Scholars Journal of Medical Case Reports
Abbreviated Key Title: Sch J Med Case Rep
ISSN 2347-9507 (Print) | ISSN 2347-6559 (Online)
Journal homepage: https://saspublishers.com

OPEN ACCESS

Surgery

Giant Pleomorphic Adenoma of the Maxillary Sinus with Orbital Extension: A Case Report

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DOI: https://doi.org/10.36347/sjmcr.2025.v13i07.053 | **Received:** 28.05.2025 | **Accepted:** 22.07.2025 | **Published:** 28.07.2025

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Abstract | **Case Report**

Pleomorphic adenoma (PA) is the most common benign neoplasm of salivary gland origin involving both major and minor glands. The most frequent site is the parotid gland. It is rare in the accessory salivary glands, preferably located in the oral mucosa. Incidence of its origin in the respiratory tract is extremely rare, and the occurrence is even lower in the maxillary sinus. We report a case of a 57 years old female presented with a swelling of the middle third of the face and an exophthalmos. Clinical evaluation and imaging studies revealed a huge mass centered on the maxillary sinus. A maxillectomy was performed and the the histopathology report was consistent with a PA. The orbital involvement due to extension as described in this case is very uncommon. These are benign salivary gland neoplasms that can grow into extensive sizes if left untreated and hence need to be diagnosed early.

Keywords: Pleomorphic adenoma, salivary glands, maxillary sinus, sinonasal, exophthalmos.

INTRODUCTION

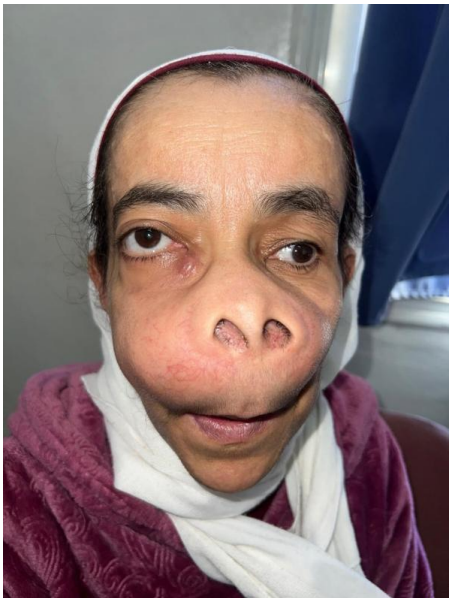
Pleomorphic adenomas (PA) are the most common benign salivary gland tumors. They arise from the major salivary glands, but they may also arise from the minor salivary glands [1]. Pleomorphic adenoma was first termed by Willis [2]. Its name comes from the architectural pleomorphism that may be seen with a light microscope. Myoepithelial and epithelial cells combine to form this benign mixed tumor [3]. In the earlier years, it was also referred as mixed tumor, enclavoma, branchioma, endothelioma, endochroma, etc. [4].

With this paper, we report a rare case of recurrent pleomorphic adenoma in the maxillary sinus, with an unusual facial presentation.

CASE PRESENTATION

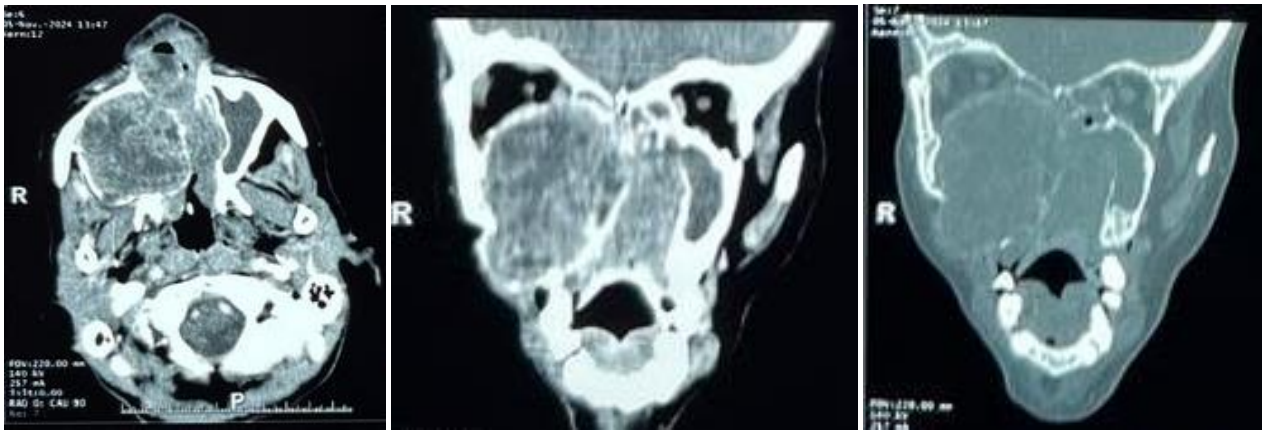
A 57-year-old female patient, with a history of resection of a pleomorphic adenoma of the palate in our department in 2018, consulted in 2024 after having been lost to follow-up, for swelling of the middle third of the face progressively increasing in size.

