

Bilateral Parotid Basal Cell Adenoma: An Unusual Case Report and Review of the Literature

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Basal cell adenomas (BCAs) are uncommon benign tumors of the salivary glands that were recognized as an independent entity in the second edition of the Salivary Gland Tumours Classification of the World Health Organization.¹ BCAs constitute approximately 1% to 2% of all salivary gland epithelial tumors.² The occurrence of bilateral basal cell adenomas is extremely rare, with only 7 cases reported in the English-language literature.³⁻⁹

We report an unusual case of metachronous bilateral parotid BCA without coexisting dermal cylindroma and present a review of the English-language literature.

A 65-year-old woman was referred in 2003 for evaluation of a palpable mass in the left parotid region. Physical examination revealed a mass measuring about 3 × 2 cm, which was hard, movable, and nontender. There was no facial palsy or regional lymphadenopathy. A fine-needle aspiration cytology report was inconclusive. Magnetic resonance imaging (MRI) showed a well-defined and homogeneously well-enhanced mass occurring in the superficial lobe of the left parotid gland (Fig 1A). No calcification or

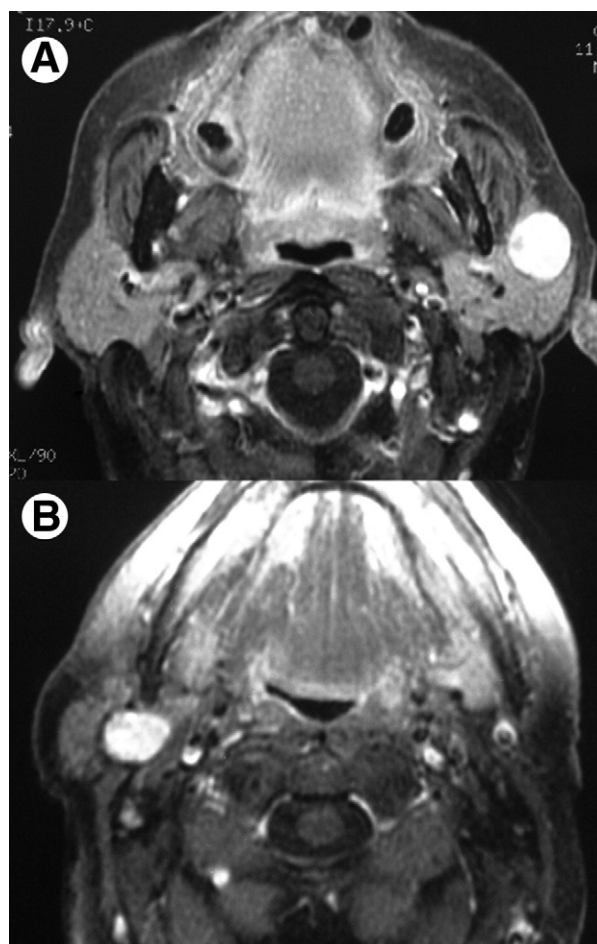


FIGURE 1. A, Magnetic resonance imaging (MRI) revealed left parotid mass in the superficial lobe. B, Second MRI 3 years after first surgery revealing a mass in the deep lobe of the right parotid gland.

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cystic component was seen within the tumor. These findings were suggestive of benign tumor of the parotid gland. The gender of the patient and clinical manifestations of the lesion suggested a mixed tumor.

