

# Juvenile pleomorphic adenoma of masticator space: The first case report

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## Abstract

Pleomorphic adenoma (PA), also called benign mixed tumor, is the most common tumor of the salivary glands. About 90% of these tumors occur in the parotid gland and 10% in the minor salivary glands. Juvenile PAs are uncommon and about 5-10% of minor salivary gland PA affects patients aged 20 years and under. The most common sites of PA of the minor salivary glands are the palate followed by lips and cheek. Other rare reported sites include the throat, floor of the mouth, tongue, tonsil, pharynx, retromolar area and nasal cavity. The masticator space is a deep facial space with a complex anatomical structure where PA is not known to occur. Here, we report an unusual case of PA of left masticator space in a 16-year-old girl patient, which to the best of our knowledge is the first reported case in English language literature.

**Keywords:** Juvenile, masticator space, minor salivary gland, pleomorphic adenoma

## Introduction

The masticator space is an anatomical and functional entity centered on the mandibular ramus dividing it into medial and lateral compartments.<sup>[1]</sup> It is separated from the adjacent face and neck fascia by a superficial layer of deep cervical fascia except superiorly, where it freely communicates with the external temporal fossa, which is considered as an upper extension of masticator space. Adjacent to the masticator space are the buccal space anteriorly, parotid space posteriorly, the parapharyngeal space medially, the submandibular and sublingual space inferiorly and the base of skull superiorly. The masticator space contains four muscles of mastication (masseter, medial and lateral pterygoid and temporalis), the ramus and posterior portion of the body of mandible, the maxillary vessels and the mandibular and maxillary branches of the trigeminal nerve.<sup>[2]</sup>

Pleomorphic adenomas (PA) are the most common benign tumors of major salivary glands representing about 3% of

neoplasms of head and neck region.<sup>[3]</sup> Neoplasms of salivary glands are mainly adult disease, being rarely reported in children. Less than 5% of all juvenile head and neck tumors have their origin in salivary glands. The incidence of juvenile salivary gland tumors in minor salivary gland is even rarer.<sup>[4]</sup> PAs arising from ectopic salivary gland tissues may have an array of unusual locations, e.g. along the cheek, along the Stenson's duct, in accessory parotid tissue, that is separated from the body of the gland, in the floor of mouth, retromolar region, throat, alveolar mucosa, in parapharyngeal and infratemporal spaces. However, PA occurring in the masticator space is unusual and yet to be reported. To the best of our knowledge, this is the first report in the English language literature of a juvenile PA presenting in masticator space.

## Case Report

A 16-year-old female patient reported to our department with the complaint of swelling in the left cheek region since 6 months. The swelling was nonpainful and was not associated with any toothache or discharge or any secondary changes [Figure 1]. Intraoral examination revealed a small swelling causing left buccal vestibular obliteration, extending from distal side of second premolar to maxillary tuberosity [Figure 2]. Computed tomography (CT) showed a hyperdense soft-tissue mass in the left masseteric space without involvement of any hard tissue. Magnetic resonance imaging (MRI) revealed a well-defined smooth capsular outlined left masticator space lesion [Figure 3]. The lesion was located lateral to left pterygomaxillary fissure and abutting lateral margin of alveolar arch of maxilla. Posterior margin was close to condylar process of mandible. No bony remodeling or erosion was seen. Medially lesion was displacing and compressing masseter. It has also resulted in mild anterior bowing of maxillary antral floor. Patient was sent to oral surgery department for incisional biopsy. Histopathology of incisional biopsy specimen revealed epithelial component consists of epithelial and myoepithelial cells with divergent growth patterns, including trabecular, tubular, solid, cystic and papillary

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architecture in a connective tissue background of myxoid in nature. A diagnosis of PA was given and excision of the whole mass was planned out. The case was taken under pre-anesthetic check-up. General anesthesia was given with nasotracheal tube using halothane and propofol. The incision was placed intraorally in the left vestibular region and was extended posteromedially using sharp and blunt dissection. The complete tumor was visualized and the surface of the tumor was firm in consistency. The complete tumor along with capsule was excised [Figures 4 and 5] and surgical site was irrigated with 10% betadine mixed with normal saline in 1:1 ratio. Closure was done in layers keeping in mind the opening of stenson's duct. The excised tumor mass was sent for histopathological evaluation. On gross examination, the tumor mass measured  $38 \times 32 \times 36 \text{ mm}^3$  in size, with a whitish, faintly lobulated and focally glistening cut surface. The histopathological findings of the excised specimen were consistent with the incisional biopsy report [Figures 6-8], confirming the diagnosis of PA. Post-operative healing was uneventful with maintenance of normal vestibular depth [Figure 9].