Clinical examination on the admission revealed an enlargement of the middle third of the face, deforming the bone structure, painless, firm on palpation, responsible for complete nasal obstruction. Ophthalmic evaluation showed a right proptosis with ocular dystopia, a restriction in the downward gaze with no other defects in ocular motility, while visual acuity and fundus examination were normal.



Photograph of the patient before surgery

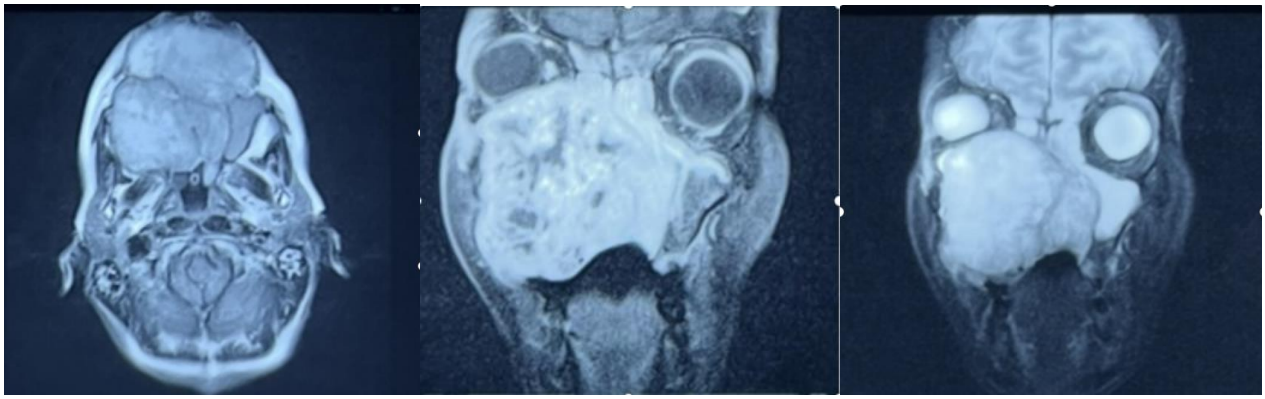
Facial computed tomography-scan revealed a voluminous right naso-ethmoidal expansive process, poorly limited, hypodense and heterogeneous, with areas of fluid density suggestive of necrosis. This process blew out the walls of the right maxillary sinus, with local bone destruction, and extended into the nasal cavity, with lysis of the nasal septum and ethmoidal bone. It extended anteriorly and laterally to the right orbit, with osteolysis of the orbital floor and grade 2 exophthalmos.



A **B** **C**

Axial (A) and frontal (B, C) CT images revealing a mass centred on the right maxillary sinus and the nasal cavity, with osteolysis of the maxillary sinus walls, the nasal septum, and orbital floor

For further evaluation of the soft tissue component of the mass, magnetic resonance imaging (MRI) was performed. MRI showed a voluminous lesional process centred on the right nasal fossa, polylobed and well limited. Characterized by an isosignal in T1-weighted MR images (with presence of a few punctiform high signals within it), and high signal intensity in T2-weighted MR images. After intra-venous injection of gadolinium, a signal intensity increase of the mass was observed, delimiting necrotic fluid areas, measuring 93x77 mm extended over approximately 63 mm.

