CASE REPORT

Soft Palatal Pleomorphic Adenoma with Dystrophic Calcifications and Upper Airway Obstruction

Ayodele Moses Akinola¹, Oluwapeleumi O. Olusoga-Peters¹, Omotayo F. Salami², Adekunle Moses Adetayo³, Taiwo O. Solaja⁴

¹Department of Surgery, Otorhinolaryngology Unit, Ben Carson School of Medicine, Babcock University, ²Department of Surgery, Anaesthesia Unit, Ben Carson School of Medicine, Babcock University, ³Department of Surgery, Oral and Maxillofacial Unit, Ben Carson School of Medicine, Babcock University, ⁴Department of Histopathology, Ben Carson School of Medicine, Babcock University, Ogun State, Nigeria

INTRODUCTION

Pleomorphic adenoma (PA) is the most common neoplasm of the major salivary glands and affects mostly the parotid gland, less frequently the minor salivary glands. It derives its name from the architectural pleomorphism seen under light microscopy.¹ It is also known as “mixed tumor of salivary gland,” which describes its pleomorphic appearance as opposed to its dual origin from epithelial and myoepithelial elements. It accounts for 73% of all salivary gland tumors.² When it occurs in the minor salivary gland, the palate is the most common site apart from the lips, oral cavity, neck, and nasal cavity.²

PAs may occur at any age but mainly affect patients in the fourth to sixth decade. Male: female affliction ratio is 2:3.³ It also ranks as the most common salivary gland neoplasm in children, representing 66%–90% of all salivary gland tumors.¹ Clinical manifestations of pleomorphic salivary adenoma (PSA) of the palate may include dysphagia, hoarseness, obstructive sleep apnea, and sometimes, difficulty breathing.⁴ Because palatal swellings involve a broad spectrum of pathologic conditions, clinical diagnosis is a great challenge.⁵ In view of this difficult diagnosis, radiographic and histopathologic investigation is indispensable as it helps in the differential diagnosis.⁶

Wide local excision with removal of the periosteum and the involved bone is the treatment of choice in the palate in order to avoid recurrence.⁵

CASE REPORT

A 49-year-old Nigerian first presented in our hospital on the 16th of April 2019 with two years' history of a firm and painless swelling with no ulceration at the roof of the mouth that has progressively increased in size from about 1cm to the present size of 6 x 8cm. There is associated muffled speech, mouth breathing, snoring with waking up and gasping for breath in the night (apnea). Diagnosis of palatal pleomorphic...
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adenoma was made, and the patient was informed of the need to do a computerized tomography scan and fine-needle aspiration cytology (FNAC). The patient, however, defaulted for a year due to financial reasons.

He represented on April 15, 2020, with progressively worsening of the voice quality and excessive snoring and sleep apnea. Physical examination showed a middle-aged man not in any respiratory distress. Mouth opening was adequate, but there was a firm, painless mass within the soft palate extending down into the hypopharynx on the left [Figure 1]. There was no area of ulceration in the oral cavity/oropharynx and no fetor oris. Cervical lymph nodes and examination of the ear and nose were essentially normal.

Computed tomography (CT) scan showed a well-circumscribed, nonenhancing isodense oval-shaped mass with multiple areas of hyperdensities that suggest amorphous calcification, bordering soft palate, and prevertebral soft tissue and extend inferiorly to just above the epiglottis at C2/C3 intervertebral disc space causing significant narrowing of the oropharyngeal airway. This mass obliterated parapharyngeal recess and displaced the ipsilateral medial pterygoid muscle laterally [Figure 2]. He was subsequently worked up for the excision of the soft palatal mass under general anesthesia. The ancillary investigations done were all essentially normal and preoperative anesthesia review showed airway assessment of Mallampati IV and necessary plan to deal with this was put in place.

Examination under anesthesia of the mass revealed a firm mass extending into the hypopharynx on the left. The mucosa overlying the mass was healthy with no area of infiltration or ulceration. An incision was subsequently made around the mass and taken deep to reach the muscle layer of the soft palate, which has a thin part of it excised with the mass, together with the overlying mucosa. Satisfactory wound healing was achieved by the 8th week postsurgery [Figures 3-5].

Microscope section shows tissue composed of stromal elements disposed in a somewhat tubular pattern. The stromal is composed of spindly stromal cells in a myxoid background. There were areas of chondroid differentiation and calcifications [Figure 6].

