

**Original Article / Orijinal Araştırma**

**Major Pleomorphic adenoma in a minor salivary gland masquerading as malignancy**

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**ABSTRACT**

Pleomorphic adenoma (PA), though a common entity, is still a challenging tumor for pathologist, radiologist, and the surgeon. Its diverse histological and clinical presentation increases the chances of misdiagnosis. We have reported a case of pleomorphic adenoma which could have been misdiagnosed as malignant tumor due to its masquerading appearance. Hence this presentation emphasizes the need for awareness of its diverse presentation and histopathological variation among the examining clinician as it greatly influence the outcome of management.

**KEY WORDS:** Pleomorphic adenoma, minor salivary gland, Histopathology

## INTRODUCTION

Pleomorphic adenoma (PA) is the most frequently encountered lesion among all salivary gland tumors, accounting for approximately 60% of all salivary gland neoplasms.<sup>[1]</sup> It accounts for 60% to 73% of the parotid gland tumours, 12% to 60% of the submandibular and 14% to 70% of the minor salivary glands tumours.<sup>[2]</sup> The importance of these lesions lies in the fact that they are more likely to be malignant when associated with minor salivary glands (50%).<sup>[3]</sup> It has a diverse histological presentation because of varying proportions of different epithelial and mesenchymal elements, and presents clinically and radiologically in various ways as it occurs at many different sites in the head and neck region.<sup>[5]</sup> We report such a case of atypical clinical presentation of pleomorphic adenoma simulating as malignancy.

## CASE REPORT

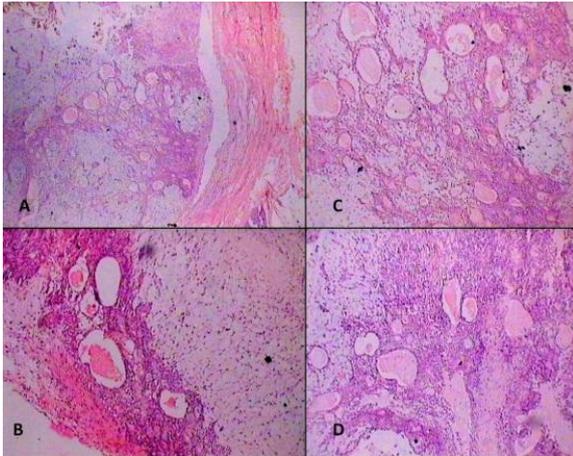
A 65-year-old female patient reported with a progressive swelling in the upper left palatal region for the past 7 months. The patient also complained of difficulty in swallowing since 3 weeks. No complaint of nasal obstruction and hearing loss was reported. History of on and off bleeding from the mass was given chiefly during intake of solid food. Her medical history was not significant and she denied drug allergy.

On intraoral examination, a single ovoid mass [Figure 1] was seen on the left side of hard palate extending posterior to obstruct the oropharyngeal area. The overlying surface on the posterior aspect of mass was erythematous with superficial varicosities and an area of ulceration. On palpation the swelling was rubbery in consistency, sessile and non tender except in the ulcerated area. Neck examination was found normal. Further patient was subjected to maxillary occlusal radiograph which revealed no evidence of bone involvement. Fine Needle Aspiration Biopsy was performed, which was suggestive of Pleomorphic adenoma.

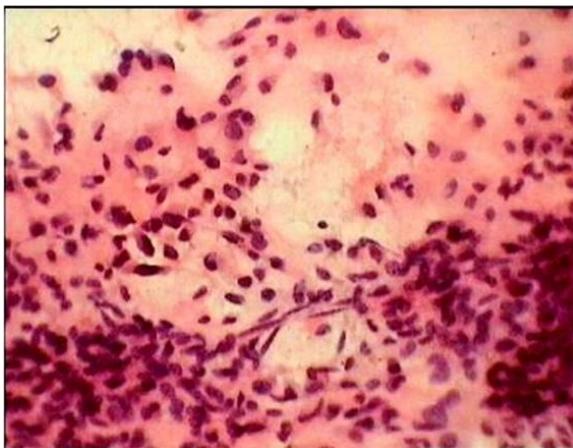


**Figure 1:** Left side of palate showing a single ovoid mass obstructing the oropharyngeal area. The surface on the posterior aspect shows erythematous surface with superficial varicosities and an area of ulceration.

After routine preoperative investigations, wide local excision of the mass and primary closure of adjacent mucosa was carried out. The excised mass was 8 × 4 cm in size which was sent for histopathological examination. The report showed the presence of a well defined lesion surrounded by a fibrous capsule separating the tumor mass from the surrounding glandular structure. The tumor mass was predominantly myxoid type with epithelial component. The epithelium shows duct formation, small cellular nests, and anastomosing cord formation. The cells were epithelioid and spindle shaped. They showed scant pale cytoplasm and vesiculated nucleus. The ductal structures were filled with eosinophilic material. Normal serous salivary gland was seen adjacent to the tumor with acinar structures, striated and excretory ducts. Blood vessels, few muscle fibres, adipose tissue and neural tissue is also seen. The presence of myoepithelial cells and ducts lined by cuboidal cells and it confirmed the lesion to be benign pleomorphic adenoma [Figure 2&3]. No recurrence was observed after a follow-up period for one year.



**Figure 2:** H&E slide shows presence of a fibrous capsule separating the tumor mass which is predominantly myxoid type with epithelial component [A&B]. The epithelium shows small cellular nests, anastomosing cord and duct formation lined by cuboidal cells and filled with eosinophilic material [C&D].



