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RESEARCH ARTICLE

BASAL CELL ADENOMA OF PALATE- A RARE OCCURENCE

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ABSTRACT

Basal cell adenoma (BCA) of the salivary glands is an uncommon type of monomorphic adenoma. It derives its name from the basaloid appearance of tumor cells and accounting for 1-2 % of all salivary gland epithelial tumors. It's most frequent location is the parotid gland, followed by the upper lip; while it is very rare in the minor salivary glands. It usually appears as a firm and mobile slow-growing mass. Histologically, it is seen as nests of isomorphic cells and interlaced trabeculae with a prominent basalmembrane. There is also slack, hyaline stroma with absence of a myxoid or chondroid component. In this article, a case of basal cell adenoma of palate with 6 month follow up has been discussed. This report emphasizes the rare site of occurrence of this tumor and briefly reviews the literature.

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INTRODUCTION

World Health Organization defined basal cell adenoma (BCA) as an idiosyncratic benign neoplasmcomposed of monomorphic population of basaloid epithelial cells, organized with a prominent basal celllayer and a distinct basement membrane like material; however, the myxochondroid stromal component characteristic of mixed tumor is absent (Gnepp *et al.,* 2000; Kratochvil, 1991). The origin of tumors of minor salivary glands account for less than 25% of all salivary neoplasms with 40– 80% occurring in the palate. [Minicucci, 2008]. In 1967, Kleinsasser and Klein introduced the term "basal celladenoma" to describe a benign salivary gland tumor comprised of uniform appearing basaloid cellsarranged in solid, trabecular, tubular, and membranous patterns, but lacking the myxoid and chondroidmesenchymal-like component as seen in pleomorphic adenoma [Takeshita *et al.,* 2004]. This tumor has been recognized as one of thenine subcategories of salivary gland adenomas in the Second Edition of the Salivary Gland TumorsClassification of the World Health Organization (WHO), and accounts for approximately 1 – 2% of allsalivary gland epithelial tumors.[4] The most common site of occurrence is theparotid gland[5-6]followed by upper lip with a decreasing incidence in palate, buccal mucosa and lowerlip [Esteves, 1997]. It usually occurs in patients over 50 years of age with slight femalepredilection [Esteves, 1997].

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Concerning itsclinical presentation, it exhibits as a slow growing, asymptomatic, movable, round or oval, normalcoloredsub mucosal mass measuring less than 3 cm in diameter (Minicucci, 2008). Microscopically, BCAs are well circumscribed and encapsulated by fibrous connective tissue (Gnepp, 2000). Tumormass consists of proliferation of terminal duct epithelial cells forming is lands or sheets supportedby a sparse fibrous stroma, and presence of a small number of myoepithelial cells (Kratochvil, 1991; Esteves, 1997). It has distinctbasement membrane and often exhibit palisading of basal layer cells. They also lack the myxochondroidareas characteristic of pleomorphic adenoma.[8] Basal cell adenoma is an uncommon tumor with palate being the rarest site for its occurrence [Esteves, 1997].

Case report

A 32 year old female was referred to our department with the chief complaint of painless swelling with respect to left side of palate, present since three months. Swelling was insidious in onset which started as a small intra-oral mass initially and gradually progressed to present size. Clinical examination revealed a well built female patient with apparently symmetrical face and no extra oral swelling. There was no local, regional or distant lymphadenopathy, or evidence of a primary tumor elsewhere. Intra oral examination revealed a diffused swelling around 1cm x 1cm in the left side of the palate extending anterioposteriorly from distal 26 to distal of 27 and mediolaterally from the free gingival margins to 1 cm short of midline.