**DISCUSSION**

PA is the most common benign neoplasm that occurs in the salivary glands. Eighty-four percent occurs in the parotid gland, 8% in the submandibular gland, and 4%–6% in the minor salivary glands. The term “pleomorphic” describes the embryogenic origin of these tumors, i.e., a benign neoplasm consisting of cells exhibiting the ability to differentiate into
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epithelial and mesenchymal elements. It is commonly seen in the fourth and fifth decades, although it can be seen in any age.

PA of minor salivary glands is usually a firm, painless, slow-growing tumor presenting as a smooth mass which was seen in our case. The tumor exhibits variation in sizes and can reach a very large proportions, an attribute that was said to be due to its slow growth which makes the patient presents late. As late as 16 years and 17 years were reported by Adetayo et al. and Bordoy-Soto et al. In contrary, the patient in this review presented after 3 years of noticing the lesion, but surprisingly, the mass was already about 7 cm × 6 cm with associated muffled speech, mouth breathing, and snoring. While this relative rapid growth may suggest malignancy, histopathologic investigation through FNAC, as highlighted by Zheng et al., was done to successfully rule out malignancy. On the other hand, it is likely that the patient failed to notice the swelling when it was small. The associated muffled speech, mouth breathing, is known to occur with palatal masses since they tend to narrow the aerodigestive tracts.

The diagnosis of pleomorphic adenoma is established on the basis of history, physical examination, cytology, and histopathology. The differential diagnoses for this case include palatal abscess, odontogenic and nonodontogenic cysts, and other soft-tissue tumors. Abscess can be ruled out because of the absence of signs and symptoms of inflammation, and cysts are not firm in consistency as seen in PSA. CT or magnetic resonance imaging (MRI) is also indispensable in assessing for the presence of bony erosion or soft tissue and nerve involvement.

CT scan and MRI can provide information of the location, size, and extension of tumor to surrounding superficial and deep structures and also the presence of tumor components, in this case, amorphous calcifications. Radiological investigation of this mass showed the presence of intratumor radiodense structures which was confirmed as amorphous calcifications on histology. Calcification as seen in this tumor is not a usual occurrence and has only been reported by a few authors. The pathophysiology is still largely unknown, but the accepted theories of formation include hypercalcemia, tumor components, dystrophic calcification of necrotic area, and calcifications of the materials secreted by the tumors. Opinions on the relevance of this calcification to the clinical behavior of PSA have also been equivocal. Some suggest that the presence of microcalcifications suggests high-grade tumors with more aggressive outcomes while others opined that the calcifications are not related to histopathological grade of differentiations and are not associated with outcome either.

The histological pictures of pleomorphic adenomas vary. PAs of the major salivary glands are similar to those in the minor
salivary glands and are composed of a mixture of epithelial and stromal elements. Three main histologic subgroups have been identified: myxoid (80% stroma), cellular (myoepithelial predominant), and mixed (classic) type. The histological pattern seen in this case was essentially stromal where areas of calcifications were seen. This finding is similar to that of Shi et al. and different from that of Kato et al. that showed more of epithelial component. This diverse histological pattern is due to the differentiation of the mesenchymal elements.

The patient underwent total excision of the mass together with the overlying mucosa. This is the treatment protocol wherein the tumor is excised up to the periosteum of bone with a cuff of normal surrounding tissue. This is aimed to prevent recurrence because PSA lacks a definite capsule. In this patient, there was significant snoring and obstructive sleep apnea as observed by his wife. We did not perform polysomnography (PSG) due to cost implications. However, we believed that if the patient’s sleep-related symptoms persisted following excision of the pleomorphic adenoma, PSG evaluation would be then be indicated. This view is supported by Casale et al. who have suggested that if there is an underlying mechanical problem with snoring, treatment of this should be done first. Snoring and obstructive sleep apnea subsided completely in our patient 3 weeks postsurgical excision of the soft palate pleomorphic adenoma.

Recurrence of PSA is usually a sequel to inability to achieve complete removal. This recurrence is attributed to implantation due to rupturing of the capsule, islands of tumor tissue left behind as a result of surgery, and to multicentricity of pleomorphic adenoma. It is also a risk factor for malignant transformation, the propensity of which has been put at 1.9%–23.3%. We performed complete excision of the tumor with overlying mucosa, and this produced an excellent result.

CONCLUSION

Large pleomorphic adenoma of the soft palate with calcification is a rare condition. Our opinion is that, in the head and neck, where anatomy is very intricate because of the presence of many structures, PSA should be included in the differential radiological and histological diagnosis of the calcifying lesions of this area.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES