



## Unusual presentation of Basal Cell Adenoma Parotid Gland: A Case Report

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### Abstract

Basal cell adenoma (BCA) of the salivary gland is a rare neoplasm consists of a monomorphic population of basaloid epithelial cells, and it accounts for approximately 1–2 % of all salivary gland tumors. Its most frequent location is the parotid gland. It usually appears as a firm and mobile slow-growing mass. Histologically, isomorphic cells in nests and interlaced trabecules with a prominent basal membrane are observed. In contrast to pleomorphic adenoma, it tends to be multiple and its recurrence rate after surgical excision is high. Due to prognostic implications, differential diagnosis with basal cell adenocarcinoma, adenoid cystic carcinoma and basaloid squamous cell carcinoma is mandatory. We report an unusual presentation of BCA of the parotid gland

**Keywords:** Membranous type, Basal cell adenoma, Parotid gland, Salivary gland tumors

### Introduction

Basal cell adenoma (BCA) of the salivary glands is a rare benign tumor with a high recurrence rate and, in general, good prognosis, is recognized as an independent entity in the Second Edition of the Salivary Gland Tumors Classification of the World Health Organization (WHO).[1] Basal cell adenoma (BCA) of the salivary gland is a rare neoplasm consisting of a monomorphic population of basaloid epithelial cells, and it accounts for approximately 1–2 % of all salivary gland tumours [2]. BCA appears most frequently in the parotid glands and in adult in 5<sup>th</sup> to 7<sup>th</sup> decade [3–5]. Clinically, BCA is usually a slow-growing, asymptomatic, and freely movable mass. We report a case of BCA in parotid gland in a 38yr old female presenting as a slow growing painful swelling in right parotid region.

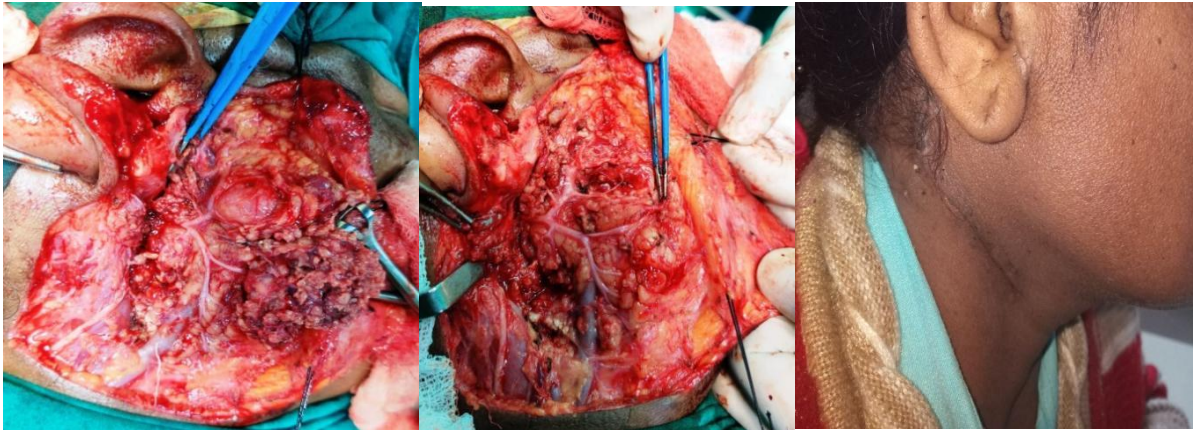
### Case Report

A 38 year old female presented to department of otolaryngology with chief complains of painful swelling in right parotid region since one year. On examination there was single, firm, tender swelling palpable in right parotid region. There was no palpable lymph node in the neck and facial nerve function was intact. On fine needle aspiration cytology small clusters of epithelial cells bound together with occasional epitheloid cells and mesenchymal cells present in stromal matrix. Myxoid stroma with fibrillar structure also seen occasionally. Cytopathological features suggestive of pleomorphic adenoma. Ultrasonography showed a well defined slightly lobulated heterogenous (22 x 20 18 mm) lesion in right parotid gland. A small cystic area noted within the lesion with posterior acoustic enhancement. Minimal internal vascularity noted on colour Doppler. Radiologist reported the mass lesion in right parotid to be suspected pleomorphic adenoma or muco epidermoid carcinoma. Patient planned for

Superficial parotidectomy after thorough investigation, anesthetist fitness for surgery and informed written consent for facial nerve injury. Under General Anaesthesia, Modified Blair's incision given, Sub myoaponeurotic system and subplatysmal flap elevated. Facial nerve with all its branches

identified and preserved. Along with tumor complete superficial part of parotid gland excised and subjected to histopathological examination. (Figure 1). Macroscopically tumor was about 2 × 2 cm in dimension, firm and grayish white in appearance. (Figure 2).