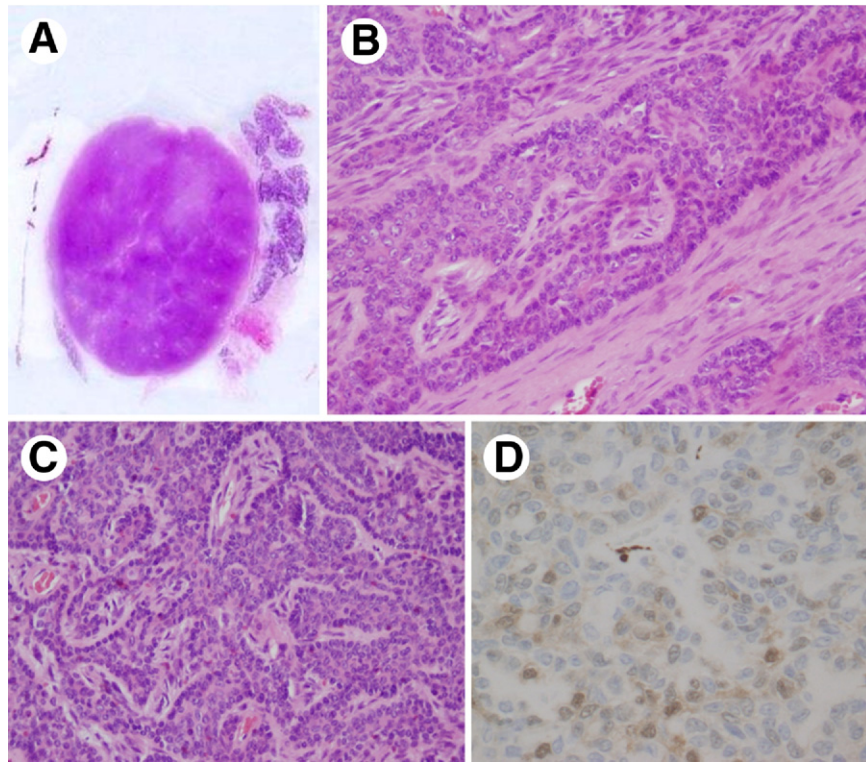


FIGURE 2. A, Left intraparotid tumor nodule revealing uniform histologic appearance dominated by basaloid cells (hematoxylin-eosin). B, Histologic examination of the left tumor showing basaloid epithelial cells in large irregular-shaped sheets (hematoxylin-eosin stain $\times 200$). C, Histologic examination of the right tumor showing characteristics typical of basal cell adenoma: aggregates of epithelial tumor separated by stromal tissue (hematoxylin-eosin stain $\times 200$). D, Immunohistochemistry revealed positivity for S-100 protein in stromal cells of the right tumor ($\times 400$).

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One month later, the patient underwent left superficial parotidectomy, including removal of the mass, with preservation of the facial nerve and its branches. Microscopic examination revealed varying sized and shaped aggregates of epithelial tumor cells separated by amounts of stromal tissue. The aggregates consisted of 2 layers of cells: dark cells with less cytoplasm and more basophilic nuclei, and light cells, some of them with 1 or more small basophilic nucleoli. Some cuboidal ductal cells surrounding small lumens were seen. Immunoreactivity to S-100 protein was localized to the peripheral tumor cells adjacent to the connective tissue stroma. The histology was compatible with basal cell adenoma with predominant solid pattern (Figs 2A,B) with small areas of trabecular pattern. The postoperative course was uneventful, without facial palsy.

The patient was carefully followed up; 3 years after surgery, she started complaining of swelling in the right parotid gland. Physical examination revealed an elastically hard and poorly defined right parotid mass. Fine-needle aspiration cytology was again nondiagnostic. MRI revealed a right parotid mass in the caudal portion of the deep lobe (Fig 1B). On T2-weighted images, the tumor showed homogeneous moderate

intensity. With the suspected diagnosis of basal cell adenoma, right conservative parotidectomy was performed with facial nerve preservation. Histology revealed small, slightly separated nodules of basaloid cells in an insular pattern, compatible again with basal cell adenoma with solid pattern (Fig 2C). Celularity in the immunohistochemical analysis was again focally positive to S-100 (Fig 2D). Postoperatively, there were no complications and no sign of recurrence after 1 year of follow-up.

Discussion

Basal cell adenoma is a benign epithelial neoplasm with a uniform histologic appearance dominated by basaloid cells and without the myxo-chondroid tissue characteristic of mixed tumor.¹⁰ More than 80% of BCAs arise in the major salivary glands, the majority occurring in the parotid gland. An incidence of 2% to 4% of all primary salivary gland tumors is often cited.¹⁰ BCAs arise almost exclusively in adults.^{2,10} The average age of patients with BCAs is 57.7 years, with a 2:1 female predominance² except in the case of membranous basal cell adenoma, which shows a male preponderance.¹¹ Similar to other benign salivary

Table 1. REPORTED CASES OF BILATERAL PAROTID BASAL CELL ADENOMAS IN THE ENGLISH-LANGUAGE LITERATURE

Reference	Age (yrs)	Gender	Dermal Cylindromas	Occurrence of BCAs
Reingold et al, ³ 1977	43	M	Yes	Metachronous
Herbst and Utz, ⁴ 1984	54	F	Yes	Metachronous
Zarbo et al, ⁵ 1985	58	M	Yes	Synchronous
Schmidt et al, ⁶ 1991	72	F	Yes	Metachronous
Katsuno et al, ⁷ 2000	65	F	No	Synchronous
Suzuki et al, ⁸ 2000	65	F	No	Synchronous
Reddy et al, ⁹ 2008	55	F	No	Synchronous
Our case 2008	65	F	No	Metachronous

Abbreviations: F, female; M, male.

