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Salivary Gland Ductal Carcinoma of Accessory Parotid Gland

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ABSTRACT

Salivary gland ductal carcinoma (SDC) is a rare, aggressive tumor with a highly malignant course and originates most frequently from the parotid gland among the major salivary glands. The incidence of accessory parotid gland (APG) varies between 21-56% and the incidence of tumor in APG is 1-8% of all parotid tumors. SDC is rarely seen in accessory glands. It was aimed to present our case, which was diagnosed with SDC in APG, in the light of the literature.

Keywords: Salivary duct carcinoma, accessory parotid gland, diagnosis, treatment

Aksesuar Parotis Bezinin Tükürük Bezi Duktal Karsinomu

Süreç

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ÖZ

Tükrük Bezi Kanal Karsinomu (TBKK), nadir, agresif bir tümördür ve genellikle büyük tükrük bezleri arasında parotis bezinden en sık köken alır. Aksesuar parotis bezi (YPB) insidansı %21-56 arasında değişir ve APG'deki tümör insidansı tüm parotis tümörlerinin %1-8'ini oluşturur. TBKK, yardımcı bezlerde nadiren görülür. Bu yazıda, APG'de TBKK tanısı alan vakamızı literatür ışığında sunmayı amaçladık.

Anahtar sözcükler: Tükrük bezi duktal karsinomu, aksesuar parotis bezi, tanı, tedavi

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Introduction

Ductal carcinoma (SDC) is microscopically similar to high-grade ductal carcinoma of the breast in both in-situ and invasive patterns[1]. It is a rare, aggressive salivary gland malignancy with high metastasis and recurrence rates. It was first described in 1968 by Kleinsasser et al[2]. This rare and aggressive tumor has 21 primary subtypes defined by the World Health Organization[3]. SDC accounts for 0.2-9% of salivary gland tumors[4]. Although there are cases reported in the submandibular gland (8%) and minor glands (4%), it often originates from the parotid gland (88%)[5].

Locoregional and distant metastasis rates are high. It is generally observed over the age of 50 (most commonly 60-65 years) and more frequently in men than in women (male/female: 4/1). Clinically, SDC is quite aggressive and typically patients present with a rapidly growing painful mass that often involves the facial nerve[6].

SDC treatment; includes total parotidectomy and ipsilateral neck dissection, followed by adjuvant chemo-radiotherapy or postoperative radiotherapy and targeted therapies[3].

However, the prognosis of SDC is poor and the mortality rate of the cases within 5 years is 60%[4].

There is no standard treatment option for patients with recurrence and metastasis.

The incidence of accessory parotid gland (APG) varies between 21-56%. they are usually less than an inch in diameter. They are located 6 mm anterior to the parotid gland, close to the Stensen canal, between or deep within the zygomatic and buccal branches of the facial nerve[7]. The incidence of tumor in APG is 1-8% of all parotid tumors[7,8]. Patients whose tumors are detected in APG usually apply to clinics with swelling in the cheek area. 26-52% of these masses observed in APG are malignant. SDC in this accessory structure is rarely seen and its incidence is not known for certain[8].

For this reason, it was aimed to present our case who was diagnosed with SDC from APG in the light of the literature.

CASE REPORT

A 76-year-old male patient was admitted to our clinic with the complaint of a painless, hard mass in the left cheek region, which has been growing steadily for two years. In the physical examination of the patient, a firm, fixed and painless mass with irregular borders, approximately 4x3 cm in size, was detected in the zygomatic region, approximately 2 cm lateral from the left preauricular region, adjacent to the masseter muscle. Facial nerve examination of the patient was normal (Figures. 1,2).

