

Pleomorphic adenoma to adenoid cystic carcinoma (Major tumor in minor salivary gland) : A case report

Abstract:

Pleomorphic adenoma is the most common neoplasm of major salivary glands and less frequently in minor salivary glands. Here, we report a case of pleomorphic adenoma of the minor salivary glands of the hard palate in a 75 year old Indian female, removed by wide local excision with adequate margins with immunohistochemistry essay suggestive of adenoid cystic carcinoma of hard palate which is a rare transformation of a benign salivary gland neoplasm to a malignant form.

Key-words: Pleomorphic adenoma, Adenoid cystic carcinoma, Immunohistochemistry, p43, p60x

Introduction:

Salivary gland neoplasms represent 1 % of all tumours and 2–6.5 % of all head and neck neoplasms. Minor salivary gland tumours are infrequent accounting for 25 % of all salivary gland neoplasms [1]. Pleomorphic adenomas may occur at any age, but mainly affect patients in the 4th to 6th decade. The treatment of choice in minor salivary gland is wide local excision with the removal of periosteum or bone if they are involved. Simple enucleation of this tumour leads to high local recurrence rate and should be avoided [2]. Hence, the diagnosis of the lesion at such a sensitive site plays a very important role in the treatment planning. Here we report a case of pleomorphic adenoma on the palate, with an immunohistochemistry(IHC) suggestive of adenoid cystic carcinoma of the same.

Case Report:

A 75 year old female patient reported to the Department of Oral and Maxillofacial Surgery, Adesh Institute of Dental Sciences and Research, Adesh University, Bathinda (Punjab), India with a slowly growing hard palatal mass on anterior hard palate region. The lesion frequently got traumatized while eating and had been present for approximately 8 – 9 months. History of attempted incisional biopsy was present for the same but no evident records were present. Clinical

examination revealed sutured, non-ulcerated palatal swelling with irregular surface on anterior hard palate region extending anteroposteriorly from anterior alveolar process region to 1st molar region and mediolaterally crossing mid palatal region [Fig. 1]. The mass was smooth, multinodular, soft to firm, non tender and non fluctuant, on palpation it measured about 4 x 5 x 3 cm[3] approximately. Radiographic evaluation reveals no intrabone radiolucency or cortical plate erosion. Complete surgical excision with a 5 mm safety margin was performed under local anaesthesia [Fig. 2]. The greater palatine neurovascular bundle was clamped before delivery of the specimen [Fig. 3]. After excision, the raw surface was covered

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with periopack and obturating plate fabricated preoperatively was fixed to cover the surgical site for 48 – 72 hours [Fig. 4a, 4b]. Obturator was removed after 48 hours and thorough saline irrigation was done. No signs of secondary haemorrhage was present at the surgical site postoperatively. The postoperative course was uneventful. Patient was recalled after 15 days for follow-up and no evidence of wound infection or secondary haemorrhage was present [Fig. 5]. The patient's postoperative course was uneventful. No recurrence was observed after a follow-up of 1 year. The excised mass was sent for histopathological examination, which revealed underlying connective tissue showed atypical tubules and glands with chondromyxoid stroma [Fig. 6a, 6b]. Full panel **IHC** was then done to rule out histopathological differential diagnosis, which confirms a most probable possibility of adenoid cystic carcinoma by showing immune-positive p63 and p40 markers.

Discussion:

Salivary gland tumours constitute about less than 4% of all head and neck tumours. Among the various histological varieties of salivary gland tumours pleomorphic adenoma happens to be the commonest one constituting about 70% of the tumors in minor salivary glands and the most common intra oral site is the palate, followed by the upper lip and buccal mucosa [3]. Most common symptom manifested by tumors of this location are dyspnea, dysphagia, acute airway obstruction, and obstructive sleep apnea. However, in our case, tumor of size of approximately 5 x 4 cm did not produce any respiratory distress although patient had history of slight dysphagia at times. Most important feature of pleomorphic adenoma of minor salivary glands is that there is no capsule, if present is only very thin [3]. This gives a false impression of infiltrating mass. The potential risk of PA becoming malignant is about 6% [4]. These tumours are encapsulated and hence complete removal ensures cure. Care should be taken to leave at least 1 mm margins around the lesion. While removing the mass, rupture of the capsule is to be avoided to minimize recurrence. Papadogeorgakis et al. [5] considered five points to be the main parameters in selecting the best approach while treating tumors of the hard and soft palate

Proximity and the projection of the tumor to the oropharyngeal wall or the neck.

Size of the tumor.

Suspicion of malignancy.

Vascularity.

Relation of the tumor to the neck neurovascular bundle.

In our case, we preferred intraoral approach as it was a cosmetic approach and offers a direct access to the hard palate. Disadvantages of this approach are that it gives a limited exposure with consequent unsatisfactory control of neck great vessels making it difficult in controlling massive hemorrhage, and increased incidence of nerve damage and capsular rupture leading to tumor recurrence [6]. Reconstruction of the palate is a challenging task. Bony defects can be treated conservatively with an obturator. The goals of maintaining speech, swallowing and anterior facial projection should be at the forefront of each surgeon's mind when approaching these difficult cases.

Immunohistochemical analytic study has been done to differentiate between polymorphous low-grade adenocarcinoma versus pleomorphic adenoma of minor salivary glands [7]. However, in our case, immunohistochemistry essay reveals most probable possibility of adenoid cystic carcinoma, which as a lesion founds to be a malignant form as far as salivary gland tumor is concerned in hard and soft palate. ACC is one of the malignant salivary gland neoplasm, first described by Theodar Bilroth in 1856 as “cylindroma” [8]. This clinical entity of carcinoma was termed as ACC by Ewing in 1954. ACC accounts for 1% of total head and neck malignancies and 22% of all salivary gland malignancies [9]. It is an uncommon malignancy of secretory glands, which is characterized by slow growth kinetics and perineural invasion, and is among the rarer tumors for which little research has been performed and for which there are few treatment options for patients with advanced disease [10].

Conclusion:

To conclude, besides considering pleomorphic adenoma as the commonest salivary gland tumors and adenoid cystic carcinoma involving minor salivary gland tumors should be considered in differential diagnosis. This malignant representation by immunohistochemistry of a benign lesion in hard palate region is quite rare. Thus, recurrence after many years of surgical excision as well as malignant transformation should be a concern and therefore long term follow-up is necessary

Declaration of patient consent:

The authors certify that they have obtained all appropriate

consent forms from patient. In the form, the patient(s) has given his consent for images and other clinical information to be reported in the journal. The patient had understood that his personal information will not be published.

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