Figure 1: Superficial parotidectomy before and after tumor excision. Note the post operative aesthetic scar.

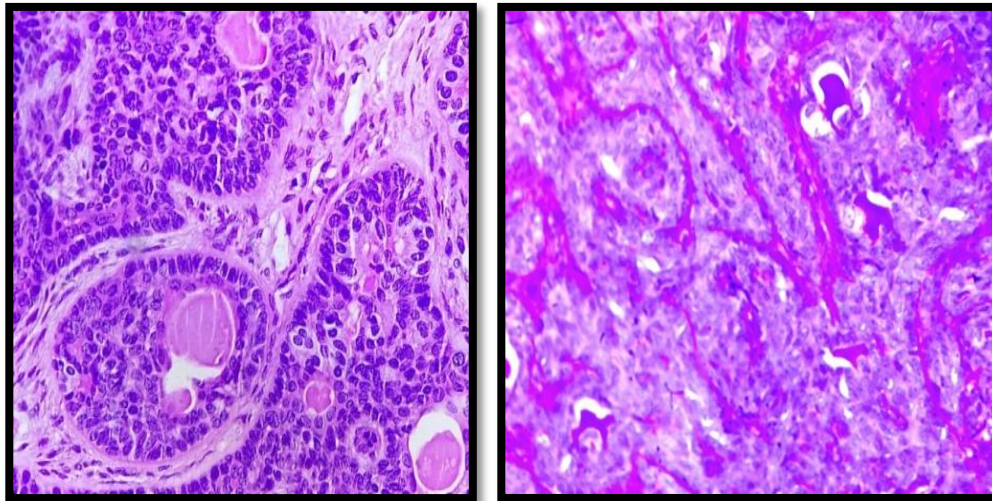


On histopathology section shows Overall findings suggestive of BCA, membranous type. (Figure 3) A special stain P.A.S was also used for confirmation. (Figure 4) Postoperative period was uneventful with a good aesthetic scar. Patient is on regular follow up.

Figure 2: MACROSCOPY :Encapsulated tumour measuring 2-cm of major diameter. It was a solid firm mass, within the superficial salivary gland body, with a brownish appearance.



Figure 3 : MICROSCOPY :This image depicts monotonous cellular growth composed of two cell types small hyperchromatic cells with scanty cytoplasm at the periphery of the anastomosing nests and larger polygonal cells with abundant cytoplasm. The stroma has scant amount of spindle shaped myoepithelial cells. A basal membrane-like structure rounds these tumoral nests, separating them from the surrounding connective tissue .



### Discussion

Basal cell adenoma, as defined by WHO, is a distinctive benign neoplasm composed of basaloid cells organized with a prominent basal cell layer and distinct basement membrane-like structure and no myxochondroid stromal component as seen in pleomorphic adenoma. Four cellular patterns occur: Solid, Trabecular, Tubular, Membranous.

The BCA was once considered to be a type of “monomorphic adenoma”. However, since 1991, according to the “Salivary Glands Tumours Histological Classification” of the World Health Organization, the name of this lesion was changed to BCA, excluding the word “monomorphic” [6]. Among the “monomorphic adenomas,” these are the following varieties: Warthin’s tumor or papillary cystadenoma lymphomatosum, oncocytoma or oxyphilic adenoma, BCA, canalicular adenoma, and sebaceous adenoma.

The salivary gland tumors are uncommon, representing less than 3 % of all neoplasms of the head and neck [7]. Although it is the most common variant in the group of “monomorphic adenomas,” BCA represents only 1 to 2 % of all salivary tumors [8].

A total of 42 cases of cytologically diagnosed BCA have been reported in the literature. False-positive and false-suspicious diagnoses accounted for 16.7 % of cases, illustrating the difficulties in distinguishing between BCA and adenoid cystic carcinoma [9].