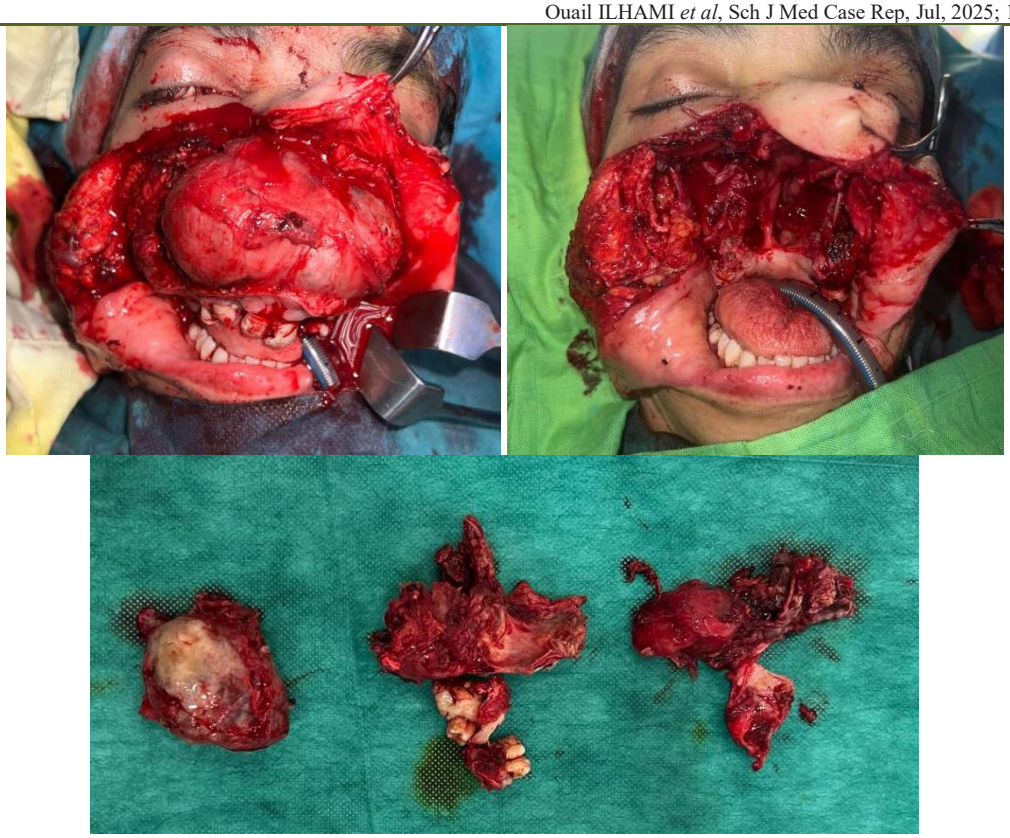


A **B** **C**

Magnetic resonance images. (A) Axial T1-weighted image demonstrating a soft tissue mass of intermediate signal intensity with a few punctiform high signals. (B) Post-contrast coronal T1 image showing enhancement of the mass, delimiting necrotic fluid areas. (C) Coronal T2-weighted image; the mass exhibits high signal intensity

The case was discussed at the multidisciplinary consultation meeting, and the decision was surgery for both diagnostic and therapeutic purposes.

The surgical plan was a medial maxillectomy, under general anesthesia, with removal of the lesion, combined with reconstruction of the right orbital floor using a mucosal flap and a titanium mesh. The tissue removed was sent for pathology examination.



Intraoperative images demonstrating a whitish soft tissue mass occupying the middle third of the face

Histopathological examination showed a pleomorphic adenoma with no signs of malignancy. The tumor limits couldn't be determined due to the fragmented nature of the material.

The post-operative period was uneventful, and the left exophthalmos resolved after surgery.

DISCUSSION

Pleomorphic adenoma (PA) is one of the most frequently seen salivary gland tumors. It is seen in the parotid gland in 80% to 90% of cases.

PA has a different embryological origin. It arises from both epithelial and mesenchymal origin. They arise from intercalated and myoepithelial cells. The mass is well demarcated from surroundings by fibrous capsule. The formation of the capsule is a result of fibrosis of the surrounding salivary parenchyma which is composed of the tumor and is referred to as false capsule. The PA is typically a well circumscribed, encapsulated tumor. The capsule may be incomplete which is more common in minor salivary gland tumours [5].

Neoplasms of the minor salivary gland may occur at any location, though the palate, upper lip, and buccal mucosa are the three most common sites comprising 82.2% of all minor salivary gland tumors [6].

PAs of the upper respiratory airway are rarely described and usually affect the nasal cavity with involvement of the septum in 82.5–90% of the reported cases, followed by the maxillary sinus and nasopharynx [7].

Pleomorphic adenomas mostly occur in the young- and middle-aged adults, between 30 and 60 years. The literature reports suggest female predilection [8]. Most cases with maxillary sinus involvement seen in the literature are adult patients, and at an advanced age [9].

