

Recurrent pleomorphic adenoma: uninodular versus multinodular disease

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Abstract

Background While treatment of previously untreated pleomorphic adenoma is relatively straightforward, recurrent pleomorphic adenoma presents a management problem with increased risk of injury to the facial nerve and an increased risk of malignant transformation in recurrent disease.

Aims The objectives of this study were to review the management of recurrent pleomorphic adenoma in our unit to identify factors that might help treatment of future cases.

Methods We reviewed the management of pleomorphic adenoma at our department over an eight-year period from 1990-1998 and present our experience of recurrent pleomorphic adenoma of the parotid gland and parapharyngeal space.

Results Twelve patients were treated with recurrent pleomorphic adenoma. In 10 of these, the site of recurrence was in the parotid gland with the remainder occurring in the parapharyngeal space. Type of recurrence was uninodular or multinodular, the former being easier to treat. Three patients required adjuvant radiotherapy. None of our patients had permanent facial nerve damage.

Conclusions In order to prevent recurrence of pleomorphic adenoma of the parotid gland, we recommend formal superficial parotidectomy for first time surgery.

Introduction

Pleomorphic adenoma is a slow growing benign salivary gland tumour that accounts for 60% of parotid gland tumours. These tumours have a pseudocapsule rather than a true capsule and frequently the tumour extends finger-like projections into surrounding tissues.¹ Enucleation is inadequate due to a high rate of recurrence.²

Surgery for recurrent or residual disease is difficult because of proximity of the facial nerve. The rate of injury to the facial nerve shows a marked increase from less than 2.5% at primary surgery to as high as 15-29% after second and subsequent surgeries.^{3,4} At surgery for recurrent disease, the nerve is frequently found ensheathed in scar tissue thus increasing the risk of damage. With repeated reoperation it becomes more difficult to get tumour clearance with retention of normal nerve function.⁵ This poses a particular dilemma for the surgeon for what is in the majority of cases a benign disease.^{5,6} Malignant transformation is more common in recurrent disease.⁵

The objectives of this study were to review the management of recurrent pleomorphic adenoma in our unit and to identify factors which might help treatment of future cases.

Patients and methods

Clinical, operative and pathological data were reviewed on all patients with pleomorphic adenoma seen between July 1990 and October 1998. A subgroup was identified whose members were secondary referrals with recurrent disease. Factors examined included age and sex distribution; previous treatment

including number of previous operations; site and type of recurrent disease (nodularity) as well as treatment of the recurrent disease.

Techniques used to locate the facial nerve were also reviewed, as were details of facial nerve function after surgery. Notes were reviewed for complications and long-term follow-up results for recurrences.

Results

Sixty-three patients with pleomorphic adenoma were seen at the otolaryngology department of the Mater Hospital between July 1990 and October 1998. There were 12 cases of secondary referrals with recurrent pleomorphic adenoma. The mean age was 48 years (range 28-64 years) and the male:female ratio of 1:1.2. The mean follow-up time was 52 months.

Clinical and surgical records revealed that 11 patients had been previously treated by enucleation and one by transoral resection. Two patients had undergone two previous operations and one had undergone three previous operations at the time of referral.

Site of recurrence

The site of recurrent disease was the parotid gland in 10 patients and the parapharyngeal space in two. Of those with recurrent disease in the parotid, three had recurrent disease in the superficial lobe alone and seven in the deep and superficial lobes. Four patients showed evidence of skin involvement also.

Type of recurrence

Recurrences occurred in two different forms either uninodular or multinodular. Eight patients presented with a uninodular recurrence, while four had multinodular recurrences. Uninodular recurrent adenoma was treated with either superficial or total parotidectomy with preservation of the facial nerve. The four patients with multinodular recurrences were treated by superficial parotidectomy with skin excision in one, radical parotidectomy with skin excision in one and radical parotidectomy with skin excision plus nerve reconstruction in two.

