Giant Myoepithelioma of the Soft Palate: Report of a case in a Teenage girl

Etetafia MO[1], Nwachokor FN[2]

Abstract:

Background: Giant myoepithelioma is a tumour arising from epithelial cells of mostly the major salivary glands. It is rare in the minor salivary glands .It constitutes less than 1% of salivary gland lesions. It is usually asymptomatic but with increase in size it can elicit pressure and obstructive symptoms as reported in our case.

Case Presentation: This 13-year-old girl presented with swelling on the soft palate for five years. Symptoms included dysphagia, hyper nasal speech, snoring and occasional sleep apnea for two years prior to presentation. Intraoral examination revealed an oval shaped swelling located at the posterior end of the hard palate extending downward and backward pressing on the dorsum of the tongue. CT showed a solid, well-circumscribed oval mass pedunculated at junction of the hard and soft palate. It extended downwards and backwards to the posterior wall of oropharynx. An excisional biopsy was carried out under general anesthesia through the transoral approach. The histology showed myoepithelioma with diffuse infiltrate of plasmacytoid cells. No mitotic figures were seen. The operative site healed without complications. No recurrence three years post op.

Discussion: Myoepithelioma should be distinguished from pleomorphic adenoma because it has been reported to be more aggressive and occasionally transforms into malignant myoepithelioma, though our case was benign.

Conclusion: Myoepitheliomas are rare salivary gland lesions in comparison to pleomorphic adenomas. When large, they can elicit uncomfortable and sometimes dangerous symptoms. They should be considered more in the differential diagnosis of oral lesions in view of their more aggressive nature.

Key words: Myoepithelioma, Plasmacytoid, Giant, Salivary gland.

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Introduction:

Myoepithelioma is a tumor of salivary glands mostly of the major salivary glands. It constitutes less than 1% of salivary gland tumors[1] and it is composed predominantly of myoepithelial cells. The component cells may be spindle-shaped, plasmacytoid, hyaline or epithelioid[2]. Myoepithelial cells are contractile cells found in normal tissues that have secretory functions like the salivary glands, sweat glands, lacrimal glands, prostate and the breasts[3]. Myoepitheliomas are composed completely or almost completely of myoepithelial cells whereas the amount is variable in pleomorphic adenoma[3] Myoepitheliomas frequently affect patients between the fourth and fifth decades of life without gender predominance[3,4].Pediatric cases of myoepitheliomas have also been reported[5,6].The case presented here is a benign myoepithelioma of a minor salivary gland of the soft palate of the plasmacytoid type.

Case Presentation: A 13-year-old girl presented to the hospital in company of her guardian with a 5-year history of swelling in her palate. The swelling was initially small in size but increased gradually until it started affecting her speech, mastication, swallowing and sleep. There was associated snoring while asleep and patient would suddenly jump up from sleep to catch some air at night. The noise of the snoring while asleep became so loud that other siblings became uncomfortable in the same room. Patient gradually resorted to semi solid diet to reduce the discomfort associated with the size of the tumor. Extra oral examination showed a healthy looking young girl who had no obvious respiratory distress but had some distortion of her speech. Intraorally, the mouth opening was good and no obvious pathology on the soft and hard tissues except for a growth located at the posterior end of the hard palate more on the right side [Fig.1]. It was oval in shape and it measured approximately 5.0cm by 4.5cm. It extended downward and backward pressing on the dorsum of the tongue anterior to the vallate papillae. It also extended laterally towards the left side almost covering the anterior wall of the oropharynx. The overlying mucosa appeared very erythematous with visible underlying blood vessels. No ulceration of the overlying mucosa. The lesion was firm in consistency. An initial diagnosis of pleomorphic adenoma was made to rule out other lesions like leiomyoma and schwannoma. The CT of the facial region revealed a solid, well-circumscribed oval mass pedunculated at the junction between the hard and soft palate [Fig.2]. It extended downwards to the posterior one third of the tongue, backwards to the posterior wall of oropharynx and upwards to the lower part of nasopharynx. There was no obvious bone involvement. The patient was prepared for general anaesthesia, consent was obtained and then she was taken to theatre for excisional biopsy. The approach was the transoral route and the lesion was excised from the base and the resultant defect was repaired. The surgical specimen [Fig.3] showed a well-circumscribed encapsulated mass measuring 7.6cm by 6.0cm by 4.0cm.The cut surface showed a variegated appearance. The histology [Fig.4 and Fig.5] showed predominant myoepithelial cells with diffuse infiltrate of plasmacytoid cells. The cytoplasm was eosinophilic with nuclei being hyperchromatic and pleomorphic. Some sections showed elaboration of extracellular mucin. No mitotic figures were present. The post-operative period was uneventful. The operation site [Fig.6] healed without any post-operative complications of defect or fistula. No recurrence up to four years post op.

