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CASE REPORT

Küttner's tumor with parotid gland involvement

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Abstract

Küttner's tumor, or chronic sclerosing sialadenitis (CSS), is a pathological process characterized by fibrosis and inflammation of the salivary glands. It is a rare disease that most commonly affects the submandibular glands. We report the case of a 36-year-old woman presented with palpable nodule in the right pre-auricular region for over a year. Ultrasound examination showed a 4 cm hypoechoic nodule in the right parotid gland. Fine needle aspiration biopsy (FNAB) presented negative result for malignancy. Before undergoing partial parotidectomy, the main diagnostic hypothesis was pleomorphic adenoma. Histopathology findings were suggestive of CSS.

Keywords: parotid diseases; parotid gland; parotid neoplasms

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Introduction

Küttner's tumor, or chronic sclerosing sialadenitis (CSS), is an inflammatory and fibrotic process of the salivary glands more frequently affecting the submandibular glands[1]. Involvement of the parotid glands is even rarer[2].

The affected glands become hardened and swollen, simulating tumors clinically. Current studies suggest a pathogenic mechanism for this disease, including this entity in the spectrum of immunoglobulin G4 (IgG4)-related sclerosing diseases[1,3].

We report a case of CSS with involvement of the parotid gland, one of the rarest locations for occurrence of this disease.

Case report

A 36-year-old female patient, previously healthy, with a complaint of a palpable nodule in the right pre-auricular region for one year. The ultrasound examination (US) showed a gnarled, hypoechoic nodule with a larger size of 4 cm in the right parotid gland. Fine-needle aspiration puncture (FNAP) presented negative result for malignancy. The patient was then submitted to partial parotidectomy following the diagnostic hypothesis of pleomorphic adenoma. At surgery, a hardened and poorly delimited lesion was observed in

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the superficial region of the parotid gland. The lesion had a neoplastic aspect, was in close contact with the upper trunk of the facial nerve, and presented thickening and absence of the cleavage plane with the nerve (Figure 1A). It was completely resected, preserving the upper trunk of the facial nerve (Figure 1B). Postoperatively, the patient presented with transient delayed facial nerve palsy and recovered all movements one month after surgery. The anatomopathological examination reported extensive areas with loss of the lobular pattern that had been replaced by dense hyalinized collagen, which presented ductal structures with low epithelium and micro-epithelial hyperplasia, lymphoplasmacytic exudation forming aggregates, and atrophy of the parotid parenchyma was identified surrounding the lesion, suggestive of chronic sclerosing sialadenitis (CSS) (Küttner's tumor) - Figure 2. The diagnosis of CSS was confirmed by immunohistochemistry (IHC). The patient is well and presents no evidence of disease recurrence. She denied any previous episodes of sialadenitis or symptoms such as xerostomia or pain in the right parotid gland.

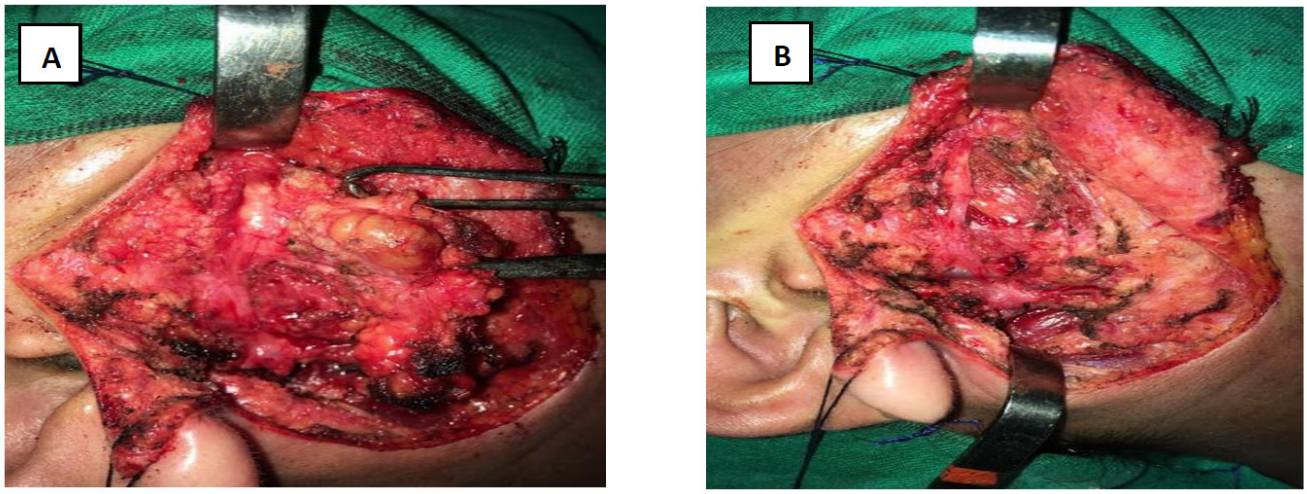

Figure 1. A - 2.4 cm lesion in the right parotid gland. **B -** Immediate postoperative after resection of the lesion.