Frequently, this slow-growing encapsulated tumour do not exceeds 3-cm of major diameter. It is a firm mobile painless mass, but our patient presented with painful swelling. It is usually superficial within the glandular body, and a brownish appearance is usually observed [10]. The most frequent location is the parotid gland, although other sites are possible, such as the upper lip, buccal mucosa, lower lip, palate and nasal septum. Epidemiologically, these tumours frequently affect patients between their fifth and seventh decades ( our patient was 38yr old female), in contrast to observations in benign mixed tumours. The diagnosis of this entity must be established by the histological study. Generally, biopsy is accepted as the most accurate method to obtain the diagnosis, although some authors advocate for FNAC if physical access to the tumour is available.

Histologically, BCA is characterized by the presence of uniform and regular basaloid cells. These cells have two different morphologies and are



intermingled. One group consists small cells with little cytoplasm and intensive basaloid rounded nuclei that are usually located in the periphery of the tumoral nests or islands. The other group is formed by large cells with abundant cytoplasm and pale nuclei that are located in the centre of the tumoral nests. A basal membrane-like structure rounds these tumoral nests, separating them from the surrounding connective tissue [11]. In our case the histopathology showed monotonous cellular growth with cells having round to ovoid hyperchromatic nuclei with pale to eosinophilic to amphophilic cytoplasm and indistinct cell border (basaloid cell) tumor showed trabecular as well as cribriform pattern. There were no mitotic figure and perineural invasion is not seen. Because BCA may have solid and cystic component, the imaging findings have been described as relatively non-specific. Although it can be cystic, was purely solid in our patient. Histo pathologically; it has four growth patterns as a solid, trabecular, tubular and membranous type. Solid type is the most common histopathologic type followed by tubular, membranous and trabecular type. Our case had membranous type which is rare. **Membranous** type is constituted by external cells in a stockade pattern and circled by an intense hyalinised basal membrane.

Immunohistochemistry: Basal cell adenoma is characterized by *CTNNB1*(beta-catenin) gene mutation and loss of heterozygosity at 16q12-13. Basal cell adenomas are amenable to conservative resection such as local excision or superficial removal of the gland, whereas the membranous subtype requires complete resection of the entire gland. The recurrence rate for the solid and trabecular, tubular variants is almost nonexistent. This contrasts with the high recurrence rate (24%) of the membranous type, which is perhaps a result of the multicentricity of this lesion. Although exceedingly rare, malignant transformation (4%) is more common in the membranous type than in the other types[15].

Among the malignant tumours, the adenoid cystic carcinoma is the lesion that shows the most histologic similarities to the BCA, suggesting that the latter is the benign homologue of the adenoid cystic carcinoma. However, characteristics such as integrity of the basal layer, decreased number of mitoses, and slow growth are typical of a benign lesion.

The basal cell adenocarcinoma is another malignant tumour that shares histologic features with the BCA. Both exhibit myoepithelial differentiation, reactivity patterns indicative of ductal epithelium, and closely similar immunohistochemical profiles. Basal cell adenocarcinoma is distinguished from BCA by the histologic features of invasion, mitotic activity, and neural or vascular involvement [6, 12,13,16]. Nuclear Beta- catenin expression was present in 97.6% of basal cell adenoma but not in adenoidcystic carcinoma. *CTNNB1* mutations were found in 60% of basal cell adenoma but not in basal cell adenocarcinoma [14].The differential diagnosis must include the pleomorphic adenoma, which is the most common benign tumour of the salivary glands, and other salivary gland tumours such as the canalicular adenoma and sebaceous adenoma. Primary treatment of BCA is surgical excision by means of a superficial or total parotidectomy in cases in which parotid affection exists. Extracapsular excision is performed in cases in which there is affection of minor salivary glands in the oral mucosa. The treatment used in this case was the same proposed in the literature.

### Conclusion

BCA is an uncommon benign tumor. As the tumor present in fifth and sixth decade of life a high index of suspicion is necessary to differentiate it from malignant tumor arising from same location. And also it is necessary to perform a complete excision of the tumor prior to the making of the final diagnosis. Vigilant comprehensive analysis of all the pertaining clinicopathologic, histopathologic features and IHC analysis are required for differentiating lesions with basaloid differentiation which can sometimes pose challenge to pathologist. With increased awareness and acceptance of this lesion as a separate entity by pathologists, we perhaps can expect an increase in its diagnosis.

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