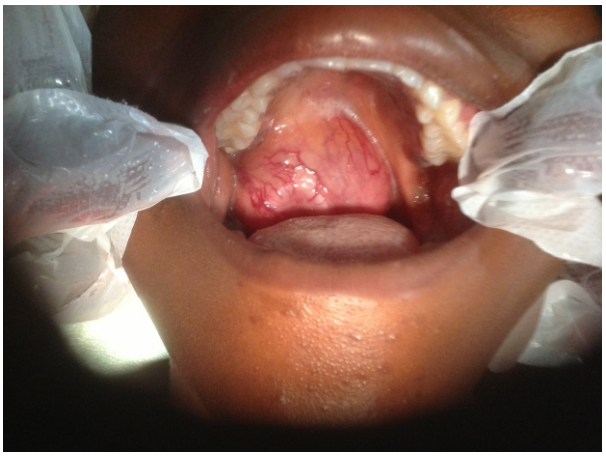

FIG.1-Palatal mass in situ

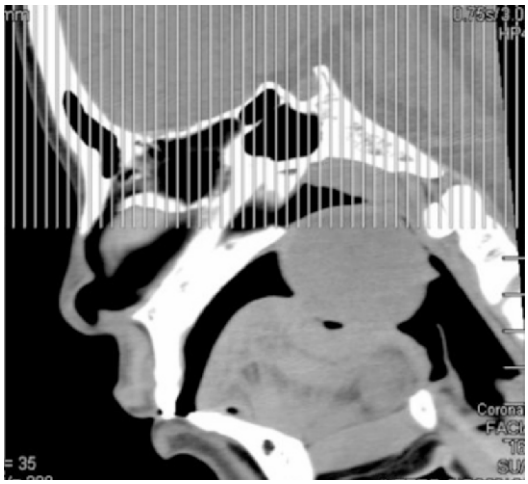

FIG.2-CT Sagittal view

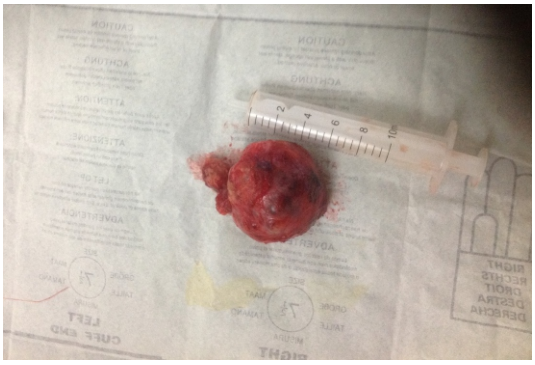

FIG.3-Surgical Specimen

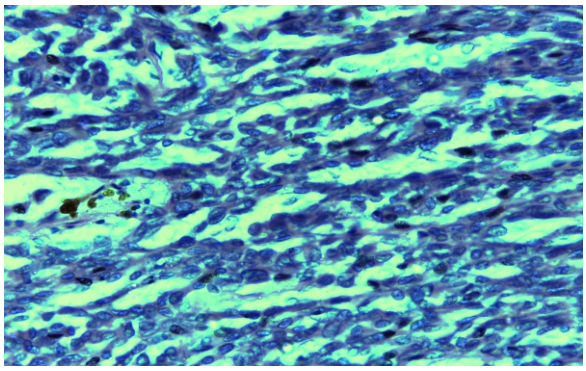

FIG.4-Histology × 200

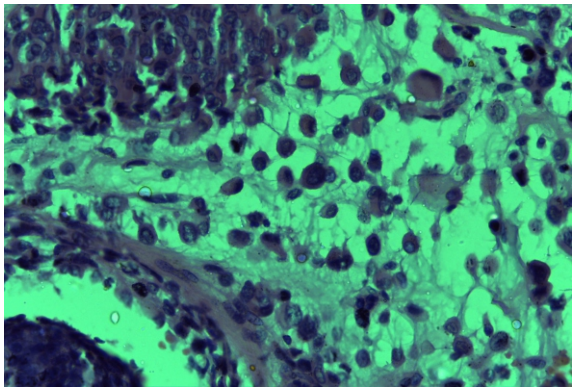

FIG.5-Histology × 400

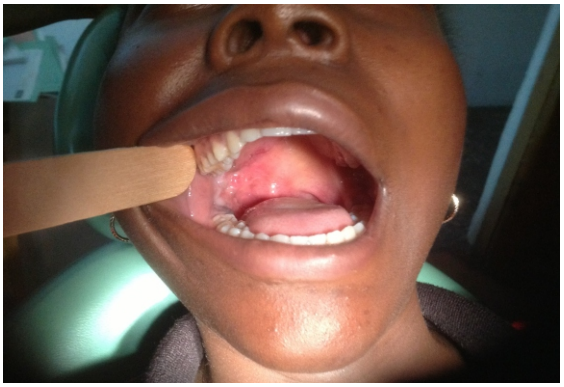

FIG.6-Post Operation site

Discussion: Myoepitheliomas are rare benign tumours of salivary glands, slow growing and asymptomatic in many cases hence the growth to a large size as seen in our case. Over a prolonged period of time the lesion located on the soft palate became very large and elicited symptoms of snoring, hyper nasal speech and sleep apnea. Although most cases of myoepitheliomas are located on the parotid glands[4], rare locations have been seen in the cheek[3], hard palate[7], soft palate[8] maxillary sinus[9], gingivae[1], tongue[10], and upper lip[11].Histopathologically, they consist of spindle-shaped, plasmacytoid, clear, or epithelioid cells.The neoplastic cells, if present, ,are arranged in sheets, irregular collections, nests, interconnecting trabeculae, or ribbons[12]. The benign histologic picture was very dominant in this patient. The neoplastic component was absent. Myoepithelioma should be distinguished from pleomorphic adenoma because it has been reported that myoepithelioma is more aggressive than pleomorphic adenoma and occasionally transforms into malignant myoepithelioma[13] hence, histologic analysis of suspected adenomas is necessary to differentiate between myoepithelioma from the more common pleomorphic adenoma. An author[5] with a contrary view stated that although myoepitheliomas in children may be invasive, it is less aggressive than other tumours. Although immunohistochemical analysis is carried out in most cases of myoepitheliomas, our diagnosis in this case was based on histologic findings. A low grade malignancy has also been reported[3] with a case of myoepithelioma of the minor salivary gland of the cheek. Surgical excision is the method of treatment of benign myoepithelioma with a long term follow up. The prognosis of benign myoepithelioma is considered to be generally good both in children[12] and in adults[3], but malignant changes and local

infiltration do occur[4,8] , hence the need for histopathologic examination and long term follow up of treated cases of myoepitheliomas. Malignant myoepitheliomas should be treated in line with other malignant lesions. A reported case[14] of malignant myoepithelioma of the submandibular gland underwent surgical excision with neck dissection and chemoradiotherapy without signs of recurrence after treatment. The surgical excision of the lesion in our reported case led to the elimination of all the presenting symptoms and there is no recurrence four years post operatively.

Conclusion: Myoepitheliomas are rare benign salivary gland lesions considered to be more aggressive in nature than pleomorphic adenoma. With that in view, they should be considered more in the differential diagnosis of oral swellings to avoid the complications arising from their neglect.

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