

Parotid Basal Cell Adenoma: A Rare Entity

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ABSTRACT

Basal cell adenoma (BCA) is a rare, benign, epithelial tumor of the salivary glands and its cytology closely mimics other conditions. Despite parotid masses being frequently evaluated by fine-needle aspiration cytology (FNAC), a technique that has been demonstrated to have good sensitivity, it is difficult to diagnose BCA with FNAC. We present a 40-year-old male with this uncommon pathology who underwent superficial parotidectomy after a provisional diagnosis of pleomorphic adenoma (PA). It was later confirmed as BCA on histopathology examination. Although BCA is a rare entity, it is a great mimicker of its other benign counterparts. The subtypes of BCA need to be addressed by the surgeon, in planning prognostication and future follow-up.



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1. INTRODUCTION

The most common parotid tumor is pleomorphic adenoma (PA) represents around 45 % – 75 % [1], followed by Warthin tumor (WT) (25-32 %) [2] and basal cell adenoma (BCA)(2% - 7%) [3]. BCA is a rare, benign, epithelial tumor of the salivary glands. It consists of a monomorphic population of basaloid epithelial cells and comprises 1-3% of all salivary gland tumors [2], [4], [5]. About 70 – 75% of these tumors occur in the parotid gland followed by the minor salivary gland from the upper lip 6% and the submandibular gland 5% [1].

The percentage of occurrence of BCA is less compared to parotid PA and WT, but the clinical presentations between these three tumors are similar, such as slow-growing, asymptomatic painless mass, and well-defined cystic or solid mass.

Hence parotid masses are frequently evaluated by fine needle aspiration cytology (FNAC), a technique that has been demonstrated to have good sensitivity before proceeding with histopathology examination (HPE) for confirmation and establishing an accurate diagnosis.

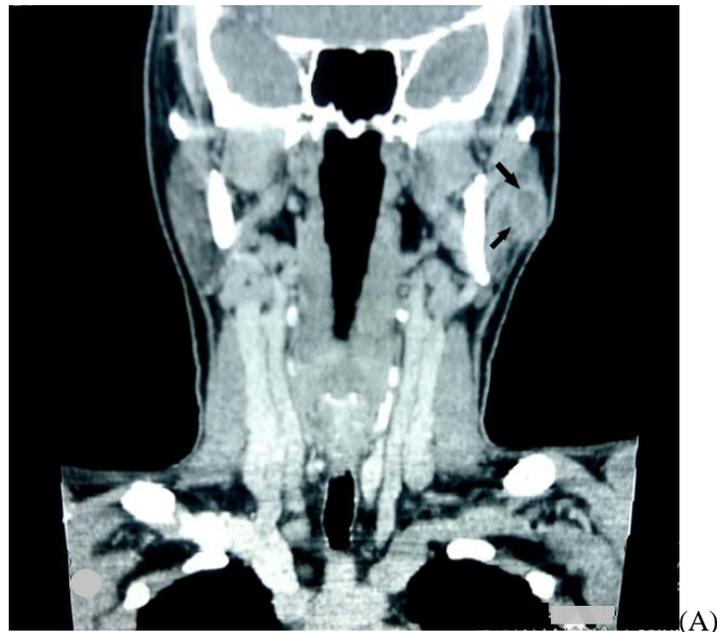
2. Case Report

A 40-year-old male smoker presented with a painless left neck swelling, which progressively increased in size over a 5-year duration. The mass was situated at the angle of the mandible and measured 1 x 2 cm. It was a well-defined firm mass, was mobile with normal overlying skin. There was no facial nerve weakness and no palpable cervical lymphadenopathy. He also denied regarding hypersalivation, pain during chewing or swallowing, increase of tumor during food intake and choking sensation during swallowing.

FNAC was reported as cystic content. Contrast-enhanced computed tomography of the neck demonstrated an ill-defined lobulated heterogeneously rim-enhancing thick-walled cystic lesion measuring 1.9 x 1.5 x 2.5 cm at the superficial lobe of parotid gland with attenuation value ranging from 30-40 HU with no calcification seen within the lesion (Figure 1).

Based on the provisional diagnosis of a benign parotid tumor, a left superficial parotidectomy was performed, and had an uneventful post-operative period with intact facial nerve functions. However, the HPE revealed BCA of monomorphic or solid subtype. The HPE findings further elaborated, trabeculae and membranous pattern which is well-encapsulated tumor composed predominantly monomorphous basaloid cells (7) (Figure 2) with eosinophilic cytoplasm, arranged in solid, trabecular (Figure 3), membranous pattern, and some ductal structures also seen. Peripheral nuclear palisading is seen in the neoplastic lobules; in some areas the stroma shows thick bands of hyaline material (Figure 4). In focal area, cystic degeneration seen and no obvious chondromyxoid component or increase in mitosis seen.

