

Deep Lobe Parotid Pleomorphic Adenoma: Ten Year Retrospective Review of Cases Treated at a United Kingdom Regional Tertiary Referral Center

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Citation

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Abstract

Objective: Retrospective review of patients with deep lobe parotid pleomorphic adenoma treated at a single regional tertiary referral center in the United Kingdom over a period of ten years (1994-2004).

Materials & Methods: A total of thirty patients were identified, including twenty-five new cases and five cases of recurrent disease that were previously treated elsewhere.

Results: Pre-auricular mass lesion was the commonest presentation (68%). All tumors were excised via an external surgical approach. Temporary facial nerve weakness was the main post-operative complication (24%). Adjuvant radiotherapy was employed in cases of macroscopic tumor spill. All new cases remain free of recurrence at a mean follow-up period of 43 months. Two of the five cases of recurrent disease relapsed following re-resection; one of these underwent malignant transformation.

Conclusion: Recurrence is rare following complete tumor excision at initial presentation. Recurrent tumors are often multifocal and difficult to excise completely, and hence more likely to relapse.

INTRODUCTION

Pleomorphic adenomas are slow growing, well-demarcated benign neoplasms that constitute more than 80% of benign parotid tumours.^{1,2} Modern surgery of the parotid gland has developed largely because of the gradual realization that although the parotid gland is a single anatomic entity, it can be cleaved into two parts in the plane of the facial nerve.³ Approximately, 80-85% of the glandular tissue lies lateral to the facial nerve, and more than 90% of parotid tumors arise in this "superficial lobe".^{1,4,5}

The "deep lobe" of the parotid gland constitutes a variable amount of parenchyma lying deep to the nerve. It can be thought of as having two arbitrary components: a small part deep to the facial nerve yet still lateral to the ramus of the mandible, and a second retromandibular portion extending posteriorly, occasionally with an extension medial to the ramus towards the parapharyngeal space.⁶ Approximately 10-12% of parotid tumors arise from the "deep lobe", with

only a small proportion (~1%) developing a significant parapharyngeal extension.^{1,5,6}

The anatomical relations of the deep lobe often make the presentation of deep lobe tumors quite atypical and confusing, and these rare tumors continue to be a diagnostic and therapeutic challenge.^{7,8}

This paper presents a retrospective review of a series of 'deep lobe' parotid pleomorphic adenoma cases encountered and treated at our institution over a period of ten years.

MATERIALS & METHODS

All cases of deep lobe parotid pleomorphic adenomas excised at our institution (a regional tertiary referral center) during a period of ten years from 1994 to 2004 were included in the review. The cases were identified via a meticulous search through histopathology records. Altogether we encountered records of one hundred and thirty-one pleomorphic adenomas of the parotid gland during

1994-2004. Thirty of these cases (22.9%) were found to be tumors originating from the deep lobe of the parotid (i.e. cases where the tumor was found to be lying deep to the facial nerve at the time of surgery).

A retrospective review of the clinical case notes of all thirty patients was undertaken. Information was gathered on patient demographics, clinical presentation, pre-operative work-up, the operative procedure, and post-operative follow-up and progress.

There were twenty-five new cases and five cases of recurrent disease that had been previously operated on elsewhere. It is important to differentiate between these two groups as the clinical presentations, associated morbidity and recurrence rates are likely to be different. Hence, for the purpose of this review the two groups have been considered separately.

RESULTS

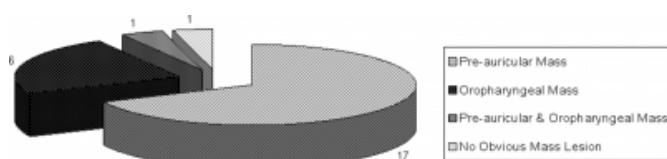
NEW CASES OF DEEP LOBE PAROTID PLEOMORPHIC ADENOMA

Twenty-five new cases of deep lobe parotid pleomorphic adenoma were identified. There was a slight female preponderance (56%), and the mean age at presentation was 47.5 years (range 28-76 years).

The duration of symptoms ranged from 1 month to 10 years, with a mean duration of 11 months. There were not any cases of pre-operative facial weakness. Twenty-four (96%) patients presented with a clinically evident mass, the majority of these (68%) being pre-auricular (see Figure 1).

Figure 1

Figure 1: Pie Chart Showing the Clinical Distribution of Deep Lobe Parotid Masses at Presentation



Three patients with oropharyngeal extension of the mass had additional symptoms of unilateral middle ear effusions and conductive hearing deficit. One of these patients also described symptoms of tongue numbness and paraesthesiae in the distribution of V1 and V2 branches of the trigeminal nerve.