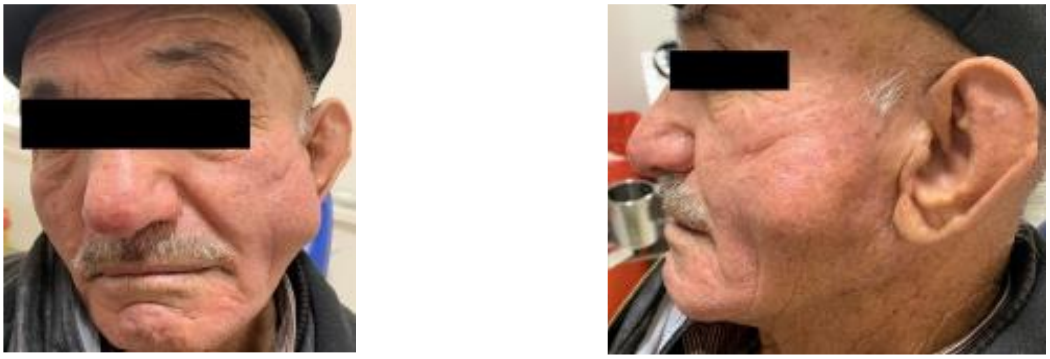

Fig. 1,2. Patient with a 4*3 cm mass in the zygomatic region

Contrast-enhanced neck-diffusion magnetic resonance imaging examination revealed that the left parotid gland is 40x33 mm in size in its widest part, adjacent to the superficial lobe anterior part and masseter muscle, heterogeneous in the T2W periphery with diffusion restriction and intensely contrasting, heterogeneous hypointense in the T2A periphery, and T1A containing cystic necrotic areas in the center, which is thought to be high proteinaceous was observed (Figures. 3, 4). No pathological lymphadenopathy was detected in the neck. Considering the clinical and radiological findings, it was thought that the patient had a pathology originating primarily from the accessory parotid gland. Fine needle aspiration biopsy (FNA) was performed on the patient.

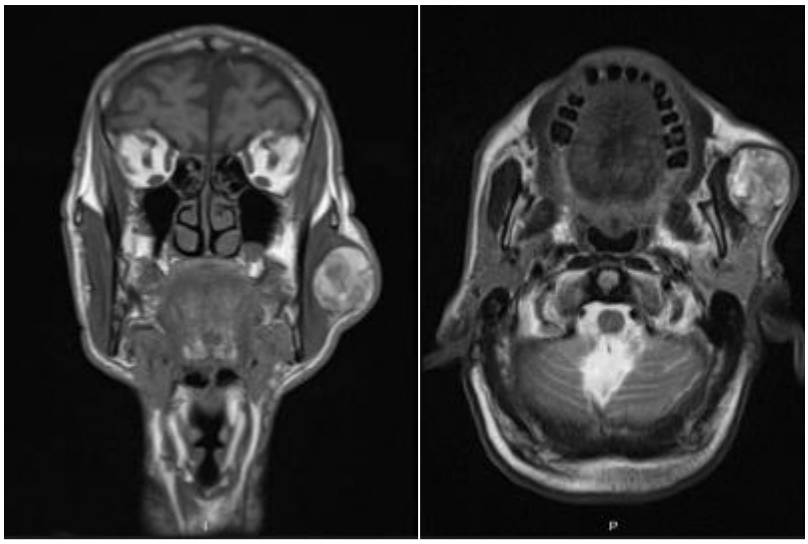

Fig. 3, 4: Contrast-enhanced neck-diffusion magnetic resonance imaging examination of the mass

In the cytopathological examination of fine needle aspiration biopsy (FNAB), atypical epithelial cells with extensive cytoplasm, hyperchromatic eccentric nuclei with prominent nucleoli with increased N/S ratio which formed three-dimensional groups and papillary structures on smears. These features interpreted in the category of "suspected malignancy" according to the salivary gland fine needle aspirations MILAN classification (Fig. 5).

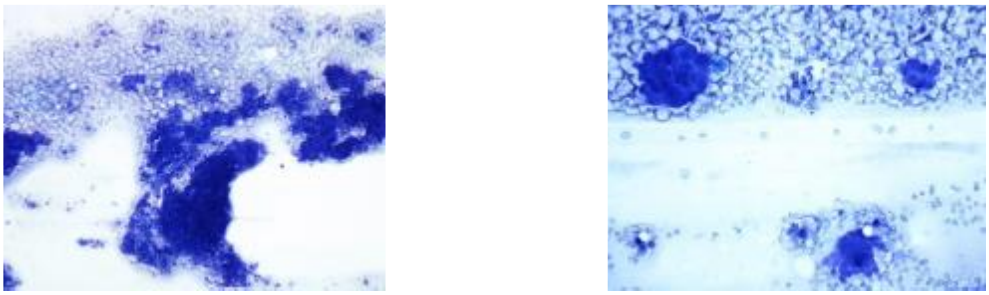

Fig. 5. Hypercellular smears consisting of atypical cells (MGGx100, MGGx200)

The patient was evaluated in the head and neck oncology council. It was decided to remove the mass in the cheek area and perform superficial parotidectomy, and to reevaluate the patient in the oncology council, according to the definitive pathology result.

