Case report:

Basal cell adenoma: a rare case report of huge parotid tumor!!

*Gore Pratibha G1, Saurabh Sabnis2, Thorat Pravin S3, Kulkarni Meena M4, Kalburge Jitendra V5

1 Sr. Lecturer, 4 HOD & Professor, Department of oral and Maxillofacial pathology & microbiology, Rural dental college, PIMS, loni.
2 Sr. Lecturer, Department of oral and Maxillofacial pathology & microbiology, Y.C.D.C dental college, Ahmednagar
3 Associate Professor, Department of Anaesthesiology & Critical Care, Rural Medical College, PIMS, Loni.
4 Professor & Head, Dept of Oral and Maxillofacial Pathology, Government Dental College & Hospital, Jamnagar- 361008.
*Corresponding author: Dr. Pratibha Gore, Sr. Lecturer, Department of oral and Maxillofacial pathology & microbiology, Rural dental college, PIMS, Loni

Abstract:
Basal cell adenoma is a benign salivary gland epithelial neoplasm composed of a proliferation of small basaloid cells. Histogenesis is most likely from intercalated ducts or the basal cells of striated ducts. These tumors composed of small, dark basaloid cells. They represent < 5% of all salivary gland neoplasms. In the parotid gland these neoplasms are clinically indistinguishable from Pleomorphic Adenomas, because they also tend to arise in the superficial lobe, are well encapsulated, and movable. Most are small tumors less than 3 cm in diameter. Here we report one of the rare case of basal cell adenoma of the parotid as it was huge in size & weight, than cases reported till now.

Key Words: Salivary gland, Salivary gland neoplasm, Basal cell adenoma.

Introduction:
Basal cell adenoma is a benign salivary gland epithelial neoplasm composed of a proliferation of small basaloid cells in solid, tubular, trabecular or membranous patterns. Histogenesis is most likely from intercalated ducts or the basal cells of striated duct. Basal cell adenoma represent usually an asymptomatic, solitary, slow growing mass, with almost 75% arising within the parotid gland. The tumor can occur at any age but is most common in middle-aged & older adults, with a peak prevalence in the seventh decade of life. The tumors appears to be more common in women, with some studies showing as high as 2:1 female-to-male ratio. Parotid tumors usually are located within the superficial lobe of parotid.

Case report:
A 65 year female presented with a chief complaint of enlarging painless mass on right side of the face. She noticed a small, non tender, slightly movable swelling over the preauricular region of the face.

[FIG.1] profile view
Eventually swelling increased in size extending till the submandibular region.
On palpation swelling was movable, soft to firm in consistency, non tender, non pulsatile, and the
overlying skin was tensed but normal in color. After clinical examination, radiographic examination with the help of CT scan & USG was done. A fine needle aspiration biopsy obtained from parotid gland mass. FNAC report revealed benign neoplasm of the salivary gland ;monomorphic adenoma. The patient was treated under general anesthesia by complete excision of the lesion with proper aseptic condition. Grossly the specimen revealed irregular surfaced, well-encapsulated mass, pale brownish in color, soft in consistency, measured 12 x 9 cm² approximately & weighed 685 gms which is very rare presentation . (Fig.2 , Fig.3) [Fig.2] Gross specimen [Fig.3] Gross Specimen

[Fig.4] :The Cut surface showed brownish fluid coming through the fragile in consistency & was separated from the capsule.

After doing the grossing; the tissue was processed & paraffin blocks were made. 3-4 um sections were cut. Sections were stained with Haematoxylin & Eosin. H & E sections showed the typical features of basal cell adenoma of parotid gland .

**Microscopic features:**

H & E stained section shows well encapsulated lesion. Uniform appearing cells arranged in islands and cords. The lesion shows uniform and regular basal cells, morphologically two forms one is the small cell with scanty cytoplasm and round nucleus , while other is large cell with pale cytoplasm and ovoid nucleus. It consists of basaloid epithelial cells which are most commonly arranged in a Tubular trabecular pattern. In the tubular pattern, the basaloid cells form islands & cords, that are broad and rounded. In epithelial cells are narrower, cord-like and interconnected with one another creating a reticular lace like pattern. The cells were sharply demarcated from the connective tissue stroma by the basement membrane. No myxomatous or chondroid areas are seen. (Fig.5)

Thus, differentiating the lesion from Pleomorphic Adenoma of the Parotid Gland.
Histopathological report given was Basal cell adenoma: tubular-trabecular type.

[Fig.5] Encapsulated lesion, uniform appearing cells arranged in islands and cords.(10x)

[Fig.6] Epithelial cells resembling basal cells in tubular pattern, the basaloid cells form islands & cords, that are broad and rounded.

[Fig.7] In the trabecular pattern, the epithelial cells are narrower, cord-like interconnected with one another creating a reticular lace like pattern.
The lesion is made up of uniform and regular basal cells showing morphologically two forms one, the small cell with scanty cytoplasm and round nucleus, while other is large cell with pale cytoplasm and ovoid nucleus. Small cells periphery & Large cells centrally.

