

## Original Research Article

# Pleomorphic adenoma of head and neck region: our institutional review

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### ABSTRACT

**Background:** Pleomorphic adenoma is the commonest benign salivary gland neoplasm but it has the tendency to reoccur if not properly removed and has ability to undergo malignant transformation. The aim of the study was to discuss the presentation and treatment of pleomorphic adenoma of the head and neck region.

**Methods:** Retrospective chart review of patients treated for pleomorphic adenoma of the head and neck region between 2011 and 2018. Data assessed included demographics, clinical presentation, imaging, treatment, complications, recurrence and follow-up.

**Results:** Seventy nine patients with pleomorphic adenoma were identified. Major salivary gland lesions were most common (n=64, 81.0%); 67% (53 of 64) arising in the parotid and 14% (11 of 64) in the submandibular glands. Minor salivary gland lesions were removed from the palate (8 of 15, 53.3%), parapharyngeal space (3 of 15, 20%), nasal septum (2 of 15, 13.3%), lower lip (1 of 15, 6.7%) and base of tongue (1 of 15, 6.7%). Preoperative imaging done was mainly computed tomography in all the cases however in parapharyngeal space cases magnetic resonance imaging was also done. Surgical excision was performed in all patients except base of tongue case.

**Conclusions:** Treatment of pleomorphic adenoma is mainly surgical with negative margin. Rate of recurrence is low, however long follow-up is recommended.

**Keywords:** Pleomorphic adenoma, Parotid gland, Submandibular gland, Minor salivary gland

### INTRODUCTION

Pleomorphic adenomas are slow growing mixed tumors accounting for 3-10% of all the neoplasm of the head and neck region.<sup>1</sup> They are formed as a result of interplay between monoclonal in origin based proliferation of mesenchymal and epithelial elements interspersed in stromal component (mixed tumor). The stroma of pleomorphic adenoma is myxoid in character making it extremely fragile.<sup>2</sup>

Pleomorphic adenomas have slight female preponderance (1.4: 1) with mean age of presentation between 43-46 years.<sup>3,4</sup> Pleomorphic adenomas account for the most common tumors of the salivary glands (50%).<sup>5</sup>

Salivary tissue is present in the major salivary glands (parotid, submandibular, and sublingual), minor salivary glands (scattered within oral cavity and upper airway) and accessory salivary glands (periparotid area). Sometimes, normal salivary tissue may occur at aberrant site termed as heterotopic salivary gland tissue (HSGT). They arise due to abnormal persistence of epithelial remnants in the branchial apparatus or faulty embryological migration. These heterotopias can be seen in tissues as diverse as the cerebellopontine angle, temporal bone, nasal cavity, lymph nodes, mandible, thyroid and parathyroid tissue, tonsils, tongue, along the anterior border of sternocleidomastoid in the upper neck and lower neck resulting in pleomorphic adenoma of ectopic salivary gland tissue.<sup>6,7</sup>

Pleomorphic adenoma of the parotid gland typically appears as slow growing, painless rounded mass with bosselated borders, usually located in the superficial lobe above the plane of facial nerve near the angle of mandible.<sup>8</sup> Occasionally it may involve deep lobe involving pre styloid area and may extend into the parapharyngeal space widening the stylomandibular tunnel. Tumors located in parapharyngeal space may present as swelling in the retromandibular area of upper neck, as a smooth bulge in the soft palate or causing medial displacement of the tonsil in lateral pharyngeal space extending from skull base to hyoid bone. Otagia, trismus or neuralgias may be seen.<sup>9,10</sup> In the cheek, a firm, bimanually palpable mobile swelling without involvement of facial tissues may be present.<sup>11,12</sup> Pleomorphic adenoma arising from minor salivary gland of the lip tends to appear as sessile masses which may ulcerate and cause difficulty in speaking and those in palate may cause mechanical obstruction of the airway.<sup>13</sup> Pain, facial weakness or palsy is uncommon and usually heralds malignant change. Cervical heterotopias are uncommon, but usually presents as painless, discharging masses or sinus openings in neck.

All salivary gland lesions should undergo pre-operative Fine needle aspiration cytology assessment for a pre-operative diagnosis without the risk associated with open biopsy of seeding tumor cells in the incision. Histological features will show variegated pattern due to ductal epithelial cells, myoepithelial cells with eosinophilic colloid.<sup>14</sup> It is usually surrounded by a thick fibrous capsule and subcapsular clefts which may be locally infiltrated. Detailed imaging yields adequate surgical information to allow safe removal. This includes high resolution ultrasonography, computed tomography (CT), magnetic resonance imaging (MRI).

