Original Research Paper

Volume - 13 | Issue - 01 | January - 2023 | PRINT ISSN No. 2249 - 555X | DOI : 10.36106/ijar

Pathology

BASAL CELL ADENOMA OF PAROTID GLAND: A RARE CASE REPORT AND REVIEW OF LITERATURE

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ABSTRACT Salivary gland tumors are uncommon and constitute about 2.0 to 6.5 % of all head and neck tumors. Basal cell adenoma (BCA), which is classified in benign salivary gland tumors by WHO in 1991, is a rare neoplasm. Incidence rate of BCA is 1.0 %–3.7 % of all salivary gland tumors with most frequent location is the parotid gland(>80% cases of BCA) and other sites such as the upper lip, buccal mucosa, lower lip, palate and nasal septum rarely affected. It usually presents as a firm and mobile slow-growing mass. Histologically, it consists of a monomorphic population of basaloid epithelial cells that are arranged in the solid, trabecular, tubular and membranous patterns. It is important to differentiate BCA from benign tumor like pleomorphic adenoma showing absence of chondromyxoid stroma in former and malignant tumor like basal cell adenocarcinoma, adenoid cystic carcinoma and basaloid squamous cell carcinoma. We report a case of BCA of the parotid gland in 59 year male. We also review the literature and discuss the diagnosis and management of this rare entity.

KEYWORDS : Basal cell adenoma, Parotid gland, Salivary gland tumors

1. Introduction

BCA is one of salivary gland tumors reported by Kleinsasseret al. in 1967 for the first time (1). It was classified as a monomorphic adenoma among salivary gland epithelial tumors in the 1972 Classification of Salivary Gland Tumors of the World Health Organization (WHO)(2). In 1991, it was subsequently classified as anindependent tumor for the first time along with Warthin tumor, myoepithelial tumor (epithelioma), etc. in the WHO Histological Classification of Salivary Gland Tumors 2nd edition(3), the same in the 3rd edition in 2005(4), and the 4th edition in 2017(5). It represents 1-3.7% of all salivary glands tumour. They are typically seen in the 7th decade with a 2:1 female predilection(5). The tumor is generally slow growing, painless, mobile without any dysfunction. The prognosis is generally very good, with a very low recurrence rate; except for membranous type, which has recurrence rate of approximately 25%(5).

2. Case report

A 59-year-old male patient presented with painless, slowly growing left cheek swelling since two year. On examination, it was 4x4cm2 size, mobile and firm in consistency. Radiological investigation was done and showed possibilities of benign lesion – most likely Pleomorphic adenoma. Patient was treated with superficial lobectomy and the specimen was sent to pathology department for histopathological.

We received excised specimen of left parotid mass which was well circumscribed globular mass measuring 3.5x3x2.5 cm3 in size, grayish white in color, firm in consistency. Cut surface shows solid whitish areas and focally cystic areas containing hemorrhagic fluid identified. sections were given from different representative areas, processed, stained with Hematoxylin & Eosin stain. Gross and histopathological examination showed well encapsulated tumor, tumor cells arranged in tubulotrabecular, solid and cribriform pattern.(Figure 2)

Tumor cells are composed of basaloid/blue cells with peripheral palisading arrangement having round/oval nuclei, indistinct cell membrane and scant amount of cytoplasm. (Figure 3 A&B) Many area shows cystic degeneration with eosinophilic secretion. Chondromyxoid stroma is not seen. No mitosis, cytological atypia/ necros is seen. Overall gross and histological findings are suggestive of Basal Cell adenoma. IHC was performed. Tumour cells are immunoreactive for Beta Catenin. Peripheral tumor cells are immunoreactive for SMA. Ductal cells are immunoreactive for CK 7(Figure 4 A,B,C). Overall gross findings, histological features and IHC findings are suggestive of Basal Cell Adenoma of Parotid gland.

Figure 1 : Cut surface showing solid whitish areas and focally cystic areas containing hemorrhagic fluid

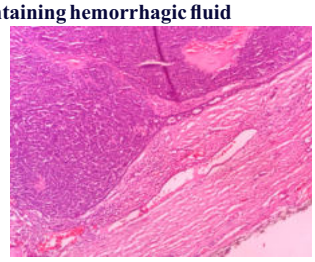

Figure 2: well encapsulated tumor, tumor cells arranged in tubulotrabecular, solid and cribriform Pattern (H&E:4x)

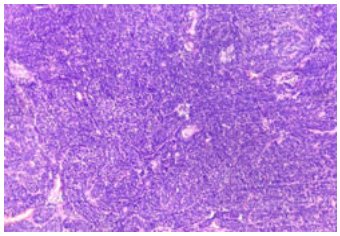

Figure 3 A: Tumor is composed of basaloid/blue cells

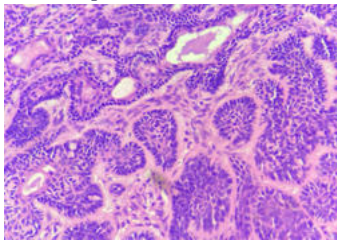

Figure 3 B: Tumor is composed of blue cells with peripheral

palisading arrangement and surrounded by basal membrane like hyaline substance

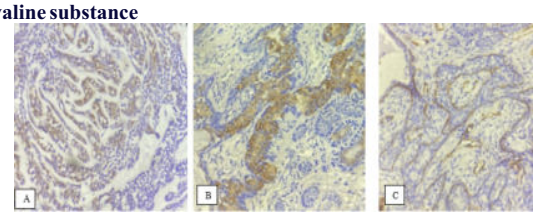

Figure 4 A,B,C: A) Tumour cells are immunoreactive for Beta Catenin. B) Peripheral tumor cells are immunoreactive for SMA. C) Ductal cells are immunoreactive for CK 7

