



Giant Pleomorphic Adenoma of Infratemporal Fossa: A Rare Case Report

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Abstract

Of all the head and neck tumors, salivary gland tumors account to 3%. Pleomorphic adenomas are one of the most common benign tumors arising from major salivary glands, although it could also develop from minor salivary glands situated at accessory sites like nasal cavity, pharynx, parapharyngeal space, lacrimal glands etc. Tumors of infratemporal fossa are quite unusual, mainly because of its hidden location in retromaxillary region. We report an unusual case of 65 years old male presenting with complaint of progressive left cheek swelling for 4 years. FNAC revealed pleomorphic adenoma of minor salivary gland tumor. Intraoperatively a giant lobulated tumor was seen occupying almost whole space of infratemporal fossa, which was removed in-toto via open approach. Patient was kept on regular follow up with no evidence of recurrence reported till date.

Keywords Pleomorphic adenoma · Infratemporal fossa · Salivary gland tumor

Introduction

Pleomorphic adenoma (PA) are benign mixed heterogeneous tumor, comprising of variable epithelial and myoepithelial components. [5] They account for 70% of all salivary gland tumours. [7] Uncommon sites reported are: retromolar region, floor of mouth, along the cheek, Stenson's duct, accessory parotid tissue and parapharyngeal space (PPS) arising either de-novo or from the deep lobe of the parotid gland. [12] Infratemporal fossa (ITF) is a deep retromaxillary area corresponding to inferior aspect of the middle cranial fossa. It is a complex region on the skull base that is affected by both benign and malignant tumors. Tumors of the ITF region are quite rare and may arise either from region itself or from surrounding tissue. However, because of its concealed anatomic location, signs and symptoms of tumour often appear late and remain unnoticed for a long time leading to delay in the diagnosis and management. [9].

Case Summary

65 years old male presented to ENT OPD with 4 years old history of painless, gradually increasing left cheek swelling. The malar mass (Fig. 1) was firm and non-tender, causing loss of left nasofacial groove and reaching upto zygoma. The overlying skin was normal with no fixity. Intraoral examination (Fig. 2) revealed firm, mucosa covered globular mass in left buccal vestibule encroaching retromolar trigone. Oropharyngeal examination, eye examination and laryngeal endoscopy was normal. No lymph nodes were palpable. Fine needle aspiration cytology of cheek swelling revealed pleomorphic adenoma arising from minor salivary gland. CECT scan (Fig. 3) showed huge expansile lobulated heterogenous enhancing mass in left parapharyngeal space, infratemporal fossa, projecting into masticator space, buccal space and oral cavity with thinning and scalloping of zygomatic bone, mandible and posterolateral wall of maxillary sinus. CEMRI (Fig. 4) revealed a partly defined, heterogeneously enhancing lobulated lesion of size 8*5*6.5 cm in left sided facial tissue with the epicentre at retro maxillary region with extension to left PPS, masticator space, left peri-mandibular and cheek regions. Erosions and scalloping of left mandible were noted with severe mass effect and compression of left maxillary sinus lumen. FNAC from the left cheek swelling revealed moderate cellularity with presence of scattered

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Table 1 Surgical approaches to infratemporal fossa

S. no.	Approach	Advantages	Disadvantages
1.	Fisch infratemporal approach	Resection of tumors with large intracranial extensions in one go	Facial nerve paresis, conductive hearing loss
2.	Transfacial/transmaxillary (our case approach)	Good exposure	facial scar, violation of nasal and oral cavities, infraorbital anaesthesia
3.	Transcervical/transmandibular	Direct approach with good access and vascular control	Trismus, infection through oral contamination
4.	Preauricular subtemporal approach	Wide exposure to tumors extending to cavernous sinus, internal carotid	Difficult hypotympanic and facial recess resection, CSF leak
5.	Endoscopic endonasal approach	Minimally invasive, excellent visualization	Limited access to lateral ITF, difficult margin status assessment

**Fig. 1** Left malar mass reaching upto the zygoma**Fig. 2** Globular mass as seen on intraoral examination

ductal epithelial cells in myxoid background, opening possibility of pleomorphic adenoma. Subsequently, the patient was taken up for excision under GA via open approach. Post tarsorrhaphy left Weber Ferguson incision was given extending to left upper gingivobuccal sulcus. Blunt dissection was done around the lobulated mass (Fig. 5) and maxillary antrum was found to be contracted. Posterior wall of

antrum was exposed and mass was seen extending from ITF, orbital floor superiorly to mandible inferiorly with temporalis muscle as lateral limit. Facial artery and vein supplying the cheek region was ligated. Blunt finger dissection was done and a huge mass was dissected free from surrounding tissue and removed in toto with preservation of adjacent vital structures and musculature of ITF and orbit. The final specimen was sent for histopathological examination. On gross examination, the tumor was solid grey white myxoid tumor of size 9*8.5*7.5 cm. Outer surface was bosselated and partly capsulated. Microscopic examination showed salivary gland tissue with an encapsulated cellular tumor disposed in sheets and lobules composed of tubular and epithelial structures enveloped with myoepithelial cells in chondromyxoid stroma suggestive of pleomorphic adenoma, with no evidence of malignancy. Postoperative period was uneventful and patient was kept on regular follow up (Fig. 6) with no recurrence reported till date.

