

Oncocytoma of the Parotid Gland in an Adolescent Female: Case Report

Adölesan Kız Çocuğunda Görülen Parotis Bezi Onkositomu

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ABSTRACT Oncocytoma is an uncommon, benign salivary neoplasm composed of mitochondria-rich oncocytes. It usually occurs in the parotid gland, seldom in submandibular gland and generally account for less than 1% of all salivary gland tumors. They are typically tumors of older adults with a slight female predominance. Clinical and radiological features of the parotid oncocytoma are not different from those of the others parotid benign tumors. Clinical diagnosis for an oncocytoma is challenging for its similar features to other benign tumours. Histopathological diagnosis is reliable with histochemical and electron microscopic conformation of the oncocytes. Surgery is the best treatment option. We report a rare presentation of oncocytoma of the parotid gland in a young adolescent female who presented with progressive swelling in the left parotid region.

Key Words: Parotid neoplasms; adolescent; parotid oncocytoma

ÖZET Onkositom, mitokondriden zengin onkositlerden oluşan ve nadir görülen benign bir tükrük bezi tümörüdür. Tüm tükrük bezi tümörlerinin %1'inden azını oluşturur ve genellikle parotis bezini ve daha nadir olmak üzere submandibuler bezi tutar. Onkositomer hafif bir kadın üstünlüğü ile birlikte tipik olarak yaşlı yetişkinlerde görülür. Parotis bezi onkositomunun klinik ve radyolojik özellikleri parotisin diğer benign tümörlerinden farklı değildir. Onkositomun klinik tanısı diğer benign tümörlerle olan benzerliği nedeniyle güçlük arz eder. Histopatolojik tanı onkositlerin histokimyasal ve elektron mikroskopik olarak gösterilmesi ile konulabilir. Cerrahi en iyi tedavi seçeneğidir. Sol parotis bölgesinde gittikçe büyüyen şişlik ile başvuran adölesan kız çocuğundaki parotis onkositomunu nadir görülmesi sebebiyle burada sunmayı uygun bulduk.

Anahtar Kelimeler: Parotis tümörleri; adölesan; parotis onkositoma

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Oncocytes are epithelial cells with granules, abundant, eosinophilic cytoplasm due to presence of numerous large mitochondria. They are present in exocrine and endocrine glands, such as thyroid, pituitary gland, pancreas, lacrimal gland, kidneys and salivary glands.¹

In salivary gland tumors, oncocytic neoplasms are found rarely and account for <1% of all salivary gland tumors.² Oncocytic neoplasms include oncocytic hyperplasia, benign oncocytoma and rarely oncocytic carcinoma. The imaging findings of oncocytic metaplasia of the salivary gland have only limited description. Also; diagnosis by fine needle aspiration biopsy (FNAB) may be very difficult due to focal sampling of the lesion, as on-

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cocytic change can occur in a large variety of neoplastic as well as non-neoplastic conditions. Standard treatment of oncocytoma is surgical excision with a margin of normal tissue. Here, we present a rare case of 14-year-old female who presented with a 4 cm diameter swelling in the left parotid region which diagnosed oncocytic lesion. Written informed consent was obtained from the parents of the patient for publication of this case report and accompanying image.

CASE REPORT

A 14-year-old female patient presented with a 6-month history of painless mass in the left parotid gland region. Physical examination revealed the swelling was firm, non-tender or not fixed to the skin or deeper structures. No facial nerve dysfunction or cervical lymphadenopathy was found. She was referred to our clinic from another hospital with a magnetic resonance imaging (MRI). MRI revealed that a 22-mm hyperintense mass located in superficial part of parotid gland (Figure 1). FNAB was done and epithelial cells and histiocytes were seen in the aspirate. The patient underwent superficial parotidectomy. Microscopic examination of hematoxylin-eosin-stained formalin-fixed paraffin-embedded sections showed epithelial cell proliferation; cells were characterised by small round



FIGURE 1: Axial section of magnetic resonance imaging reveals hyperintense tumour mass of the left parotid gland.

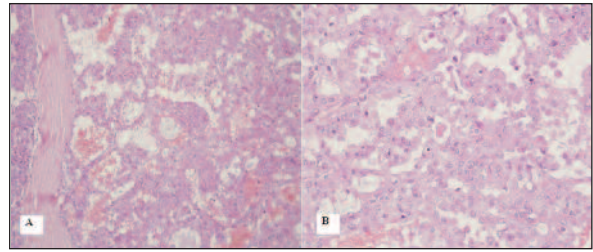


FIGURE 2:

A: Tumor is separated with a capsule from salivary gland tissue (HE, x100)
B: Tumor composed of epithelial cells with granular, abundant, eosinophilic cytoplasm (HE, x 400).