Discussion:

The incidence rate of BCA is 1.0%-3.7% of all salivary gland tumours(5). The parotid gland is the most common site for BCA with 40-80% incidence(6), while the upper lip, buccal mucosa, lower lip, palate, and nasal septum being less frequently affected(5).In 1991, 160 cases of BCA were registered at the Armed Forces Institute of Pathology (AFIP), which constitute 1.8% of all benign epithelial salivary gland tumors, out of those BCA 75% were reported in the parotid gland and 20% in the minor salivary glands of the upper lip(7). BCA mostly arise in older age with the mean age of the patient is 57.7 years with peak incidence in seventh decade of life with slight predominance of this tumor in females(5), although membranous variant has an equal gender distribution(8). They typically present as slowly growing , solitary painless mass(9).

In order to diagnose BCA, a biopsy is considered to be the most accurate method. Most BCAs are well circumscribed and well encapsulated by fibrous tissue. Although the aetiology is uncertain, the multiple cylindroma and trichoepithelioma (Brook-Spielger Syndrome)are associated with membranous type, while the tubulotrabecular type of BCA, is linked to a mutation in the CTNNB1 gene which encodes for beta-catenin(9).

The gold standard test for confirming a BCA diagnosis is histopathological examination(10).

Histologically, BCA consists of 2 types of cell populations (basaloid cell and luminal duct cells). Basaloid cells are small cuboidal or columnar shaped, present peripherally in a palisading arrangement within the tumor nests or cords, with round deeply stained nuclei and scant cytoplasm. While, luminal cells are located centrally, larger with modest cytoplasm, indistinct cell borders and a pale staining oval nucleus. Tumor nests are surrounded by basement membrane like hyaline material(8). The solid variety is the most prevalent of the four histologic subtypes(trabecular, tubular, and membranous) of basal cell adenoma that Gardner and Daley described(9). However, each tumor has combination type.

Despite the fact that immunohistochemistry (IHC) tests are non-specific, inconsistent, and largely reliant on the histologic subtype of BCA(8), it can be used as an adjunct in the differentiation of these tumors. The peripheral tumour cells of BCAs exhibit immunopositivity for the S-100, SMA, and Vimentin, While luminal cells show immunoreactivity for cytokeratin, epithelial membrane antigen (EMA), and carcino embryonic antigen (CEA). Tubulotrabecular type of BCA may show nuclear positivity for beta-catenin.(9). Hemachandran et al. showed selective pancytokeratin, S-100, and smooth muscle actin (SMA)positivity, which implies the involvement of myoepithelial cells in the histogenesis of BCA(6).

Our case showed histological and IHC findings similar to those reffered in the literature, consisting of basaloid cells and luminal cells, arranged in tubular, trabecular and solid pattern showing Immunoreactivity for B-catenin(outer tumor cells), SMA (myoepithelial cells) and CK 7(luminal cells).

It is crucial to distinguish BCA from pleomorphic adenoma (PA), adenoid cystic carcinoma (ACC), basal cell adenocarcinoma (BCAC), and basaloid squamous cell cancer(Basaloid SCC) despite the fact that BCA has a unique appearance.

Lack of myxochondroid stroma and immunonegativity for glial fibrillary acidic protein(GFAP) helps to differentiate BCA from pleomorphic adenoma(6). Our case did not show any chondromyxoid stroma. The adenoid cystic carcinoma(ACC)(9) is the lesion that shows the most histologic similarities to the BCA, However, characteristics such as integrity of the basal layer, decreasednumber of mitoses, lack of perineural invasion and slow growth are typical of a benign lesion. Our case showed well circumscribed tumor without perineural invasion.

Differentiation of BCA from its malignant counterpart BCAC is crucial as shares histologic features, however an infiltrative growth, more mitotic figures (>4 mitotic count/10 HPF) and may show angio lymphatic and perineural invasion along with Ki67 index greater than 5%(8) suggests BCAC. Our case did not show features of BCAC. Basaloid SCC is characterized by the presence of solid cells in a lobular fashion, close to the superficial mucosa, cells are small and have scant cytoplasm with hyper-chromatic nuclei without nucleoli. In this latter entity, both populations of basal cells are not observed, in contrast to BCA. Continuity of tumoral cells with epithelium of the surface and squamous dysplasia are also observed, in contrast to BCA(8).

4) Conclusion:

Basal cell adenoma is a specific type of monomorphic benign tumor of salivary glands. It is important to differentiate BCA from benign tumor like Pleomorphic adenoma, It's malignant counterpart like Basal cell adenocarcinoma and other malignant tumors like adenoid cystic carcinoma, and basaloid squamous cell carcinoma due to prognostic implications. For this reason, careful, thorough examination of all relevant clinicopathological, histological, and IHC findings are necessary for its diagnosis, which can occasionally present a challenge to pathologists. We would anticipate a rise in diagnoses as a result of pathologists' greater understanding and acceptance of this lesion as a distinct entity.

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