Olgu Sunumu
Case Report

Psammoma body–rich acinic cell carcinoma of maxillary sinus

MAKSİLLER SİNÜSÜN PSAMMOM CİSMİNDEN ZENGİN ASİNİK HÜCRELİ KARSİNOMU

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ABSTRACT

Acinic cell carcinoma (ACC) is a rarely seen, slowly growing, low-grade malignant neoplasia of the salivary gland. ACC is most frequently detected in the parotid gland, rarely it is localized in a minor salivary gland. Rarely psammoma bodies are seen in neoplasic, and nonneoplasic lesions of the salivary gland. Herein a case with psammoma body-rich ACC is presented together with its differential diagnosis. A 59-year-old male patient consulted our hospital with complaint of stuffy nose persisting for a year. His radiological examination revealed the presence of a mass lesion in the maxillary sinus which expanded neighbouring bony structures. Macroscopically a tumoral mass with a diameter of 6 cm was observed in maxillectomy specimen in sections prepared from the tumor, acini-like structures containing microcalcifications were observed. Tumor cells had basophilic granular cytoplasm, and round or oval shaped nuclei. Tumor cells stained with PAS and dPAS demonstrated cytoplasmic granules. The case was reported as "psammoma body-rich ACC".

Keywords: acinic cell carcinoma, maxilla, psammoma body

ÖZ

Asinik hücreli karsinom (AHK) nadir görülen, yavaş büyüme hızına sahip, düşük dereceli malign tükürük bezi neoplazisidir. Tümör en sık parotiste saptanmakta olup minör tükürük bezi yerleşimi nadirdir. Tükürük bezinin neoplastik ve nonneoplastik lezyonlarında psammom cismi varlığı nadirdir. Burada maksiller sinüs yerleşimli, psammom cisimlerinden zengin, AHK olgusu ayırıcı tanıları ile birlikte sunulmaktadır. 59 yaşında erkek hasta bir yıldır devam eden burun tıkanıklığı şikâyeti ile hastanemize başvurdu. Hastanın radyolojik görüntülemelerinde maksiller sinüsde, komşu kemik yapılarda ekspansiyona neden olan kitlesel bir lezyon görüldü. Maksillektomi materyalinde makroskopik olarak 6 cm çapındaki tümör izlendi. Tümörden hazırlanan kesitlerde, arada psammom cismi özelliğinde mikrokalsifikasyonlar içeren, asinüs benzeri yapılar izlendi. Tümör hücreleri bazofilik granuler sitoplazmalı, yuvarlak oval nukleuslu idi. Tümör hücreleri PAS ve dPAS ile sitoplazmik granuler tarzda boyandı. Olgu psammom cisimlerinden zengin AHK olarak raporlandı.

Anahtar Sözcükler: Asinik hücreli karsinom, maksilla, psammom cismi

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Acinic cell carcinoma (ACC) is a rarely seen, slowly growing, low-grade malignant neoplasia of the salivary gland. Tumor is most frequently (90-95%) detected in the parotid gland, rarely it is localized in a minor salivary gland [1]. Rarely psammoma bodies or nonlaminated calcifications are seen in neoplasic, and nonneoplasic lesions of the salivary gland [2]. Here in, a case with psammoma body-rich acinic cell carcinoma is presented together with its differential diagnosis.

CASE

A 59-year-old male patient consulted our hospital with complaint of stuffy nose persisting for a year. Computed tomography, and magnetic resonance imaging of the patient revealed a mass lesion in the maxillary sinus which extended into nasal cavity, and caused expansion of the neighbouring bony structures. Its radiological findings were priorly reported to be "consistent with inverted papilloma" (Figure 1). Cell groups with granular cytoplasm which formed acini-like structures in a hyalinized stroma were observed in cross-sections of punch biopsy specimens. Calcifications some of which resembled psammoma bodies were dispersed between acini-like formations that were stained positively with alfa 1 antitripsin (AAT). With these findings the tumor detected in the biopsy material was reported as consistent with salivary gland-type tumor, mainly ACC. Maxillectomy material of the patient whose systematic screening did not reveal any evidence of metastasis was sent to our department. Macroscopically on medial surface of the maxillectomy material measuring 7x4,5x4cm which faced the sinus, a yellow-white, patchy areas of bleeding, irregular contours, 6x4x3.5cm solid tumor was seen. Histopathological examination of the specimens displayed the presence of a tumor below the superficial respiratory epithelium. Underlying tumor was solid growth pattern with acini-like formations. Microcalcifications were also accompoined which demonstrating characteristic features of a psammoma without papilla formation (Figure 2). Tumor cells had basophilic granular cytoplasm with round or oval shaped nuclei (Figure 3). Necrosis, lymphovascular invasion, and perineural invasion were not seen. Mitotic activity was counted in 10 High power fields (HPF). Less than one mitosis was seen within 10 HPFs without any atypical mitoses. Tumor cells demonstrated cytoplasmic granules staining positively with PAS ve dPAS. Immunohistochemically, tumor cells yielded negative reaction with smooth muscle actin, p63, S100, TTF1 stains, but stained positively with alfa-1 antitrypsin, and DOG 1 (Figure 4). In the light of these findings the tumor was reported as"psammoma body-rich ACC".