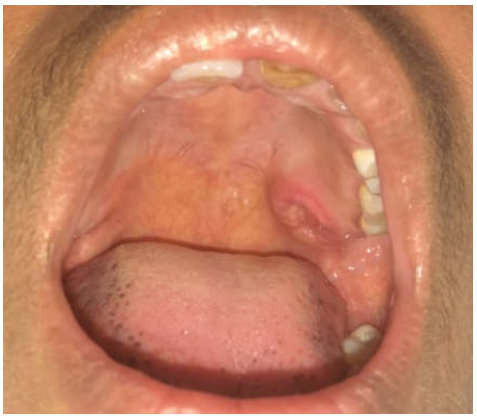

Fig 1. Pre op

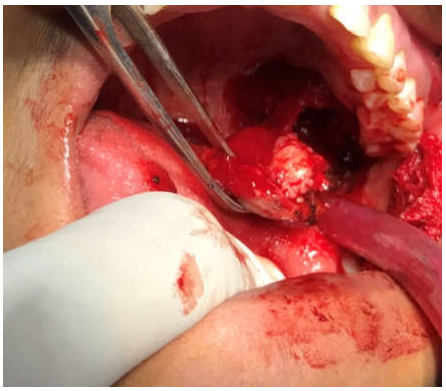

Fig. 2. Tumor Excision

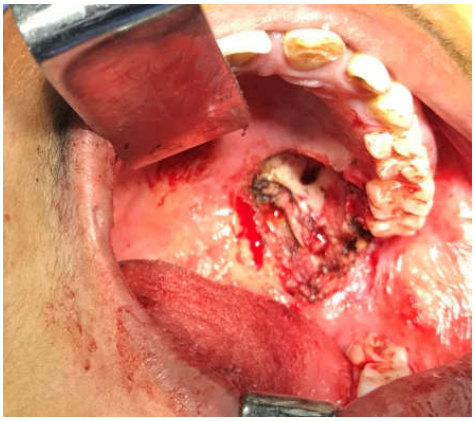

Fig 3. Surgical defect

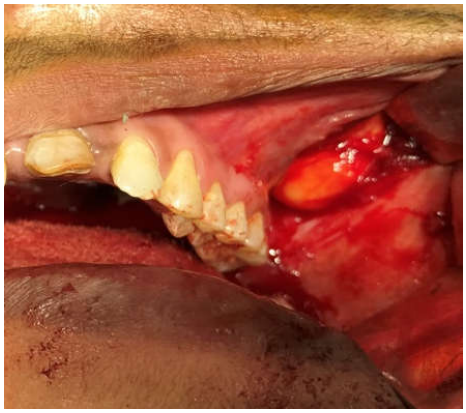

Fig 4. Harvesting of buccal fat pad

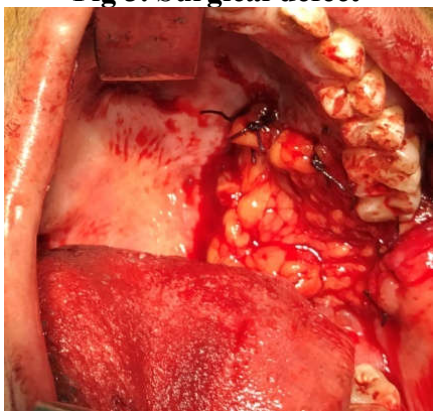

Fig 5. Buccal fat pad sutured to defect

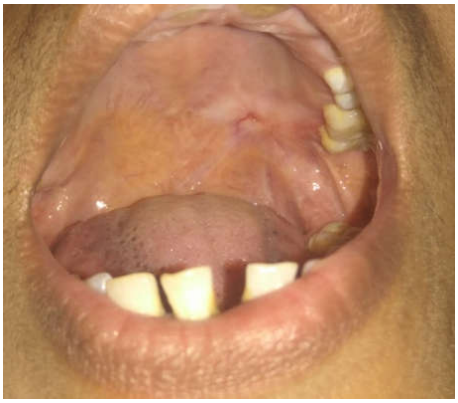

Fig 6. Post op 6 months

On aspiration negative pressure was observed suggestive of solid lesion. An incisional biopsy was performed and the section showed tumor arranged in islands, cords and tubules admixed with eosinophilic basement membrane like material. The slide possesses glands with epithelial and myoepithelial cells better appreciated in tubules. There was no evidence of matrix production or atypia. The report was suggestive of basal cell adenoma.

The patient was prepared for tumor excision under general anesthesia. Under sterile aseptic conditions, the tumor was excised with 5 mm margin and overlying palatal mucosa. A vestibular incision was given on left maxillary vestibule and a pedicled buccal fat pad was harvested. Sufficient amount of fat pad was exposed and transposed over the defect created after tumor excision. The patient made an uneventful recovery and is under regular follow up. Excisional biopsy confirmed basal cell adenoma histopathologically.

