

# Pleomorphic Adenoma of Minor Salivary Gland Arising *de novo* in the Parapharyngeal Space- A Rare Case Report

RAHUL LATURIYA<sup>1</sup>, JENDI SHOEB KASIM<sup>2</sup>, AJIT SURYAKANT JANKAR<sup>3</sup>, SYED AHMED MOHIUDDIN<sup>4</sup>

## ABSTRACT

Parapharyngeal space tumours are rare. Pleomorphic adenoma is the commonest salivary gland tumour that is found in this space. This tumour may arise in the deep lobe of parotid gland and extend into the parapharyngeal space or may arise *de novo* from the aberrant minor salivary glands in parapharyngeal space. The latter entity is an extremely rare finding. CT scan and fine-needle aspiration cytology form the important diagnostic tools in case of these tumours. Surgery is the mainstay for the treatment of these tumours. The strategic location and also the extension of these tumours may at times demand to alter the surgical procedure for their excision. This article presents a case of a 27-year-old female, who presented with a mass in the right submandibular region that was excised successfully using transcervical approach in conjunction with transoral approach without mandibulotomy. The biopsy report suggested it to be "pleomorphic adenoma of minor salivary gland".

**Keywords:** Minor salivary gland tumour, Parapharyngeal space tumour, Transcervical approach

## CASE REPORT

A 27-year-old female reported to the Department of Oral & Maxillofacial Surgery with a chief complaint of difficulty in swallowing since 15 days. She was apparently alright seven months before then, she noticed a small swelling in her neck on right side that increased in size gradually. She underwent Fine-Needle Aspiration Cytology (FNAC) one month after the appearance of the swelling. She then, visited to an ENT surgeon almost five months after the appearance of the swelling where she was given only medical treatment and was advised for Computed Tomography (CT) scan. The patient then reported to us complaining about the difficulty in swallowing. The patient neither had any significant medical or dental history nor any deleterious habit. On clinical examination, a large swelling was present in the right submandibular region that was oval, measuring approximately about 8cm X 5cm in size and was firm and painless [Table/Fig-1]. The overlying skin appeared normal and was freely movable. The swelling was neither adherent to the skin nor to the underlying structures and moved on deglutition. None of the cervical lymph nodes were palpable. The patient did not have fever or sore throat. On intraoral examination, the uvula was pushed towards the left side with bulging of the soft palate with a smooth overlying mucosa. The right tonsil and the airway were pushed towards the midline of the oropharynx [Table/Fig-2]. The clinical examination did not reveal involvement of any cranial nerve.



[Table/Fig-1]: Extraoral view showing a swelling on Right Submandibular region.

[Table/Fig-2]: Intraoral view showing the displacement of soft palate and airway on left side.

On CT scan a large well defined mass in the right parapharyngeal region measuring about 6.5cm X 5.5cm X 4.2cm in size was seen [Table/Fig-3]. CT scan was suggestive of neurogenic tumour in the right parapharyngeal space. FNAC suggested it to be a "pleomorphic adenoma". It was decided to excise this tumour via a transcervical approach under general anaesthesia.

