Rare carcinoma ex pleomorphic adenoma of the buccal minor salivary gland causing a therapeutic dilemma

YOGESH KINI, CHIRAG DESAI, UMA MAHINDRA, JITENDRA KALBURGE

Abstract

Carcinoma ex pleomorphic adenoma (CXPA), as a group, constitutes 12% of malignant salivary gland tumors. We present a case of CXPA of the buccal mucosa in a 17-year-old patient. The buccal mass was of a size of 3.0 cm located in the right cheek. Pleomorphic adenoma was the provisional diagnosis. The tumor was excised under local anesthesia. Histopathological evaluation revealed a pre-existing pleomorphic adenoma. However, on magnification, certain areas showed islands of dysplastic epithelial cells invading the fibrous capsule and CXPA was diagnosed. The patient was recalled and secondary surgery of the site performed. No tumor tissue could be detected in the secondary resection specimen. There is no sign of recurrence since 2 years.

Keywords: Carcinoma ex pleomorphic adenoma, minor salivary gland, pleomorphic adenoma

Introduction

Malignant mixed tumors are usually classified as one of three main subtypes: (a) carcinoma ex pleomorphic adenoma (CXPA), (b) true malignant mixed tumor (carcinosarcoma), and (c) metastasizing mixed tumor.[1] CXPA is defined as pleomorphic adenoma from which an epithelial malignancy is derived.[2] CXPA is reported to represent approximately 3-5% of all major salivary gland neoplasms and 5-15% of major salivary gland malignancies.[3,4] CXPA is exceedingly rare in minor salivary glands of the oral cavity. We present here a case of CXPA arising in the buccal mucosa.

Case Report

A 17-year-old female reported to us with a slowly enlarging mass in her right side of buccal mucosa [Figure 1]. She had first noticed a small swelling six months back. Since then, it had increased markedly in size. The mass was not painful and did not trouble her, except that it had become increasingly noticeable.

Bimanual palpation revealed a firm mobile, 3-cm well-circumscribed mass in the right buccal mucosa region lateral to the upper premolars. The mass could be felt in between the mucosa and skin. The overlying mucosa and skin were of normal colour and texture. Clear saliva was expressed from the right parotid duct. There was no trismus or any associated lymphadenopathy. The clinical features were highly suggestive of a benign lesion.

Ultrasonography showed a well-defined, round; predominantly hypo echoic mass lying underneath the right buccinator muscle. A differential diagnosis of sebaceous cyst or epidermal cyst was suggested. Fine needle aspiration cytology was suggestive of epidermal cyst. All other routine haematological investigations were within normal limits. The tumor was enucleated under general anesthesia [Figure 2]. On gross examination, the encapsulated lesion measured 4 × 2.5 cm, with a whitish, faintly lobulated surface.

Histopathological examination, (×20, HandE stain) showed a neoplasm with an admixture of epithelial and stromal components. Tumor epithelial cells arranged in the form of ducts containing eosinophilic material, islands and sheets [Figure 3]. Outer myoepithelial cells were surrounded by a chondromyxoid stroma, consistent of pleomorphic adenoma.

On magnification (×40, HandE Stain) certain areas showed tumor epithelial cells invading the fibrous capsule [Figure 4]. Cellular details revealed moderate nuclear pleomorphism, loss of chromatin, prominent nucleoli, and moderate mitotic activity. Keratin formation by epithelial cells is also seen [Figure 5]. Due to the presence of atypia and capsular infiltration, the diagnosis of minimal invasive variant of CXPA was made. This rare histopathological diagnosis warranted a
therapeutic reconsideration as some of the margins showed infiltration. The patient was recalled after a month and wide local excision done. The second histopathological report was free of tumor cells, and exhibited connective scar tissue. After a 1 year follow-up period, the patient is symptomless and shows no signs of recurrence.

Discussion

About 1.6-7.5% of pleomorphic adenoma show malignant changes in its natural course. Pleomorphic adenoma is very rarely reported in the minor salivary glands and CXPA involving the buccal minor salivary gland is even rarer. CXPAs are subclassified into: noninvasive, minimally invasive (about 1.5-mm penetration of the malignant component into the extra capsular tissue), and invasive (more than 1.5-mm penetration into the extra capsular tissue). Depending on this subclassification the treatment may vary. However, due to the rarity of occurrence, it is not usually considered in the differential diagnosis, as in our case where the histopathological finding necessitated a second surgery. We should, therefore, always advise an incisional biopsy in large lesions of the buccal mucosa followed by thorough histopathological evaluation and definitive radical treatment. In smaller lesions, complete excision with wide margins should be undertaken.

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