



Giant Pleomorphic Adenomas of Parotid Gland: a Case Series

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Abstract The parotid gland is the largest salivary gland in the body. Pleomorphic adenomas (PA) are most common benign tumors of parotid gland. If left untreated, they can gradually attain the size which can weigh several kilograms. This paper highlights a case series of 15 cases diagnosed as giant pleomorphic adenoma arising in the parotid gland along with their management. A hospital based, observational study of 15 patients of PA of the parotid gland, > 10 cm in its widest dimension considered as giant, done at tertiary care ENT hospital over period of 9 years. The age of the patients ranged from 30 to 81 years with mean age 50.33 ± 15.30 years. There were 5(33.33%) males and 10(66.67%) females. The time duration of having tumor ranged from 5 to 20 years with mean duration of 10.4 ± 4.17 years and the largest tumor was 25 cm in its largest diameter. Mean weight of tumors was 2.72 ± 0.52 kg. One out of fifteen cases was detected as malignant in nature. All cases were treated by surgical excision without any complications. Pleomorphic adenoma of parotid gland can assume a giant proportion when timely surgical intervention is not done. We can build up confidence and hope for life in patients after complete surgical excision of giant PA.

Keywords Parotid · Pleomorphic adenoma · Giant · Surgery

Introduction

Parotid gland neoplasms comprise 3% of all head and neck tumors and about 70% of all salivary gland tumors [1]. Approximately 85% are benign and out of these, 80% are pleomorphic adenomas (PA) [2]. The exact etiology is obscure but it has been suggested that prolonged exposure to radiation and the simian virus (SV40) may play a role in the development [3]. The adenomas are neglected and untreated so they get enlarged; it is the most common cause to become giant.

Most cases of giant PA were seen before the 1980's, but some cases have been published recently [4, 5]. In 1989, Schultz-Coulon reviewed 31 cases of giant pleomorphic adenomas of the parotid gland [6]. PA usually manifests as a slowly progressive asymptomatic, parotid gland swelling without facial nerve involvement [7]. It has a glandular origin in the head and neck region and usually manifests as a mobile, slowly progressive, asymptomatic firm swelling that does not cause ulceration of the overlying skin [8]. The tumor may weigh from several grams to more than 8 kilograms [9].

PA has been documented to occur in sizes ranging from 1 to 10 cm but can attain a grotesque proportion and weigh several kilograms or centimeters in size when timely surgical intervention is not sought. If untreated, this tumor can cause facial disfigurement and can impact negatively on patient's psychology [10]. The risk increases in tumors with long time of evolution, recurrences, advanced age of the patient and location in a major salivary gland [11].

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Ninety percent of all parotid neoplasms are found in the superficial lobe and lie lateral to the facial nerve; it is the relationship of the gland and facial nerve that has shaped the techniques of parotid surgery through the years [12].

The late presentation appears to be a major factor in the size of the PA presented. Oji [13] has observed that some of the factors affecting late presentation include ignorance and poverty. Olaitan et al. [14] also identified low socioeconomic status, distance to the hospital, and different myths related to fear of hypodermic needles and nasogastric tube used in the hospitals as other reasons for delayed presentation in patients with oro-facial tumors.

In this case series we reported 15 cases of giant pleomorphic adenomas with their management to shed light on the fact that size is not a criterion to get worried either by the patient for seeking treatment or by surgeons to remove it.

Materials and Methods

This was a hospital based study of some case files (our institution utilizes case files for the documentation of patient's records and follow up findings) and observation of some ongoing patients who presented to our institution, over a 9 years period (from January 2010 to December 2018) with diagnosis of Pleomorphic Adenoma.

The information assembled included the socio-demographics as well as clinical characteristics such as the anatomical site, side, presenting features, and onset of symptoms, duration and type of treatment carried out as well as treatment outcomes including collection of all the data of ongoing cases. PA of sizes > 10 cm in its largest diameter was considered to be "giant" (considering that most literature do not report lesions > 10 cm).

On clinical examination of PA of parotid gland those presented with macroscopic features consisting of an irregular to ovoid mass with well-defined borders and mostly incomplete fibrous capsule; also, microscopic features consisting of demonstrable combinations of glandular epithelium and mesenchyme-like tissue with varying proportion of each component in individual tumors was considered diagnostic. Specific investigations for tumors like Fine Needle Aspiration Cytology, Ultrasonography, Computerized Tomography scan and Magnetic Resonance Imaging done in all patients to assess the extent of mass and its vascularity. All the routine investigations also had done which were required for general anesthesia like complete blood count, blood sugar, coagulation profile, liver and kidney function tests, viral markers, electrocardiography and chest X-ray.

The tumours were excised under general anaesthesia. Despite the size of the mass, a clear plane of dissection was

found in all cases of giant pleomorphic adenoma. Facial nerve was avoided in all cases without difficulty. The postoperative course was uneventful. Subjects with missing or incomplete data were excluded and recurrent cases of PA were also excluded.

