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ENTCase

A VERY RARE CASE: BASAL CELL ADENOCARCINOMA IN THE SUBMANDIBULAR GLAND AT A YOUNG AGE

ÇOK NADİR BİR VAKA: GENÇ YAŞTA SUBMANDİBULAR BEZDE BAZAL HÜCRELİ ADENOKARSİNOMA

Baş Boyun Cerrahisi

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Özet

Bazal hücreli adenokarsinom, tükürük bezlerinin nadir görülen bir tümörüdür. Bazal hücreli adenokarsinomların çoğu parotis bezinden (>%80) kaynaklanırken, diğerleri sublingual ve submandibular, minör tükürük bezlerinden kaynaklanır. Bazal hücreli adenokarsinom tipik olarak 60 yaşın üzerindeki yetişkinlerde ortaya çıkar ve cinsiyet üstünlüğü göstermez. Submandibular bezin bazal hücreli adenokarsinomu, bilinen literatürde nadiren tartışılmıştır. Submandibular bezden kaynaklanan bazal hücreli adenokarsinom tanılı 38 yaşında bir erkek olguyu bu vaka raporunda sunuyoruz. Bazal hücreli adenokarsinomun klinik ve patolojik özelliklerini, tedavi seçeneklerini ilgili literatürü gözden geçirerek tartışacağız

Anahtar kelimeler: Tükürük bezi, Bazal hücreli adenokarsinoma

Abstract

Basal cell adenocarcinoma is a rare tumor of the salivary glands. . Most BCACs arise from the parotid gland (> 80%), whereas others originate from the sublingual and submandibular glands ,minor salivary glands .BCAC typically arise in adults over the age of 60 and do not show a sex predominance. Basal cell adenocarcinoma of submandibular gland has rarely been discussed in the English literature. In this case report . We present an unusual case of a 38-year-old male with BCAC that arise from the submandibular gland . We describe the clinicopathological features, the the treatment choices of BCAC with a review relevant literature.

Keywords: Salıvary gland, Basal cell adenocarcinoma

Introduction

Basal cell adenocarcinoma is a rare tumor of the salivary glands. . Most BCACs arise from the parotid gland (> 80%), whereas others originate from the sublingual and submandibular glands ,minor salivary glands .BCAC typically arise in adults over the age of 60 and do not show a sex predominance. Basal cell adenocarcinoma of submandibular gland has rarely been discussed in the English literature. In this case report . We present an unusual case of a 38-year-old male with BCAC that arise from the submandibular gland . We describe the clinicopathological features, the the treatment choices of BCAC with a review relevant literature.

Case Report

A 38-year-old male patient applied to our clinic with swelling left submandibular region. On physical examination, there is a painless, hard-fixed mass that measured approximately 5 cm in the left submandibular region. That mass has been growing slowly for the last year. The patient has no known chronic disease. Contrast enhaced magnetic resonance imaging (MRI) showed a mass measured 5x3.5x5 cm in size, located in the submandibular gland. An isointense lightly lobulated contoured area was detected in the central part of the lesion in T1W and T2W examination, and it was interpreted as hyperintense in T1W and T2W examinations in the peripheral parts [Figure 1,2].

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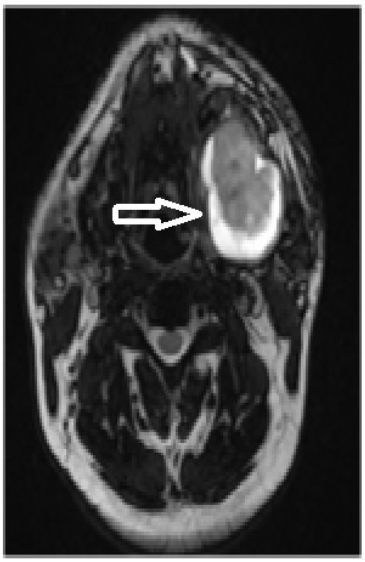

Figure 1
t2 weighted axıal cross-sectıonal ımage

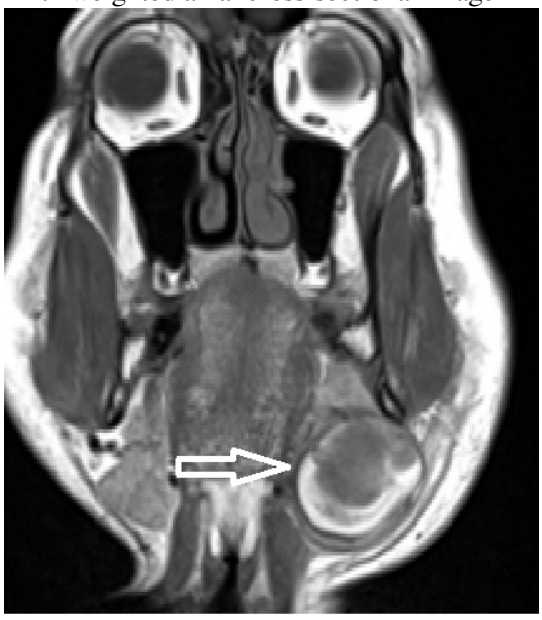

Figure 2
T1 weighted coronal cross-sectional image

Then, fine needle aspiration biopsy was performed with US guidance. Biopsy material was reported as Milan classification category 4A, lymphoepithelial cyst and whartin tumor for preliminary diagnosis. According to the biopsy result, submandibular gland excision was performed after obtaining written informed consent from the patient. Specimen pathology is reported that basal cell adenocarcinoma, low grade[Figure 3,4].