Although PAs are benign, local recurrence is a potential risk. Following resection, the recurrence rates reported vary from 0 to 8%, while multiple recurrences are associated with malignant transformation [10]. Metastasis can also occur particularly in cases where enucleation or incomplete excision is performed [11].

The main symptoms of sinonasal/nasopharyngeal PA are usually unilateral nasal congestion and epistaxis; other symptoms may include nasal swelling, mucous purulent rhinorrhea, external deformities, otalgia, hearing loss, and otitis media [12].

The diagnosis of PA in the sinonasal/nasopharyngeal regions is challenging because symptoms are not characteristic and radiologic findings are usually nonspecific. CT generally shows bony alterations and expansive or destructive type changes, providing reliable clues for differentiating between benign and malignant lesions. PA usually presents with well-defined, homogeneous soft tissue masses and

expansile bony changes. An aspect of osteolysis is an indirect sign of malignancy [13].

MRI manifestations are varied but often well defined. The signal intensity of T1-weighted images is low to moderate and that of T2-weighted images is high [14].

An extended biopsy with debulking through an endonasal, endoscopic approach when easily accessible, may be optimum for establishing a correct diagnosis especially in cases of large tumors [15].

Establishing the scope and method of resection requires a balance between limiting recurrence due to radicality and ensuring quality of life by minimizing the invasiveness of the procedure. Apart from an open surgery, which consist of an anterior approach with an incision through the upper lip and surrounding the nose wing, an endoscopic intervention can be performed but the effectiveness of this method is not yet assessed due to insufficient observations [16].

Histologically, the pleomorphic adenomas are limited by the outer connective tissue capsule which helps to distinguish the tumor from the surrounding normal tissue. The tumor comprises diverse patterns with three basic structures: Tubuloductal, solid and myxoid areas. While the ducts are made up of cuboidal cells, spindle and myoepithelial cells comprise the solid areas. Myxoid areas are characteristically hypocellular in nature. The tumor may show sheets and strands of cells in a mucoid or chondromyxoid background [17].

Radiotherapy's function is still debatable. Jackson et al. administered postoperative radiation in the patients whose histology revealed that the specimen's resection margins were not clear of tumor or in those who had tumor spilling during surgery. It showed good results, and also good control rates in recurrent tumours treated with a combination of surgery and radiotherapy. This suggests that microscopic residual tumour may be radiosensitive. However, follow-up of the patients is still short in terms of late sequelae of radiotherapy [18].

As far as maxillary sinus PAs are concerned, there are only a few case studies available. Martis and Karakasis were the first to report a case of maxillary sinus PA presenting with cheek swelling in 1971 [19].

In a recent systemic review about PA in maxillary sinus by Maciej and al. only seven described cases of PA originally localized in maxillay sinus. Therefore, it cannot be guaranteed for any of the included case reviews that PA actually originated from the mucosa of the maxillary sinus [20].

In the report by Ishikawa *et al.,* the tumor entered the maxillary sinus presumably from the nasal cavity [21].

In the description by Gupta *et al.,* of an unspecified swelling of the palate that was surgically treated 2 years before the first examination in the authors' clinic, an oral origin was suggested [22].

The cases presented by Lygeros *et al.,* and Ray *et al.,* seem to be the only ones in which tumor emergence from the mouth or nose was not indicated [23].

Analyzing the study of Sınacı et al. it may be questionable whether the original orally derived tumor crossed the bottom of the maxillary sinus or just deformed it. The later presence of PA in the maxillary sinus could be due to the destruction of the bone barrier during the first surgery [24].

We report the case of a patient who underwent surgery for a PA of the palate, and presented 6 years later with a PA centered in the maxillary sinus, with proptosis due to erosion of the orbital floor and invasion of the orbit by the tumor. Our tumour is therefore probably of buccal origin.

We proceeded surgically with an extended resection, which confirm the diagnosis or PA. However, a limitation that applies in our case is the lack of long-term follow-up.

CONCLUSION

Pleomorphic adenoma is known as a benign neoplasm of the major salivary gland tumors. But when affecting the accessory salivary glands, there is a high risk of malignancy and recurrence.

This requiers early detection, accurate diagnosis, effective treatment, and a strict follow-up.

Complete excision of the lesion is a definitive treatment protocol for these cases, and postoperative radiotherapy is recommended due to the high risk of recurrence. However, one should try and prevent breach in the continuity of lesion and remove the entire lesion, to minimize recurrence and transformation into malignancy.

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