Discussion

Due to complex anatomy of infratemporal fossa and its occult nature, tumors of the infratemporal fossa pose a diagnostic and surgical challenge. [8] Tumors of ITF can be primary, secondary or metastatic. Nasopharyngeal carcinoma, juvenile angiofibroma, and adenoid cystic carcinoma are among the most common lesions occurring in the infratemporal fossa. Pleomorphic adenoma arising in the infratemporal region has been reported an unusual entity with less than ten cases reported till now. [15] In cases of ITF tumors, signs and symptoms are generally noticed late, when the disease has already progressed, making surgical planning far difficult. [3].

In our case pleomorphic adenoma in ITF appeared as cheek swelling along with buccal vestibule mass. Jeyanthi K et al., Hadi et al. reported with similar cheek symptoms of pleomorphic adenoma arising from ITF. [9] However, in a case report by Gurey LE et al., ITF pleomorphic adenoma

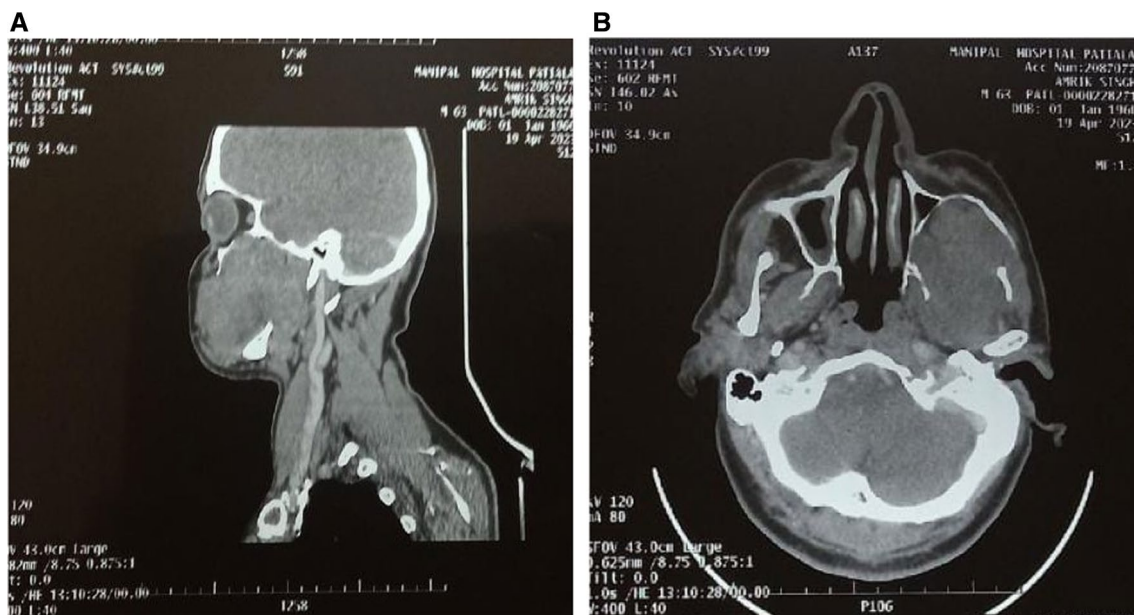


Fig. 3 a, b: CECT showing expansile lobulated heterogenous enhancing mass in left parapharyngeal space, infratemporal fossa, projecting into masticator space, buccal space and oral cavity with thinning and scalloping of mandible and posterolateral wall of maxillary sinus

was noted incidentally. [8] CT scan remains a substantial image modality in diagnosing tumors of the ITF as it helps in determining the location of the tumor, its extensions and its connections with neighbouring structures. Furthermore, Magnetic resonance imaging (MRI) is considered whenever there is suspicion of the vascular lesion, atypical presentations or to determine any skull base/orbital invasion. [6] Nevertheless, actual diagnosis is concluded only after processing of final histopathological specimen. Here, frozen sections play crucial role in early diagnosing the true benign/malignant nature of tumors so that it can be managed in time accordingly intraoperatively. On histopathological examination, pleomorphic adenoma is characterized by a mixture of epithelial, myoepithelial, and stromal elements with myxoid/chondroid stroma. [6].

