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Soft Palate Pleomorphic Adenoma: Triple Upper Aerodigestive Tract DisordersNgamdu YB^{1*}, Adamu AS², Ngadda HA³

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Abstract: Pleomorphic adenoma composed of epithelial and mesenchymal elements, the predominant salivary gland tumours, represents 3-10% tumours of the Head and Neck region. Majority of the pleomorphic adenomas arise from the major salivary glands. The minor salivary glands account for few cases and the soft palate by far is the predominant site, followed by the lips and cheeks. Other rare sites in the upper aero-digestive tract include the hypophaynx and the larynx. A giant pleomorphic adenoma is very rare, but presents mainly with upper aero-digestive obstructive symptoms. Here, we are reporting an unusual giant pleomorphic adenoma of the soft palate in a 28-year old man presenting with triple upper aero-digestive tract disorders. The mass was excised completely with adequate margins under General Anaesthesia and prior tracheostomy. He was followed-up for one- year with no evidence of recurrence.

Keywords: Pleomorphic adenoma, Soft palate, Triple aero-digestive disorder, Tracheostomy.

INTRODUCTION

Pleomorphic adenoma (PA) is a benign tumour that made of both epithelial and mesenchymal elements. It is the most frequent benign neoplasm of the salivary gland. It accounts for about 10% of the tumours in the head and neckregion and 60% of all salivary gland neoplasms [1-5]. Among the salivary glands, parotid is the commonest site, accounts for 80% and 5-10% [1, 6] in minor salivary glands; although the tumours of the minor salivary gland are malignant [6]. In the minor salivary gland, thecommonest site involved by PA in the body is the palate (42.8-68.8%). Upper lips and the cheeks account for 10.1% and 5.5% respectively [2, 5, 7]. The PA of the sublingual salivary gland is exceptionally rare [8]. The variability of the PA is as a result of interplay between the epithelial and the mesenchymal elements. There are reports that these elements are from the same cell clone; myoepithelial or ductal reserve cells [2, 3]. It has a slow and continuous growing pace; presents initially as painless, firm swelling [4, 5], without intervention can reach very large size. Ademar et al. reported a giant PA measuring 28cm and weighed 4kg in the parotid gland [9]. The giant PA of the palate is very uncommon.

We present an unusual case of a giant PA of the soft palate presenting with triple upper aerodigestive tract disorders; severe snoring, difficulty in swallowing and speech problem.

CASE REPORT

A 27year old male applicant presented to the ENT clinic with four years history of a lump in the throat and was progressive. He had snoring (disturbed his neighbours), sleep apnoea and mouth breathing. Other symptoms included difficulty with swallowing (tolerate liquid only), speech problems (barely understood his statements) and a progressive weight loss. No associated nasal discharge, disturbance of smell, odynophagia, cough, fever, neck pain or swelling. He neither smoked nor drank alcohol.

On examination he was found to be wasted (weigh 48 kg), no nasal air current. The intra-oral examination, which revealed a huge non-tender mass (measured 6 x 7cm), as shown in Fig. 1a. The mass was sessile, firm, aroused from the soft palate projected anteriorly and downward, seated on the posterior two-third of the tongue, completely obscured the oropharynx. The overlying mucosa was smooth and intact; no contact bleeding. Dentition and hard palate appeared grossly normal. The cervical lymph nodes not significantly enlarged.

X-rays of the jaw and soft tissue of the neck showed a rounded soft tissue shadow completely obscuring the nasopharyngeal and oropharyngeal air columns, no bony destruction noted. The biochemistry and haematological investigations were within normal limits. Provisional clinical diagnosis of PA of the soft palate was entertained and he was planned for excisional biopsy.

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Anaesthesiologist evaluation was found to have mallampati class IV. Informed consent for preliminary tracheostomy under local anaesthesia obtained and excision of the soft palatal mass under general anaesthesia carried out. The mass was completely excised. It was multi-lobulated with dimension of 8 x 7cm x 5 as shown in Fig. 1b. The soft palateprimarily repaired in two layers. An intra-oral

haemostatic gauge pack left in place for 24 hours. Recovery and immediate post- operative period were uneventful. Tracheostomy decanulated after 48 hours. Histopathological report revealed pleomorphic adenoma as shown in (Fig. 1c). Patient discharged on the sixth post-operative day and followed- up for six months (Fig. 1d), no evidence of recurrence.

