and a high degree of clinical suspicion this entity may be pre operatively distinguished from a malignancy saving the clinician and the patient from a lot of dilemma. However, histopathology and immunohistochemistry would be the key in establishing the diagnosis.

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