Case Report / Olgu Sunumu

Oncocytoma of the parotid gland complicated by hypercalcemia: a case report

Hiperkalsemi ile komplike parotis bezinin onkositomu: Olgu sunumu

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ABSTRACT

Salivary gland tumors are rare head and neck tumors. The majority of these tumors are benign and include pleomorphic adenoma, monomorphic adenoma, oncocytoma, and papillary cystadenoma lymphomatosum. Oncocytoma is a rare benign salivary gland tumor. In this article, we report a 69-year-old female case of oncocytoma of the right parotid gland in whom fine needle aspiration cytology result was reported as a Warthin's tumor.

Keywords: Hypercalcemia; oncocytoma; salivary gland.

ÖZ

Tükürük bezi tümörleri, nadir baş ve boyun tümörleridir. Bu tümörlerin büyük bir çoğunluğu iyi huylu olup pleomorfik adenom, monomorfik adenom, onkositom ve papiller kistadenom lenfomatozumu içermektedir. Onkositom nadir iyi huylu bir tükürük bezi tümörüdür. Bu yazıda, ince iğne aspirasyon sitoloji sonucu Whartin tümörü olarak bildirilen, sağ parotis bezinde onkositom olan 69 yaşında bir kadın olgu sunuldu.

Anahtar Sözcükler: Hiperkalsemi; onkositoma; tükürük bezi.

Oncocytomas are rare tumors of the neck and head region that account for nearly 2% of benign salivary gland tumors. They are seen most often in the sixth decade of life with no clear sex predilection.[1] Oncocytomas affect the parotid gland in 85% to 90% of cases.[2] The superficial lobe of the parotid gland is primarily involved with almost benign nature.[3] These tumors may indicate an underlying metabolic disorder with malignant potential depending on the origin. Herein, we present a case of a parotid oncocytoma with associated undiagnosed hypercalcemia that resolved after surgery.

CASE REPORT

A 69-year-old female patient was admitted to our clinic with a history of hemicolectomy due to gastrointestinal cancer 10 years ago. Positron emission tomography computed tomography that was performed due to suspected metastatic disease showed no metastatic finding. However, a hypermetabolic lesion was viewed in the right

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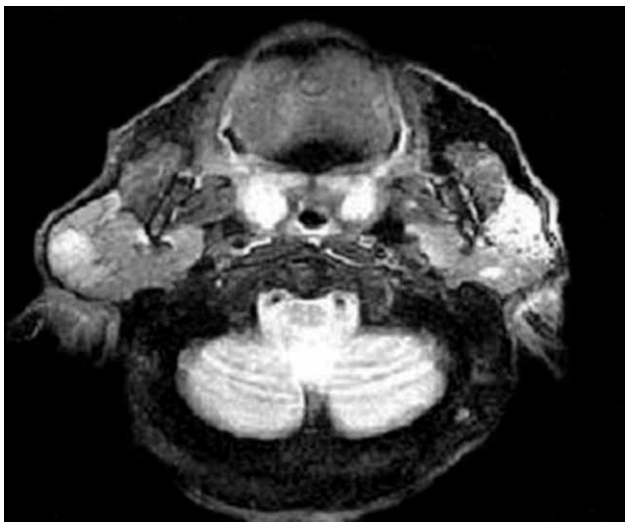

Figure 1. *Axial magnetic resonance image reveals a mass in the right superfacial parotid gland.*

parotid gland. Physical examination revealed a 3x2 cm irregular and painless mass on palpation, which had grown within the past few months. Facial nerve examination was normal. Magnetic resonance imaging showed a 24x16 mm mass of the right superficial parotid lobe superolateral in the largest transverse diameter on fat-suppressed T_2-weighted images (Figure 1). Fine needle aspiration biopsy (FNAB) showed findings consistent with a Warthin's tumor. Based on the patient-reported symptoms including fatigue, constipation, diarrhea, and urinary frequency, the patient was diagnosed with hypercalcemia (12.5 mg/dL) at the internal medicine outpatient clinic. However, parathyroid hormone levels were found to be normal, indicating no hypercalcemia etiology. She was administered calcitonin (5 IU/kg).

The patient was operated on for a benign tumor of the parotid gland. The mass was excised through superficial parotidectomy under general anesthesia, preserving the facial nerve. Pathological examination of the tissue sections was reported as a neoplasm (oncocytoma) which was isolated by a thin fibrous capsule and contained oncocytic cells with microfollicular appearance in the surrounding tissues. No capsular or vascular invasion was observed (Figures 2, 3). Chemotherapy and radiation therapy were not recommended. No recurrence was seen during a 12-month follow-up period postoperatively. Calcitonin treatment was reduced by titration by the treating physician. Hypercalcemia resolved postoperatively (9.2 mg/dL).