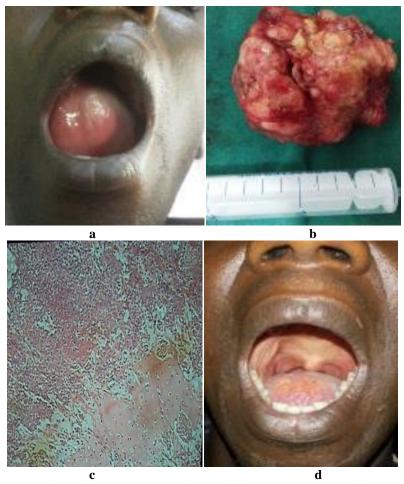


Fig 1: (a). Showing a mass arising from the soft palate filling the oropharynx, encroaching the oral cavity, (b) the mass was shell out: weighing 300g, (c) Photomicrograph showing Pleomorphic adenoma (H&EX160), (d) Showing oropharynx 6 months post-operative period.

DISCUSSION

PA account for 2.4 – 3.05 cases/100, 000 population annually [10, 11]. It is commonly seen 40-60 years old,[5] but affect patients from the first decade to tenth decade of life [7]. There is male to female ratio of 1:1.9 [5, 7]. The commonest sites are the oropharynx and the oral cavity. Other sites that are rare in the upper aero-digestive tract include: Sino-nasal tract, nasophaynx, hopharynx larynx and the cervical oesophagus [2, 5]. PA of the minor salivary gland usually manifests as painless, sub-mucosal swellings, if allowed unattended will reach a very large size. In this index case, the patient also presented long standing painless slow growing firm mass. The most common

site is the meeting point of the hard and soft palate on one side. Here the tumour is often adhered to the adjacent mucoperiosteum. The size of most of the tumours varies from 2-6cm [12], hardly causes upper aero-digestive tract disorders as in contrast to the index case that presented with severe snoring, speech and swallowing disorders. Malignant tumours were detected in 6% PA and the recurrence was found to be 2-44% [6]. Plain radiograph and haematological assessments have no role in the diagnosing PA of the soft palate. Computed Tomography scan is better than Magnetic resonance imaging (MRI) in assessing pressure effect on the bony palate and other contiguous structures nose, maxillary antrum. PA has high signal intensity onT2-

weighted images, whereas the mass of intermediate or low signal intensity on T2-weighted images raises the possibility of malignancy. The MRI features of PA on T2-weighted images are attributed to myxoid tissue within the tumour. In addition, PA usually shows homogeneous enhancement following gadolinium administration, although heterogeneous enhancement may observe in large lesions or cystic tumours [2, 13]. These imaging modalities are valuable in surgical planning especially in giant PA. In the index case, the patient could not afford any of the two radiological investigations because of financial constraint.

PA is a mixed benign tumour of the salivary glands is of variable capsulation characterised microscopically by architectural rather than cellular pleomorphism. Epithelial and modified myoepithelial elements intermingled most commonly with tissue of mucoid, myxoid or chondroid appearance [14], as evident in index case, Figure 1c.

The differential diagnosis are many but the usually ones are palatal abscess, lipoma, fibroma, salivary gland cyst and malignant tumour [2, 4-6]

The PA treated by wide surgical excision with clear margins that involve the periosteum and the associated mucosa. Then this is followed by curettage of the underlying bone with a curette or bur under copious, sterile, normal saline irrigation [14]. The overlying mucosa sometimes can be repaired with a flap.The main problems inthe management of pleomorphic adenomaare the tendency of tumours to recur following surgery and the risk of malignant transformation. In PA of minor salivary glands, recurrence rates are rare, and most of the problems are seen in PA of theparotid gland. A metaanalysis of parotid PA showed 3.4% of tumours recurred afterfive years and 6.8% after ten years [15]. Our patient has not experienced any recurrence after a one- year of follow-up as shown in Figure 1d.

CONCLUSION

Pleomorphic adenoma is benign tumour of the salivary gland. Giant cases are usually uncommon in the upper aero-digestive tract. This case represents a rare example of pleomorphic adenoma of the soft palate. Computed tomography aids in evaluating the extent of the lesion and in guiding the surgical strategy. A long-term follow-up needed to detect the risk of recurrence.

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