Ultrasound typically shows a hypoechoic lesion, well defined contour with or without posterior acoustic enhancement. It differentiates diffuse from local disease, solid from cystic mass, assess vascularity of the tumor and determine nodal staging. It is a preferred first modality in pregnant females and children.<sup>15</sup>

MRI is the method of choice in patients with palpable mass. It can be T1 or T2 weighted sequences, dynamic contrast enhanced MRI (DC-MRI), diffusion weighted MRI (DW-MRI) or proton MR spectroscopy (MRS). High probability criteria of MRI for pleomorphic adenoma include bright T2 signals, dark T2 rim, nodular enhancement and sharp margins. MRI addresses to perineural invasion, bone spread along with localization and extent of the disease.<sup>16</sup>

CT can be used to assess the extension and relationships to nearby structures in cases with contradiction for MRI or associated inflammatory disease. Ideally, both pre and post contrast CT imaging must be performed. For small tumors, they show homogenous attenuation with

prominent enhancement. Foci of necrosis and regions of calcifications may be present for large tumors.<sup>17</sup>

Although, pleomorphic adenoma is benign tumors with good prognosis, the treatment of choice is surgical excision with free margins to prevent recurrence or malignant transformation. Removal of tumor along with superficial lobe of parotid gland is adequate for major cases. In case of deep lobe, total conservative (leaving facial nerve intact), total parotidectomy should be done.<sup>18</sup> Pleomorphic adenomas in parapharyngeal space due to its complex anatomical region are traditionally approached by trans-cervical and parotid approaches as they provide wide intra-operative visibility and minimal functional after-effects. Rarely, trans-oral approach is used. Tumor spillage, capsular tears, dehiscences, female gender is critical for recurrences.

The objectives of the study are to evaluate the frequency of distribution of pleomorphic adenoma in head and neck region in our hospital and to study their surgical outcome, its complication and recurrence.

## METHODS

### *Study design*

Retrospective study design was followed.

### *Study place*

The study was done in department of otorhinolaryngology and head and neck surgery at GGS Medical College Faridkot.

### *Study period*

The study was conducted retrospectively from 1st January 2011 to 31st December 2018.

### *Inclusion criteria*

Histological confirmed cases of pleomorphic adenomas of head and neck region were included in our study.

### *Exclusion criteria*

All other variant of benign and malignant lesion and patients with previous surgical history due to pleomorphic adenoma were excluded.

The retrospective data analysis after obtaining ethical clearance was carried out in the Department of Otorhinolaryngology at our institute from 1st January 2011 to 31st December 2018. Patients presented with longstanding swelling in the head and neck region which was proven as pleomorphic adenoma by histopathological examination were included in the study.

A total of 79 patients selected for study were further statistically analyzed for demographics, clinical presentation, imaging i.e., ultrasonography or CT over the region of the swelling, anatomical location, surgical treatment, complications, recurrence, and follow-up.

### Procedures

All patients underwent surgical excision under general anesthesia. Patients with pleomorphic adenoma in the parotid gland underwent superficial parotidectomy or total conservative parotidectomy through standard parotidectomy approach. In case of submandibular gland, standard submandibular approach and for parapharyngeal caes standard transcervical approach was followed. The cases of pleomorphic adenoma on palate, nose and lip were undergone for wide local excision, whereas case of base of tongue underwent radiotherapy due to inoperability.

The collected data was analysed using Microsoft Excel and presented in number and percentages.

### RESULTS

The seventy nine patients who underwent surgical excision and HPE proven as pleomorphic adenoma were included in the study. There were 45 females and 34 males with the ratio being 1.3: 1. The age of the patients ranged from 11 to 70 years.

The most number of cases were in the fourth decade followed by fifth decade (Table 1). Most common presentation of pleomorphic adenoma regardless of the site is painless, slow growing, firm swelling in all patients.

Pleomorphic adenoma affected the parotid gland in 53 patients (67%), the submandibular gland in 11 patients (14%), the palate in 8 cases (10%), parapharyngeal space in 3 cases (4%), 2 cases in nasal septum (2.5%) and one each on lip and base of tongue (1.25%) (Table 2).

Amongst pleomorphic adenoma of parotid gland, the

swelling was noted in the infra-auricular region in 38 patients (72%) and pre-auricular region in 15 patients (28%). The duration of the symptom varied from 1 month to 16 years.

The intra-operative tumor was localized in the superficial lobe in 46 (86.8%) patients, deep lobe in 4 (7.5%) and both superficial and deep lobe in 3 (5.7%) patients. A total of 46 (86.8%) patients underwent superficial parotidectomy except cases in which tumor was in deep and both lobes that underwent total conservative parotidectomy i.e., 13.2%. All other cases of tumor also underwent surgical excision except tumor of base of tongue that was inoperable.

