Kuttner tumor (KT) also known as chronic sclerosing sialadenitis (CSS) was first described in 1896 by Kuttner in Germany. It is an uncommon, benign chronic inflammatory condition affecting submandibular gland. Kuttner tumor mimics a malignant neoplasm clinically because of presentation as hard swelling. This lesion has been recognized as a distinct clinicopathological entity in the latest edition of World Health Organization Classification of tumor like lesions of the salivary glands.6

In order to raise the awareness of this inflammatory condition, we report a case of Kuttner tumor of submandibular gland in a 50 year old female.

To the best of our knowledge this is the first case report in Indian literature.

Case Summary

A 50 year old female, nondiabetic, nonhypertensive presented with a painless, hard nodule involving the left submandibular gland since 15 months. There was no history of dryness of mouth. Local examination revealed a 2.0x1.8x1.3 cm, nontender swelling of left submandibular gland with normal overlying skin. There was a palpable single, small, mobile preauricular lymph node of 1 cm in diameter. 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. She was negative for both HBsAg and HIV. Clinically the diagnosis of malignancy of submandibular gland was based on hard submandibular swelling with lymphadenopathy and fine needle cytology (FNAC) was advised. As the FNAC did not yield any material, biopsy was carried out and sent for histopathology.

Pathological Findings

The biopsy specimen was grayish white, firm to hard, measuring 1.0x0.5x0.5 cm.

Microscopically the lobules showed dense lymphoplasmacytic infiltrate with acinar atrophy and periductal concentric fibrosis (Fig. 1) and lymphoplasmacytic inflammation with lymphoid follicle formation (Fig. 2). There was no evidence of lymphoepithelial lesions, granulomas, ductal atypia or malignancy.

Discussion

Kuttner tumor is an under recognized entity and still a rare lesion despite it has been known for more than a century; with only a few publications about this entity, to our knowledge, in the literature.1,5

This is a benign process of uncertain etiology. The mechanisms which have been postulated are sialolithiasis, secretary dysfunction with ductal inspissation, duct abnormalities,
infectious agents, and autoimmune reaction.4,5

Kuttner tumor usually presents more commonly in middle aged adult males with characteristic clinical presentation of firm or hard nodule in the submandibular gland.6 Our patient was a 50 years female who presented with a painless swelling of left submandibular gland. Microscopically there is loss of acini and ducts and eventually marked sclerosis of the salivary gland.3,5 Similar histomorphology was observed in this case.

The differential diagnosis of KT includes chronic sialadenitis, granulomatous sialadenitis, inflammatory pseudotumor, benign lymphoepithelial lesion and fibrohistiocytic tumors.2-5

Prognosis is very good as these are benign lesions that do not tend to recur.3,4 There have been reports nevertheless supporting the opinion that this condition may provide a state in which malignancy can arise.4,7 Patient was followed for 6 months and she was perfectly alright without any recurrence of tumor. Excision of affected gland should be performed as a treatment of KT. In our case following the diagnosis of KT the remaining gland was completely excised.

Conclusion

Kuttner tumor is a rare disease which mimics malignant neoplasm particularly in the submandibular gland. KT is usually diagnosed by histopathologist when lesion is surgically biopsied to confirm clinical diagnosis of malignancy.

References