Adjuvant therapy

Three patients were referred for postoperative radiotherapy because of microscopic residual disease and fears for further recurrences. Figure 1 shows a mass in the deep lobe of the parotid gland presenting as a parapharyngeal swelling obvious on intra-oral examination.

Figure 1: Clinical photograph of an intra-oral swelling which proved to be a pleomorphic adenoma in the deep lobe of the parotid gland presenting as a parapharyngeal swelling.



Location of the facial nerve

A number of methods were used to locate the facial nerve. In nine cases, it was possible to locate the main trunk of the nerve using a combination of the tragal pointer and the posterior belly of the digastric muscle. In two cases, the distal branches were located first and traced back to the main trunk and, in one, the nerve was located using the tympanomastoid suture. The location of the nerve was confirmed in all cases using a nerve stimulator (either Xomed or Mag-Stim).

Facial nerve function

Review of results of facial nerve function revealed normal function in nine patients and slight lower lip weakness in two. The marginal mandibular branch was sacrificed and reconstructed with a great auricular cable nerve graft resulting in a mild facial nerve paresis (House-Brackmann stage 2) in one patient. No patients who had surgery for primary pleomorphic adenoma had permanent facial nerve injury. No patient operated on for primary pleomorphic adenoma developed recurrence.

Follow-up

Follow-up on patients post surgery for recurrent disease revealed that two developed further recurrences. One patient developed a unifocal recurrence 75 months post-op while a second developed a multifocal recurrence 43 months after surgery. Two patients developed Frey syndrome with postgustatory sweating. No patient developed salivary gland malignancy. All patients were alive and well after a mean follow-up of 52 months (range 3-99 months).

Discussion

The cause of recurrence following resection of pleomorphic adenomas is multifactorial and may be due to incomplete excision, tumour spillage at the time of the initial surgery or non-recognition of the multicentric origin of some of these tumours. Pleomorphic adenomas have a pseudocapsule rather than a true capsule. Incomplete excision may occur due to dislocation of the tumour bud as it protrudes through this pseudocapsule.¹ Tumour spillage can occur following rupture of the pseudocapsule which may lead to seeding of the surgical bed.⁷ Multicentricity is uncommon but may be a contributing factor for recurrence on occasion.¹

Enucleation results in a recurrence rate of 10-35% as opposed to formal parotidectomy which is associated with recurrence rates of 0-2.5%.^{2,4} There is a significant increased rate of recurrence after reoperation for pleomorphic adenoma.

Recurrent disease presents either as uninodular or multinodular recurrences.^{4,8} Uninodular recurrences may be treated by surgery alone. In this study, uninodular recurrent adenoma was treated with either partial or total parotidectomy with preservation of the facial nerve. The four patients with multinodular recurrences had treatment appropriate to the extent of recurrence.

In our experience, multinodular recurrences warrant more aggressive surgery and some patients may benefit from postoperative radiotherapy in order to reduce future recurrences. This must be balanced with any risk of malignant transformation.^{9,10} Nerve preserving surgery is recommended but, on occasion, is not possible (in such instances, cable grafting is used). In all cases, a facial nerve monitor was used and we would recommend its use for surgery for recurrent disease.

Malignant transformation is rare and has not occurred in any of our patients. However, it can occur in long-standing pleomorphic adenoma.¹¹ Furthermore, the risk of malignant transformation is thought to be increased in recurrent disease.⁵

Conclusion

In summary, treatment of recurrent pleomorphic adenoma is difficult, has an increased risk of facial nerve damage and is associated with both a risk of further recurrences and a possible higher risk of malignant transformation. We recommend wide anatomical excision at the first procedure to reduce recurrence and would advocate that enucleation has no role to play in pleomorphic adenoma of the parotid gland.

When dealing with primary pleomorphic adenoma, a cuff of normal tissue should be removed and care taken to avoid tumour spillage in order to reduce recurrence.¹² This will normally necessitate formal superficial parotidectomy for pleomorphic adenomas of the superficial lobe of the parotid gland.

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