

# Basal Cell Adenoma Occurring at an Uncommon Site: Masquerading as a Mucous Retention Cyst

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**Abstract**— Basal cell adenoma is a rare benign salivary gland neoplasm with a monomorphic basaloid tumour cell pattern, most commonly occurring in the parotid. We present a rare case of basal cell adenoma arising in an uncommon site of minor salivary glands of the upper lip in a 74-year-old male. The patient presented with a mobile, non-tender mass on the upper lip, which was gradually increasing in size. The tumour was excised under local anaesthesia with a clinical diagnosis of mucous retention cyst. Histopathological examination revealed a circumscribed tumour composed of mixed tubular, membranous and trabecular patterns of basaloid cells with lumen of tubules containing eosinophilic material. Immunohistochemically, the tumour showed positivity to SMA and BCL2 while it was negative for CD117. Basal cell adenoma was diagnosed based on these results. The postoperative course was uneventful and there has been no recurrence.

**Keywords**— Basal cell adenoma; histopathology; monomorphic; palisading; salivary gland.

## I. INTRODUCTION

Initially described by Kleinsasser and Klein in 1967, Basal cell adenoma (BCA) is a rare tumor that constitutes about 1-3% of major salivary gland tumors.<sup>[1]</sup> A subtype of monomorphous adenomas, it constitutes around 54% of adenomas of this group in the WHO classification.<sup>[1,2]</sup> Upto 80% are found in major salivary glands, the most common site being parotid gland.<sup>[3]</sup> Other rare sites include nasal septum, palate, upper and lower lip and buccal mucosa.<sup>[1,2]</sup> It usually presents in elderly patients in the fifth and seventh decades. Here, we report an interesting case of basal cell adenoma presenting at an uncommon location on upper lip.

## II. CASE REPORT

A 74-year-old male presented with a history of painless swelling on the upper lip since one year. Clinical examination revealed a mobile, firm, non-tender, submucosal mass located on upper lip measuring approximately 01x01 cm with normal-looking overlying mucosa. Based on the above findings a clinical diagnosis of mucous retention cyst was made. The patient underwent wide local excision of the mass and the excised specimen was received in our department for histopathological examination. Grossly, the specimen measured 1x1x0.4 cm with a cut surface showing a well-circumscribed solid greyish-white tumour throughout the tissue. There were no cystic/ gelatinous/ mucinous areas, haemorrhage or necrosis in the tumour. Microscopic examination of hematoxylin and eosin (H&E) stained sections from the submitted tissue showed a circumscribed tumour arranged in trabecular, tubular and membranous patterns. Numerous tubules consisting of central lumen and lined by cuboidal and basaloid cells with peripheral palisading were noted with lumen of few tubules containing eosinophilic secretions (Fig.1a). Prominent hyaline material was noted within the tumour with occasional foci of squamous metaplasia (Fig.1b). Mitosis, atypia and perineural invasion

were absent and there were no areas of necrosis. To further substantiate our diagnosis, the sections were subjected to immunohistochemical analysis, which revealed strong positivity to smooth muscle actin (SMA) and BCL2 whereas CD117 was negative (Fig.2a,b). In view of the histopathological and immunohistochemical findings, the patient was diagnosed as a case of BCA. The postoperative period was uneventful and the patient shows no recurrence of swelling after one year.

