Chronic Sclerosing Sialadenitis : An Under Recognized Entity for Salivary Gland Swellings

Dr. Bhuvan Nagpal
Consultant Oral Pathologist & Chief Administrator
Manglam Diagnostics
Hansi-125033 (Haryana)

Dr. Priyanka Mittal
Consultant Pathologist
Sapra Multi-Speciality Hospital
Hisar – 125005, Haryana

Dr. Tazeen Rahman
Consultant Dental Surgeon,
Patna-800014, Bihar.

Dr. Archana S.
Consultant Dental Surgeon & Oral Pathologist
Bengaluru, Karnataka.

Dr. Anupam Nagpal
Junior Resident
Post Graduate Institute of Dental Sciences (PGIDS)
Rohtak – 125005, Haryana

Dr. Anuradha Nagpal
Consultant General Physician
Hisar – 125005, Haryana

Abstract

Chronic sclerosing sialadenitis which is also known as Kuttner tumor is an uncommon chronic, benign, inflammatory lesion, involving submandibular salivary glands mostly. Clinically, it affects middle aged individuals majorly and presents as a firm to hard mass. Histologically, lobular architecture of the salivary gland is well preserved with thickening of interlobular septa by sclerotic tissue and preservation of ducts and periductal fibrosis. It is also characterized by dense lymphoplasmacytic infiltrate and variable loss of acini.

This article discusses in detail about chronic sclerosing sialadenitis and this lesion should be considered in the differential diagnosis for salivary gland swellings.

Key Words: Chronic sclerosing sialadenitis, Kuttner tumor, benign salivary gland lesion, hard palate, minor salivary gland, periductal fibrosis, lymphoplasmacytic infiltrate.

Introduction

Kuttner tumor (KT) is a benign tumor like lesion involving the salivary gland with unknown etiology. It was first described in 1896 by a German physician, Dr. H Kuttner.[1]

The first case of KT was reported a century ago, only a few publications about this entity exists in the literature.[5-9] It has recently been stressed that this lesion is an under recognized entity and still a rare lesion.

Clinical Presentation

It presents commonly in middle aged adult males.[8,9] Clinically, it presents as stony hard mass which mimics malignant lesion. Most of the cases are asymptomatic. It affects mainly submandibular salivary gland, but some cases of parotid gland, minor salivary glands and bilateral submandibular and lacrimal gland involvement have been reported in the medical literature.[1,2] Literature reports few cases of regional lymph node adenopathy in Kuttner tumor of submandibular gland.[4] The characteristic clinical presentation is firm or hard swelling affecting submandibular gland predominantly. It varies from being asymptomatic to recurrent painful swelling.[8]

There is unilateral involvement of the gland more commonly, but less frequently both glands as well as the parotid and other minor salivary glands can be involved.[6,8,9,11,12]

A case that affected submandibular and lacrimal glands bilaterally has also been described.[6]

Fig. 1 showing submandibular gland swelling

Etiopathogenesis

The etiology of the lesion is uncertain, which includes sialolithiasis (in 29-83% of the affected glands), secretory dysfunction with ductal inspissations, abnormalities of duct, infective agents and autoimmune reaction.[3,8,9] It was suggested that immunoglobulin G4 (IgG4) antibodies are involved in pathogenesis of sclerosing sialadenitis.[3]

Histopathological features

There is marked sclerosis of salivary gland, periductal fibrosis, dense infiltration with lymphocytes along with lymphoid follicles, loss of acini, acinar atrophy, ductal squamous metaplasia and sialoliths in salivary ducts.[2,4] The Histopathologic features of KT may evolve through four histologic stages according to Seifert: Stage 1: There is mild, focal chronic lymphoplasmacytic cell infiltration, usually in periductal area with periductal fibrosis.

The lobular architecture of the gland is preserved. Stage 2: Loss of lobular architecture with dense lymphocytic infiltration and severe periductal fibrosis.

Stage 3: More prominent lympho-plasmacytic infiltrate with reactive lymphoid

follicle formation, extensive fibrosis with acinar atrophy, periductal hyalinization and ductal dilatation. Stage 4: destruction of lobular architecture with sclerosis and parenchyma loss.[8,10,15]The features on FNAC suggestive of KT are low cellularity, scattered ductal structures with paucity or absence of acini, ducts intimately surrounded by fibres or lymphoid cells, fragments of fibrous stroma and moderate to large number of lymphoid cells lacking atypia.[14] Immunohistochemical markers used are CD 4, CD 8 and CD 20. CD 4 is expressed on the surface of T helper cells/lymphocytes, monocytes, macro-phages and dendritic cells. CD 8 is also a T cell receptor. It is expressed on the surface of cytotoxic T cells but also found on the natural killer cells and dendritic cells. CD 20 is expressed on the surface of all B cells beginning at pro B phase and increasing in concentration until maturity.[13,16]

Fig. 2A (H & E, 40X) shows dense infiltration with lymphocytes with lymphoid follicles; 2B (H & E, 100X) shows periductal fibrosis

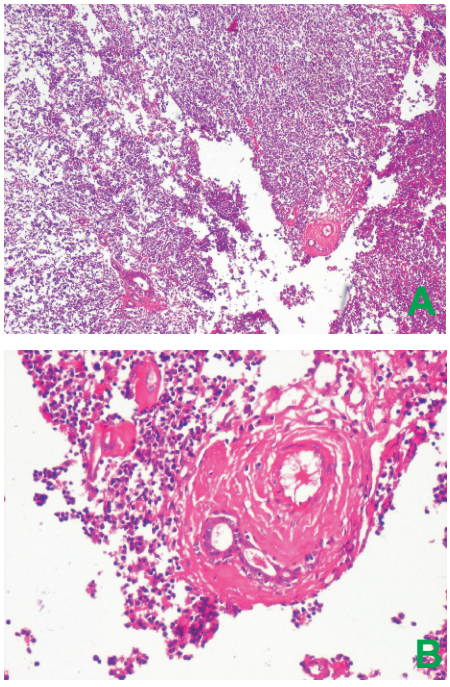
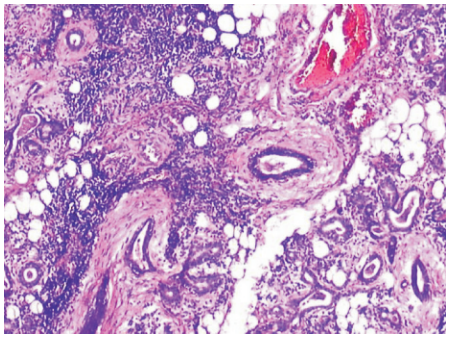


Fig. 3 (H & E, 100X) showing periductal fibrosis, dense infiltration with lymphocytes, blood vessels, acinar atrophy and ductal squamous metaplasia



Treatment

Management of KT can be done through conservative method by adopting "wait and watch" approach in cases where the mass is otherwise asymptomatic. In symptomatic cases, the mass is surgically excised. Prognosis is good as it is a benign fibro inflammatory lesion that has fewer tendencies to recur.[17]

Conclusion

Kuttner tumor is a rare and underrecognised entity which mimics malignancy usually affecting submandibular gland, but may also involve other salivary glands. Hence, it should be considered in the differential diagnosis of firm to hard swellings of the salivary glands. With increased awareness of this disease and support of FNAC findings, an accurate preoperative diagnosis is possible, but has to be confirmed by histopathological findings and by immunohistochemical examination in certain cases.

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