Case Report

Basal Cell Adenocarcinoma Ex Pleomorphic Adenoma

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Abstract

Basal cell adenocarcinoma (BCAC), a salivary gland tumor, has been theorized to originate either from monomorphic adenomas undergoing a malignant transformation or to arise de novo. Here we present the first case report of a BCAC tumor that arose within a pleomorphic adenoma.

Keywords
basal cell adenocarcinoma; parotid gland; pleomorphic adenoma; salivary glands

1. Introduction

In 2007, a review of the literature revealed that fewer than 100 cases of basal cell adenocarcinoma (BCAC) had been reported [8]. A slow-growing malignant neoplasm of the salivary glands, BCAC usually affects the major salivary glands, specifically the parotid gland [1,8]. When arising in minor salivary gland, BCAC occurs most frequently in the oral cavity and upper respiratory tract [3,6]. BCAC does not show sex predilection and predominates in patients in their seventies [3].

The WHO classification of salivary gland tumors first included BCAC in 1991 as the malignant counterpart of the monomorphic adenoma [8]. For more than 40 years, monomorphic adenomas had been recognized formally as a class of salivary gland tumors, apart from pleomorphic adenomas [3]. The prevailing theory is that BCAC tumors have two different origins: monomorphic adenomas undergoing a malignant transformation or the tumor arisen de novo [8]. BCAC is considered a low-grade malignant tumor. This is supported by diploid DNA content [1]. In 1993, one-third of the reported cases had recurred, and approximately 10% metastasized on follow-up. The most common metastatic sites were lymph nodes and lungs. Metastatic tumors all had solid growth patterns [1].

To date, there have been several case reports describing the characteristics and behavior of BCAC, with no definitive consensus regarding treatment [8]. Here we present the first case report of a BCAC that arose within a pleomorphic adenoma (carcinoma ex pleomorphic adenoma).

2. Case report

A 79-year-old man with a past medical history significant for hypertension, GERD, COPD, and OSA was referred for the management of a growing left-sided facial mass, which was first noticed by the patient about 3–4 years ago. He denied any pain, and his only complaint was that it was causing problems with retention of his dentures.

Physical examination revealed a left-sided preauricular mass, about 7 cm in the largest dimension. The surrounding soft tissue was not fixed to the lesion. There was no noted facial nerve weakness, and no cervical adenopathy was noted.

An MRI was obtained and revealed a complex mass within the left parotid gland with multiple areas of cyst formation or necrosis within the mass, involving both the superficial and deep portions of the left parotid gland. There was no evidence to suggest perineural spread (Figure 1).

After a discussion of the risks including damage to the facial nerve, the patient opted to have the lesion removed. An MRI was obtained and revealed a complex mass within the left parotid gland with multiple areas of cyst formation or necrosis within the mass, involving both the superficial and deep portions of the left parotid gland. There was no evidence of regional adenopathy. There was no evidence to suggest perineural spread (Figure 1).

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After a discussion of the risks including damage to the facial nerve, the patient opted to have the lesion removed. A left subtotal parotidectomy and abdominal fat graft were performed. A modified Blair incision was made, extending from the zygomatic arch almost to the midline of the neck. Working between the sternocleidomastoid muscle and then the posterior belly of the digastric and the tragal pointer, the facial nerve was identified. The lower branch was found to be pushed inferiorly. The mass grew deeper than this branch.
Figure 1: Axial T1 weighted image with gadolinium showing an enhancing mass of the left parotid gland with multiple area of cyst formation or necrosis with in the mass.

It was freed from the overlying mass and required dissection out to its most peripheral aspect. The upper division was then addressed, which was again intimately adherent to the mass. This upper division was also followed to the periphery, and then once this was achieved, the soft tissue in between those two areas was progressively taken as the mass was being pulled from the deep part of the gland. The mass was totally excised and was sent for histopathological analysis. After performing an abdominal fat graft and placing a drain, the wound was closed. His postoperative course was unremarkable, and there was no facial nerve deficit.

Pathologic examination revealed a parotid gland containing a $5.3 \times 4.2 \times 4$ cm mass. The mass was tan-white, multilobulated, and firm. No necrosis was identified grossly. On histopathologic examination, sections show foci of residual benign pleomorphic adenoma surrounded by basal cell adenocarcinoma (Figure 2). Residual pleomorphic adenoma is comprised of chondromyxoid stroma containing benign myoepithelial cells. The pleomorphic adenoma is largely replaced by a proliferation of basaloid cells with peripheral palisading, areas of squamous differentiation (Figure 3), frequent mitotic figures, necrosis (Figures 4(a) and 4(b)), and expansile growth with focal invasion.

3. Discussion

Kleinsasser and Kline proposed monomorphic adenoma as a separate entity from pleomorphic adenoma [11]. Batsakis later subclassified monomorphic adenomas into basaoid basal cell adenoma and non-basaloid type [4]. To date, basal cell adenocarcinoma has been described to be a malignant counterpart of basal cell adenoma. Thus far, two different origins of the tumor have been described; as a malignant transformation of monomorphic adenoma (carcinoma ex monomorphic adenoma) and de novo, which seem to be responsible for the majority of these tumors [3,9,12].

In 1988, Ellis and Gnepp described the histologic features, and in 1990, Ellis and Wiscovitch’s report of 29 patients with BCAC at AFIP defined the clinicopathologic features of this rare disease [7,5]. BCAC typically arises
in adults older than 60, with no gender predominance [13]. The 2005 WHO classification of tumors categorizes BCAC as a low-grade tumor with a favorable prognosis [2]. There are four major histologic growth patterns of BCAC: solid, tubulotrabecular, cribriform, and membranous, with solid being the most common and also the most likely to present with perineural invasion.

The differential diagnosis of BCAC includes basal cell adenoma and adenoid cystic carcinoma. BCAC appears very similar to basal cell adenoma histologically, but is distinguished from this benign entity by displaying invasion of local structures, perineural invasion, and/or angiolympathic invasion [15]. Adenoid cystic carcinoma has a poorer prognosis and displays dark hyperchromatic angulated nuclei, which differ from the vesicular nuclei of BCAC. BCAC also often have prominent peripheral palisading of the outer layer that adenoid cystic carcinomas lack [14].

Treatment consists of surgical excision with a wide margin of tissue to reduce the chance of recurrence. These tumors are locally destructive, and recurrence has been reported to be 28% in the largest series published [5]. Radiotherapy is suggested for lesions originating in the minor salivary glands due to a higher likelihood of vascular and neural invasion [10].

4. Conclusion

The origin of BCAC is not well defined. Although most appear to arise de novo or from monomorphic adenoma, here we report evidence of a BCAC ex pleomorphic adenoma. Differentiating BCAC from basal cell adenoma and adenoid cystic carcinoma is important due to the difference in prognosis that each of these diseases possess. Surgical excision is the preferred method of treatment for most of these lesions.

References