DISCUSSION

Oncocytic lesions of the salivary glands were first reported by Schaffer in 1897.[4] These lesions may affect kidneys, pancreas, thyroid gland, pituitary gland, lacrimal gland and salivary gland. It has been considered that oncocytes arise from normal and abnormal mitochondria clusters of the acidophilic granules in their cytoplasm, thereby indicating abnormal or impaired metabolic-respiratory function.[3]

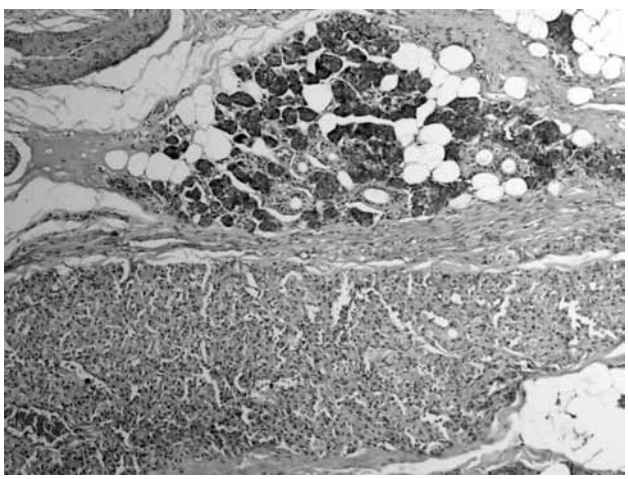

Figure 2. *Normal salivary gland (above), oncocytoma (below) with a fibrous capsule separating two components (H-E x 100).*

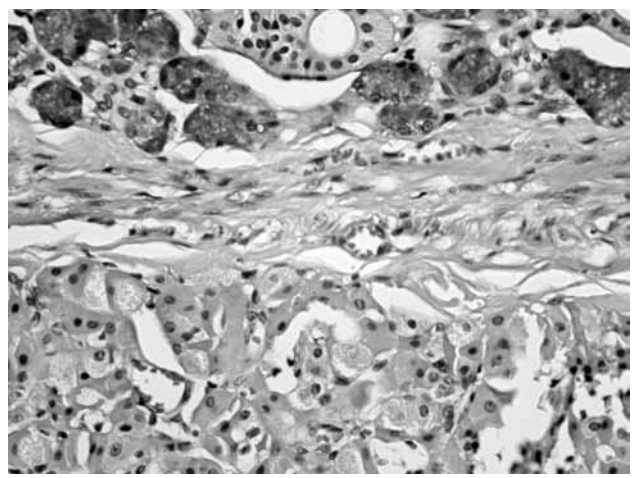

Figure 3. *Gross examination of a normal salivary gland (above) and oncoyctoma (below) with a fibrous capsule separating two components. A neoplasm containing large eosinophilic circular cytoplasma with moderately marked nucleoli and mild hyperchromatic, centrally localized nucleoli oncocytic cells (H-E x 400).*

Oxidative phosphorylation defects have been also reported in mitochondria of these cells.[5]

Oncocytomas primarily affect the parotid salivary gland. Oncocytic metaplasia increases with aging.[6] They are usually unilateral, limited and capsulated with a size of ≥3 cm. Despite their mostly asymptomatic nature, pain is severe in symptomatic patients.

Fine needle aspiration biopsy is a valid preoperative diagnostic method for masses of the salivary gland. Its diagnostic sensitivity has been reported to be 80.9% in the literature. In our case, FNAB showed findings consistent with a Warthin's tumor. This can be attributed to the fact that oncocytomas are rarer tumors of the salivary gland and oncocytomas, pleomorphic adenomas, and Warthin's tumor share common tumor histology.[7]

Surgical excision is the optimal treatment method for oncocytomas. The salivary gland should be excised simultaneously if oncocytomas invade or are associated with the parotid and submandibular salivary gland. Almost no recurrence is seen.[6] In our case, the mass was excised through superficial parotidectomy under general anesthesia, preserving the facial nerve. No recurrence was observed.

Hypercalcemia is a serious complication of several malignant and benign diseases. Malignant diseases and primary hyperparathyroidism may present in over 90% of patients. Squamous cell endometrial, breast, and renal carcinomas may also develop. Although the disease may be asymptomatic depending on the severity of hypercalcemia, it may present with varying symptoms such as nausea, vomiting, dehydration, and even death.[8] Nearly 20 to 30% of all cancer patients have been reported to have hypercalcemia. Of these patients, 80% have hypercalcemia induced by parathyroid hormone-related protein released by tumor cells. This is termed as humoral hypercalcemia of malignancy. In a study, half of the cancer patients with hypercalcemia died within 30 days due to high calcium levels.[9,10]

In our case, hypercalcemia with unknown etiology resolved following parotidectomy. To the best of our knowledge, there is no case report of parotid gland oncocytoma-induced hypercalcemia in the literature. However, Popalzai et al.[11] reported a case of metastatic mucoepidermoid carcinoma-induced hypercalcemia.

In conclusion, patients with hypercalcemia should be examined for a mass of the salivary gland. Oncocytoma should be also considered in such patients with a mass. Our case was previously diagnosed with idiopathic hypercalcemia followed by oncocytoma that was treated with medical therapy. This indicates that the designation of idiopathic should be made more carefully after secondary causes are ruled out.

Declaration of conflicting interests

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