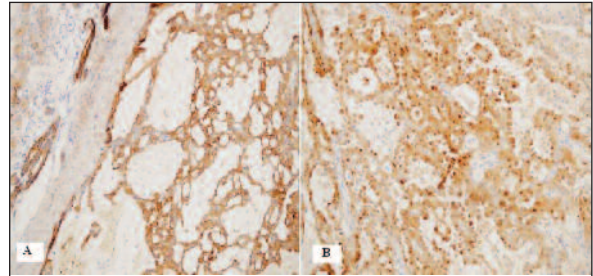


FIGURE 3:

A: Tumor cells stained positive for S-100 (S-100X200).
B: Tumor cells stained positive for cytokeratin (CytokeratinX200).

nuclei and microgranular, eosinophilic cytoplasm. A mitotic count was negative. The mass was surrounded by a thin fibrous capsule. Tumor cells stained positive for S-100 and cytokeratin. These findings were consistent with oncocytoma of the parotid gland (Figure 2, 3). No complications were recorded during the post-operative course. There was no recurrence three years after surgery.

DISCUSSION

Oncocytes are cells which can be seen in various tissues and a variety of conditions ranging from hyperplastic changes to malignant conditions. In 1894, Hurtle first described these granular cells in normal canine thyroid. The term "oncocyte" was coined by Hamperl in 1931.² Tandler et al. revealed by electron microscopy that the oncocytes contained large number of mitochondria.³ Oncocytic cells are thought of as metaplastic cells formed in response to adverse changes such as aging. Studies have showed that solitary oncocytes appear in aging sa-

linary tissue.⁴ According to authors, Warthin tumor can be differentiated from oncocytoma. Both Warthin tumors and oncocytomas show intense uptake of the nuclide on technetium-Tc99m perchnetate radionuclide scans of the salivary glands.

World Health Organisation defines 3 categories of oncocytic neoplasms of the salivary gland: nodular oncocytic hyperplasia (or oncocytosis), oncocytoma, oncocytic carcinoma. Oncocytosis is considered a hyperplastic change which may present with generalized enlargement of salivary gland. Oncocytoma is well-circumscribed, encapsulated benign tumor composed of monotonous sheets of oncocytes, frequently with a central scar. Oncocytic carcinoma is the rarest of the oncocytic neoplasms. It accounts for 5% of all oncocytic tumors and less than 1% of all salivary gland tumors.⁵

Oncocytomas are rare tumors in the head and neck region. They occur commonly in salivary glands, especially in parotid glands and rarely affect the submandibular gland. Bilateral oncocytoma is reported to be extremely rare. Uzunkulakoglu et al. reported a case of bilateral oncocytoma of parotid gland.⁶ The prevalence of oncocytoma is 0.5% to 1.2% in parotid neoplasms. Oncocytomas usually occur in the elderly and affect the parotid glands in 80%.⁷ Their occurrence in young individuals is rarely seen. On review of literature, submandibular oncocytoma in an adolescent has rarely

been reported.⁸ This case is unique because of the younger age, as the patient was a 14-year-old female with a parotid gland oncocytoma. Oncocytomas present as slow growing unilateral mass in the age group of 50-70 years with a slight female preponderance. The aging mitochondrial hypothesis could explain the predilection between 50-70 years.

Oncocytic lesions demonstrate decreased signal intensity on both T1 and T2- weighted MR images, attributed to the high cellularity and low free water content. Fine needle aspiration cytology has increasingly been used a diagnostic tool for salivary gland tumors with high sensitivity and specificity. But, diagnosis by cytology alone often become difficult because of histopathological variety of salivary gland tumors.^{9,10} Surgical management with radical or superficial parotidectomy represents the cornerstone of therapy. Recurrence is less than 20%, mainly because of incomplete surgical resection.^{7,10} After parotidectomy for oncocytic lesions, patients should be followed for recurrence arising from residual parotid tissue. In our case, there was no recurrence three years after surgery.

In conclusion, oncocytic neoplasms should be considered as a possible diagnosis in patients with parotid enlargement. Due to the lack of large series, assiduous study of the cases reported in the literature may lead to better understanding of this rare disease.

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