Original Research Paper

Volume - 13 | Issue - 08 | August - 2023 | PRINT ISSN No. 2249 - 555X | DOI : 10.36106/ijar

ENT

A UNIQUE CASE OF BASAL CELL ADENOCARCINOMA EX PLEOMORPHIC ADENOMA

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ABSTRACT Basal Cell Adenocarcinoma (BCA) is a low-grade malignancy usually found in parotid gland. It is theorized to originate either from pleomorphic/monomorphic adenomas undergoing a malignant transformation or to arise de novo. Basal Cell Adenocarcinoma (BCA) is a carcinoma arising from a primary or recurrent benign pleomorphic adenoma. It often poses a diagnostic challenge to clinicians and pathologists. This study intends to review the literature and highlight the current clinical and surgical perspectives about this entity. The proportion of adenoma and carcinoma components determines the macroscopic features of this neoplasm. The entity is difficult to diagnose pre-operatively. Pathologic assessment is the gold standard for making the diagnosis. Treatment for BCA often involves surgical procedure which may be followed by radiotherapy. Diagnosis and aggressive surgical management of patients presenting with BCA can increase their survival rates. Molecular studies have showed that development of BCA follows a multi-step model of carcinogenesis, with the progressive loss of heterozygosity at chromosomal arms 8q, then 12q and finally 17p. There are specific genes in regions that are associated with particular stages in the progression of BCA. Many genes which regulate tumor suppression, cell cycle control, growth factors and cell–cell adhesion play a role in the development and progression of BCA.

KEYWORDS : BCA, Parotid Gland, Salivary Gland, Pleomorphic Adenoma

INTRODUCTION

A slow-growing malignant neoplasm of the salivary glands, BCA usually affects the major salivary glands, specifically the parotid gland. When arising in minor salivary gland, BCA occurs most frequently in the oral cavity and upper respiratory tract. BCAC does not show sex predilection and predominates in patients in their seventies. The WHO classification of salivary gland tumors first included BCA in 1991 as the malignant counterpart of the monomorphic adenoma. For more than 40 years, monomorphic adenomas had been recognized formally as a class of salivary gland tumors, apart from pleomorphic adenomas. The prevailing theory is that BCA tumors have two different origins: monomorphic adenomas undergoing a malignant transformation or the tumor arisen de novo. BCA is considered a low-grade malignant tumor. This is supported by diploid DNA content. In 1993, one-third of the reported cases had recurred, and approximately 10% metastasized on follow-up. The most common metastatic sites were lymph nodes and lungs. Metastatic tumors all had solid growth patterns. To date, there have been several case reports describing the characteristics and behavior of BCAC, with no definitive consensus regarding treatment. Here we present a case report of a BCA that arose within a pleomorphic adenoma (carcinoma ex pleomorphic adenoma).

Case Study

A 65-Year-old female patient came to ENT Department of Dr. SCGMC Nanded with a 4-year history of enlarging mass over right parotid region. Patient have history of Tobacco chewing for 25-year 5-10 gm per day. Patient also have history of femur nail fixation few months back.

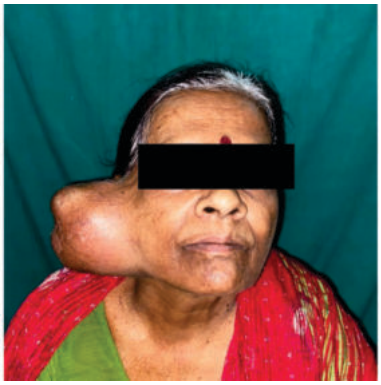

Figure-1

She complained of pain over the swelling and that swelling was causing cosmetics problem. Physical examination revealed an enlarging mass over the right parotid gland of approximately 10cm x8cm x6cm which was a superficial, hard, painful and fixed. The skin overlying the surface of the mass was tense with ulceration just Infront of right external auditory canal. There was no cervical lymphadenopathy and the facial nerve was intact.