Treatment of pleomorphic adenoma of the infratemporal space is mainly surgical, with adjuvant radiotherapy advocated for inoperable tumors. Many surgical approaches (Table 1) have been illustrated in literature, keeping in mind the proximity to neighbouring vital structures, neuromusculature and difficult access to ITF. The approach should be selected in accordance to age, gender, facial growth or shape, biological characteristics of pathology,

and surgeon's experience. [10]

In 1987, Al-Mefty and colleagues described a combined infratemporal and posterior fossa approach, a modification of the Fisch infratemporal Approach which allowed tumors with large intracranial extensions to be resected in one go. However, the anterior transposition of the facial nerve and conductive hearing loss remains the concern. [1] Sekhar

et al. elaborated the sub-temporal preauricular ITF approach that provides an extensive exposure but difficult resection of facial recess and hypotympanic areas when invaded by the tumor and CSF leak were few limitations. [14] Al-Mefty and Anand described the zygomatic approach, a preauricular approach which provides an excellent exposure of the ITF while keeping vascularity of temporalis muscle intact. [2] In general, the ITF approaches with preauricular incision are insufficient to expose the mastoid portion of the facial nerve and the jugular bulb. [4] Kassam and coworkers proposed that for lesions medial and inferior to neurovascular structures, endoscopic expanded endonasal approach can be advocated. [11].

The transmaxillary approach and intraoral approach is considered when the tumor has a moderate volume with limited local propagation whereas lateral facial approach is indicated when the mass is enormous and extensive. Some of the limitations include facial deformity, infraorbital anaesthesia, violation of the nasal and oral cavities and vascular complications from the pterygoid venous plexus/maxillary artery. [13]

In a first case report by Jeyanthi et al. [9], zygomatic arch access osteotomy and coronoidotomy was done for tumor mass excision of size 6*5 cm². Haldi et al. performed transmaxillary approach through superior vestibular incision for removal of 4*3.5*2 cm tumor. [6] Gurey et al. [8] performed typical anterior antrostomy to approach ITF space whereas Sequoria et al. [15] used intraoral approach for debridement of pleomorphic adenoma co-existing with actinomycosis in ITF. Thus, a teamwork of otorhinolaryngologist,

