

CASE REPORT

Küttner's Tumor of the Submandibular Gland: A Rare Case Report

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ABSTRACT

Küttner's tumor (KT) remains an underdiagnosed medical identity and is often confused as a malignancy or a metastatic lymph node. It is a benign identity involving the salivary glands and is also known as chronic sclerosing sialadenitis (CSS). Histopathological examination of these tumors demonstrates features of chronic inflammation and fibrosis. We report a case of KT of the left submandibular gland in view of developing insights about diagnosing and managing this rare clinical entity.

Keywords: Chronic sclerosing sialadenitis, Kuttner's tumor, Salivary gland, Submandibular gland.

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INTRODUCTION

Küttner's tumor (KT) is a rare benign fibroinflammatory condition affecting the salivary glands, predominantly the submandibular gland. It has been also termed as chronic sclerosing sialadenitis (CSS). It was first described by H. Küttner, a German physician in 1896.¹ It is often clinically misdiagnosed as a malignant lesion owing to the hard indurated consistency on palpation. Histopathological examination holds the clue where lymphoplasmacytic infiltrates are seen surrounding the ducts along with periductal fibrosis in the background of fibrous stroma. Treatment includes surgical excision of the involved gland along with the mass. We report a case of KT of the left submandibular gland in a 30-year-old gentleman so as to raise awareness among the surgeons about this rare benign clinical entity.

CASE REPORT

A 30-year-old gentleman presented to our outpatient department with a slowly progressive painless swelling below the angle of left mandible of 3 months duration.

He was a chronic "paan" chewer (a preparation combining betel leaf, areca nut, and tobacco) for the past 10 years. There was no history of dryness or difficulty in opening the mouth. Local examination revealed a 4 × 3 cm, nontender bidigitally palpable swelling in the area of left submandibular gland with normal overlying skin. No cervical nodes were palpable. The examination of the oral cavity, right submandibular gland, and both the parotid glands did not reveal any abnormality. Systemic examination was normal, and blood investigations revealed normal parameters. A provisional diagnosis of malignancy of submandibular gland was made based on the hard submandibular swelling.

Ultrasonography (USG) of neck demonstrated well-defined heterogeneous lesion (Fig. 1) with ill-defined margins seen in the left submandibular region, measuring 2.2 × 2.1 cm, showing central vascularity (Fig. 2).



Fig. 1: Heterogeneous lesion in the left submandibular gland

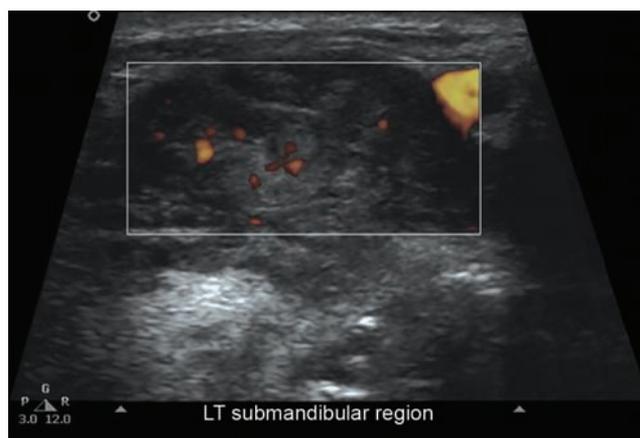


Fig. 2: Lesion showing central vascularity

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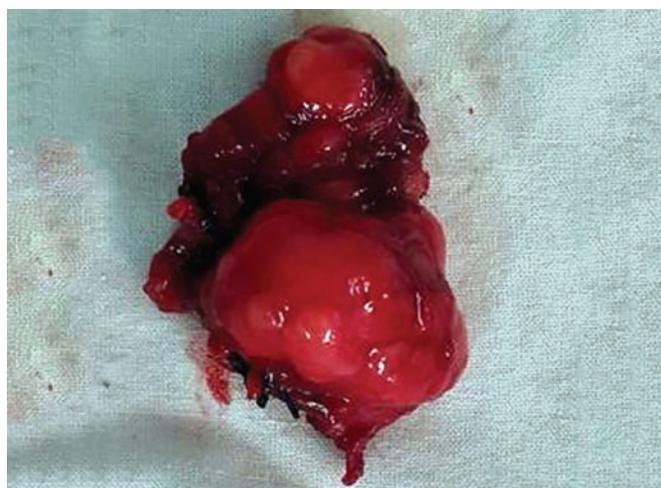


Fig. 3: En bloc excision of specimen

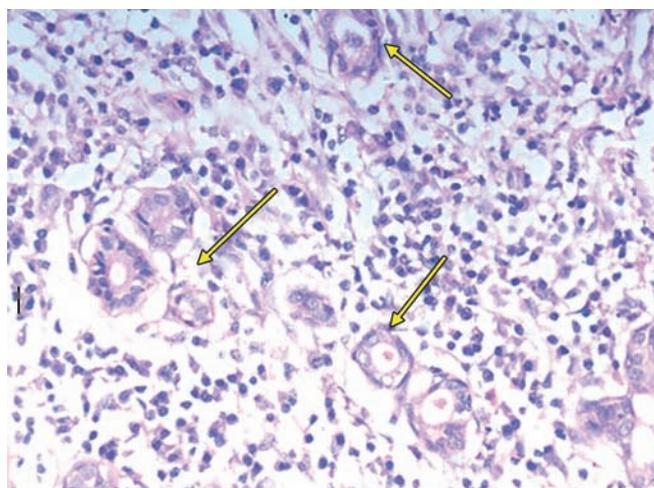


Fig. 4: Magnification showing plasma cells (40×)