## III. DISCUSSION

BCA is a rare tumour that constitutes about 1-3% of major salivary gland tumours with a peak in fifth to seventh decade of life.<sup>[1]</sup> Mostly BCA arises in the parotid gland with other sites that can be involved being submandibular glands and rarely minor salivary glands of buccal mucosa, upper and lower lip, nasal septum and palate.<sup>[2,3]</sup> On radiology, BCA shows a round, well-defined solid mass with or without cystic component. Clinically, the most common presentation seen is a slow-growing, asymptomatic, movable, well-circumscribed firm mass, usually solitary and measuring less than 3 cm.<sup>[4,5]</sup> Histopathologically, BCAs are benign tumours composed of relatively monomorphic basaloid cells, a conspicuous basal cell layer with peripheral palisading and distinctive basement membrane-like material. Most are well circumscribed and encapsulated, although a multinodular microscopic pattern may be found in any of the subtypes. Morphologically, it can be divided into four subtypes: solid, trabecular, tubular, and membranous; with the solid variant being the most common.<sup>[6]</sup> The solid subtype has large islands with hyperchromatic palisading peripheral cells with central cells that are squamous and form keratin pearls.<sup>[4,7]</sup> The solid subtype is composed of epithelial cells supported by a small amount of fibrous stroma. In trabecular subtype, the basal cells are in an elongated, ribbon-like pattern.<sup>[4,7]</sup> Tubular subtype has multiple small duct-like structures lined by columnar-appearing cells with uniform, hyperchromatic and round to oval nuclei. In contrast to other subtypes, the membranous pattern has a jigsaw

arrangement of epithelial islands and is multilobulated. The islands are surrounded and separated by eosinophilic, hyalinized material representing reduplicated basal lamina.<sup>[4]</sup> Malignant transformation is highest in association with the

membranous subtype.<sup>[4,7]</sup> On immunohistochemistry (IHC), tumour cells are positive for SMA, S100 and BCL2.

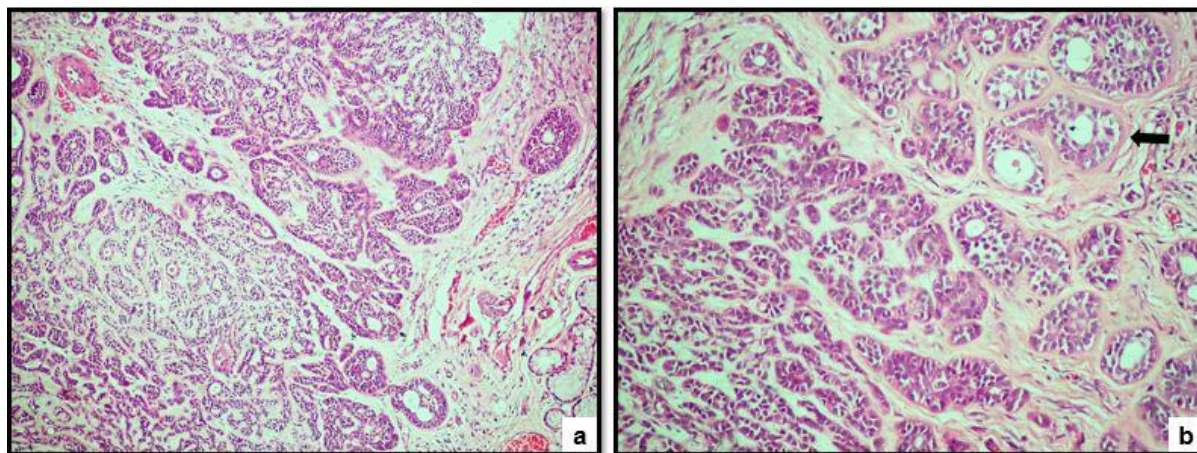


Figure 1(a-b): Hematoxylin and eosin stained microphotographs: (a) Tumour cells arranged in tubular and trabecular pattern with few tubules showing luminal eosinophilic secretions(100X); (b) Peripheral palisading of basaloid cells with hyaline basement membrane-like material (arrow) (200X).