Discussion:
Salivary tumors are said to be relatively uncommon lesions. The incidence is 3 to 5% of the tumors. In major salivary glands the most common site is parotid (64-80%) whereas submandibular gland & sublingual glands show incidence of 8 to 11% & 1% respectively. Among all these tumors monomorphic adenoma is a group of benign salivary gland tumors composed of a proliferation of a single epithelial cell type. The two most common types are basal cell adenoma & canalicular adenoma. Monomorphic adenomas lack the wide cellular diversity encountered in Pleomorphic Adenoma. They are composed of single cell type, hence the term Monomorphic Adenoma.

Basal cell adenoma, as defined by WHO, is a distinctive benign neoplasm composed of basoloid cells organized with a prominent basal cell layer and distinct basement membrane-like structure and no myxochondroid stromal component as seen in pleomorphic adenomas. Basal cell adenoma was first described by Kleinsasser and Klein in 1967. In 1991, the World Health Organization (WHO) separately classified basal cell adenoma and its malignant counterpart, basal cell adenocarcinoma, as well as canalicular adenoma. Malignant transformation occurs in 4.3% of BCA that to in membranous variety.

Histogenesis:
The isocellular cells resemble the reserve cells of intercalated duct. Batsakis is credited with reporting the first case in the American literature in 1972, and suggested that the intercalated duct or reserve cell is the histogenetic source of basal cell adenoma.

Clinical features:
The tumor can occur at any age but is most common in middle-aged & older adults ,with a peak prevalence in the seventh decade of life. The tumors appears to be more common in women,with some studies showing as high as 2:1 female -to - male
ratio. Clinically, the basal cell adenoma appears as a slow growing, freely movable mass similar to a pleomorphic adenoma. Most tumors are less than 3 cm in diameter. Parotid tumors usually are located within the superficial lobe of parotid.

Membranous basal cell adenoma: This occur in association with skin appendage tumors such as dermal cylindromas and trichoepitheliomas. Multiple bilateral tumors may develop within the parotids. Because these tumors often bear a histopathologic resemblance to the skin tumors, they also have been called dermal analogue tumors.

**Pathologic features:**
Grossly the tumor is well circumscribed, pink–brown and smooth in texture, simulating an enlarged lymph node. Size may be large, but is generally < 3 cm in diameter. On cut section, they are typically uniform and solid, without necrosis. They can, however, occasionally be cystic. They have characteristic, numerous endothelial lined vascular channels, in which small capillaries and venules are prominent within the microcystic areas of the adenoma. These vascular structures can cause intratumoral hemorrhage. The membranous type is noteworthy in that it can be multifocal, with a multinodular growth pattern.

In many cases, cystic formations containing mucinous fluid are present in the center of a tumor. This can explain why basal cell adenomas are well enhanced and why hemorrhagic components are found within the cystic portion. In our case the size of the tumor was appx. 12x9 cm^2 which represented as a giant tumor & thus the rarest one.

**Histopathologic findings:**
The tumors are firm and well circumscribed with a distinct capsule. Cystic change is common. No matter what the pattern, the individual cells have sparse eosinophilic to clear cytoplasm and round nuclei. The nuclei are not atypical but are prominent because there is minimal cytoplasm. Basal cell adenomas can be divided on the basis of their morphologic appearance into four subtypes:

- **Solid**
- **Tubular**
- **Trabecular**
- **Membranous**

The most common subtype is the solid variant, which consists of multiple islands and cords of epithelial cells that are supported by a small amount of stroma. The peripheral cells of these islands are palisaded and cuboidal to columnar in shape, similar to the microscopic appearance of basal cell carcinoma.

The tubular subtype is characterized by the formation of small, round, duct-like structures.

The trabecular subtype demonstrates narrow cord-like epithelial strands. Frequently mixture of histopathologic subtypes is seen.

The membranous type exhibits multiple large lobular islands of tumor that are molded together in a jigsaw puzzle fashion. These islands are surrounded by a thick layer of hyaline material, which represents reduplicated basement membrane.

**Immunohistochemical features:**
More recently evaluations, including electron microscopic and immunohistochemical studies have shown that basal, ductal, and myoepithelial cell differentiation occurs to variable degrees in basal cell adenomas. 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. Morinaga et al. have reported focal reactivity for myosin. Dardick et 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. 

**Fine needle aspiration:** 
Aspirates consists of sheets or syncytial fragments of bland oval cells with scanty cytoplasm and round to oval nuclei. Nests or groups may be surrounded by a bright green (papanicolau) or pale magenta (May-Grunwald – Giemsa) hyaline band. 

**Differential diagnosis:** 
The Differential diagnosis may include malignancies such as basal cell adenocarcinoma or adenoid cystic carcinoma. However, cytologic atypia, infiltration, and perineural invasion are seen in carcinoma but not in adenoma. Benign entities such as canalicular adenoma, cellular pleomorphic adenoma or myoepithelioma may also be considered, but usually have different patterns of growth, and thus differentiating from the same.

**Treatment:** 
Basal cell adenoma are benign and conservative surgical excision including a margin of normal uninvolved tissue is adequate therapy in most patients. Surgical excision, to possibly include parotidectomy, is the treatment of choice.

**Conclusion:** 
Among all salivary gland neoplasms, pleomorphic adenoma is the commonest one and basal cell adenoma is the second most common occurring in the parotid gland, but a tumor of such a size is an exception. However to rule out malignant transformation excision followed by histopathological examination is essential along with follow up.

**References:**
1. Head and neck pathology. Lester D. R. Thompson foundations in Diagnostic pathology.pg 300,303