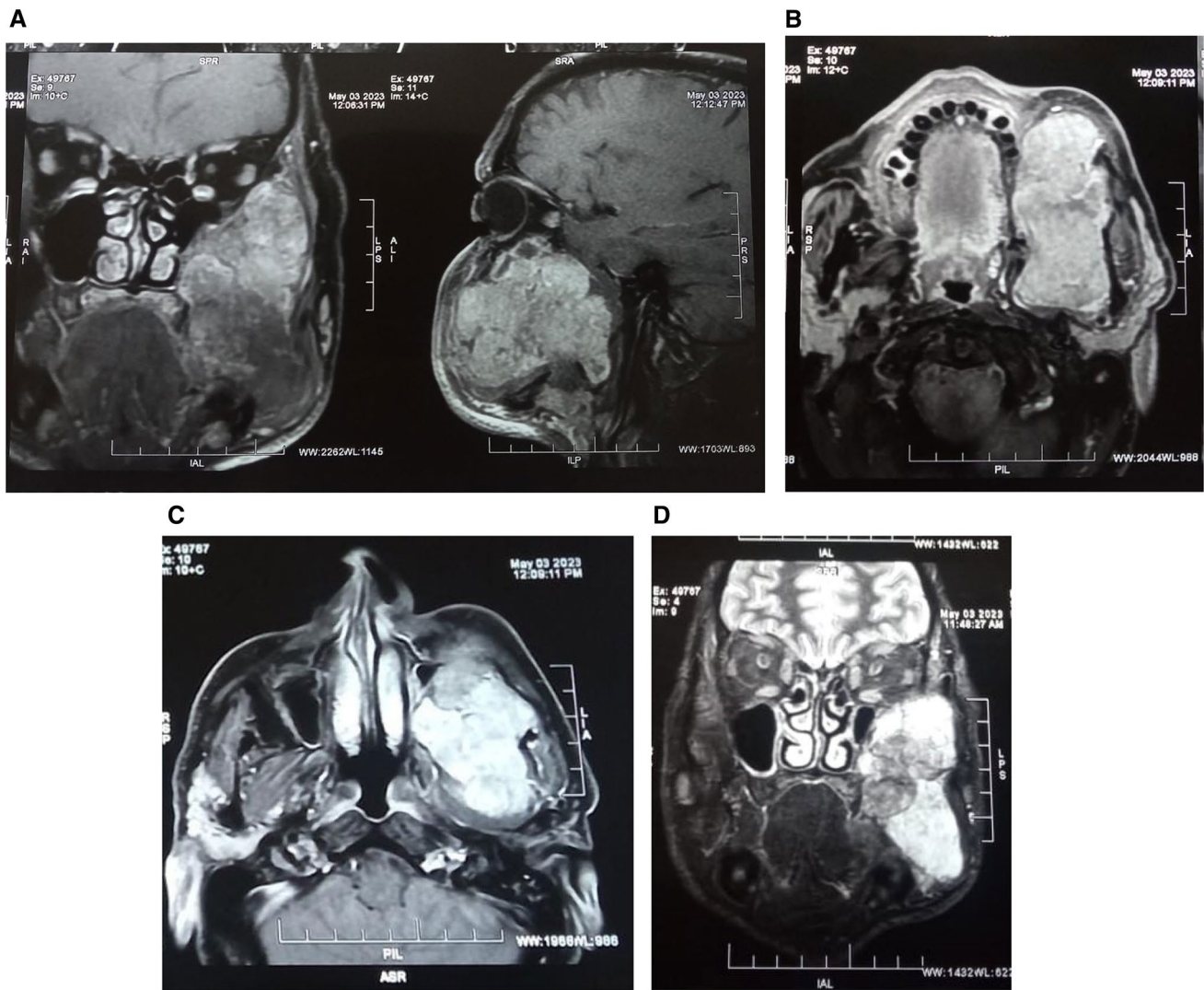


Fig. 4 a, b, c, d: CEMRI showing partly defined, heterogeneously enhancing lobulated lesion of size 8*5*6.5 cm in left sided facial tissue with the epicentre at retro maxillary region with extension to left

PPS, masticator space, left peri-mandibular and cheek regions. Erosions and scalloping of left mandible was noted with severe mass effect and compression of left maxillary sinus lumen

maxillofacial surgeon and neurosurgeon is required to accomplish a good prognosis. [6].

In spite of good prognosis of pleomorphic adenoma, it remains a tumor which has a very high potential of local recurrence ranging from 2.4 to 10%. [6] Long-term follow-up is therefore mandatory, even if the tumor is completely resected and appears to be clinically and histologically non-malignant.

Conclusion

Pleomorphic adenoma in the infratemporal space is a rare entity. Thorough clinical examination along with CT and MRI are the key investigations and incisional biopsy guides the diagnosis. Surgical resection is the main treatment, although concealed location of ITF and its proximity to adjacent vital structures makes surgical planning

Fig. 5 a, b: Intra-operative image (Left); Specimen (Right)

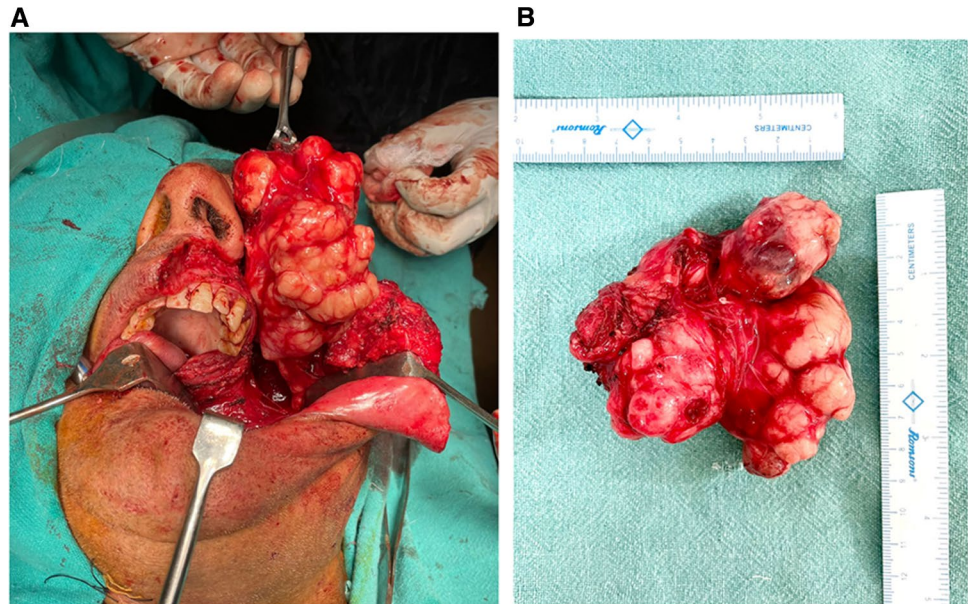


Fig. 6 Post operative clinical picture at 1 month follow-up

difficult. The high potential of recurrence of pleomorphic adenoma makes the follow-up mandatory after surgery.

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Declarations

Conflict of interest The authors have no conflicts of interest.

Ethical Approval Ethics approval has been obtained from the ethics committee of Government Medical College, Patiala.

Informed Consent Informed consent has been taken from the patient for publication.

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