Endoscopic resection of a giant basal cell adenoma of the nasal septum: A case report

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ABSTRACT

Introduction: Basal cell adenoma is a rare, benign epithelial neoplasm and represents 2.4% of all benign salivary gland tumors. Basal cell adenoma of the nasal septum is extremely rare and only one case had been reported in literature. We describe the clinical history, imaging findings, surgical resection technique and pathology for the case of this extremely rare sinonasal pathology. Case Report: We report a case of a giant sepal basal cell adenoma resected using the endoscopic endonasal technique. The basal cell adenoma was resected using the endoscopic endonasal technique without complication. The patient is disease free two years following surgery. Conclusion: Basal cell adenoma of the sinonasal cavity is a rare, not previously reported, benign salivary gland tumor. Resection using an endoscopic endonasal technique can provide a safe and complete resection, with adequate margins to prevent recurrence of the disease.

Keywords: Basal cell adenoma, Nasal septum, Nasal cavity, Benign salivary gland tumor, Minor salivary gland tumor, Sinonasaltumor

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INTRODUCTION

Basal cell adenoma (BCA) is a rare, benign epithelial neoplasm and represent 2.4% of all benign salivary gland tumors [1]. They are generally slow growing and painless [2]. Histologically BCA consists of isomorphic cells with a prominent basal cell layer and are characterized by a distinct basement membrane-like structure but are lacking the chondro-myxoid matrix which is characteristic of plasmacytoma adenoma [3]. Here we report a case of a giant sepal basal cell adenoma treated by endoscopic resection along with a review of literature.

CASE REPORT

A 64-year-old male not known to have any medical problems presented with a 12 month history of bilateral nasal obstruction unresponsive to medical treatment. He was referred by an otolaryngologist for evaluation of a nasal mass found on endoscopy. There were no associated symptoms of pain, epistaxis, facial deformity or visual changes. Endoscopy showed a submucosal mass of broad base arising from posterior nasal septum completely obstructing the nasal cavity bilaterally. The mass compressed and laterally displaced the posterior half of each of the inferior turbinates (figure 1).

A computed tomography (CT) scan showed a large soft tissue mass involving the posterior nasal septum filling each of the nasal cavities. The mass demonstrated partial rim calcification and measured 3.5 cm in
diameter. The MRI demonstrated a mass arising in the posterior nasal septum causing mild lateral displacement of the medial walls of the maxillary sinuses (figure 2). The mass was of homogeneous low T1 signal intensity, isointense to skeletal muscle, and was of slightly higher T2 signal intensity. It demonstrated intense homogeneous enhancement.

A biopsy was done under local anesthesia and revealed a cellular tumor with fairly closely spaced tubules with some intervening basophilic stroma. There was no significant pleomorphism present and no increased mitotic activity. The features were suggestive of a cellular adenoma.

This patient underwent endoscopic resection of the septal tumor. After adequate tumor had been removed for pathology, the entire mass was decompressed to its mucosal flaps by using the double suction technique as is commonly used for soft pituitary tumors. The walls could then be collapsed to the midline and endoscopic scissors were used to excise all the tumor walls with a small margin. The site of contact with the floor of the sphenoid and nose was then drilled with a diamond bur (figure 3). No packing was used at the end of the procedure but septal splints were placed over either side of the cartilaginous septum to prevent synechiae formation. The postoperative course was uneventful and the patient at two years follow-up has not shown any evidence of recurrence.

The histopathology sections showed portions of nasal mucosa with a cellular monomorphic 'basaloid' tumor with palisading at the periphery of epithelial nests. The tumor also showed areas of acinar differentiation. There is no evidence of marked nuclear atypia or increased mitoses. There was no evidence of tumor necrosis. Immunohistochemical stains showed that the tumor cells were positive for AE1,3 and vimentin. S100 was only focally positive. P63 staining shows diffuse positivity in the basal/myoepithelial cells. These findings were consistent with solid trabecular basal cell adenoma (figure 4).

**DISCUSSION**

There are an estimated 600-1000 minor salivary glands in head and neck region. Seventy to ninety percent of them are distributed in the oral cavity and oropharynx. The remainder are localised to the nose, pharynx and larynx. Tumors of the minor salivary glands represent 10-15% of all salivary gland neoplasms.
Pleomorphic adenoma is the most common benign tumor of minor salivary glands. BCA was first defined as a separate entity in 1967 by Kleinsasser and Klein. It usually occurs in the major salivary glands of elderly patients (75% in the parotid and 5% in the submandibular gland) [5]. In minor salivary glands, BCA occurs in or adjacent to the upper lip (80%), palate (8%), buccal mucosa (4%) and in the lower lip (3%) with a mean age of 61 years and without a sex predilection [6]. Basal cell adenoma of the nasal septum is extremely rare and only one case had been reported in literature [7].

BCA have been classified into four subtypes: solid, trabecular, tubular and membranous [3]. Solid type is the most common, followed by the trabecular, the membranous and rarely the tubular type [5]. The solid type is composed of solid nests or islands of basoloid cells usually exhibiting peripheral palisading [2]. Some centers of the islands may show expansions by cells that have a whirled, basal squamous appearance with possible keratin pearl formation [8]. In trabecular type, the basoloid cells grow in a solid, ribbon-like pattern with variable thickness which may approach a thickness reminiscent of the solid pattern. The tubular type consist of multiple duct-like structures lined by basoloid cells, it is usually found with trabecular patterns so called tubulo trabecular variant [8].

Membranous or dermal analogue type arises mainly in parotid gland [5] and is commonly associated with autosomal dominant syndrome called Brooke-Spiegler syndrome (cylindromas, trichoepitheliomas, and spiradenomas). It consists of basoloid nest and island of cells that arranged in a jigsaw pattern and surrounded by thick eosinophilic hyaline membrane [5]. This type is usually multifocal and often not encapsulated with up to 28% incidence of malignant transformation and high rate of recurrence 25% [5, 8]. The other types have a low rate of recurrence [8]. BCA arising from parotid are encapsulated while those arising from minor salivary glands are not encapsulated, particularly those in the palate [9].

The diagnosis and classification of the BCA and its types can be difficult despite advances in immunohistochemistry and the availability of a variety of differentiation markers because of lack of specificity of many markers for specific histologic tumor type [5].

The differential diagnosis of BCA includes pleomorphic adenoma, adenoid cystic carcinoma and cutaneous basal cell carcinoma [8]. Unlike pleomorphic adenoma, BCA has a distinct basement membrane-like structure and lacks the chondroid-myxoid matrix that is characteristic of pleomorphic adenoma [3]. Although it is not easy, adenoid cystic carcinoma can be distinguished by true invasion of surrounding tissue, perineural invasion and lacking vascularity in the microcystic areas [6].

CONCLUSION

Basal cell adenoma of the nasal septum is a rare benign lesion with low a recurrence rate. We recommend endoscopic excision of such a tumor given its favorable location and its amenability for clear resection margins. Even with a relatively large tumor, as occurred in this patient, endoscopic techniques afford excellent visualization and exposure, which are keys for successful surgical resection whether performing open or endoscopic surgery.

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Author Contributions

Dakheelah M Al Mutairi – Substantial contributions to conception and design, acquisition of data, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Don Wang – Substantial contributions to conception and design, analysis and interpretation of data, Drafting the article, Final approval of the version to be published

Shaun J Kilty – Substantial contributions to conception and design, Drafting the article, revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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