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Case Report

Giant pleomorphic adenoma of submandibular salivary gland: a case report

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ABSTRACT

The most common benign salivary gland tumor is the pleomorphic adenoma (PA). They can attain grotesque proportions and weigh several kilograms. They can cause facial disfigurement and, if untreated, could lead to airway compromise. Authors report a case of a large PA arising from the right submandibular salivary gland in a 48-year-old male. The lesion measured 9cmx8cmx5cm.

Keywords: Carcinoma ex pleomorphic adenoma, Facial deformity, Giant pleomorphic adenoma, Mixed tumor, Submandibular tumor

INTRODUCTION

Pleomorphic adenoma (PA) is the most common salivary gland tumor in both the major and minor salivary glands. They are also referred to as "benign mixed tumors." They represent 60 to 80% of all benign tumors in major salivary glands and 40 to 70% of minor salivary glands.[1] These tumors are slow-growing and painless. However, failure to seek early treatment can result in grotesque facial deformity.[2]

They are also reported to occur in the main bronchus, columella, larynx, pharynx, trachea, lacrimal gland, sinonasal tract, and maxillary sinus, albeit less commonly.[3] The purpose of this article is to emphasize that although the PA is a well-reported entity, they can attain very large sizes and, if left untreated, could also lead to the airways being compromised.

Authors report a case, which to the best of the author's knowledge is the largest PA of the submandibular gland.

CASE REPORT

A 48-year-old male presented to ENT department seeking medical attention for a large swelling on the right side of his jaw (Figure 1 and 2). The swelling was painless with a history of having increased in size over one and half year period. He noticed the swelling for the first time at the age of 20. The patient was otherwise healthy with no other significant medical history or findings. On examination, the weight of the swelling tilted the patient's head to the left. The swelling was multinodular, nontender, hard, and mobile. Movement of the tumor mass could be elicited and palpated bimanually both intraorally (in the region of the right submandibular salivary gland) and extra orally over the mass simultaneously.

Submandibular lymph nodes could not be palpated due to the tumor mass. No other supraclavicular nodes were palpable. The lesion was diagnosed clinically as a PA. FNAC was done which showed features of a chondro

myxoid matrix and sheets of epithelial and myoepithelial cells suggestive of PA (Figure 3 and 4). The cartilage and myxoid areas grew in nodules with cellular areas in between. No mitotic activity was present .

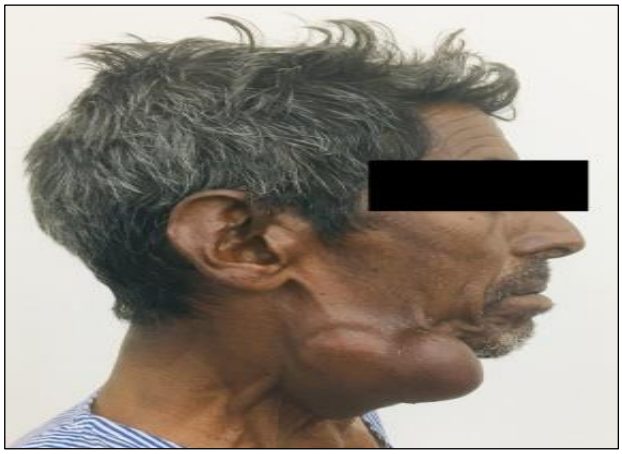

Figure 1: Front and profile view of tumor mass.

Figure 2: Front and profile view of tumor mass.

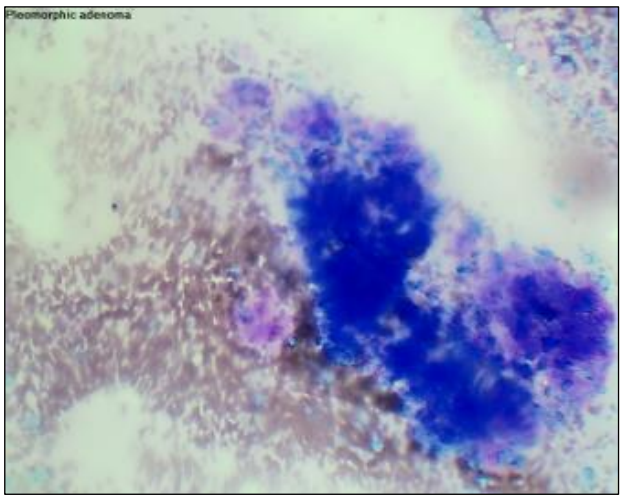

Figure 3: FNAC images of PA.

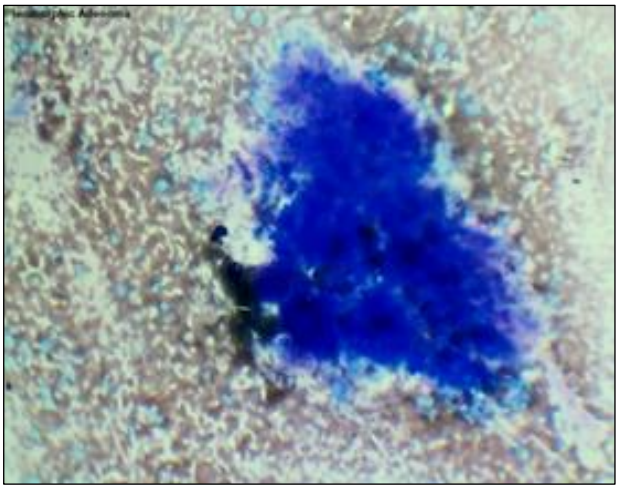

Figure 4: FNAC images of PA.