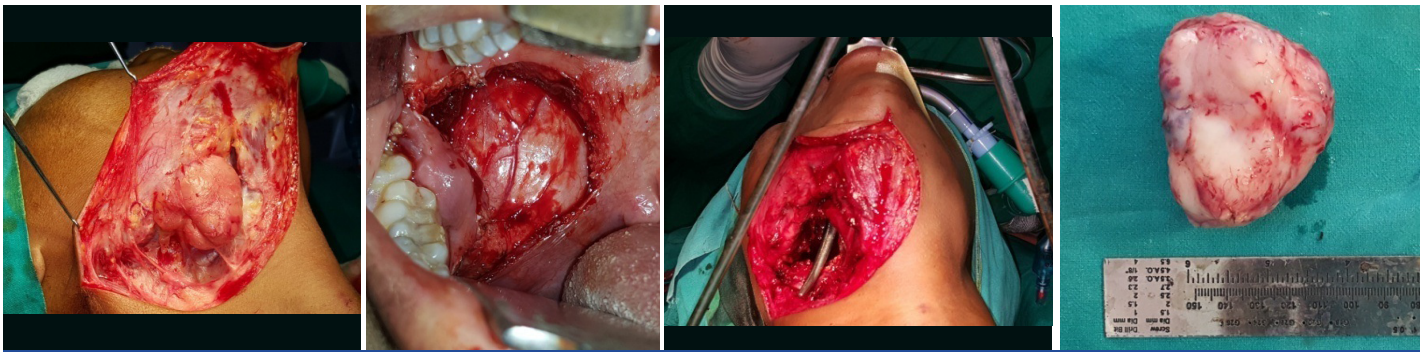
The incision was made two fingers breadth below the inferior border of right mandible in the skin crease extending from the right angle of mandible to the mental protuberance [Table/Fig-4]. After reflecting the flap, the mass was identified and was carefully separated from the adjacent vital structures. The facial artery and vein were ligated. The right submandibular gland and lymph node were excised. The marginal mandibular nerve was identified and reflected superiorly. The stylohyoid muscle and mylohyoid muscle were transected. To avoid the mandibulotomy procedure, an intraoral incision was made over the right tonsillar region to expose the tumour intraorally [Table/Fig-5]. After exposure of the tumour intraorally, it was carefully separated from the underlying structures to deliver it extraorally [Table/Fig-6]. Haemostasis was achieved and closure was done layer-wise. The surgical specimen was cut to expose the internal structure [Table/Fig-7&8]. The final diagnosis "pleomorphic adenoma of minor salivary gland" was confirmed on histopathological examination [Table/Fig-9-11].

## DISCUSSION

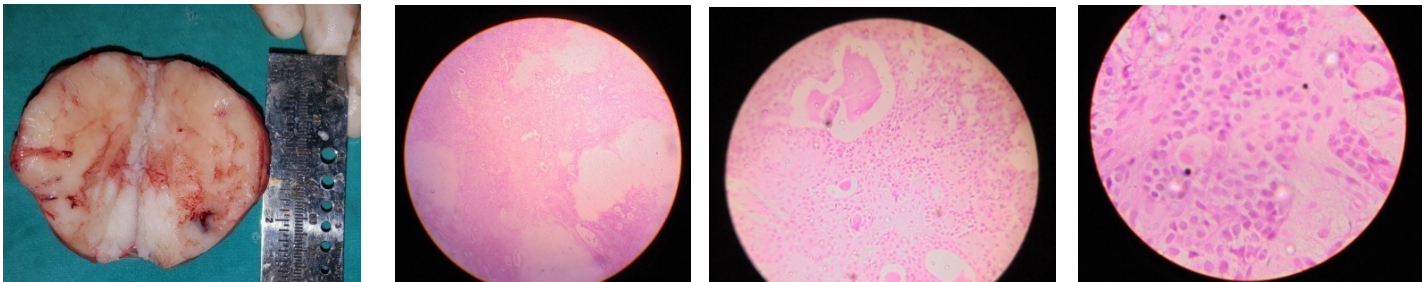
Parapharyngeal Space (PPS) tumours are infrequent that account for about 0.5% of all the head and neck neoplasm. About 70-80% of them are benign and others are malignant in nature [1-3]. About 40-50% of these tumours arise in the salivary glands [4,5].



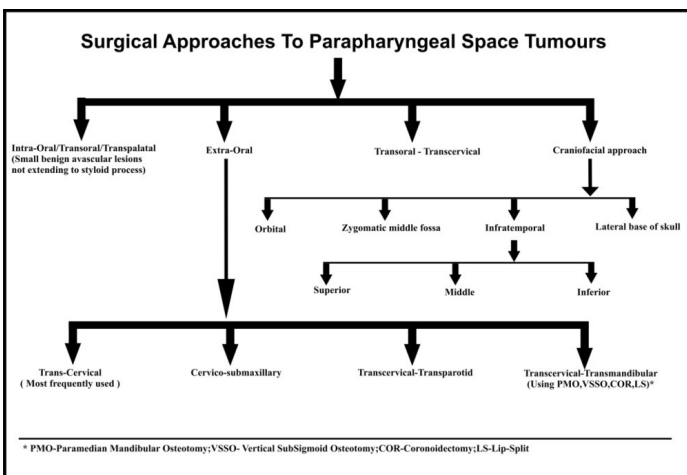
[Table/Fig-3]: CT Scan showing a well-defined mass in the right parapharyngeal space suggesting a tumour of benign origin.