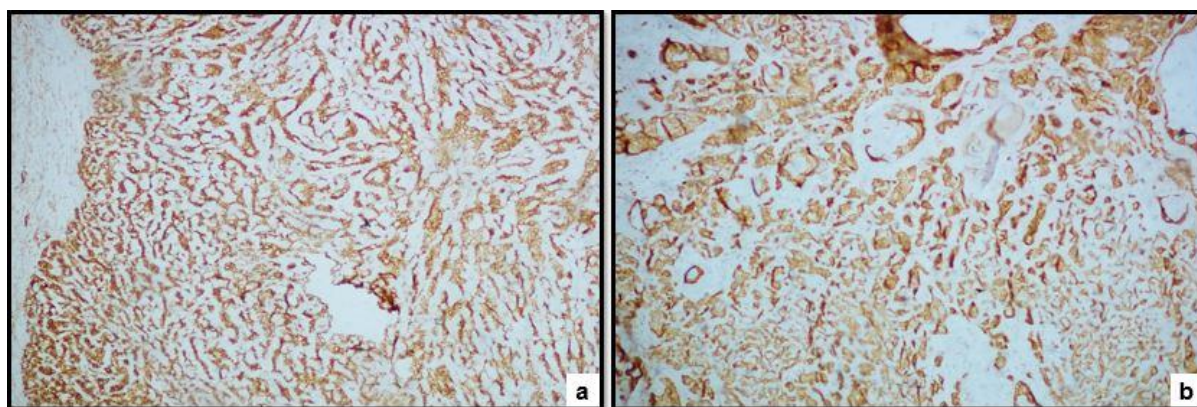


Figure 2(a-b): Immunohistochemistry: Strong and diffuse positivity for BCL2 (a; 100X) and SMA (b; 100X).

The various differential diagnoses to exclude are: mucous retention cyst, canalicular adenoma (CA), pleomorphic adenoma (PA), lymphangioma, hemangioma, adenoid cystic carcinoma (ACC), and basal cell adenocarcinoma (BCAC), basaloid squamous cell carcinoma (BSCC) and carcinoma ex pleomorphic adenoma (CEPA).

Mucous retention cysts are more common in older age on the upper lip and can be differentiated clinically from BCAs and other salivary gland tumours as these cysts usually present as an asymptomatic swelling that is soft in consistency (as opposed to firm swellings in case of salivary gland tumours), mobile and fluctuant due to presence of mucinous contents.<sup>[8]</sup> The overlying mucosa may appear normal or bluish in colour due to vascular stasis and accompanying cyanosis causing stretching of the mucosal lining. On imaging, mucous retention cysts appear as cystic masses that sometimes may contain septae within the mucinous area.<sup>[8]</sup> Our patient presented with a slow growing non-tender firm swelling over the upper lip which was mobile with normal looking overlying mucosa.

CA is also more common on the upper lip and histologically, shows bilayered strands or ribbons of basaloid cells enclosing ductal spaces/tubules. There is absence of myoepithelial cells and on IHC, CAs shows strong and diffuse positivity to pan-cytokeratin and S100.<sup>[9]</sup> Presence of hyaline material and squamous metaplasia along with the predominant trabecular and membranous pattern of tumour cells ruled out canalicular adenoma in our case.

Most common site for the occurrence of PA is parotid gland which shows epithelial and myoepithelial cells along with areas of myxoid, cartilaginous, osseous or hyaline differentiation in the stroma. Our case showed absence of the characteristic chondromyxoid stroma.<sup>[6,10]</sup>

BCA and BCAC are difficult to differentiate only on cytomorphological features and histology along with imaging plays an important role as invasive growth patterns with areas of necrosis and evidence of metastasis favours BCAC.<sup>[11]</sup>

ACC can be differentiated from BCA as BCA predominantly has a nested pattern with an inner layer of cells having a paler cytoplasm and peripheral palisading of the

outer layer of cells which are smaller and darker (basaloid). On the other hand, nuclei of ACC cells are hyperchromatic and angulated with absence of palisading.<sup>[11]</sup> Also, ACC is positive for CD117 on IHC.

