Kuttner’s Tumor of Submandibular Gland: A Case Report

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ABSTRACT

Kuttner’s tumor is called chronic sclerosing sialadenitis, a benign inflammatory condition affecting the salivary gland. It mimics a malignant neoplasm clinically because of its presentation as a hard mass. This is an under-recognized entity in the surgical and pathological literature.

Kuttner’s tumor was first described by Kuttner, a German physician, more than a century ago, in 1896.1

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. We are presenting here a case of Kuttner’s tumor of the submandibular gland.

CASE DESCRIPTION

A 71-year-old male patient came to our ENT outpatient department with complaints of swelling in the right side of neck for 2 months. It was started with a small swelling which enlarged progressively to reach the present size. The swelling was painless and not associated with fever, cough, etc. The patient was hypertensive and nondiabetic.

On local examination, there was a swelling of size 4 cm × 4 cm confined to the right submandibular triangle. The mass was firm in consistency, nontender with irregular surface, and not fixed to the skin or underlying structures. The mass was palpable bimanually. Skin over the swelling was normal. Clinically, the patient was diagnosed as “chronic submandibular adenitis” and sent for fine needle aspiration cytology (FNAC) and ultrasonogram (USG) of the neck. FNAC came as chronic adenitis of submandibular gland (right). USG neck came as submandibular adenitis (right). So the right submandibular gland was excised under general anesthesia and sent for the histopathological study. The microscopic study reported as Kuttner’s tumor (chronic sialadenitis with sialometaplasia). This is a rare benign condition of the submandibular gland simulating a tumor and is because of chronic inflammation. This case is presented here because of its rare occurrence.

Keywords: Salivary gland, Sialoadenitis, Sialometaplasia.


INTRODUCTION

Kuttner’s tumor is otherwise called chronic sclerosing sialadenitis, a benign inflammatory condition affecting the salivary gland. It mimics a malignant neoplasm clinically because of its presentation as a hard mass. This is an under-recognized entity in the surgical and pathological literature.

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Clinically, the patient was diagnosed to be chronic submandibular adenitis and sent for fine needle aspiration cytology (FNAC) and ultrasonogram (USG) of the neck.

The FNAC report came as chronic adenitis of the right submandibular gland. Ultrasound neck report came as submandibular adenitis (right).

So we planned for excision of the right submandibular gland under general anesthesia. A 10 cm long incision was given about 1 inch below the lower border of the mandible in a skin crease. The incision was deepened and the platysma muscle was incised. The skin flap along with the platysma muscle was elevated superiorly and inferiorly. The tumor mass was identified. A horizontal incision was given over the mass at the level of the hyoid bone and the capsule was separated. Then the skin, platysma along with the gland capsule were elevated up to the lower border of the mandible to avoid injury to the mandibular branch of the facial nerve. The gland was mobilized by ligating the facial artery at two sites, that is, posterior to the gland and superiorly near the lower border of the mandible. Digastric muscle, hypoglossal nerve, and lingual nerve were identified and the gland along with the mass was removed by ligating and dividing the submandibular duct (Fig. 1). The wound was closed in layers.

The mass along with the gland was sent for the histopathological study. The postoperative period was uneventful. The patient was discharged on the 7th postoperative day after removal of sutures.

Histopathological report showed that there were multiple sections revealing salivary tissue, majority of the salivary tissue showing cystically dilated spaces filled with mucinous secretions and the gland showing squamous metaplasia. Stroma revealed plenty of lymphocytic aggregates and congested vessels. This microscopic feature is suggestive of chronic sialadenitis with sialometaplasia, i.e., Kuttner’s tumor (Fig. 2).
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Discussion

Kuttner's tumor is a chronic inflammatory condition commonly found in the submandibular gland either unilaterally or bilaterally. But it is also found in other major and minor salivary glands.

In general, salivary gland tumors are relatively rare (approximately 2.5–3 occurrences per 100,000 population per year) seen in the Western world. Salivary gland malignancy account for 3–5% of all head and neck cancers. Approximately 20–25% of parotid tumors, 35–40% of submandibular tumors and more than 90% of sublingual gland tumors are malignant. This underscores the clinical diagnosis of Kuttner's tumor. Being a benign condition, the tumor mimics a malignant tumor of the salivary gland.

Kuttner's tumor was first described by Kuttner more than a century ago in 1896. Chan described it as one of the most common diseases affecting the submandibular gland. Surprisingly, it is still under-recognized, and there are few publications on this entity in the English-language literature.

Kuttner's tumor is a benign lesion of uncertain cause. Postulated mechanisms of the tumor include sialolithiasis (demonstrated in 29–83% of the affected glands), dysfunctional secretory function leading to ducral inspissations and autoimmune reaction. The presence of abundant immunoglobulin G4 (IgG4) associated with plasma cells infiltrating into the salivary glands, as well as serum IgG4 concentration has been noted in patients with Kuttner's tumor. However, in recent times, administration of steroids, which shrinks the inflammatory lesion and is known to reduce the serum IgG4 level, has been found to be useful in younger patients or those who refuse surgery.

So Kuttner's tumor is called chronic sclerosing sialadenitis which simulates malignant tumor especially when presented in elderly patients. We should be able to diagnose the condition by using preoperative USG, FNAC and postoperative histopathological study. Till now a large number of cases are under-reported and under-recognized.

Acknowledgments

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References