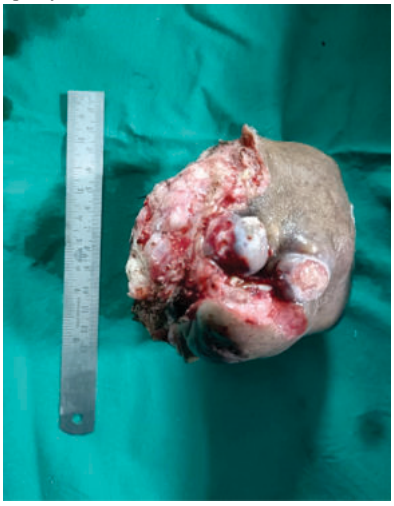

Figure-2

An FNAC was performed which came back as PLEOMORPHIC ADENOMA. After a discussion of the risks including damage to the facial nerve, the patient opted to have the lesion removed. A right superficial parotidectomy was performed. An elliptical incision was made, around the swelling blunt dissection was done superficial temporal artery was identified and clamped cut and ligated, dissection continued till swelling separated from surrounding structure. Working between the sternocleidomastoid muscle and then the posterior belly of the digastric and the tragal pointer, the facial nerve was identified. It was freed from the overlying mass and required dissection out to its most peripheral aspect. The mass was totally excised and was sent for histopathological analysis. After placing a drain, the wound was closed. Her postoperative course was unremarkable, and there was no facial nerve deficit. Pathologic examination revealed a globular tissue

mass containing a 12cm x 9cm x 9cm irregular and cystic at places with skin flap. The mass was multilobulated, and firm. On c/s mucinous fluid oozes out, multiple cystic spaces noted hemorrhagic at places, partially autolyzed. On external surface 3 nodules noted one of size 2x2x2cm second of size 1.5x1x1cm and third of size 1x1x1cm. On histopathologic examination, sections medial margins section shows capsular and vascular invasion, on superior margin section only skin with dermis, no tumor cells seen, on inferior margins section show skin lining with underneath plenty of inflammatory cells and in deep dermis salivary duct seen. On three nodule section presence of capsule with chondromyxoid tissue with small myoepithelial cells, also seen the tumor cells round to oval with slight pleomorphism and scanty cytoplasm arranged in nodule like form with peripheral palisading seen. Also, mitotic activity seen to be present.

DISCUSSION

BCA typically arises in adults older than 60, with no gender predominance. The 2005 WHO classification of tumors categorizes BCA as a low-grade tumor with a favorable prognosis. There are four major histologic growth patterns of BCA: **solid, tubulotrabecular, cribriform, and membranous,** with solid being the most common and also the most likely to present with perineural invasion. The differential diagnosis of BCA includes basal cell adenoma and adenoid cystic carcinoma. BCA appears very similar to basal cell adenoma histologically, but is distinguished from this benign entity by displaying invasion of local structures, perineural invasion, and/or angiolymphatic invasion. Adenoid cystic carcinoma has a poorer prognosis and displays dark hyperchromatic angulated nuclei, which differ from the vesicular nuclei of BCA. BCA also often have prominent peripheral palisading of the outer layer that adenoid cystic carcinomas lack. Treatment consists of surgical excision with a wide margin of tissue to reduce the chance of recurrence. These tumors are locally destructive, and recurrence has been reported commonly. Radiotherapy is suggested for lesions originating in the minor salivary glands due to a higher likelihood of vascular and neural invasion.

CONCLUSIONS

The origin of BCAC is not well defined. Although most appear to arise de novo or from monomorphic adenoma, here we report evidence of a BCAC ex pleomorphic adenoma. Differentiating BCAC from basal cell adenoma and adenoid cystic carcinoma is important due to the difference in prognosis that each of these diseases possess. Surgical excision is the preferred method of treatment for most of these lesions. Management of BCA involves either simple excision with superficial parotidectomy or simple excision with total parotidectomy with facial nerve preservation. With post-surgery regular follow-ups to rule out case of recurrence despite surgical excision, or further need of radiotherapy.

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15 Case Report Basal Cell Adenocarcinoma Ex Pleomorphic Adenoma John Heaphy Journal of Case Reports in Medicine Vol. 2 (2013), Article ID 235732