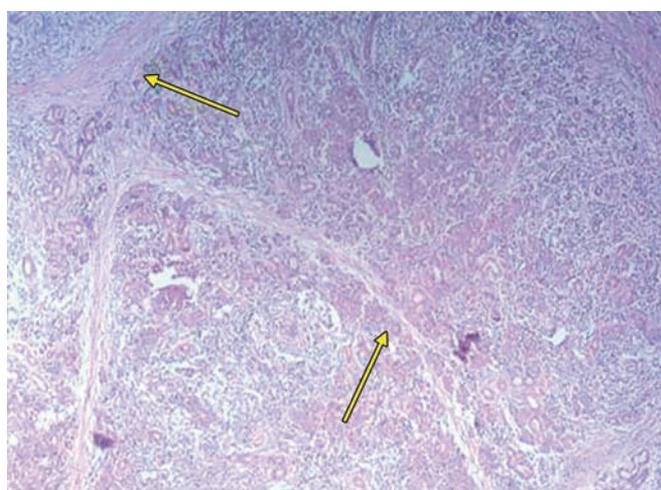


Fig. 5: Periductal sclerosis with interlobar inflammatory infiltrates

Thyroid gland, bilateral parotid gland, and right submandibular glands were normal.

The patient was posted for a left submandibular gland excision under general anesthesia. Intraoperatively, the mass was seen arising from the left submandibular gland measuring approximately 4 × 3 cm and was hard in consistency (Fig. 3).

Histopathological examination demonstrated seromucinous glands with interlobular and interlobar stroma showing chronic inflammatory cells in the background of fibrous stroma and periductal sclerosis (Figs 4 and 5). These features were consistent with KT.

DISCUSSION

Küttner's tumor also known as CSS was first described in 1986. It is a benign fibroinflammatory condition affecting the salivary glands, especially the submandibular gland. Clinical presentation may vary from an asymptomatic hard mass in the neck to recurrent episodes of postprandial pain. It usually affects males in the middle age and is very rarely seen in adolescents.

The exact etiology of KT has not been established till date; however, various theories like salivary duct obstruction, salivary stasis, sialolithiasis, secretory dysfunction, and autoimmune response have been proposed.² Immunoglobulin G4 disease has also been postulated to be one of the probable theories involved in KT.³

Apart from malignancy, the other differentials that should be considered for KT are simple chronic sialadenitis, granulomatous sialadenitis, Kimura disease, necrotizing sialometaplasia, sialolithiasis, sarcoidosis, Sjögren syndrome, inflammatory pseudotumor, benign lymphoepithelial lesions, and low-grade lymphomas.⁴

According to Seifert and Donath,⁵ the histopathological characteristics can be subcategorized into various stages based on the progressiveness and severity of inflammation. The various histological stages are listed as follows:

- *Stage 1:* Focal chronic inflammation with periductal lymphocytosis. The ducts are moderately dilated with inspissated secretion.
- *Stage 2:* Lymphocytic infiltration is denser and increases periductal sclerosis. Apart from inspissated secretion, the ductal system shows features of focal metaplasia and proliferation of ductal epithelium. Central fibrosis in the lobules may be seen with demonstrable acinar atrophy.
- *Stage 3:* Apart from increasing lymphocytic infiltration, atrophy of parenchyma of the glandular tissue, hyaline deposition around ducts, and sclerosis are seen. Ducts may show metaplastic squamous and goblet cells.
- *Stage 4 (end stage):* This is also termed as burnt-out phase, which demonstrates extreme sclerosis and parenchyma loss.

Earlier, it was conceptualized that KT only occurs in one submandibular gland, but recent reports have suggested that it occurs in bilateral submandibular glands and also in parotid glands. It had been also reported to affect submandibular and parotid gland simultaneously.

Malignant transformation is extremely rare; however, few cases have been reported demonstrating malignant transformation of these lesions.^{6,7}

Ultrasonography features suggest diffuse heterogeneous echogenicity with multiple hypoechogenic shadows. These features give a picture very similar to that of liver cirrhosis.

Treatment of choice for symptomatic lesions is surgical excision; however, in asymptomatic lesions, wait and watch policy also serves the purpose.⁸

CONCLUSION

A firm to hard submandibular swelling is often clinically diagnosed as a malignant lesion; however, CSS (KT) is a rare possibility upon histopathological confirmation. Symptomatic cases are best managed by surgical excision. The above case was presented aiming at developing insights about clinical presentation and diagnosis of such rare benign lesions.

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