The data were entered into Statistical Package for Social Sciences version 15.0 (SPSS Inc., Chicago, IL, USA) and analyzed. Absolute numbers and simple percentages were used to describe categorical variables. Quantitative variables were described using measures of central tendency (mean, median) and measures of dispersion (range, standard deviation) as appropriate. No tests of significance were done.

Results

A total of 90 patients with a diagnosis of PA of parotid gland were seen within the period of study, only 18 were considered to be of significant size and met our inclusion criteria for the study. Three of the patients were excluded on account of missing or incomplete information. Thus, 15 cases of PA of the parotid gland met all the inclusion criteria. Prevalence of the giant PA of the parotid gland was found to be 16.7%. Among those affected by giant PA 5(33.33%) patients were males and 10 (66.67%) were females and their ages ranged from 30 to 81 years with a mean of 50.33 ± 15.30 years. The 9 patients were of right sided swelling and 6 on left sided. Only two patients presented with ulceration on the overlying skin. The sizes, duration, and weight of the lesions are shown in Table 1. All the patients were of rural population.

It was noticed that some patients have always dressed in a manner to obscure the tumor or developed minimal outdoor interactions because of their tumors due to poor socioeconomic status, ignorance and poverty.

All the patients had undergone surgery. Enucleation was done in one case, superficial parotidectomy done in 3 cases and total parotidectomy done in 11 cases. In one case neck dissection was also done with total parotidectomy and patient was advised for radiotherapy post operatively, after confirmation of histopathology examination that was malignant in nature. 14 out of the 15 patients had a postoperative histology of benign lesion whereas only one postoperative diagnosis of a malignant lesion in a parotid gland was made.

Precautions were taken to ensure the preservation of the facial nerve. No recurrence was observed throughout the follow-up period. The follow-up period was of minimum 6 months. Our patients were treated by surgical excision of the lesion while paying adequate attention to preserve facial nerve and achieve hemostasis as these tumors may

Table 1 Demographic, clinical and histologic characteristics of all patients with giant pleomorphic adenoma (n = 15)

Age (years)	Sex	Size (cms)	Duration (years)	Ulceration	Weight (kgs)	Histology
30	M	12*10*2	5	No	2.0	Benign
40	F	10*11*5	10	No	2.2	Benign
81	F	12*10*7	20	Yes	2.1	Benign
55	F	17*12*10	15	No	3.1	Benign
45	F	10*9*9	12	No	2.0	Benign
31	M	14*12*10	8	No	2.6	Benign
39	F	15*10*8	6	No	3.0	Benign
61	M	13*10*12	11	Yes	3.1	Benign
54	M	20*18*10	10	No	3.3	Benign
48	F	20*14*13	7	No	3.0	Benign
74	M	25*22*15	14	No	3.5	Benign
46	F	23*20*15	6	No	3.4	Benign
35	F	15*12*8	8	No	2.9	Benign
48	F	12*10*6	9	No	2.2	Benign
68	F	15*14*10	15	No	2.5	malignant

**Fig. 1** Clinical photograph of patient 1

have well-developed feeder vessels (Figs. 1, 2, 3, 4, 5, 6, 7, 8).

Study characteristics are shown in Table 2.

Discussion

World Health Organization (1972) defined PA as a well-defined tumor characterized by its pleomorphic or mixed appearance. In our study 5 patient were males (33.33%)

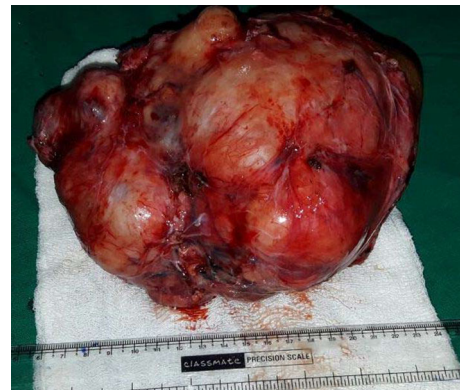
**Fig. 2** Radiological image of patient 1**Fig. 3** Tumour of patient 1



Fig. 4 Post-operative photograph of patient 1



Fig. 7 Tumour of patient 2



Fig. 5 clinical photograph of patient 2



Fig. 8 Post-operative photograph of patient 2



Fig. 6 Radiological image of patient 2

and 10 females (66.67%). This number is comparable with the following studies. In a review of 31 giant PAs occurring in the parotid gland over a period of 140 years by Schultz-Coulon in 1989 [6] most occurred in females (64.5%) and only 35.5% occurred in males which is consistent with the gender variation of other salivary gland tumour and particularly with that of PA.

Ages ranged from 30 to 81 years with mean of 50.33 ± 15.30 years. A mean age of 56.2 years was reported in a review of the 10 largest pleomorphic tumors before the 1980's in English literature [4, 6].