**[Table/Fig-4]:** Surgically exposed lesion through transcervical approach. **[Table/Fig-5]:** Transoral incision exposing the tumour intraorally. **[Table/Fig-6]:** Transpharyngeal tunnel after the delivery of the tumour. **[Table/Fig-7]:** Excised tumour.



**[Table/Fig-8]:** Cut section of the tumour showing a rubbery glistening surface with a small area of infarction. **[Table/Fig-9]:** Photomicrograph showing epithelial components that form ducts seen in a chondromyxoid stroma. (H&E X10). **[Table/Fig-10]:** Photomicrograph showing epithelial components in a cystic arrangement around mucoid stroma. (H&E X40). **[Table/Fig-11]:** Photomicrograph showing interspersed islands of myo-epithelial and ductal epithelial cells showing nuclear hyperchromatism and amorphous myxoid stroma. (H&E X100).



**[Table/Fig-12]:** Surgical approaches for parapharyngeal space tumours advocated in the literature- An Overview.

The most common tumour found in the PPS is pleomorphic adenoma (61-80%) followed by Schwannoma and Paraganglioma [6]. Pleomorphic adenoma may arise from parotid (deep lobe) and enter into PPS or can also arise from aberrant salivary gland in PPS [7]. In a study conducted by Zhi K et al., the most common tumour found in the PPS was salivary gland tumour (45.68%), of which pleomorphic adenoma accounted for about 35.19% [8]. Carrau RL et al., reported that neurogenic tumours were most common in the PPS accounting for about 57.41% [9]. The study of Papadogeorgakis N. et al., reported the occurrence of pleomorphic adenoma in the PPS to be about 62% [4], Basaran B. et al., reported it to be about 29.5% [1], while Vanessa Suarez-Fente et al., reported about 39% of its occurrence [10].

Pleomorphic adenoma of minor salivary gland arising *de novo* in the PPS is extremely rare that presents as a neck or a pharyngeal mass. The symptoms of this lesion include dysphagia, dysarthria, hoarseness or it may be asymptomatic [4,5,11]. In our case, the patient presented with a neck mass along with dysphagia and hoarseness. The diagnosis of PPS tumours is difficult clinically and therefore appropriate imaging is essential for making a provisional diagnosis. The most common helpful imaging modalities include CT scan and MRI. They not only provide information about the exact

location, size and relations of the tumour with great vessels of the neck but also depict the benign or malignant nature of the tumour. If a high vascularity of the tumour is shown on CT scan or MRI, an angiography must be performed that will tell about the relation of the tumour to the great vessels in the neck and also its blood supply. A magnetic resonance angiography may be performed additionally for supportive evidence in case of vascular tumours [4]. In our case the CT scan revealed a large well defined mass in the right parapharyngeal space with intact fat planes around this mass. CT scan suggested it to be a neurogenic tumour.

FNAC forms an important diagnostic tool in the PPS tumours. Using FNAC, one can know about the nature of the tumour (benign/malignant) as well as about its origin. Papadogeorgakis N. et al., reports 100% accuracy of FNAC in detecting the nature of the tumour [4] while F Bozza et al., reports its accuracy to be about 90-95% [12]. FNAC has been found to be reliable in the diagnosis of pleomorphic adenoma [13]. However, its application in the PPS tumours is much controversial. This is due to the localization of these lesions and their relation with the surrounding vessels and nerves that can be damaged by this blind procedure. According to Sergi B et al., FNAC is a reliable procedure in the PPS tumours because it can guide the surgeon to choose the right surgical procedure [14], although it cannot be used as a first diagnostic tool, it should be performed after the diagnostic imaging so as to exclude a vascular lesion. It can be performed intraorally or percutaneously. Incisional biopsies should be avoided, because any damage to the tumour capsule increases the risk of recurrence [8] and more importantly to avoid the risk of fatal complications such as that of a carotid body tumour rupture [15]. In our patient the FNAC yielded a whitish granular material which on staining with PAP stain, showed myoepithelial cells, round plasmacytoid ductal cells with round nucleus and eosinophilic cytoplasm arranged in follicular pattern, clusters and sheets with a background of abundant chondromyxoid material and RBCs suggesting it to be a “pleomorphic adenoma”