The right submandibular space and right anterior cervical space with superior abutment of the right parotid gland. Diffuse calcifications were found within the lesion. The epicenter of the tumor was within the submandibular gland. There was medial extension to the left parapharyngeal space. Bony elements of the maxilla and mandible were within normal limits. An en bloc resection of the tumor was done with preservation of the overlying nonulcerated skin (Figure 5).

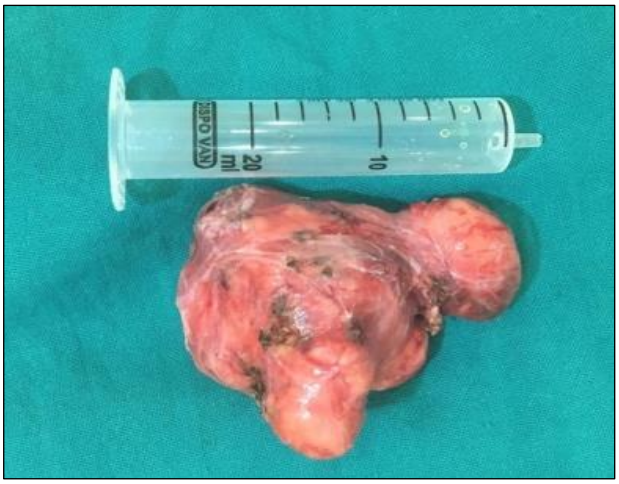

Figure 5: Specimen after removal.

The patient recovered well and was discharged on day 4 postoperatively. Patient review was uneventful, and the incision wound healed well .

DISCUSSION

There is consistency in published reports showing the PA to be the commonest of the salivary gland tumors. Certainly in one of the largest studies involving 6982 primary salivary gland tumors in a Chinese population by Tian Z, PA was the most common tumor (69%) of which only 20% were located in minor salivary glands.[4]

They have been reported to be as large as 28x20x16 cm in the parotid gland.[5] Gupta M in her study reported weights of PAs ranging from 1 kg to 27 kg.[6] The occurrence of very large PAs in the submandibular gland are rare and reports of 8x6x4 cm have been documented.[6] This case measured 9x8x5 cm. They are epithelially derived and typically present as a cytologically benign circumscribed mass with variable encapsulation. Histomorphologically, it is characterized by a variegated architecture comprising epithelial elements admixed with a mucoid, myxoid, fibrohyaline, or chondroid stroma (Figure 6).

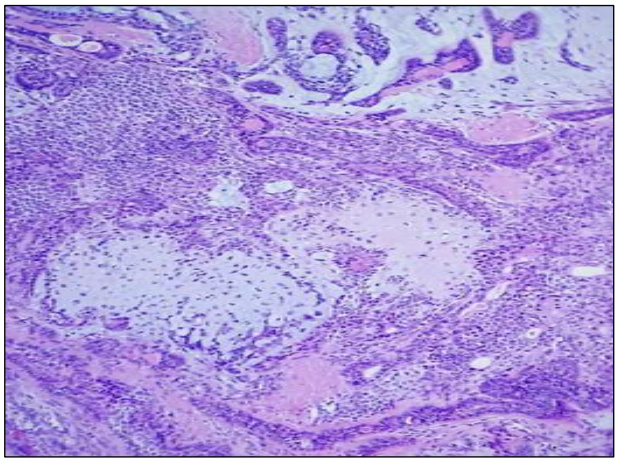

Figure 6: Histopathology picture of PA.

Historically, the main clinical problems with PA have been the risk of recurrence and progression to a clinically or histologically based malignancy.[7] The rate of tumor recurrence seems to depend on surgical techniques used as this could contribute to intra surgical rupture and tumor spillage with resultant seeding of tumor cells. Histopathologic features may also impact on the rate of recurrence. In a study of 225 patients with PA, Henriks son et al, found 56% of the PAs that had recurred showed finger like tumor extensions or pseudopodia outside the pseudo capsule microscopically as compared with the 25% of recurrences in patients who had intraoperative rupture of the tumor.[8] Over recent years, there has emerged a rare but nonetheless well-documented subgroup of PA entities that require additional awareness and precise recognition in terms of their propensity for future aggressiveness. These include features of vascular invasion, focal dysplasia, and noninvasive (intracapsular) carcinoma ex pleomorphic adenoma.[7] Generally, malignant transformation can be suspected with a sudden increase in size accompanied by local signs of malignancy, pain, ulceration, spontaneous bleeding, and superficial and deep tissue invasion. Furthermore, multiple recurrences and the size of the tumor may play a role in the malignant transformation of the PA. The clinical characteristics of malignant transformation have been reported in the literature as (1) a long history of PA, (2) advanced age, (3) location in a major salivary gland, and (4) history of rapid growth associated with pain or ulceration.[7] Age in our case was to the patient's advantage. Representative microscopic sampling of large PAs is important to rule out malignant change, which could increase in frequency with large tumors as malignant sites could be missed. Reporting a case of this nature has significance as delayed intervention allows for rapid tumor growth (believed by some to be due to mutated ras genes) ,poor aesthetics, psychological trauma, and a possible threat to life especially with the presence of tumor pressure over the parapharyngeal space and oropharynx, which could be compounded by infection thus threatening the airway.[9] The expanding mass could further distort the anatomy thus displacing vital structures and making the dissection more challenging.

CONCLUSION

The presentation of cases such as the one being reported highlights the plight of many patients especially in the developing nations. Whatever the reasons; such disease progression should never be condoned, certainly not in the twenty-first century.

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