One patient did not have a clinically evident mass lesion at presentation. This patient was investigated with an

ultrasound scan to exclude thyroid pathology and was found to have a 2cm 'swelling' of parotid origin, a subsequent MRI scan confirmed the lesion to be originating from the deep lobe of the parotid.

Fine needle aspiration cytology (FNAC) was performed in eighteen (72%) of the cases and confirmed pleomorphic adenoma in fourteen cases. The results were inconclusive in four patients.

Pre-operative radiological assessment was carried out in twenty cases (80%). Eighteen patients underwent magnetic resonance imaging (MRI) and two underwent computerized tomography (CT). Twelve (67%) of the eighteen cases undergoing MRI and one of the two cases undergoing CT evaluation, were correctly identified as having deep lobe tumors pre-operatively.

Five patients did not undergo any pre-operative radiological evaluation. All five had presented with pre-auricular parotid masses, and four of these had FNAC confirmation of pleomorphic adenoma prior to surgery. One patient did not undergo any pre-operative cytological or radiological investigations, the clinical findings, however, were strongly indicative of a pleomorphic adenoma of the parotid gland.

Two patients had undergone open biopsies of their oropharyngeal masses prior to referral to our institution, confirming the diagnosis of pleomorphic adenoma.

In all twenty-five cases the tumor was excised via an external approach and intra-operative facial nerve monitoring was used. Four cases required a mandibulotomy, and in one case the parapharyngeal mass was excised via an upper lateral cervical approach through the submandibular fossa. Fracture of the styloid apparatus was carried out in three cases. The tumor size ranged from 2cm to 7.5cm in the largest dimension.

Macroscopic tumor spill was noted intra-operatively in two cases. One of these cases had undergone a previous intra-oral biopsy and tumor spill occurred due to capsule breach at the site of the biopsy incision.

The facial nerve was dissected and preserved in twenty-one cases. In three cases the tumors were excised in completion without any formal dissection of the facial nerve (two via mandibulotomies and one via submandibular fossa approach). In one case the marginal mandibular branch of the facial nerve had to be sacrificed.

Temporary facial weakness (mainly involving the lower branches) was noted in six patients (24%), with complete recovery of nerve function subsequently. One patient had mild permanent weakness of the marginal mandibular branch following intra-operative nerve sacrifice.

Two patients developed post-operative haematomas requiring evacuation, one under general anesthetic and one under local anesthesia. There were two cases of Frey's syndrome post-operatively and one of these two patients also developed a keloid scar and complained of scar discomfort. There were no cases of salivary fistulae.

The two cases with evidence of macroscopic tumor spillage underwent post-operative radiotherapy. All twenty-five patients remain free of recurrent disease, at mean and median follow-up period of 43 and 42 months respectively (range 11 months – 7 years).

RECURRENT CASES OF DEEP LOBE PAROTID PLEOMORPHIC ADENOMA

Five patients with recurrent /residual pleomorphic adenoma of the deep lobe of the parotid, previously treated elsewhere, were operated on at our institution over the ten year period of our review. The salient features of these cases at the time of referral are shown in Table 1.

Figure 2

Table 1: Table Showing the Salient Features of Cases of Recurrent/Residual Deep Lobe Parotid Pleomorphic Adenoma at Referral

Patient	1	2	3	4	5
Number of previous excisions	2	3	4	1	1
Tumor Spill at 1 st Resection	Yes	Yes	Yes	Yes	Yes
Radiotherapy after 1 st Resection	Yes	No	Yes	No	No
Time to Recurrence* (years)	1	10	3	<1	11
Type of Recurrence	Multifocal	Multifocal	Multifocal	Solitary	Multifocal
Facial Weakness	No	No	No	No	No

* Time from last procedure elsewhere to episode of recurrence resulting in referral to our institution

Three of the five patients had undergone multiple previous resections, and macroscopic tumor spillage was noted in all five cases at the time of their original resection. However, only two patients had received post-operative radiotherapy prior to referral.

In all five cases the extent of the recurrent disease was assessed pre-operatively with MRI. All five tumors were excised using an external approach. Mandibulotomy was required in one case for complete excision.

Complete macroscopic tumor clearance was achieved in three cases, and macroscopic tumor spill was noted in the remainder two (both multifocal recurrences; in one case the tumor was adherent to stylomastoid foramen).