The mainstay for the treatment of pleomorphic adenoma in the PPS is surgical excision [5,7]. These tumours are well encapsulated, however there may be microscopic pseudopod-like extensions of these tumours extending into the surrounding tissues due to

dehiscence in the false capsule and hence they should be excised including adequate margin of normal surrounding tissue to prevent the local recurrence [5]. There are various surgical approaches advocated in the literature for the excision of PPS tumours [Table/ Fig-12] including pleomorphic adenoma [4,8,13]. The intraoral approach, also known as transoral or transpalatal approach is useful when a small benign avascular tumour is presented in oropharynx that does not extend to styloid process. However, this approach does not give satisfactory control over the great vessels and cranial nerves in the neck and severe complications may occur. Also, if there is a massive bleeding during intraoperative procedure, achieving haemostasis becomes very difficult due to limited access. The limited access also increases risk of nerve damage, thus increasing the postoperative morbidity. Also, the limited access can cause rupture of the tumour capsule and tumour seeding during surgery thus increasing the risk of recurrence [4]. M. Iseri et al., has recently described endoscopic-assisted transoral approach to the parapharyngeal space tumours that gives an advantage of less operative trauma by providing increased operative exposure [11].

The transcervical approach is the most frequently used approach for the excision of PPS tumours [1,4]. This approach was first described by Morfit in 1955 [1]. Using this approach, large tumours measuring upto 11cm can be excised successfully [1,8]. This approach provides a direct access to the PPS and gives adequate control over the neurovascular bundle in the neck. Vanessa Suarez-Fente et al., has described this approach as an adequate and sufficient approach for complete complication-free excision of most parapharyngeal tumours [10]. In this approach, a transverse, curvilinear incision is placed in the natural skin crease, two fingers breadth below and behind the ramus and the angle of the mandible at the level of hyoid bone. This incision is so designed as to permit ease in conversion to a more extensive operation that may be necessary in cases like malignancy necessitating lymphadenectomy. Subplatysmal flaps are elevated, preserving the submandibular gland and marginal mandibular nerve that are reflected superiorly. The access is improved by the division of the stylohyoid ligament. The submandibular gland may be excised if necessary. The dissection can then be easily accomplished under direct vision, that offers excellent control over the common carotid artery, internal jugular vein and IX, X and XI cranial nerves [8]. In cases of larger tumours, surgical access can be improved by performing mandibulotomy after splitting the lower lip [1]. Using the combination of transcervical along with transoral approach we were able to excise the tumour in toto without causing damage to any of the vital structures and avoiding the need for mandibulotomy.

The larger tumours arising from the deep parotid lobe require partial parotidectomy. The marginal mandibular nerve should be preserved during this procedure [1]. The cervico-submaxillary approach, where the submandibular gland is either excised or reflected superiorly provides better access to the antero-inferior

aspect of PPS, offers same benefit as cervical approach [13]. Cranio-facial approaches are reserved for the tumours that extend into the infratemporal fossa, the base of skull, temporal bone or nasopharynx. They include orbital, zygomatico-middle fossa, lateral base of skull and infratemporal approaches [4].

## CONCLUSION

Parapharyngeal tumours are rare and their treatment should begin with a careful preoperative diagnostic procedure. The advantage of CT scan or MRI should be taken before going for FNAC. Whenever possible, FNAC should be performed prior to the surgical treatment planning because this provides guidance for choosing a proper surgical approach. The goal of the treatment should be to excise the tumour in toto or completely. The surgical approach chosen should be safe with least complications, aesthetic and functional damage and risk of recurrence.

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### PARTICULARS OF CONTRIBUTORS:

1. Professor, Department of Oral and Maxillofacial Surgery, MIDSR Dental College, Latur Maharashtra, India.
2. Senior Resident, Department of Oral and Maxillofacial Surgery, MIDSR Dental College, Latur Maharashtra, India.
3. Professor, Department of Prosthodontics, MIDSR Dental College, Latur Maharashtra, India.
4. Professor and Hod, Department of Oral and Maxillofacial Surgery, MIDSR Dental College, Latur Maharashtra, India.

### NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Rahul Laturiya,  
Professor, Department of Oral and Maxillofacial Surgery, Midsr Dental College,  
Latur Maharashtra-- 413531, India.  
E-mail: jendijcdr@gmail.com

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