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Basal Cell Adenoma of Parotid Gland: A Rare Case Report and Review of Literature

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Abstract: ***Basal cell adenoma of salivary gland is an uncommon type of monomorphic adenoma. Approximately 70% of basal cell adenomas occur in the parotid gland, the rest involve the minor salivary glands, especially those in the upper lip. It accounts for approximately 1-3% of all salivary gland tumors. It usually occurs in adult with slight predilection for females. We report a case of 35 year old female who presented with 3x2 cm painless firm fixed swelling in her right parotid region since 6 months. On Fine needle aspiration cytology (FNAC),a diagnosis ofbasaloid neoplasm was given. Histopathologically, the tumor was characterized by tubular and trabecular areas composed of small isomorphic cells with hyperchromatic, round nuclei and an eosinophilic cytoplasm. The tumour cells were clearly separated by a well-defined basement membrane, and the diagnosis was made as tubular-trabecular type of basal cell adenoma of parotid. On immunohistochemistry,the inner cells of tubular nests were positive for cytokeratin (CK) 7 and that of outer basaloidcells were positive for smooth muscle actin (SMA) and the stromal cells were positive for S100.***

Keywords: Basal cell adenoma, Immunohistochemistry, parotid, Salivary gland

1. Introduction

Salivary gland tumours constitute about 3–4% of all head and neck neoplasm.[1] Basal cell adenoma (BCA) accounts for only 1 to 3% of all Salivary gland tumours and demonstrates a female predominance of 2:1.[2] Age group most commonly affected are between sixth to seventh decades.[3] The basal cell adenoma was once considered to be a type of "monomorphic adenoma". However, since 1991, it was recognised as an independent entity in the Second Edition of the "Salivary Gland Tumours Histological Classification" of the World Health Organization (WHO).[4] The most frequent location is the parotid gland (80%), although other sites are possible, such as the upper lip, buccal mucosa, lower lip, palate and nasal septum.[5] Basal cell adenoma is a benign salivary gland tumour comprised of uniform appearing basaloid cells arranged in solid, trabecular, tubular, and membranous patterns, but lacking the myxoid and chondroidmesenchymal-like component as seen in pleomorphic adenoma. However, basal cells are found in several primary salivary gland tumors either as a major component presents in a mixture of cell types or as pure basal cell neoplasms.[2] In this regard, the distinction between the true neoplasm and other primary tumors with basal cell features pose diagnostic difficulties.

For several years many reviewers erroneously interpreted the „‟monomorphous‟‟ appearance to indicate isomorphism of the epithelial cell type and absence of myoepithelial differentiation.[6-12]More recently evaluations, including electronmicroscopic and immunohistochemical studies have shown that basal, ductal, and myoepithelial cell differentiation occurs to variable degrees in basal cell adenomas. [13-22]

Different investigators reported varying results of immunohistochemicalstaining in basal cell adenomas.[17, 20, 21, 23, 24]

Cytokeratin is demonstrable in nearly all tumors, but the number of reactive cells varies from few to many. Similarly, immunoreactivity to S-100 protein, smooth muscle actin (SMA), and vimentin can be demonstrated in most basal cell adenomas but is typically localized tothe peripheral tumor cells adjacent to the connective tissue stroma.[21, 23–25]

Morinaga et al.[27] have reported focal reactivity for myosin. Williams et al[34] have even found rare, faint staining for glial fibrillary acidic protein (GFAP), a tumor marker most often associated with mixed tumors, others have not seen this.[22, 24, 26]
Carcinoembryonic antigen and epithelial membrane antigen reactivity is mostly confined to luminal cells. [17, 20, 21]

Dardicket al. described some tubulo-trabecular basal cell adenomas with S-100 protein immunoreactivity of spindled "stromal" cells that they interpreted as myoepithelial cells with electron microscopy. [25]

2. Case Report

A 35 year old female patient who presented to the Department of ENT KIMS Hubballi with a complaint of swelling in her right parotid region since 6 months, which was measuring nearly 3x2 cm. On physical examination the swelling was firm, non-tender. No other lymph nodes or masses were palpable in the head and neck region.There was no evidence of facial nerve involvement. CT scan revealed a well-defined focal hyperdense lesion in the right parotid region, wassuspicious of adenocarcinoma. On Fine needle aspiration cytology (FNAC), features were consistent with basaloid neoplasm.

