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Pleomorphic adenoma and severe obstructive sleep apnoea

C E B Giddings, MRCS(Eng), D Bray, MRCS(Ed), J Rimmer, MRCS(Eng), P Williamson, FRCS(Ed)

Abstract
Two cases of deep lobe parotid tumours extending into the parapharyngeal space and causing obstructive sleep apnoea are described. Post-operatively, marked improvements in nocturnal hypoxic episodes and the symptoms of obstructive sleep apnoea were seen. Although minor salivary gland pleomorphic adenomas have been described as a cause of airway compromise, pleomorphic adenomata arising from the deep lobe of the parotid, causing proven obstructive sleep apnoea, have not previously been documented. The anatomy and common pathologies of the parapharyngeal space are discussed.

Key words: Pleomorphic Adenoma; Parotid Tumours; Obstructive Sleep Apnoea; Airway Obstruction; Parapharyngeal Space

Introduction

We report two cases of pleomorphic adenoma of the deep lobe of the parotid gland extending into the parapharyngeal space. Both patients presented with obstructive sleep apnoea (OSA), 13 years apart. In each case, removal of the adenoma resulted in resolution of the symptoms of OSA. Space-occupying lesions within the parapharyngeal space are rare, accounting for 0.5 per cent of head and neck neoplasia,[1] but can have significant implications for the airway. Most parapharyngeal space tumours arise from salivary glands, but schwannomas, lipomas and carotid body tumours, among others, may all arise within or encroach into this potential space. Pleomorphic adenomata of minor salivary glands causing airway obstruction have been described previously.[2] However, pleomorphic adenomata arising from the deep lobe of the parotid, that extend into the parapharyngeal space and impinge on the airway causing severe obstructive sleep apnoea, do not appear to have been previously described in the literature.

Case reports

Case 1

A 35-year-old man presented in 1990 with severe snoring and apnoeic episodes. This had begun shortly after his marriage and become progressively worse over the subsequent 2 years. He attended the clinic at the request of his spouse. Examination revealed an asymmetrical soft palate, with narrowing of the nasopharyngeal isthmus on the left. Transoral fine needle aspiration resulted in cytology consistent with a pleomorphic adenoma. Computed tomography (CT) scan revealed a mass arising from the deep lobe of the left parotid with compression of the airway (Figures 1 and 2).

The patient underwent total parotidectomy via a standard external approach and made an unremarkable recovery. Post surgery, his snoring resolved completely, as did the apnoeic episodes. Unfortunately he developed a recurrence and a second resection was performed in 1996, with sacrifice of his facial nerve. He received radiotherapy post-operatively, and has had no further recurrence to date of the tumour or his symptoms.

Case 2

A 67-year-old lady presented in 2003 with severe obstructive sleep apnoea that had deteriorated over a 2-year period. Her main complaints were of daytime somnolence and early morning headaches; she lived alone so was unaware of any snoring. She would often fall asleep during meals, and once awoke to the sound of car horns whilst stationary at traffic lights. She did not seek medical attention at this time, despite requests from her family. On elective attendance for a total knee replacement, her operation was cancelled after the anaesthetist took a history, elicited her obstructive symptoms, and examined her oropharynx.

General examination showed her to be obese, with a body mass index (BMI) of 36 and a large bull neck. Oral examination revealed a large, firm, non-tender, non-pulsatile swelling projecting into the right oropharynx, causing deviation of the uvula to the contralateral side and narrowing of the nasopharyngeal isthmus (Figure 3). She was referred to the otolaryngology clinic, and to the respiratory physicians for polysomnography.

This sleep study confirmed severe sleep apnoea, with an apnoea/hypopnoea index of 93.2 per hour, and desaturation events of approximately 109.5 per hour. Nadir oxygen saturation was 57 per cent, with frequent episodes below 60 per cent and a baseline of 87.6 per cent.

Transoral fine needle aspirate confirmed the diagnosis of a pleomorphic adenoma. A contrast-enhanced CT scan of the neck showed a mass arising from the deep lobe of the

From the Department of Head and Neck Surgery, St George's Hospital, London, UK.
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FIG. 1
Case 1: Coronal CT scan of left parapharyngeal mass.

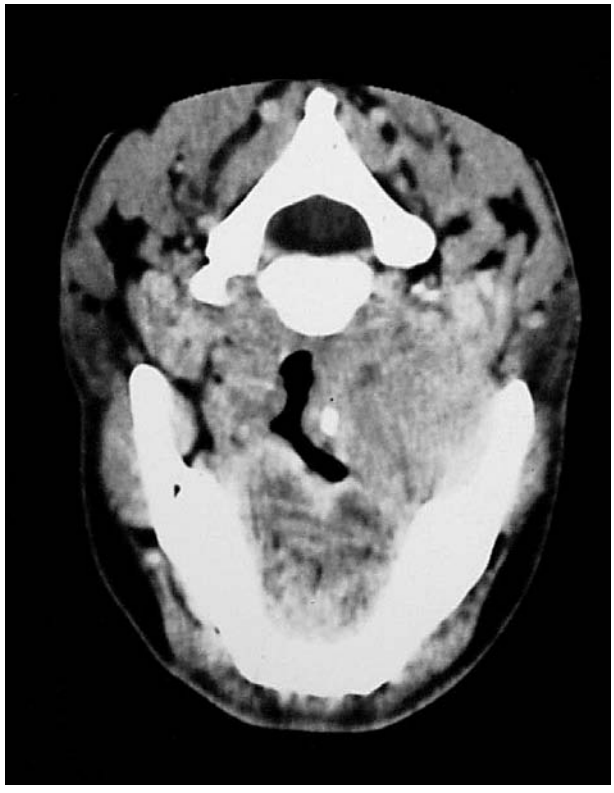

FIG. 2
Case 1: Axial CT scan of left parapharyngeal mass.