Macroscopically, it weighs 22 g, measures 4.2x4x3.5 cm, has encapsulated nodular appearance, and consists of gray-white, brown-colored gelatinous areas with lobulation on the section face and heterogeneous yellow-orange-colored areas (Fig. 6). Histopathological examination of hematoxylin-eosin sections with light microscope dilated ductal structures with papillary areas, "Roman-bridge", or solid growth accompanied by comedo necrosis, tubular and cribriform structures with scirrhous pattern, apocrine appearance with large pleomorphic nuclei and abundant eosinophilic cytoplasm seen (Fig. 7). Surgical margins were negative. No perineural and lymphovascular invasion was observed. Immunohistochemical (IHC) stains, HER2 positive (score 3), Androgen (AR) positive, GATA3 positive, GCDFP-15 positive, CK5/6 focal positive, P63 negative were found (Fig. 8). The case was reported as ductal carcinoma of salivary gland with morphological and IHC features.

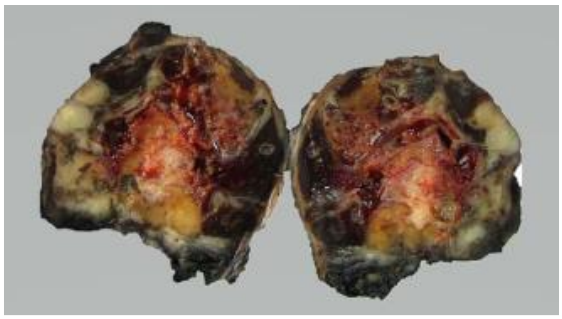

Fig. 6. Filling the entire gland hemorrhagic, grey-white-yellow solid and cystic areas were observed.

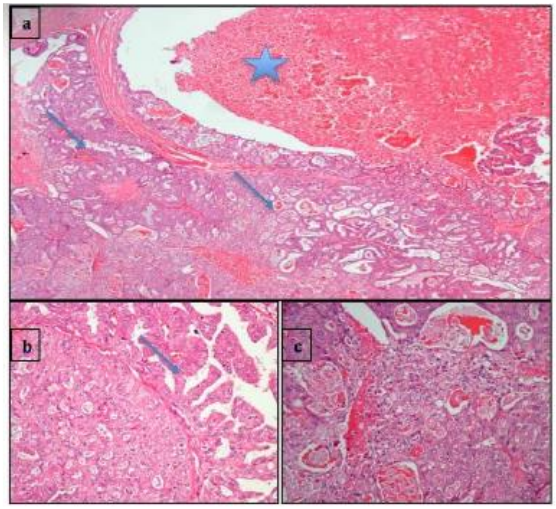

Fig. 7. a Tumor with cribriform ( ), papillary and solid patern and necrosis areas ( ) are observed (HEx40) b. Apocrine appearance and papillary structure ( ) (HEx200) c. There is pronounced pleomorphism and numerous mitoses (HEx200)

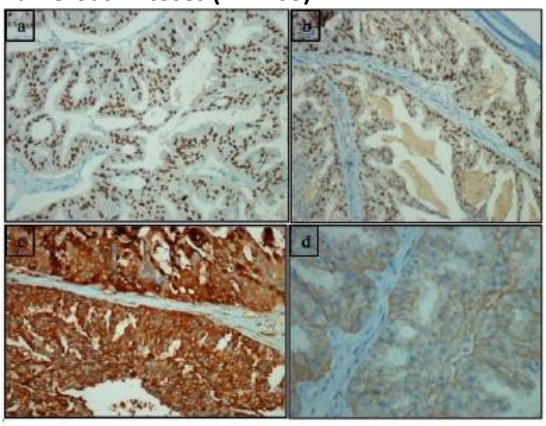

Fig. 8. Significant immunohistochemical stains (X200)
a. Nuclear Androgen expression b. Nuclear GATA 3 expression
c. Cytoplasmic GCDFP 15 positive d. Complete membranous HER 2 expression

Considering the tendency of TDC to make distant metastases, the case was evaluated radiologically. No distant metastases were detected.