Gross Examination

After surgical excision specimen was sent to the department of pathology KIMS Hubballi. The specimen obtained was of 2x2 cm globular well encapsulated mass with attached normal parotid tissue (Fig 1). The cut surface of the mass

was solid, homogenous with focal dark brownish areas (Fig 2).

Figure 1: Gross specimen showing 2x2 cm globular encapsulated mass with attached normal parotid tissue.

Figure 2: Cut surface of mass showing solid, homogenous with focal dark brown areas.

Microscopic Examination

Sections studied from the tumor shows a well-definedtumor covered with fibrous capsule. The tumor cells are arranged in tubular and trabecular pattern and are sharply demarcated from the stroma by basement membrane(Fig 3).The basaloid cells making up the bulk of the tumor arefound to be monomorphic. The peripheral cells are palisaded with a cuboidal-to-columnar shape, while the central cells are relatively rounded. These peripheral cells are hyperchromatic, while the central cells had pale staining nuclei (Fig 4). There is no cartilage formation, mucous stroma or necrosis in the tumor. There is no nuclearatypia and mitotic figures. With these classical histopathological findings, the tumor wasdiagnosed as tubular-trabecular type of basal cell adenoma of parotid gland.

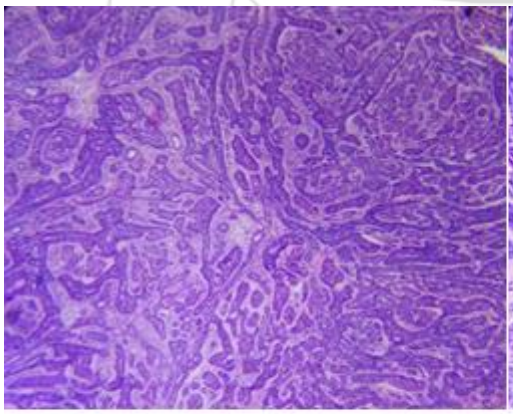

Figure 3: Microscopy showing tubular and trabecular arrangement of basaloidtumor cells sharply demarcated by basement membrane. (H & E 10x10)

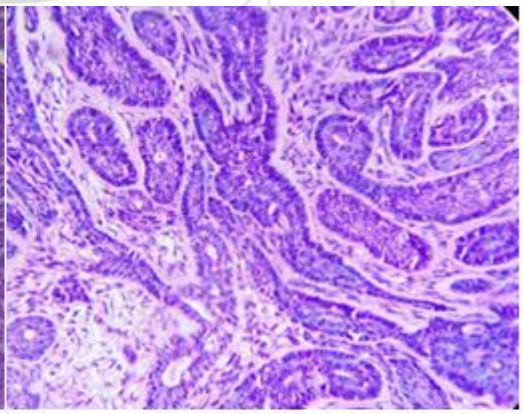

Figure 4: Microscopy showing peripheral palisading of basaloid cells with central round cells. (H & E 40x10)

Immunohistochemistry

On immunohistochemistry (IHC) the tumor shows that the inner epithelial cells of tubular components is positive for cytokeratin (CK) 7 and negative for smooth muscle actin (SMA). On the contrary, the outer basaloidcells are positive for SMA and negative for CK7. The stromal cells are positive for S100 (Fig 5, 6 and 7).

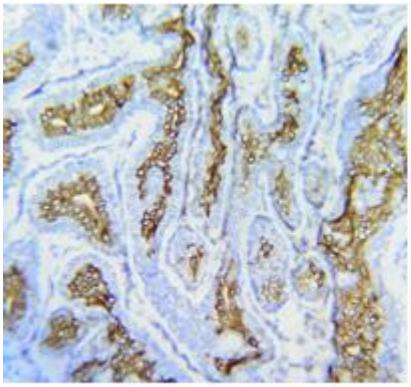

Figure 5: IHC showing inner epithelial cells positive for CK7.

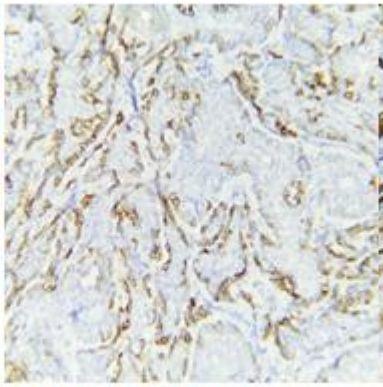

Figure 6: IHC showing outer basaloid cells positive for SMA.