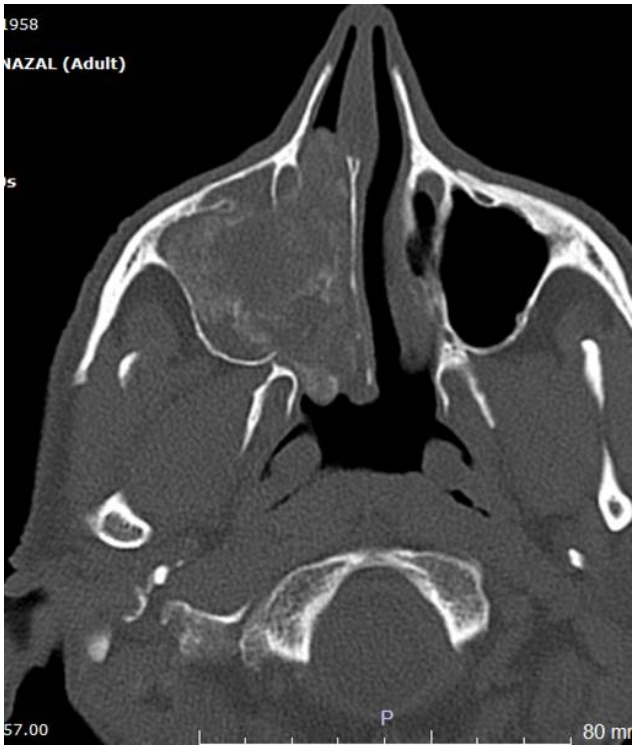

Figure 1. A mass lesion in the maxillary sinus, which extended into nasal cavity, and caused expansion of the neighbouring bony structures

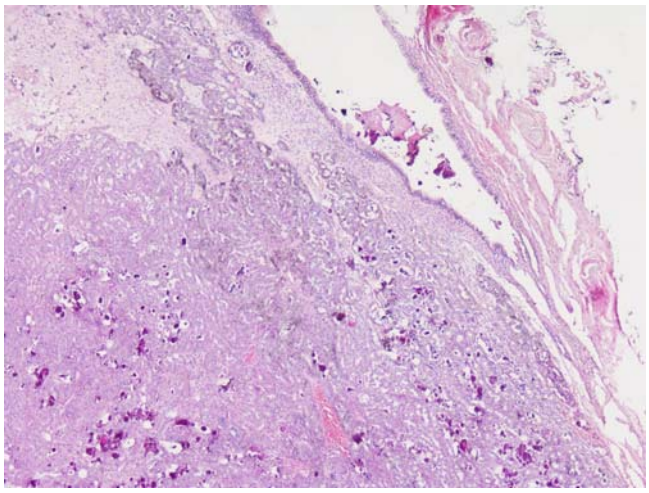

Figure 2. A tumor with superficial respiratory epithelium, and underlying acini-like formations and microcalcifications some of which demonstrating characteristic features of a psammoma (X200)

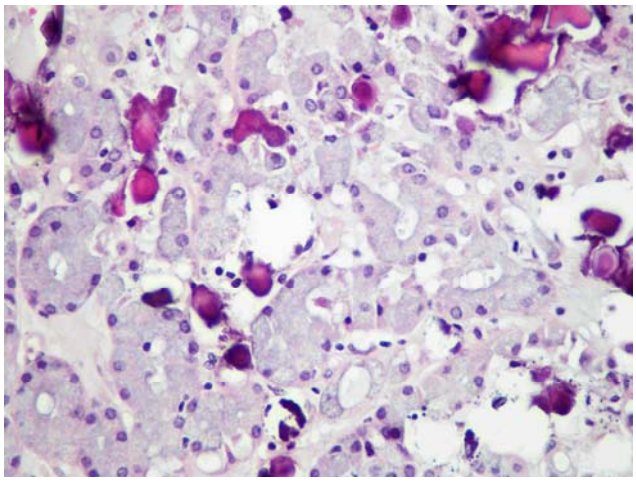

Figure 3. Basophilic granular cytoplasm with round or oval shaped nuclei of tumor cells with psammom bodies (X400)