**Table 1: Age distribution of patients with pleomorphic adenoma (n=79).**

Age of patient (in years)	No. of patients	
	Male	Female
11-20	1	0
21-30	2	3
31-40	11	13
41-50	9	14
51-60	5	7
61-70	6	8
<b>Total</b>	34	45

**Table 2: Distribution of pleomorphic adenoma according to the site of involvement (n=79).**

Site of pleomorphic adenoma	No. of patients	%
<b>Major salivary gland</b>	<b>64</b>	<b>81</b>
Parotid gland	53	67
Submandibular gland	11	14
<b>Minor salivary gland</b>	<b>15</b>	<b>19</b>
Palate	8	10
Parapharyngeal space	3	4
Lip	1	1.25
Nasal septum	2	2.5
Base of tongue	1	1.25

**Table 3: Treatment modality of pleomorphic adenoma.**

Site of lesion	Presentation	Frequency	Treatment (%)
<b>Parotid gland</b>	Painless pre auricular swelling	46	Superficial parotidectomy (86.8)
	Infra auricular region swelling	7	Total conservative parotidectomy (13.2)
<b>Submandibular gland</b>	Swelling in submandibular region	11	Excision of gland (100)
<b>Palate</b>	Swelling on palate	8	Excision (100)
<b>Parapharyngeal space</b>	Intraoral swelling and bulge in submandibular region	3	Trans cervical excision (100)
<b>Lip and nasal septum</b>	Painless swelling	3	Excision (100)

Post operatively 11 (20.75%) patients who underwent parotidectomy have facial paresis out of which 5 (9.4%) recover fully and rest persists i.e., 11.3%, 3 (5.7%) have marginal mandibular paresis which recovers fully, 13 (24.5%) cases have paresthesia of ear lobule.

Post operatively no complication was seen in patients underwent surgical excision for other site of lesion. No reoccurrence was seen after two year of follow up.

## DISCUSSION

Pleomorphic adenoma occurs mostly between the ages of 31 to 50 years of age with female preponderance the ratio being varying from 1:1.4 to 1:1.7.<sup>19-21</sup> In our study also pleomorphic adenoma was most commonly seen in 4<sup>th</sup> and 5<sup>th</sup> decade of life with female to male ratio of 1.3:1.

The pleomorphic adenoma accounts for 63% from parotid gland, 11% from submandibular and only 6.5% from minor salivary glands from which palate is considered to be the most common site (42.8-68.8%), followed by lip (10%) and cheek (5.5%).<sup>22</sup> In our data, the parotid gland pleomorphic adenoma was seen in 67% followed by submandibular gland in 14% and from minor salivary gland 19% of which palate is most common site in 53.3% of cases.

Painless slow growing mass is the most common feature of pleomorphic adenoma. The firmness of the pleomorphic adenoma varies with the nature and the amount of the stromal component. So, it ranges from soft in the case of the more mucinous form to hard with extensive chondroid or collagenous component.<sup>23</sup> All patients in our study also presented with painless, slow growing swelling which was firm in consistency.

The best mode of management for pleomorphic adenomas in parotid and submandibular gland is wide excision with negative margins. The majority arises in the parotid gland and for these the preferred treatment is superficial parotidectomy or total conservative parotidectomy and in submandibular gland excision. Although wide margins are preferred, the majority of parotid pleomorphic adenomas about the main trunk or its branches so that a limited enucleation or capsular dissection is necessary in most cases.<sup>24</sup> In our setup also same treatment of method was adopted. Again for management of palatal pleomorphic adenomas is a wide excision with negative margins.<sup>25</sup> In our study palatal cases were managed by wide local excision with negative margins with no reoccurrence. The transcervical approach is the most widely used approach for the excision of parapharyngeal space tumours, using this approach, large tumours measuring upto 11 cm can be excised successfully and same technique was used for the excision of parapharyngeal space tumors in our study.<sup>26</sup>

Laccourreye et al reported on 229 patients who underwent total parotidectomy with 7th nerve preserving

the complications rate of hemorrhage, 1.7%; hematoma, 6.1%; seroma, 4.8%; and skin necrosis, 0.9%. Temporary 7th nerve paresis and paralysis occurred in 64.6% and 5.6% of patients, respectively, where as in our study seroma was seen in 6% and skin necrosis in 1.5% of patients. Temporary facial nerve paresis is seen in 20% of cases.<sup>20</sup>

## CONCLUSION

Pleomorphic adenoma is the most common benign salivary gland neoplasm. Recurrence and malignant transformation are concerns necessitating proper treatment. Regardless to the site of occurrence of Pleomorphic adenoma surgical treatment is the choice of treatment. The goal of surgery should be complete surgical excision with negative margins.

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