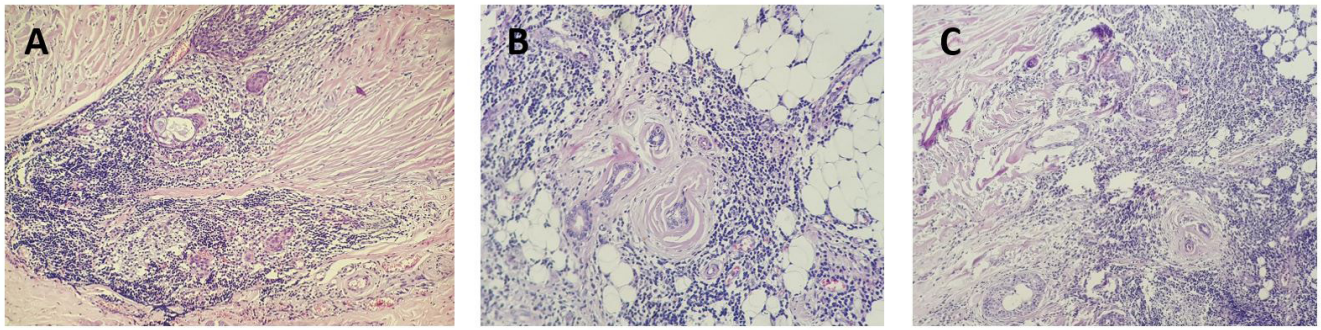

Figure 2. A - Intermediate 10x enlargement of the parotid gland. Dense lymphomononuclear infiltrate associated with fibrosclerosis and severe atrophy. **B -** Intermediate 10x enlargement of the parotid gland. Highlight of the inflammatory infiltrate forming periductal aggregates associated with intense atrophy. **C -** Intermediate 10x enlargement of the parotid gland. Highlight of the sclerosis area on the left of the photo and the dense lymphomononuclear inflammatory infiltrate on the right, associated with atrophy.

Discussion

Küttner's tumor, also known as chronic sclerosing sialadenitis (CSS), was initially described by Hermann Küttner in 1896, who reported four cases of patients with a hardened and painful mass on the submandibular gland topography[4]. It was described as a terminal fibrotic result of chronic sialadenitis in a major submandibular salivary gland[3]. The affected glands become hardened and swollen, simulating tumors clinically. It mainly affects adults and there is a slight predominance in males. The first changes in the gland include periductal lymphoplasmacytic infiltrate followed by periductal fibrosis. This process progresses over time and eventually replaces the entire lobe or group of lobules. It causes acinar atrophy and lymphoid follicles are frequently found[3].

Current studies suggest an autoimmune pathogenic mechanism for this disease, including this entity in the spectrum of immunoglobulin G4 (IgG4)-related sclerosing diseases[1]. CSS can occur as a localized or systemic process with bilateral involvement of the salivary glands[1]. It may be associated with sclerosing cholangitis, retroperitoneal fibrosis, or sclerosing pancreatitis, all of which are IgG4-related sclerosing diseases[5]. The submandibular gland is the most common site of this entity, with rare reports of occurrence in the parotid gland[2].

The most common histological findings are lymphoplasmacytic inflammation, fibrosis, and acinar atrophy to varying degrees. Sclerotic areas are more evident between the lobes and intralobular ducts, with formation of thick fibrous trabeculae. Inflammation tends to form periductal aggregates. Follicles and lymphoid aggregates with expanded germinal centers are common. When there is marked acinar atrophy, acini are completely absent. Ducts and acini often appear ectatic. The ductal epithelium is prone to metaplastic changes, including squamous and mucinous cell metaplasia. Oncocytic changes are more common in the minor salivary gland. Granulomatous inflammatory response due to extravasation of saliva secondary to duct rupture may also be observed[4]. The patient's histological findings are shown in Figure 2.

Clinically, CSS manifests as a predominantly unilateral palpable mass of the affected salivary glands that can be symptomatic (pain and excessive salivation) and, many times, difficult to differentiate from salivary gland neoplasms[2]. In this case reported, it manifested itself as a unilateral, painless mass without excessive salivation.

Due to the difficulty in differentiating it from neoplasms, treatment usually involves surgical excision of the affected gland(s), and there may be transient or permanent paralysis of the facial nerve ipsilateral to the affected gland, considering the possibility of invasion of the facial nerve by the Küttner's tumor or as a result of nerve manipulation during surgery. In addition to this type of treatment, there are reports of patients undergoing corticotherapy in the literature. Most patients present good prognosis after treatment[1,2].

The literature presents few cases of Küttner's Tumor involving the parotid gland(s). It is worth highlighting that this case report involved a female patient, although higher prevalence of CSS has been observed in males. The clinical findings of this case report largely corroborate the analyzed cases described in the literature. In addition, some characteristics such as absence of permanent

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sequelae after surgical treatment, despite the involvement of the facial nerve, reinforce the relevance of the present case report.

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