The patient was consulted again in the oncology council. Radiotherapy was planned after total parotidectomy and neck dissection. Since the patient did not accept the operation, the treatment was completed as chemo-radiotherapy. After surgery and chemo-radiotherapy, follow-up continues for approximately one year, and no local recurrence and/or distant metastasis were observed during this period. No pathological finding was detected in the radiological follow-ups.

Discussion

SDC arises from the ductal epithelium of the salivary gland tissue and histologically resembles high-grade breast ductal carcinoma[9]. Most cases of SDC develop de novo, but some may arise from pre-existing carcinoma ex

pleomorphic adenoma[1]. Clinically, SDC presents as a firm, painful, rapidly growing mass that causes facial paralysis and tends to metastasize to the temporal bone via perineural spread. Although SDC is most commonly seen in the parotid gland; It can also be seen in the submandibular and sublingual glands, minor salivary glands, larynx, parapharyngeal region and sinonasal region[10]. Patients often present with a rapidly growing mass, facial paralysis, pain and local regional metastases. At the time of diagnosis, 46-60% of the cases have multiple lymph node metastases[11]. Clinically, SDC is characterized by cervical lymph node involvement, high recurrence rate, distant metastasis, and high mortality. Distant metastases are frequently found in the bones and lungs, but liver and brain metastases can also be observed in these cases[12]. Local regional metastases are frequently observed in cervical lymph nodes. Cervical lymph node involvement in SDC at the time of diagnosis varies between 50-82.5% in the series in the literature[13]. It has been 50 years since SDC was first described and numerous cases have been reported in the literature. These case reports generally focused on the clinical features of the cases, diagnosis and treatment approaches, prognosis and genetics of the tumor. In our literature review, SDC studies and case reports, including clinical series, show that the tumor usually involves the parotid gland and that the most prominent clinical symptoms are facial paralysis and pain. SDC also has strong neuronal tropism. Due to this feature, there are case reports in the literature that draw attention to the possibility of metastasis to the mandible and intracranial region by holding the facial nerve or its branches[12-13]. FNA has become widely accepted as an efficient first line diagnostic test in the management of salivary gland lesions. The reported overall sensitivity of salivary gland FNA in most series ranges from 86% to 100%, and the specificity ranges from 90% to 100%[14]. It was evaluated with the MILAN classification, an evidence-based system that associates the FNA diagnostic categories of the case with the risk of malignancy and clinical management strategies. SDC are easily recognized cytologically as high-grade carcinomas, but often require additional methods for more specific classification. The gene expression pattern of SDC is quite similar to that of molecular apocrine breast carcinoma such as AR, HER 2[15]. Masubuchi et al. AR positivity and EGFR positivity were associated with better DFS in patients with SDC, and they stated that anti-AR, anti-HER2 and/or anti-EGFR targeted therapies may be beneficial. In our case, AR, HER 2 positivity was also present[16]. In addition, diagnostically, GATA 3 and GCDFP 15 positivity and p63 negativity are also helpful in diagnosis. The FNAB of our case was evaluated with the MILAN classification, and it was decided by the oncology council to use the histopathological result instead of the FNAB result in the surgical planning, since it was interpreted in the "suspicious malignancy" category. Ductal carcinoma of APG is very rare, so we could not reach an incidence reported in the literature. The biggest problem in APG tumors, which are very rare, is the preoperative diagnosis stage; The most important diagnostic criterion is APG and the physician's suspicion of malignancies in this region[17]. There is currently no consensus on the surgical treatment of APG; however, superficial parotidectomy is a safe surgical procedure with high survival and low morbidity rates[18].

Conclusion

Due to the rarity of salivary duct carcinoma, there is a lack of comprehensive documentation in the literature regarding its clinical features, treatment, and clinical outcomes. In a number of case reports and small case series, it has been pointed out that aggressive clinical behavior, regional lymph node involvement and a tendency to distant metastasis are present. The most important difference of our case from SDC cases in the literature is the diagnosis of SDC of accessory parotid gland while investigating the origin of the fast growing, painless and non-facial paralysis mass detected in the zygomatic region. In addition, the mass did not show neuronal tropism as expected, and it differed from other cases in the literature.

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