**Figure 3:** H&E slide shows epithelioid and spindle shaped cells with scant pale cytoplasm and vesiculated nucleus present in a myxoid stroma

## DISCUSSION

Pleomorphic adenoma remains one of the most interesting benign tumours encountered in the head and neck surgery.<sup>[2]</sup> The term pleomorphic describes the embryogenic origin of these tumors, which contains both epithelial and mesenchymal tissues.<sup>[6]</sup> It can appear at any age and appear between the fifth and seventh decades of life with a female predilection.<sup>[1]</sup>

Clinical, radiological, and histopathological presentations are diverse as it may occur in a variety of anatomical sites and be

composed of varying proportions of different epithelial and mesenchymal elements. Consequently, there are important considerations for both radiological and surgical approaches to these head and neck tumours.<sup>[5]</sup>

### Diversity in clinical presentation:

Pleomorphic adenoma usually presents as a mobile, slowly growing, painless, firm swelling that does not cause ulceration of the overlying mucosa.<sup>[7]</sup> It is usually well demarcated or encapsulated but extension of tumor into the capsule is a common feature and sometimes lobules of tumor may appear to be completely separated from the main tumor mass.<sup>[1]</sup> Although uncommon painful symptoms have been associated with pleomorphic adenoma in some studies.<sup>[2]</sup> Most tumors occur originate in superficial lobe but more rarely may involve deep lobe of parotid growing medially and occupying parapharyngeal space.<sup>[4]</sup> In 1989, Schultz-Coulon reviewed 31 cases of giant PAs of the parotid gland and found a female predominance (64,5%), with an age range of 20 to 40 years old, and weight of the tumor between 1 to 27 Kilograms.<sup>[9]</sup> The mechanical symptoms most commonly manifested by tumors of this location are dyspnea, dysphagia, acute airway obstruction, and obstructive sleep apnea.<sup>[3]</sup> In rare cases, pleomorphic adenoma can develop simultaneously with other salivary gland tumors or can develop at different sites simultaneously.<sup>[8]</sup> These tumors are also able to invade and erode adjacent bone, causing radiolucent mottling on the x-ray of the maxilla.<sup>[6]</sup>

**Diagnosis:** The choice of imaging with ultrasound, magnetic resonance imaging (MRI), or computed tomography (CT) depends on the site and size of the tumour. On ultrasound it appears typically as a hypoechoic, homogenous, well-circumscribed mass with posterior acoustic enhancement.<sup>[5]</sup> The information of the location, size and extension of tumor to surrounding superficial and deep structures can be detected in CT scan and MRI.<sup>[6]</sup> CT is superior to MRI in evaluating the erosion and the perforation of the bony palate, or the involvement of the nasal

cavity or the maxillary sinus.<sup>[7]</sup> On MRI it has an intermediate or low T1 signal and a more variable T2-weighted signal of which the intensity varies from high in cellular areas to very high (higher than cerebrospinal fluid) in myxoid areas. There is a variable pattern of enhancement on MRI depending on the myxoid and cellular composition of the tumour.<sup>[5]</sup>

Histopathological sampling procedures include Fine Needle Aspiration Cytology (FNAC) and core needle biopsy.<sup>[6]</sup> Fine Needle Aspiration (FNA) operated in experienced hands, can determine whether the tumor is malignant in nature with 90% sensitivity. FNA can also distinguish primary salivary tumor from metastatic disease. Core needle biopsy is more invasive but is more accurate compared to FNA with diagnostic accuracy greater than 97%.<sup>[6]</sup> The risks of seeding many different tumours along the needle tract have been described and this increases with a large bore of needle and the number of passes made.<sup>[5]</sup>

**Histologic variation:** Pleomorphic adenoma is a benign tumour arising from cells of salivary gland tissue.<sup>[5]</sup> The mixed aspect of PA is constituted by two tissue-specific findings: the sub-differentiation of epithelia and modified myoepithelia and the amount and constitution of the stroma. Two subtypes are distinguished on routinely processed slices, i.e. the stroma-rich and the stroma-poor.<sup>[8]</sup> The highly variable morphology of this neoplasm is the result of the interplay between these elements.<sup>[7]</sup> Formation of the capsule is as a result of fibrosis of surrounding salivary parenchyma, which is compressed by the tumour and is referred to as false capsule.<sup>[6]</sup>

It is usually encapsulated when it arises in the major salivary glands, but not in the minor salivary glands.<sup>[5]</sup> The variants of pleomorphic adenoma include pleomorphic adenoma with a lipomatous change, myxolipomatous pleomorphic adenoma, pleomorphic adenoma with a squamous differentiation and benign metastasizing mixed tumour.<sup>[7]</sup>

**Management:** The treatment consists of wide local excision with clear margins which

involves the periosteum and the associated mucosa, followed by curettage of the underlying bone with a curette or bur under copious, sterile, normal saline irrigation.<sup>[7]</sup>

Local recurrence after surgery has been attributed to different factors:-

- 1) type of surgery (i.e. enucleation or lateral parotidectomy),
- 2) lesions of the pseudocapsule due to intraoperative maneuvers, and
- 3) insufficient preoperative diagnostics leading to an underestimation of tumor extension, in particular in cases with multifocal origin of the tumor.<sup>[4]</sup>

The propensity for malignant transformation is documented to be 1.9-23.3%.<sup>[3]</sup> The malignant transformation has been linked to recurrence and multiple excisions, laminin, and collagen IV deposition. Recently, it has been found that loss of b-catenin adhesion molecule is one of the factors responsible for development of pleomorphic adenoma and cytoplasmic accumulation of this molecule causes malignant transformation in pleomorphic adenoma.<sup>[10]</sup>

Thus, the awareness of its varied clinical and histopathological presentation, recurrence, and malignant potential is instrumental on the part of the examining clinician or treating surgeon to avoid incorrect diagnosis or treatment.

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