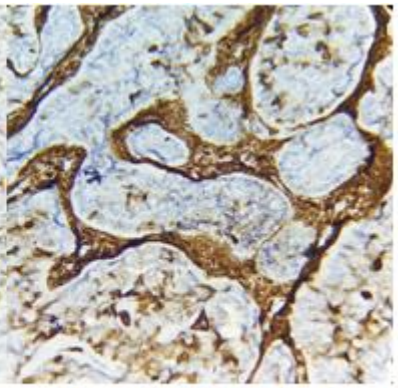

Figure 7: IHC showing stromal cells positive for S100.

3. Discussion

Most of the parotid tumours (70-80%) are benign and within this group, pleomorphic adenoma is most frequent. Within the adenomas group, monomorphic tumours are very uncommon. They are defined as epithelial benign tumours of salivary glands which are not pleomorphic adenomas. Within this group, basal cell adenoma must be signalled. This tumour is subdivided into solid, trabecular, tubular and membranous subtypes. Frequently, this slow-growing encapsulated tumour do not exceeds 3cm of major diameter. It is firm mobile painless mass. It is usually superficial within the glandular body and brownish appearance is usually observed.[7]The diagnosis of this entity must be established by histopathological study. Generally, biopsy is accepted as the most accurate method to obtain the diagnosis, although some authors advocate Fine needle Aspiration (FNA) if physical access to the tumour is available.

Histologically, basal cell adenoma is characterized by the presence of uniform and regular basaloid cells. These cells have two different morphologies and are intermingled. One group consists in small cells with little cytoplasm and intensive basaloid rounded nuclei that are usually located in the periphery of the tumor 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 tumor nests. A basement membrane-like structure surrounds these tumor nests, separating them from the surrounding connective tissue. Globally, as it has been referred in classic texts, the tumor adopts an ameloblastoma-like pattern.[28]

Solid basal cell adenomas are formed by small cells organized in a compact manner. In the trabecular and tubular subtypes, important groups of cells exist they are disposed in narrow bands and ductal structures or in a combination of both. Membranous subtype is constituted by external cells in a stockade pattern and by an intense hyalinized basement membrane material. Some authors have referred the existence of an association between this type and cutaneous cylindroma, trichoepithelioma or eccrinespiradenoma of the scalp. This association has been observed in one third of the cases. Equally, it has been referred an association of glandular and cutaneous tumours in this Basal cell adenoma subtype, as an autosomal dominant disease, due to the presence of affected subjects within the same family.[29-31]

Differential diagnosis must be established with cellular pleomorphic adenoma and also with some unfavourable entities such as basal cell adenocarcinoma, adenoid cystic carcinoma. The monomorphic appearance and the absence of chondroid tissue and myxoidstroma differentiate basal cell adenoma from pleomorphic adenoma. The basal cell adenocarcinoma, the malignant counterpart of basal cell adenoma needs to be excluded that shares similar histologic features. Both exhibit myoepithelial differentiation, reactivity patterns indicative of ductal epithelium and has similar immunohistochemical profiles. Basal cell adenocarcinoma is distinguished from basal celladenoma by the histologic features of invasion, mitoticactivity, and neural or vascular involvement.[33]

The basal cell adenomas are some time mistaken for adenoid cystic carcinoma. There are two features that help to distinguish these lesions. One is the circumscription of the basal cell adenoma, which contrasts with the invasive pattern of adenoid cystic carcinoma and the other is the lack of vascularity in the microcystic areas of adenoid cystic carcinoma, in contrast with the numerous endothelial-lined channels in basalcell adenoma.[35] It is interesting that basal cell adenomas have microscopic features that may help in the differential diagnosis. Tumor nests are clearly differentiated from inter-epithelial stroma because of an intact basement membrane. This delimitation is observed neither in the pleomorphic adenoma nor in the adenoid cystic carcinoma.[32]

Basal cell adenoma may rarely transform (4%) to malignant conditions like basal cell adenocarcinoma,adenoid cystic carcinoma,salivary duct carcinoma,or adenocarcinoma NOS. This transformation rate is higher in membranous subtype, being upto 28%.[34]

4. Conclusion

Basal cell adenoma is a very rare benign parotid tumor. FNAC is inconclusive but the confirmatory diagnosis can be made by histopathological examination and immunohistochemistry.

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