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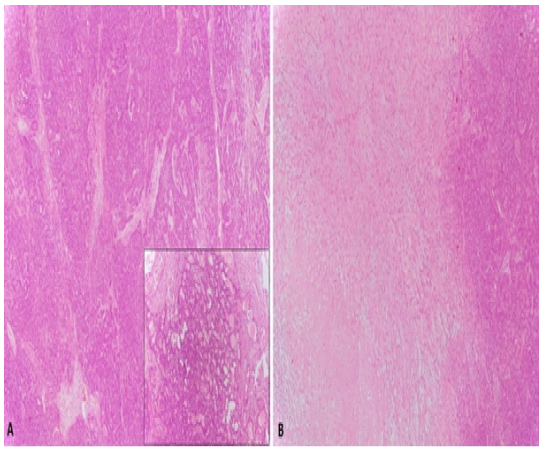

Figure 3

A: Neoplasia consisting of basaloid cells forming solid islands in a trabecular pattern, an area forming tubule formation in a focal area in the small square is observed (H&E, x40). B: Widespread

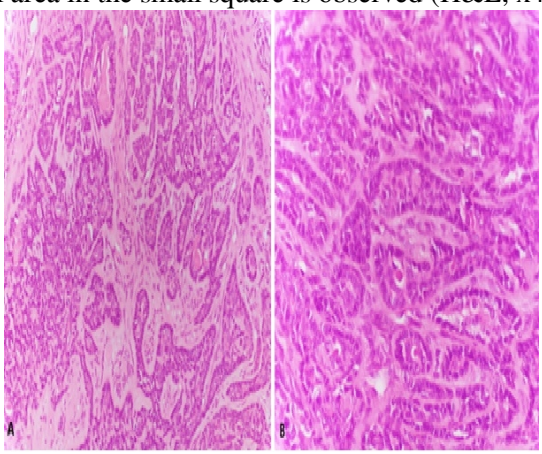

Figure 4

Basaloid cells with narrow cytoplasm showing peripheral palisade(green arrow) and ductal cells with polygonal wide cytoplasm in the central(A:H&E, x200; B:H&E, x400)

Any metastases were not detected on Positron Emission Tomography -Computerized Tomography (PET /CT) scan. There was no recurrence of the disease in the one-year follow-up of the patient.

Discussion

Basal cell adenocarcinoma (BCAC) are rare salivary gland tumors. BCAC comprising 1% to 3% of malignant salivary gland tumors[1]. Synonyms include carcinoma ex-monomorphic adenoma, basaloid salivary gland carcinoma, and malignant basal cell tumor[2].Klima et al. are credited for the first report of basal cell adenocarcinoma, the malignant counterpart of the basal cell adenoma(BCA). Their description was that of a parotid gland neoplasm that appeared to be similar to basal cell adenoma but showed necrosis and mitoses[3].Although basal cell adenocarcinoma has rarely been reported to arise from an underlying basal cell adenoma, most basal cell adenocarcinomas are thought to occur de novo.[4].Gender balance in each shows an almost equal proportion and occurs over a wide age range, mostly in the sixth to seventh decade. BCAC can occur

in the major or minor salivary glands, but the parotid gland is the most common site. Second most common is regional submandibular gland, followed by the sublingual and then the minor salivary glands in the head and neck, including the buccal mucosa, the palate, the tongue, and even the parapharynx. BCAC is composed of the following 2 cell types: smaller basaloid cells with scant cytoplasm and dark nuclei and slightly larger polygonal basaloid cells with eosinophilic cytoplasm. BCACs have 4 major histologic growth patterns consisting of solid, tubular-trabecular cribriform, and membranous. The solid pattern is the most common and the most likely to manifest with perineural invasion [5].In the histopathological examination of our patient's submandibular gland excision specimen, it was seen to be compatible with BCAC, which is a tubular-trabecular pattern [Figure 3,4]. The most common symptoms associated with BCAC are enlargement and tenderness of the salivary glands. There may also be inability to move some facial muscles and facial numbness. Our patient had only painless swelling. He did not have any other complaints such as facial numbness,facial paralysis.,tenderness. Surgical excision with a wide margin to ensure complete removal has been suggested as the primary treatment for BCAC[6].Radiotherapy has been proposed for lesions in the minor salivary glands because of the higher likelihood of vascular and neural invasion.Radiotherapy has also been used for tumors with a diffuse infiltrating pattern to adjacent tissue[7]. Recurrence rates have been reported between 15% and 50%, with most reported rates of around 30%. Less than 10% develop cervical lymph node metastases and less than 5% develop distant metastases and die. While local recurrences are common, regional lymph node and distant metastases are rare.Thus, the World Health Organization categorizes BCAC as a low-grade tumor with a favorable prognosis. The differentiation of BCACs from BCAs is based on invasive proliferation into the nerves, blood vessels, and surrounding tissues, necrosis, mitotic figures, and anisonucleosisThe BCAC is not fully circumscribed and, if encapsulated, may show invasive areas pushing the capsule. Perineural invasion may also occur but is not present in all cases. Adenoid cystic carcinoma (ACC) is basaloid tumors with morphologic similarities to BCA and BCAC, which have a biphasic cellular arrangement The nuclei of ACC are angular rather than rounded, and the cell arrangements are not palisaded. ACC has an infiltrative border as small cells at low power, whereas BCA has a rounded border[1].

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