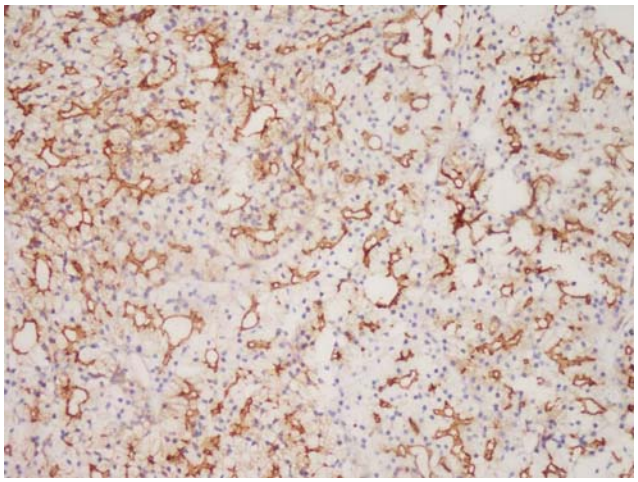

Figure 4. Apically and membranous DOG 1 positivity in tumor cells (X200)

DISCUSSION

Acinic cell carcinoma represents nearly 6% of all salivary gland tumors, and less than 17 % of primary malignant salivary gland tumors [2]. The tumor is most frequently (90-95%) detected in the parotid gland, and it is considered to be a low-grade malignant salivary gland neoplasia with a slow growth rate [1].

Tumor cells consist of intercalated ductal cells with eccentric, round nuclei, basophilic, granular cytoplasm surrounding luminal areas. Histologically solid, microcytic, papillary cystic and follicular growth patterns have been described [2]. Any relation between these four architectural patterns and prognosis has not been reported so far [2, 3]. Rarely, mitoses, necrosis, and atypia were seen.

Psammoma bodies are deposits of concentric layers of calcium salts [2]. It is mostly observed in neoplasms of papillary thyroid carcinoma (PTC), meningioma and papillary serous cystadenocarcinoma [4]. They are often associated with papillary architecture nevertheless they may appear in benign non-neoplastic, inflamed and neoplastic processes of many organs [2]. Frierson et al indicate that psammoma bodies or nonlaminated calcification was noted in the normal submandibular gland and the area of chronic sialadenitis [5]. In the literature, the

presence of psammoma bodies has been described in polymorphous adenocarcinoma, salivary duct carcinoma gland, benign/malignant mixed tumors, and oncocytic adenocarcinoma [2]. Psammoma bodies accompanying acinic cell carcinoma are very rarely reported in the literature [2, 6, 7, 8]. Negahban et al. reported a psammoma-rich papillary cystic ACC localized in the parotid gland in a 24-year-old female patient [2]. Peng Zhang et al. reported a case of ACC associated with psammoma bodies in the lungs in a 31-year-old male patient [8].

In the differential diagnosis of acinic cell carcinoma, secretory carcinoma (SC) demonstrating ETV6-NTRK3 gene fusion takes the lead. Secretory carcinoma has been newly included in the classification of the World Health Organization. In cases with secretory carcinomas, lymph node metastasis is slightly more frequent, also it is more often detected in minor salivary glands when compared with ACC. Histologically luminal, and intracytoplasmic mucin deposits are observed, while in ACC intracytoplasmic zymogen granules are detected. Staining with histochemical marker PAS reveals granular staining in ACC, while globular staining in SC. Immunohistochemically, positive reaction with S100, and mammaglobin, and and DOG1 negativity were detected in SC, while completely reverse staining pattern is observed in ACC [9].

Because of the presence of psammoma bodies in the lesion, in the differential diagnosis, metastasis of PTC should be kept in mind. In the histological examination presence of flat layers, absence of ground glass nuclei in tumor cells, and also intranuclear inclusions, and TTF1 or thyroglobulin-negativity in immunohistochemical staining may exclude the possibility of PTC metastasis [2].

Five-year survival rates are above 80% after surgical resection of ACC while incidence rates for disease recurrence, and metastasis have been reported as 30%, and 15%, respectively [10]. Histologically, gross invasion, desmoplasia, atypia, and increased mitotic activity have been indicated to be predictive indicators of the disease progression [6].

In Conclusion, Psammoma body-rich acinic cell carcinoma has been very rarely reported in the literature. Our case was presented with its clinical, and histomorphologic features together with striking characteristics which should be taken into consideration in the differential diagnosis from secretory carcinoma of the salivary gland included in the latest classification of the World Health Organization.

*The authors received no refund

**We declared that there is no conflict of interest with any institution about the subjects discussed in the article.

***Informed consent was obtained from the patient.

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