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gland tumors, BCAs present as a slowly enlarging, asymptomatic, freely movable mass. Their greatest dimension is usually less than 3 cm.

The bilateral occurrence of parotid tumors is rare, accounting for 1% to 3% of all parotid tumors. The most common tumor to occur bilaterally is Warthin's tumor, reported to represent 5% to 10% of all Warthin's tumors.¹² The occurrence of bilateral BCAs of the parotid glands is unusual, and only 7 cases have been reported so far in the English literature (Table 1).³⁻⁹

There is a notable similarity between dermal eccrine tumors (eccrine spiradenoma and cylindroma) and BCAs, and a diathesis of both tumors has been reported. It has been suggested that the histologic features of BCAs and skin tumors were similar and their synchronous occurrence may result from a single pleiotropic gene that acts on ontogenetically similar stem cells.¹³ Most of the BCAs associated with dermal tumors were the membranous type, and most of the dermal tumors were cylindroma.¹⁰ Four of the 7 cases reported in the English-language literature of bilateral BCAs coexisted with dermal cylindromas (Table 1).³⁻⁶

Histologically, BCAs have various variants, including solid, trabecular, tubular, and membranous variants. Although the most common type is the solid variant, individual tumors commonly display a combination of several growth patterns.^{2,10} BCAs are composed of 2 types of cells. The first type is a small cell with scant cytoplasm and a round basophilic nucleus. The second type presents large cells with eosinophilic cytoplasm and oval nuclei. The precise cell origin of this peculiar tumor has also been studied by immunohistochemistry. BCA subtypes appear microscopically basaloid and monomorphic in architectural patterns compared with pleomorphic adenoma, periductal, epithelioid, and spindled (stromal-like) myoepithelial cells contribute to the proliferation of these tumors. Positivity for S-100 protein indicates the myoepithelial nature of these tumor cells.¹⁴

BCAs are amenable to conservative resection such as superficial parotidectomy. The recurrence rate for the solid and trabecular-tubular variants is almost nonexistent.¹⁰ The membranous type is the most commonly associated with recurrence (25% to 37%), and this may be a result of the multicentricity of this lesion rather than to true recurrences,² although rare, malignant transformation is more common in the membranous type than in the other types. Male preponderance, multicentric development, absence of capsule in 50% of cases, association with eccrine tumors, and malignant transformation have led some investigators to suggest that membranous type of BCA should be classified separately.¹⁰

Differential diagnosis with entities of varied prognosis such as pleomorphic adenoma, adenoid cystic carcinoma, and basal cell adenocarcinoma makes necessary the consideration of this entity in the field of glandular tumors of the maxillofacial area.

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Management of Facial Penetrating Injury—A Case Report

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Penetrating injuries are described as involving body parts either partially embedded or fully transected by a foreign body.¹ These injuries often have devastating consequences to the patient. However, case reports

of facial penetrating injuries generally describe these injuries with less morbidity to the patient.¹⁻¹⁹ The face has protective reflexes that help divert it from incoming objects.⁹ The face also has a smaller surface area than the trunk or extremities. Furthermore, the structure of the face and cranium are suited to absorb shocks owing to the presence of resistant pillars, buttresses, and the presence of pneumatized cavities.^{3,7} These anatomic differences might explain why facial penetrating injuries generally result in less concomitant injury to the patient. In addition, fatal facial penetrating injuries generally penetrate the intercranium and are seldom described in published reports. The following case report presents an outline for the management of a facial penetrating injury.

Report of a Case

On November 19, 2007, at approximately 3:30 PM, a 10-year-old boy arrived at the Fresno Community Regional Medical Center emergency room with 2 large pieces of metal impaled in his neck and flank (Figs 1, 2). The injury occurred after a manure spreader was inadvertently driven while the boy was playing beside the machinery. The pieces of metal were heavily soiled with dirt and manure and had T-prongs off the main stem. It was only possible to transport the patient after the metal had been trimmed down enough by the paramedic team to put the boy into the ambulance. His initial Glasgow Coma Scale was 15, and he was answering questions appropriately. His mouth opening was measured at approximately 1 cm, and he was in no respiratory distress at the time. The Fresno Fire Department was enlisted to further cut and remove the metal so the patient could be positioned appropriately on a gurney (Fig 3). The primary and secondary surveys were then performed by

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