The facial nerve was dissected and preserved in all cases. In one case the facial nerve was microdissected proximally in its course through the mastoid, due to the difficulty in identifying the nerve more distally because of the disseminated nature of the recurrence.

There were two cases of temporary facial nerve weakness post-operatively. There was also one case of a post-operative salivary fistula, requiring reconstructive surgery for closure.

Both patients with evidence of macroscopic tumor spill received post-operative radiotherapy (neither had undergone radiotherapy previously).

Two patients in this group developed further recurrences. In the first case there was evidence of macroscopic tumor spill at the time of revision surgery and despite post-operative radiotherapy, a further recurrence (1cm) was noted 2 years later on MRI. Repeated imaging after a further 12 months did not show any change in the size of the lesion, and the case is being managed conservatively at present.

The second case had four previous tumor excisions and radiotherapy prior to referral. At the time of the fifth excision (at our institution) macroscopic clearance was achieved, however, the patient developed three further recurrences after 6, 8 and 9 years. In the first two instances the recurrent disease was histologically confirmed as benign pleomorphic adenoma. The resection specimen from the last recurrence, however, was found to be malignant. Twenty-three years after the original presentation, the patient succumbed to carcinomatosis with diffuse lung metastasis.

The remainder three patients remain free of further recurrence at 5, 6, and 10 years of follow-up.

DISCUSSION

Deep lobe parotid pleomorphic adenomas are rare tumors that present a diagnostic and therapeutic challenge. Approximately 10-12% of pleomorphic adenomas of the parotid are thought to arise from the deep lobe of the parotid.^{1,5} In our review the percentage of deep lobe tumors is slightly higher (22.9%), this may be accounted for by the fact that being a tertiary referral center cases of deep lobe tumors (if recognized pre-operatively) are frequently

referred to us via other institutions in the region, thus skewing the results somewhat.

These tumors have a variety of distinct clinical presentations, most commonly arising in the portion of the gland deep to the facial nerve yet lateral to the mandible, and present as pre-auricular masses. This is reflected in our results with seventeen (68%) of the previously untreated patients presenting with pre-auricular masses alone (see Figure 1).

Not all deep lobe tumors extend through the stylomandibular tunnel to the parapharyngeal area.¹⁰ Tumors that do so may be predominantly pharyngeal or have a combination of pre-auricular and pharyngeal components as in the 'dumb-bell' variety. Due to the anatomical relations and restrictive boundaries of the deep lobe, parapharyngeal tumors may remain asymptomatic until reaching a very large size.⁵ Eustachian tube dysfunction, serous otitis media, conductive hearing loss, and rarely cranial nerve and the sympathetic cervical chain impairment may result from very large parapharyngeal extensions.¹⁰ Three of the seven cases with parapharyngeal extension in our series, had additional symptoms of middle ear effusions, and one patient displayed symptoms of tongue numbness and trigeminal paraesthesiae.

Deep lobe parotid tumors presenting as pre-auricular masses are often clinically indistinguishable from lesions of the superficial lobe, thus making it difficult to ascertain a pre-operative diagnosis of lesion lying deep to the facial nerve. In our series thirteen (52%) of the twenty-five previously untreated tumors were correctly identified as arising from the deep lobe of the parotid pre-operatively. All thirteen of these patients had undergone radiological evaluation. It is interesting to note, however, that only twelve (67%) of the eighteen cases undergoing MRI and one of the two cases undergoing CT evaluation, were correctly identified as having deep lobe tumors.

Two of our cases had undergone diagnostic open oropharyngeal biopsies prior to referral. In one of these the tumor capsule was found to be adherent to the pharyngeal mucosa at the biopsy site and in the course of excision this lead to tearing of the mucosa and tumor capsule rupture and tumor spill. Incisional biopsy of parapharyngeal masses suspected of being pleomorphic adenomas is contraindicated unless malignancy is suspected.¹¹ The procedure is fraught with risks of hemorrhage, tumor seeding and providing a portal of entry for infection into the parapharyngeal space.¹¹

Deep lobe parotid tumors of all types should be removed by an external approach so that the facial nerve can be identified with precision and preserved undamaged.¹² Access can be greatly improved by division of the stylomandibular ligament and styloid process, mandibulotomy, or by using an upper lateral cervical approach through the submandibular fossa.^{5,6,10,11,12} Excision via an intra-oral approach may be disappointing and risks damage to major neurovascular structures.¹²