After 3 years of follow up, no complications or recurrence were noted.



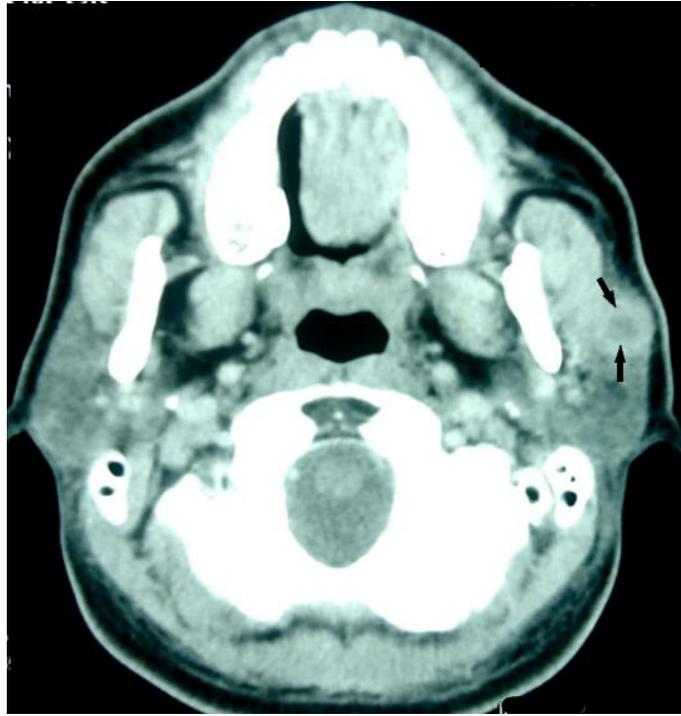


Figure 1 An ill-defined lobulated lesion (black arrow) in the superficial lobe of the left parotid in the coronal (A) and axial (B) views.

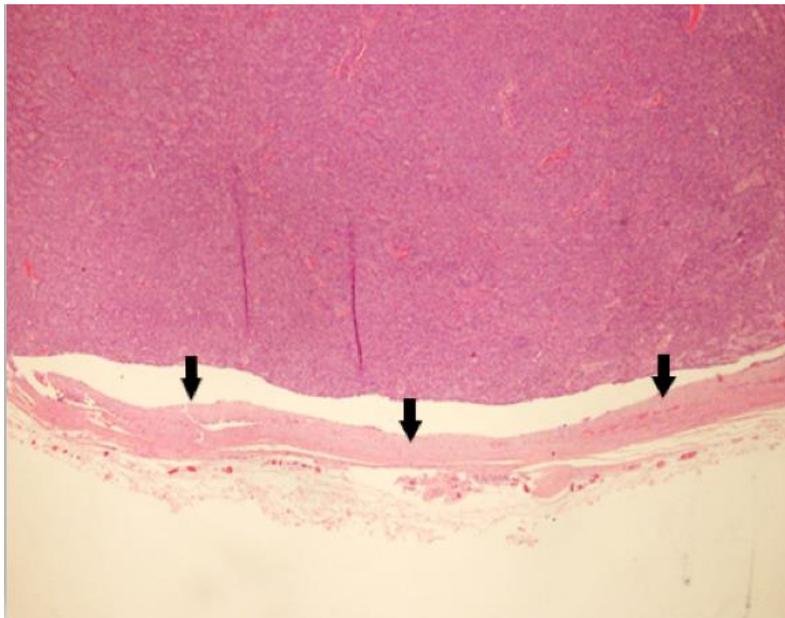


Figure 2 Low magnification x4: well circumscribed tumor composed predominantly epithelial cells