DISCUSSION

The basal cell adenoma was once considered to be a type of monomorphic adenoma.[5]Kleinsasser andKlein were the first to designate the term basal cell adenoma and to establish it as a distinct clinical andpathologic entity in 1967 (Gnepp, 2000; Kratochvil, 1991; Jang, 2004). Gardner and Daley described the distinguishing features of basal celladenoma and canalicular adenoma to document these as two separate entities [Gardner, 1983]. In the revised WHO classification of salivary gland tumors (1991), basal cell adenomas were included in the benign epithelialneoplasm, excluding the word monomorphic (Gnepp *et al.,* 2000; Esteves, 1997) and define it as a tumor of isomorphic basaloid cellsorganized with a noticeable basal cell layer and separate basement membrane like structure and myxochondroid stromal component of mixed tumor was not present (Mărgăritescu *et al.,* 2005). Gardner and Daley described the histologic subtypes of Basal cell adenoma (BCA)

which include solid, trabecular, tubular, and membranous (Kratochvil, 1991), with the solidvariant being the most common (Kudoh, 2014). Batsakis reported the first case of BCA in the American literature in1972, and suggested that the intercalated duct or basal cell is the histogenetic source of BCA (Gardner, 1983). Salivary gland tumors are uncommon and constitute 2-6.5% of all the head and neck neoplasms[5,13] and tumors of minor salivary gland account for less than 25% of all salivary neoplasm (Esteves, 1997). Palate is the most common site for minor salivary gland tumors with 40-80% of incidence (Siddaraju, 2013). But it isrelatively an uncommon site for BCA [Fantasia, 1980] which usually arises in the major salivary glands with theparotid being the most frequent site of occurrence followed by minor salivary glands of upper lip. [Kudoh, 2014; Ishibashi *et al.,* 2012]. In the opinion of Fantasia and Neville, BCA typically occurs in older patients with mean age of61 years andmost commonly involves the upper lip [Fantasia, 1980]. 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 (Kratochvil, 1991; Nakabayashi *et al.,* 2010). In our case the tumor presentation was in a female patient in her third decade of life. It was present in minor salivary glands of the palate which isconsidered as a rare site for BCA (Gupta, 2009; Siddaraju *et al.,* 2013; Ishibashi *et al.,* 2012; Pesic, 2009).

Biopsy is recognized as the most precise method for diagnosis of BCA. BCAs are mostly well circumscribed and encapsulated by fibrous connective tissue. Histologically, it has four variants named as solid, tubular, trabecular, and membranous; solid being the most common, but each tumor has combination type (Gnepp, 2000; Kratochvil, 1991). These variable patterns of BCA consists of 2 types of cell populations (basaloid cell and luminal duct cells) (Mărgăritescu, 2005). The first cell type is small cuboidal or columnar shaped, present peripherally in a palisading arrangement within the tumor nests or cords, with round deeply stained nuclei and little discernible cytoplasm. The second cell type, presenting centrally, is larger with modest cytoplasm, indistinct cell borders and a pale staining oval nucleus. Sharp demarcation is present between the neoplastic cells and the surrounding stroma (Gnepp, 2000; Kratochvil, 1991; Kim, 2012). In spite of distinctive appearance of BCA, other primary tumors which can simulate its basal cell featuresand causes difficulty in diagnosis are pleomorphic adenoma (PA), adenoid cystic carcinoma (ACC), basalcell adenocarcinoma (BCAC). Lack of myxo-chondroid stroma and parenchyma well demarcated by adistinctive basal membrane differentiate BCA from pleomorphic adenoma (Gnepp *et al.,* 2000; Kratochvil, 1991). BCA is differentiated from BCAC by two characteristics. One is the circumscription of the basal cell adenoma which differs frominvasive pattern of adenoid cystic carcinoma. The other is the absence of vascularity in themicrocysticareas of adenoid cystic carcinoma, which differs from the numerous endothelial lined channelsin basal cell adenoma (Jang *et al.,* 2004). Other distinctive features of BCA are lack of perineural invasion andincreasedbland tumor cell population. The recurrence rate is 25-37% for the membranous variant of BCA (Kudoh, 2014), possibly related to its multifocalnature, which impairs complete removal. Although exceedingly rare, malignant transformation of BCAhas been reported (Gnepp, 2000). Therefore, it is necessary to perform complete tumor excision. The approach used in our case and the postoperative period was uneventful with no signs of recurrence even after a follow up of 6 months.

Conclusion

Definitive diagnosis by clinical and imaging techniques alone is difficult. The final diagnosis requires histopathological analysis, which can be inverterated by immunohistochemical examination. In any suspected neoplastic salivary gland lesion, due to prognostic implications, differential diagnosis with malignant counterparts is mandatory. This paper is an addition of one more case of this rare tumor arising from minor salivary glands of posterolateral aspect of hard palate to the literature.

It is important to differentiate BCA from pleomorphic adenoma, adenoid cystic carcinoma, canalicular adenoma and basal cell adenocarcinoma, to which it may bear resemblance. Treatment should aim at complete excision of tumor and allowing secondary healing for small lesions of palate or coverage with local flaps if the lesion is large or requires considerable excision or fenestration of soft palate. There is no role of post operative chemotherapy or radiation therapy if adequate excision is performed.

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