Six (24%) of the twenty-five previously untreated cases in our series developed temporary post-operative facial nerve weakness. This is comparable to previously reported figures of 31%.¹³ Ischemia is though to be the most important factor in causing this functional paralysis. Other factors include edema and stretching of the nerve, in particular of the finer branches.¹⁴ It is important to emphasize that in spite of all precautions the risk of functional paralysis cannot be completely eliminated.¹⁴

Peri-operative electromyographic facial nerve monitoring was used in all cases in our series. There have been suggestions in the literature that use of facial nerve monitoring reduces the incidence of temporary facial weakness post-operatively.¹⁵

In one case in our series the marginal mandibular branch of the facial nerve was sacrificed during tumor excision, leaving the patient with a very mild permanent weakness. The slight degree of facial paralysis resulting from sacrifice of the lower divisions may be attributable to interconnecting branches.¹⁵

There were two reports of Frey's syndrome (8%) amongst our series of patients, this is comparable to other reports of 11.1%.⁹ Post-operative morbidity patterns following excision of recurrent/residual pleomorphic adenoma have been reported to be different from those in new cases.⁹ The patterns observed in our series are shown in Table 2.

Figure 3

Table 2: Post-operative Complications Following Excision of New and Recurrent Deep Lobe Pleomorphic Adenoma

Post-operative Complication	New Cases	Recurrent Cases
Temporary Facial Weakness	6/25 (24%)	2/5 (40%)
Permanent Slight Weakness of Marginal Mandibular Branch (Secondary to Intra-operative Sacrifice)	1/25 (4%)	0
Frey's Syndrome	2/25 (8%)	0
Salivary Fistula	0	1/5 (20%)
Other Wound Complications	2/25 (8%)	0

Risk of recurrence of pleomorphic adenoma is thought to be related to the adequacy of surgical excision. The ideal method excision would be to remove the tumor with a margin of normal tissue, thereby reducing the risk of recurrence as no marginal areas of tissue are left behind and no spilling of tumor cells occurs from breaches of the tumor capsule.¹¹ Simple enucleation of the tumor is thought to be associated with higher recurrence rates.¹⁶

Deep lobe tumors, however, can rarely be excised with a complete covering of normal parotid tissue. The incidence of enucleation is increased by the proximity of the facial nerve, carotid artery and internal jugular vein. This does not, however, appear to compromise prognosis or recurrence rates in these patients.^{1,16} It has been postulated that this may be due to the deep lobe tumors having thicker capsules and thus making it less likely for the tumor cells to penetrate the full thickness of the capsule. These histological variations of the capsule are thought to be due to the anatomical relations of the deep lobe rather than due to innate biological behaviour.¹⁶

Once tumor spillage has occurred there is no way of successfully removing all the cellular tumor elements and a high rate of recurrence is to be expected in this instance.¹⁰ In the former years, post-operative radiotherapy was recommended in all cases with recognizable tumor spillage at operation, however, the long-term morbidity of postoperative radiotherapy should not be underestimated; and radiotherapy should therefore only be considered after uncontrollable tumor spill.¹⁷ In our series of new cases of deep lobe parotid pleomorphic adenoma, there were two cases of macroscopic tumor spillage resulting in incomplete tumor clearance. Both of these underwent post-operative radiotherapy and remain disease free.

Scar tissue and the loss of anatomical landmarks make excision of recurrent tumors more difficult. Exposure of the

facial nerve in its bony canal can often aid the surgeon in identifying the nerves position, even in dense scar and recurrent tumour.¹⁰

In our group of recurrent tumors, macroscopic tumor spill was noted in all five cases at the time of the primary resection. Two of these patients had also received post-operative radiotherapy. Four of the five cases demonstrated multifocal recurrences, with one case having a solitary recurrent lesion. Multifocal recurrences are thought to be associated with a higher risk of relapse.¹⁸ There were two cases of relapse after excision of recurrent disease in our series, and in both cases the recurrence was multifocal.

One case in our series underwent malignant transformation, following multiple recurrences, twenty-three years after the original presentation. Malignant transformation of benign pleomorphic adenomas is an area of controversy and it has been postulated that risk of such a transformation is higher in cases of recurrent disease.²

SUMMARY

Deep lobe parotid pleomorphic adenomas are rare benign tumors that are often clinically indistinguishable from lesions of the superficial lobe of the parotid. Recurrence is rare following complete surgical excision at initial presentation via an external approach.

Recurrent tumors are often multifocal and complete surgical excision may not always be possible. The likelihood of further relapse in these cases is higher.

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