BCA is primarily treated with surgical excision: superficial or total parotidectomy is done where the parotid gland is involved. For BCA of minor salivary glands, extracapsular excision is performed. In the membranous subtype, total parotidectomy is advised as this subtype is associated with multicentricity, chances of recurrence and occasional risk of malignant transformation. Long-term follow-up is recommended so that rare cases of recurrences are diagnosed in time.<sup>[2]</sup>

#### IV. CONCLUSION

There have been very few cases of BCAs of minor salivary glands reported in literature. Majority of monomorphic adenomas exhibit non-aggressive behaviour. Mixed subtype is not a frequent finding. Surgical excision of the lesion along with surrounding normal tissue is the definitive mode of treatment and it generally has a good prognosis. Histopathological and immunohistochemical analysis in correlation with clinical and radiological inputs proves a great help in correct diagnosis and management. Ours is an interesting case of BCA with occurrence in an unusual location in a 74-year-old male diagnosed only after surgical excision followed by histopathological and IHC evaluation.

#### ABBREVIATIONS

BCA: Basal Cell Adenoma  
H&E: Hematoxylin and Eosin  
SMA: Smooth Muscle Actin  
IHC: Immunohistochemistry  
CA: Canalicular Adenoma  
PA: Pleomorphic Adenoma

ACC: Adenoid Cystic Carcinoma  
BCAC: Basal Cell Adenocarcinoma  
BSCC: Basaloid Squamous Cell Carcinoma  
CEPA: Carcinoma Ex Pleomorphic Adenoma

#### REFERENCES

1. Luna MA, Eugenia Tortoledo M, Allen M. Salivary dermal analogue tumors arising in lymph nodes. *Cancer* 1987;59:1165-9.
2. González-García R, Nam-Cha SH, Muñoz-Guerra MF, Gamallo-Amat C. Basal cell adenoma of the parotid gland. Case report and review of the literature. *Med Oral Patol Oral Cir Bucal* 2006;11:206-9.
3. Douglas R Gnepp, Alena Skalova, Silvana Di Palma, Roderick HW Simpson, Toshitaka Nagao, Elizabeth Ann Bilodeau. Salivary glands. In: Douglas R Gnepp, editor. *Diagnostic surgical pathology of head and neck*, 3<sup>rd</sup> ed. Canada: Elsevier; 2015. p. 432-605.
4. Karim AB, Sumarriva L, Sharabi A *et al*. Basal cell adenoma of the upper lip: a case report. *J Dent Health Disord Ther* 2015;2:140-2.
5. Paker I, Yilmazer D, Arikok AT, Saylam G, Hucumenoglu S. Basal cell adenoma with extensive squamous metaplasia and cellular atypia: a case report with cytohistopathological correlation and review of the literature. *Diagn Cytopathol* 2012;40:48-55.
6. Seifert G, Brocheriou C, Cardesa A, Eveson JW. WHO International Histological Classification of Tumours. Tentative Histological Classification of Salivary Gland Tumours. *Pathol Res Pract* 1990;186:555-81.
7. Nagao K, Matsuzaki O, Saiga H, Sugano I, Shigematsu H, Kaneko T *et al*. Histopathologic studies of basal cell adenoma of the parotid gland. *Cancer* 1982;50:736-45.
8. Tandon A, Sircar K, Chowdhry A, Bablani D. Salivary duct cyst on lower lip: A rare entity and literature review. *J Oral Maxillofac Pathol* 2014;18:151-6.
9. Khodaei M, Amani M, Mirinezhad S, Rafieyan S. Canalicular adenoma of the hard palate: A rare case report. *Dent Res J* 2021;18:15.
10. Jain S, Hasan S, Vyas N, Shah N, Dalal S. Pleomorphic Adenoma of the Parotid Gland: Report of a Case With Review of Literature. *Ethiop J Health Sci* 2015;25:189-94.
11. Wilson TC, Robinson RA. Basal cell adenocarcinoma and Basal cell adenoma of the salivary glands: a clinicopathological review of seventy tumors with comparison of morphologic features and growth control indices. *Head Neck Pathol* 2015;9:205-13.