Table 2 Study characteristics

Study characters	Number (%)
<i>Gender</i>	
Male	5 (33.33%)
Female	10 (66.67%)
<i>Laterality</i>	
Right	9 (60%)
Left	6 (30%)
<i>Lobe involved</i>	
Superficial alone	4 (26.67%)
With Deep lobe	11 (73.33%)
<i>Type of surgery</i>	
Enucleation	1 (6.67%)
Superficial parotidectomy	3 (20%)
Total parotidectomy	11 (73.33%)
<i>Histopathological finding</i>	
Benign	14 (93.33%)
Malignant	1 (6.67%)
<i>Post-operative complications</i>	
Facial weakness	2 (13.33%) recovered within 6 months
Others	Nil

Laccourreye et al. [15] reported on 229 patients treated at the University of Paris V (Paris, France) and observed a 1:1.4 male-to-female ratio. It occurs most often between the ages of 30 and 60 years and is found more commonly in females than in males. O'Brien reported on 254 patients with parotid pleomorphic adenoma operated at the Royal Prince Alfred Hospital (Sydney, Australia) and observed a median age of 46 years and a 1:1.7 male-to-female ratio. Patients usually presented with a painless, slow-growing mass [16]. Male-to-female ratio in this study is 1:2 which is consistent with above studies.

In this study weight of the tumours ranged from 2 to 3.5 kg and the mean was 2.72 ± 0.52 kg. Takahama Jr et al. [17] in 2008 presented a tumour of weight 4 kg. Sajid et al. [18] in 2015 presented a tumour of weight 1.8 kg.

The largest tumor in the study was 25 cm in its largest diameter. The literature also documents PA to occur in sizes up to 10 cm in widest diameter [19] and PA of 16 cm, has also been reported [20]. Omeje et al. [10] reported PA of parotid gland of size 23 cm in its largest diameter.

In this study, time duration of having tumor ranged from 5 to 20 years with mean duration of 10.4 ± 4.17 years. The duration of symptoms is variable in the studies. Dawson and Orr [21] reported a mean duration of symptoms of 5 years (range 1 month–34 years) in a series of 311 patients treated at the Western General Hospital and Royal Infirmary (Edinburgh, Scotland). Although

progression is slowly, left untreated, the tumor can cause significant morbidity and, rarely, death [22, 23].

The surgical approach to parotid tumors has undergone considerable evolution. In the early part of the last century, fear of injuring the facial nerve bedeviled parotid surgeons, and inadequate operations, mainly enucleations, were widely practiced. Complete superficial parotidectomy soon became the operation of choice for benign parotid neoplasms, and this procedure was associated with a low recurrence rate and a reduced incidence of permanent facial paralysis. Enucleation, superficial or total parotidectomy with preservation of the facial nerve formed the mainstay of surgical treatment [24]. Parotid masses are excised more easily today due to the advances in technology and surgical techniques.

The results of which review of 363 operations performed for benign parotid tumors over a period of 14 years show that superficial parotidectomy is associated with low morbidity. Although it is necessary to expose the main trunk of the nerve in every case, only the branches immediately adjacent to the tumor require close facial dissection [16].

Facial nerve paralysis has a significant functional and emotional impact on patients. The most frequent complication is temporary facial nerve paresis after parotidectomy, and facial paralysis is observed to be the most important complication [25]. Facial nerve weakness is an infrequent sign in parotid tumors, although large neglected tumors may present with facial nerve weakness [17, 26]. A clear plane of dissection was found in all cases of giant pleomorphic adenoma and facial nerve was preserved meticulously.

Maahs GS et al. [27] showed an incidence of 15% of cases with postoperative facial paralysis and only 1.9% of cases with permanent paralysis and concluded that cases of permanent paralysis are rare, and are associated with severe malignant cases with prior involvement of adjacent structures. In this study, only two patients suffered with facial weakness post operatively which was recovered within 6 months.

The incidence of malignancy frequently shows a correlation between the length of the history of PA and the development of a carcinoma [28]. The incidence of malignancy is correlated with the duration of pleomorphic adenomas, the risk of developing malignancy is only about 1.5% for a duration of < 5 years but increases to 9.5% for a duration of > 15 years [29].

The classical clinical history of carcinoma ex-pleomorphic adenoma is of a slow-growing mass for many years, with a recent fast growth [30]. In this study only one case was malignant in nature, this may be due to small sample size and study in a limited area.

Key Messages

Early diagnosis and surgery of parotid pleomorphic adenomas is desirable. Untreated PAs can enlarge gradually up to several kilograms in weight. Neglecting even a benign parotid tumour carries an increased risk of facial nerve injury when surgery is performed for giant PA and risk of malignant transformation is also increased, thus an extensive histopathological assessment of tumor is mandatory after early surgical intervention to rule out malignant transformation in these giant pleomorphic adenoma cases.

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no competing interests.

Ethical Approval Ethical approval taken from institute ethics committee.

Informed Consent Written and informed consent taken from all participants.

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