## Discussion

PA or benign mixed tumor is the most common tumor affecting major salivary gland. Approximately 90% of PAs occur in the major salivary glands and 6% in the minor salivary glands.<sup>[5]</sup> The most common site of PA of a minor salivary gland is palate followed lip, buccal mucosa, floor of mouth, tongue, tonsil, pharynx, retromolar area and nasal cavity.<sup>[6]</sup> Even though, PAs are more prevalent in the 4<sup>th</sup> and 6<sup>th</sup> decade of life, cases of PAs occurring in the 1<sup>st</sup> two decades have been reported. Case series of juvenile PAs have been presented by Krolls *et al.*, Byars *et al.*, Yamamoto *et al.*, Fonseca *et al.*, Jorge *et al.* but none of them had occurred in masticator space.<sup>[7]</sup> Hence, it is essential to point out that this case report of PA arising from masticator space in a 16 year old juvenile is the 1<sup>st</sup> of its kind to be reported in English Language literature.

Due to their unique anatomic location the tumors of masticator space remain silent for very long time, until they attain a very large size or until they hinder any function. The frequent clinical presentation of tumors of masticator space has been reported in superior gingivobuccal sulcus.<sup>[8]</sup> Similarly, our patient also presented a swelling in the left cheek region, with obliteration of left maxillary buccal vestibular region, which only became more prominent while opening the mouth. Differential diagnoses in these cases include neural and vascular benign lesions, e.g. schwannoma, neurofibroma, hemangioma, lymphangioma along with other entities such as lipoma, leiomyoma, rhabdomyoma, rhabdomyosarcoma and chondrosarcoma.<sup>[2]</sup> Masticator space is difficult to explore only by means of clinical examination; imaging techniques, such as MRI and CT, are, therefore, essential in order to correctly evaluate this region.<sup>[9]</sup> CT scan and MRI were proved as significant diagnostic tool in our case



**Figure 1:** Extraoral view showing mild facial asymmetry on the left cheek region (black arrow)



**Figure 2:** Intraoral view showing slight obliteration of the left buccal vestibular region (black arrow)



**Figure 3:** Magnetic resonance imaging showing a well-defined smooth capsular outlined lesion in left masticator space

determining the extent and boundaries of lesion along with local spread and bony erosion of adjacent tissue.

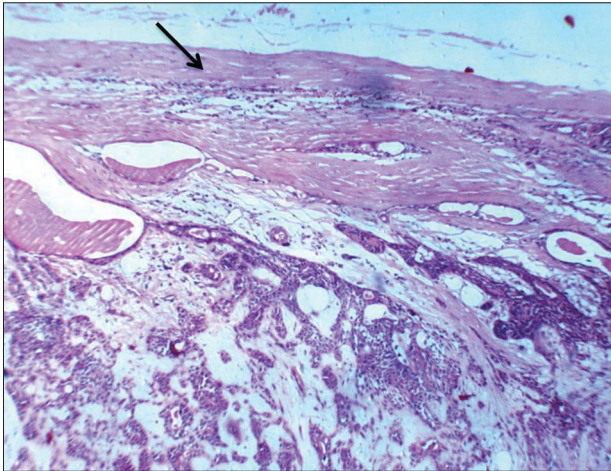




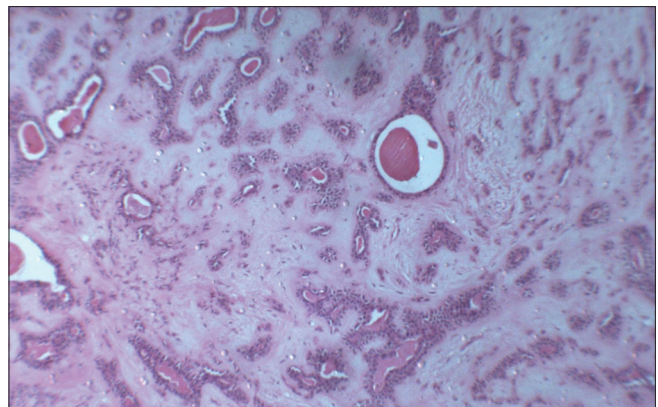
**Figure 4:** Intra-operative photograph showing encapsulated tumor mass dissected out from the masticator space