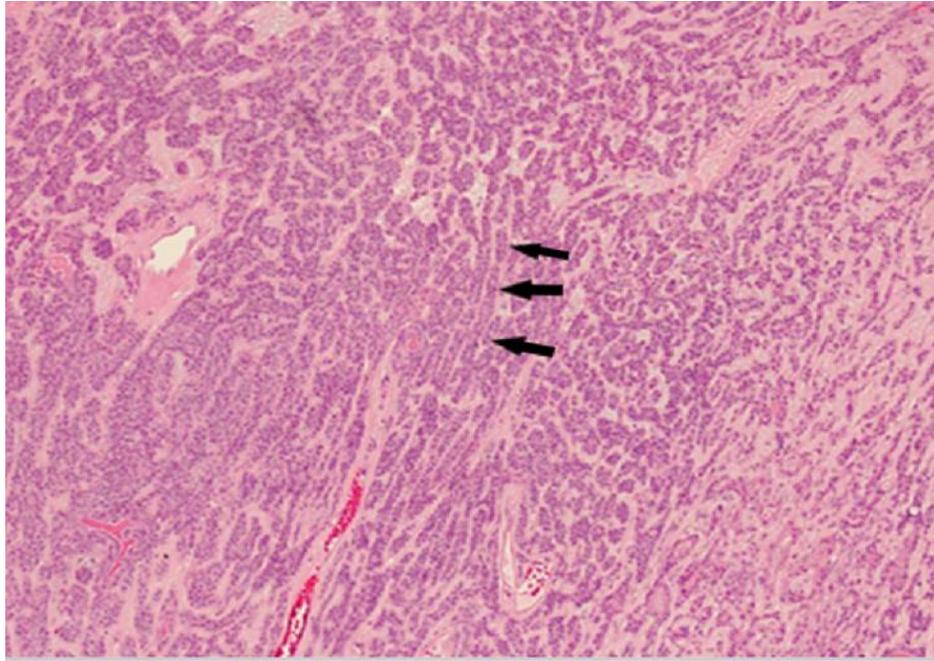


Figure 3 Most of the cells are arranged in trabecular pattern

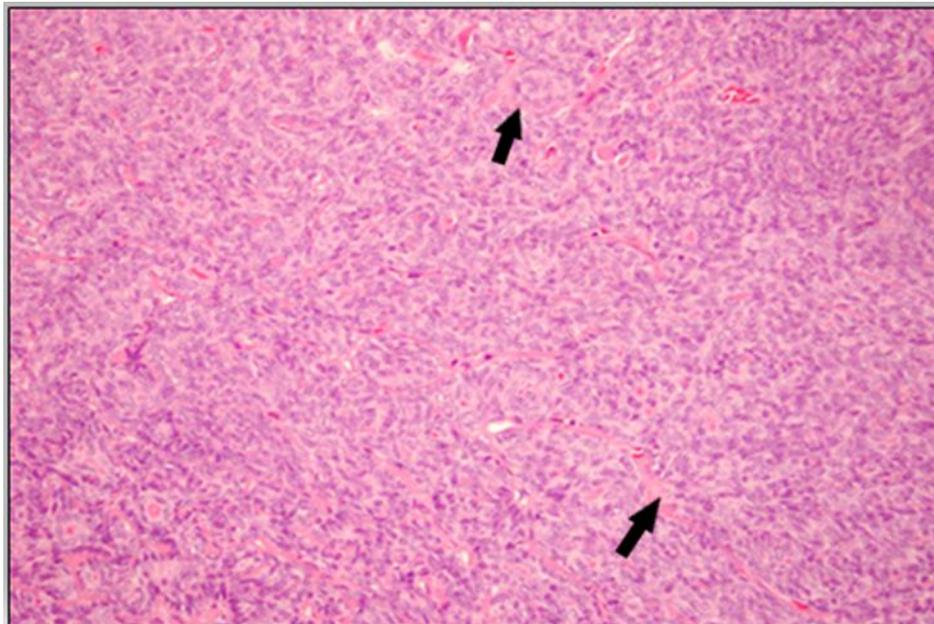


Figure 4 The epithelial cells are separated by hyalinized stroma

3. Discussion

The classical presentations for benign tumors (BCA, PA, WT) are painless, slow-growing mass, mobile, not fixed to the skin, and generally do not cause neural palsies (facial nerve dysfunction, pain, hoarseness) [8]. The parotid gland is the most commonly affected gland.

In our case, WT is the closest differential diagnosis because of a few similarities. BCA and WT usually occur in adults in the 5th to 7th decade of life, with the average patient age being 57.7 years [8]. There is a 2:1 female predominance for both tumor [9], [10]. Meanwhile, the pathogenesis of WT is not clear, because it may originate from inclusions in the lymph nodes or may result from an inflammatory reaction, as patients

who smoke show increased and increase tendency due to retrograde flow of substances from tobacco smoke. Another common differential diagnosis is the most common benign parotid lesion, the PA. PA occurs in all age groups, but the most common is in the 3rd to 6th decades. PA incidence is slightly higher in females than in males in a 2:1 ratio. It affects commonly in the superficial lobe of the parotid gland. Etiology of PA is unknown, but the incidence for the last 15-20 years has increased in trend due to exposure to radiation [1].

