Case Report

Oncocytic changes in pleomorphic adenoma: Report of a rare case

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Abstract

Pleomorphic adenoma is the most common benign salivary gland tumor, accounting for almost three-fourths of all such tumors. Cells with oncocytic change are a common finding in salivary glands and in salivary gland tumors. When found within pleomorphic adenomas, cells with oncocytic changes may be perceived as evidence of malignancy, and lead to a misdiagnosis of carcinoma ex-pleomorphic adenoma. A case of pleomorphic adenoma arising de novo in the minor salivary glands with oncocytic changes is discussed here.

Key words: Minor salivary gland tumors, oncocytes, pleomorphic adenoma

INTRODUCTION

Pleomorphic adenoma is the most common benign tumor to arise in the minor salivary glands. However, the majority of minor salivary gland tumors are of the malignant variety. Oncocytic metaplasia is a common finding in neoplastic and nonneoplastic salivary gland specimens. In normal salivary glands, the presence of oncocytic metaplasia, also known as “oncocytosis,” is regarded as a feature of an “ageing” salivary tissue.[1] A case of a pleomorphic adenoma of the minor salivary glands arising de novo in the hard palate in a 15-year-old male patient is described.

CASE REPORT

A 15-year-old male patient reported to the outpatient department with the chief complaint of swelling in the middle of his upper jaw since 6 months. Patient also complained of pain in the right upper back tooth since 6 months. History of present illness revealed that asymptomatic swelling in the upper jaw initially appeared 1-year back which then itself bursted with blood discharge. It again reappeared 6 months back.

Patient was having pain in right upper back tooth since 6 months which was severe, intermittent, throbbing type, nonradiating, increased while taking hot and cold food items and has decreased a little after initiation of root canal treatment (2 months back) and medication (antibiotic and pain killer). Medical and family history was not relevant.

Intra oral examination [Figure 1] revealed a well-demarcated, solitary, sessile, roughly ovoid, red-colored, dome-shaped, lobulated swelling on the right side of palate in relation to 13, 14, 15, 16, crossing midline, measuring approx. 3 cm anteroposteriorly and 2 cm transversely in its greatest dimensions. On palpation swelling was nontender, lobulated, soft, nonfluctuant, nonreducible, noncompressible, and fixed to underlying structures. There was no discharge on palpation.

Hard tissue examination revealed proximal dental caries involving pulp with respect to 16 and 26 with tenderness on vertical percussion. Based on the above findings, a provisional diagnosis of dental caries involving pulp in relation to 16 leading to palatal abscess was given.

The main differential diagnosis which is to be considered includes any odontogenic cyst like radicular cyst in relation to 16, any benign tumor of minor
salivary glands or a low-grade malignant tumor of salivary gland like mucoepidermoid carcinoma.

An intraoral periapical radiograph (Figure 2) was advised which showed a radiolucency involving enamel, dentin, and pulp in 16 on the mesial side suggestive of mesial proximal caries involving pulp with no lytic areas in the periapical region. Occlusal radiograph (Figure 3) also showed a normal trabecular pattern of palatal bone. Fine-needle aspiration was done which showed epithelial cells, myoepithelial cells, and chondromyxoid material suggestive of a pleomorphic adenoma. Hence, a working diagnosis of pleomorphic adenoma of a minor salivary gland of palate was given.

Excision of the lesion was done under general anesthesia, and the tissue was sent for histopathological examination. The given section showed (Figure 4) sheets of cells with less fibrous stroma surrounded by connective tissue capsule. At the periphery epithelial cells with few angular darker myoepithelial cells were arranged in tubular and ductular pattern with hyalinized and myxoid stroma. In center, oncocytic cells (Figure 5) were round to polygonal with eosinophilic cytoplasm and vesicular nuclei with prominent nucleoli.

Based on the following features, a final diagnosis of “oncocytic variant of pleomorphic adenoma” of minor salivary glands of the palate was given.

The prognosis of the case was good and on a follow-up of 2 years, no signs of recurrence were reported (Figure 6). The patient is still under regular follow-ups.

**DISCUSSION**

Pleomorphic adenoma is a term suggested by Willis. It is the most common type of all benign tumors, accounting three-fourth of all salivary gland tumors.
Onset is usually seen in fourth and sixth decade with slight female predilection. Pleomorphic adenoma usually presents as a mobile slowly growing, painless firm swelling. It may grow to form large giant lesions.[4]

The diagnosis of pleomorphic adenoma is established on the basis of history, physical examination, cytology, and histopathology.[5] Computed tomography scan and magnetic resonance imaging can provide information of the location, size, and extension of tumor to surrounding superficial and deep structures.[6] Pleomorphic adenomas are usually well-demarcated or encapsulated, but an extension of tumor into the capsule is a common feature.[7]

In 1931, hamper coined the term “oncocyte” to describe the cells with abundant eosinophilic granular cytoplasm and central hyperchromatic nuclei. Ultrastructural analysis has revealed the presence of an increased number of normal and abnormal mitochondria in the cytoplasm of oncocytic cells. Tumors of salivary glands formed by oncocytic cells include oncocytoma, oncocytic carcinoma and warthin’s tumor. In pleomorphic adenoma, eventual oncocytes are common, but extensive oncocytic changes are unusual.

Rarely, nononcocytic tumors present a predominance of oncocytes, and this has been reported in myoepithelioma, acinic cell carcinoma, mucoepidermoid carcinoma, basal cell adenoma, polymorphous low-grade adenocarcinoma, and sebaceous adenoma.[8] Oncocytic changes in myoepithelial cells in myoepitheliomas and pleomorphic adenomas can cause diagnostic pitfalls in the differential diagnosis of salivary gland tumors.[9]

According to Palma et al. 2007, pleomorphic adenoma cells show the typical immunohistochemical profile, including positivity for Ck5/6, Ck8/18, Ck14, vimentin, alpha-smooth muscle actin, S100 protein, p63, epidermal growth factor receptor and b-catenin, whereas oncocytic cells shows a luminal phenotype, expression of anti-mitochondria antibody and reduced b-catenin staining.[10]

The surgical treatment for the oncocytic pleomorphic adenoma is complete wide surgical excision with good safety margins.[10,11]

A key area of difficulty is in the diagnosis and significance of the intra-capsular change in pleomorphic adenoma. The WHO classification recognizes an entity termed noninvasive or intra-capsular carcinoma ex pleomorphic adenoma provided the capsule has not been breached or if tumor does not extend further than 1.5 mm from the main tumor mass, then the lesion appears to have the same prognosis as a conventional benign pleomorphic adenoma.[7]

Learning points
• Juvenile oncocytic pleomorphic adenoma of the palate is a rare neoplasm and, therefore, its diagnosis requires a high index of suspicion
• Oncocytes found within pleomorphic adenomas may be perceived as evidence of malignancy, and lead to a misdiagnosis of carcinoma ex-pleomorphic adenoma.

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