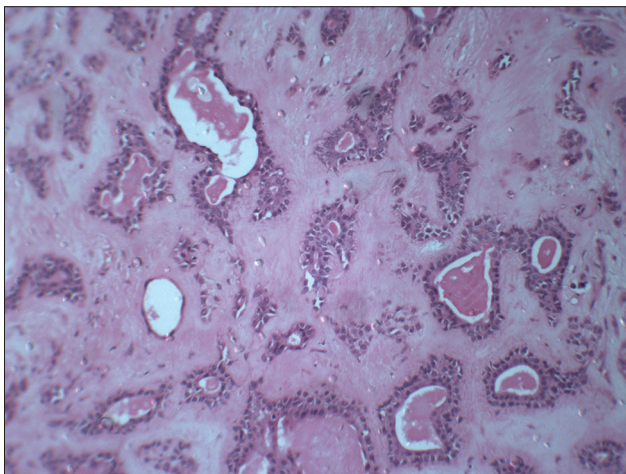
**Figure 5:** Excised surgical specimen



**Figure 6:** H and E, section showing encapsulation (black arrow), duct like structures and myxomatous stroma (x10)



**Figure 7:** H and E, section showing numerous duct like structures with eosinophilic coagulum and epithelial, myoepithelial cells arranged in tubular and ductular patterns against a myxomatous stroma (x20)



**Figure 8:** H and E, section showing hyalinization surrounding duct like structures, lined by double layered cuboidal epithelium (x40)



**Figure 9:** Post-operative intraoral photograph showing maintenance of normal vestibular depth along with healed site from where excisional biopsy had been taken, which is marked by hyperkeratotic line (black arrow)

The term “pleomorphic” refers to both histogenesis and histology of the tumor. PA is a benign salivary gland tumor

with wide cytomorphic architectural diversity. The tumor has three components: an epithelial cell component;

myoepithelial cell component; and a stromal (mesenchymal) component. The identification of these three components, which may vary quantitatively from one tumor to another, is essential to the recognition of PA. Fine-needle aspiration biopsy can be done for diagnosis of PA.<sup>[10]</sup> Histologically PA presents with variable pattern of epithelium in a loosely fibrous stroma, which may be myxoid, chondroid or mucoid. The epithelium is usually arranged in sheets or strands and ductal structure, often bilayered, are atypical. The myoepithelial cells are often polygonal with a pale eosinophilic cytoplasm. These cells are as typical as to almost diagnostic and their presence in small biopsies is helpful. In most instances, the diagnosis of PA is a straightforward microscopic identification. However, immunohistochemistry may be supportive and helpful in delineating the different cell types and components, as well as in differentiating PA from other tumors. The following immunohistochemical stains have proven to be helpful; Keratin-positive in luminal epithelial and abluminal basal/myoepithelial cells, Cam 5.2 and epithelial membrane antigen positive in luminal epithelial cells, P-63-positive in abluminal basal and myoepithelial cells, calponin, maspin, S-100 - positive in myoepithelial cells.<sup>[11]</sup>

Regarding the origin of PA in the typical location as in our case, it can be suggested by the presence of heterotrophic salivary gland tissue in masticator space. Ferlito *et al.*, in his study, have suggested various locations for ectopic salivary gland tissues such as pituitary glands, external auditory meatus, nasal fossa, sterno-clavicular joint, mandible and cervical soft tissue.<sup>[12]</sup> Even though, there exists an anatomic proximity between deep lobe of parotid and masticator space, but the probability of deep lobe PA spreading to masticator space can be ruled out in our case through revelation done by CT scan images, which suggested tumor arising *denovo* in masticator space region by showing a fine lucent line representing compressed layer of fibroadipose tissue between tumor and deep lobe of parotid. In our case, a modified approach was made with vestibular incision without any osteotomy and taking the whole capsulated tumor in toto.

The prognosis of PA is good and the chances of recurrences are rare in PA arising from minor salivary glands (3.4% recurrence in 5 years).<sup>[13]</sup> Long-term follow-up is mandatory in younger patients to ensure disease free period. However in our case due to its location in masticator space, only periodic clinical follow-up is insufficient and it should be supported by radiological and CT scans evaluation to detect

any recurrences. In our case, patient was disease free after surgical excision and was under regular follow-up.

## Conclusion

We are reporting the first case of juvenile PA presenting in the masticator space.

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