It is a common practice to perform FNAC to diagnose type of parotid tumor prior to the surgery decision. Diagnosing BCA via FNAC remains challenging or misinterpreted. This difficulty is usually due to their rarity, sampling limitations [11] of FNAC and the overlapping characteristics of another benign salivary tumor.

Hence, after routine investigations, the patient was subjected to superficial parotidectomy. Surgery is decided depending on the size, location, and extension of the tumor. It serves as a diagnostic and therapeutic purpose as FNAC is not sensitive [4], [8]. Unlike PA, BCA rarely reoccurs post-surgery with a negative margin.

On gross examination BCA mass was encapsulated, lacked extensions outside the capsule, and could be easily enucleated. For PA, it appears an irregular-ovoid mass with well-defined borders. It may have an incomplete fibrous capsule or are unencapsulated. Cut surfaces may be rubbery, fleshy, mucoid, or glistening in color. Areas of hemorrhage and infarction may be noted also. WT meanwhile, spherical or oval, well-circumscribed, encapsulated mass. Once the mass is cut open, solid areas and multiple cysts with papillary projections are identified. The cystic spaces often contain mucoid creamy brown or white fluid, may be fixed to overlying skin, or may undergo hemorrhagic infarction, particularly after fine needle aspiration.

Histologically, BCA has 4 morphological types such as trabecular, trabecular-tubular, membranous or tubular [1,4,7,9] growth, of epithelial cells.

In PA, it contains both epithelial and mesenchymal (or stromal) components with differentiated mesenchymal areas. The epithelial component typically forms structures similar to ducts or cystic structures associated with cells with variable morphology may appear as ducts, tubules, strands, trabeculae, and solid sheets. PA also may exhibit different types of stroma in the same tumor, such as fibrous stroma (most found), followed by myxoid, hyaline, and chondromyxoid stroma.

For WT, histologically, these tumors are well-encapsulated lesions with cystic and solid areas. These tumors consist of an oncocyctic epithelial cell component arranged in double layers, which develops cysts and papillary projections, and a variable amount of lymphoid tissue, often with germinal centers [11]. The immunochemistry of the lymphocyte subtype is similar to that in normal or reactive lymph nodes. A few WT (about 8%) show areas of squamous cell metaplasia and regressive changes [11].

Table 1 Histopathological differences amongst the three common benign parotid gland tumors

	PA	WT	BCA
%	45% - 75%	25% - 32%	2% - 7%
AGE	3-6 decades of life	5 – 7 decades of life	5 – 7 decades of life
F:M	2 : 1	2 : 1	2 : 1
GROSS EXAMINATION	<ul style="list-style-type: none"> - Irregular ovoid mass with well-defined borders - Incomplete fibrous capsule or unencapsulated 	<ul style="list-style-type: none"> - Spherical or oval, well circumscribed - Encapsulated 	<ul style="list-style-type: none"> - Irregular ovoid mass - Encapsulated

CUT SURFACE	- Rubbery - Fleshy - Muroid - Glistering color	- Solid area - Multiple cyst - Papillary projection	- Solid - Homogenous, -Focal dark brownish area
HISTOLOGY	- Mixture of epithelial, myoepithelial and stromal components - Epithelial cells: nests, sheets, ducts, trabeculae - Stroma: myxoid, chondroid, fibroid, osteoid	-Papillary projection into cystic spaces surrounded by lymphoid stroma - Epithelium: Double cell layer (luminal and basal cells) - Stroma: mature lymphoid follicles with germinal centers	- Mixture of solid, trabecular, tubular and membranous - Composed of basaloid cells
RECURRENCE	High	Low	Rare

Treatment for BCA is surgical, either a superficial parotidectomy or a total parotidectomy, depending on the tumor location.

The prognosis of BCA is generally good, with a very low recurrence rate or almost not exist for solid and trabecular-tubular variants, except for the membranous type (recurrence rate of approximately 25%) [10], hence patient was subjected for routine surveillance after the operation up to 3 years, to look for any recurrence.

4. Conclusion

Although BCA is a rare entity, it is a great mimicker. Diagnosing the disease via FNAC alone for further treatment is not conclusive. Thus a proper and detailed HPE of the excised mass is required for differentiating with other pathology. In view of that, histopathology investigation is crucial for accurate diagnosis and subtypes, for further management and prognostic implication.

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