FIG. 3
Case 2: Right parapharyngeal mass and uvula deviation.

right parotid with compression of the airway (Figure 4). The lesion extended from the right pterygoid plate and pterygopalatine fossa inferiorly, to the level of the right vallecula and tonsil above. It abutted the pterygoid muscles anteriorly, and the longus coli muscle posteriorly. Inferiorly, there was extension to the superior border of the submandibular gland. The lesion had a maximum diameter of approximately 4 cm, and a necrotic centre. It encroached severely on the pharynx and appeared to have caused an element of pressure necrosis of the pterygoid plates.

The patient underwent a right total parotidectomy, via an external approach, with a mandibular condylotomy and enucleation of her deep lobe tumour. She made an unremarkable post-operative recovery.

Histological examination of the resected specimen revealed a mixture of epithelial cells forming strands and glandular structures, with myxoid stroma, consistent with a pleomorphic adenoma. A specimen taken deep to this showed connective and adipose tissue, but no evidence of salivary gland tissue.

Three months post-operatively she confirmed an improvement in her symptoms, especially her early morning headaches and daytime somnolence. However,

FIG. 4
Case 2: Right parapharyngeal mass arising from the deep lobe of the parotid and displacing the carotid sheath posteriorly.

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she was still mildly symptomatic. A repeat sleep study performed at that time showed a significant improvement with an apnoea/hypopnoea index of 29.5 per hour. However, she was still experiencing a large number of desaturation events, approximately 64.8 per hour, as well as having a high BMI of 36, and was therefore commenced on a trial of continuous positive airway pressure (CPAP). This improved her symptoms further, and to this date she has had no recurrence.

- **Two patients with neoplasia of the deep lobe of the parotid are presented in this paper**
- **Both had obstructive sleep patterns that were relieved by surgery**
- **The authors recommend that obstructive sleep apnoea should be considered in such cases**
- **The anatomy of the parapharyngeal space is discussed**

Discussion

The parapharyngeal space is a deep neck space in the shape of an inverted pyramid, with the greater cornu of the hyoid bone as its apex. The floor is the skull base. The anterior limit of the space is the pterygomandibular raphe, and the posterior border is the pre-vertebral fascia. The lateral wall is formed by the mandible, and the medial wall by the superior pharyngeal constrictor with the overlying buccopharyngeal membrane. As this is the only non-osseus border, masses within the parapharyngeal space extend infero-medially causing medial displacement of the tonsillar fossa and tonsil, and downward displacement of the soft palate. The stylomandibular tunnel is bounded by the posterior border of the ramus of the mandible, the skull base and the stylomandibular ligament, and extension of a parotid mass through this tunnel produces a characteristic radiological appearance.

The parapharyngeal space can be subdivided into pre-styloid and retro-styloid compartments by the tensor veli-styloid fascia. Structures found in the pre-styloid compartment include a portion of the retro-mandibular parotid gland, fat and lymph nodes. The retro-styloid compartment contains the internal carotid artery, internal jugular vein, cranial nerves IX–XII, cervical sympathetic chain and lymph nodes.

Tumours of the parapharyngeal space can be primary, metastatic or extensions from adjacent structures. Approximately 80 per cent are benign and 20 per cent malignant.[3] Salivary gland tumours are the most commonly found neoplasms, with pleomorphic adenoma the most frequent histological type.[4] They may arise within the parotid or minor salivary glands, and in the latter are thought to originate from aberrant salivary gland tissue within lymph nodes[5] or from remnants along the parotid's developmental tract.[6] These primary minor gland tumours are anatomically distinct from those arising medial to the pharyngeal constrictor muscles. Other primary tumours including schwannomas, carotid body tumours, and lymphomas have also been described involving the parapharyngeal space.[4] Any nearby head and neck tumour may encroach into the parapharyngeal space, including nasopharyngeal, oropharyngeal, neck and bony lesions. Metastases are rare.

Such tumours tend to present with a neck or oropharyngeal mass, and may reach a considerable size before becoming clinically obvious. Although minor salivary gland tumours causing airway obstruction have been documented at this site,[2] there have been no previous reports of parotid tumours presenting in this manner. Intra-oral minor salivary gland tumours have also been noted to present with respiratory obstruction,[7] as well as with dysphagia and dysphasia.[8] Masses within the deep lobe of the parotid gland may extend into the parapharyngeal space either by passing into the space posterior to the stylomandibular ligament or through the stylomandibular tunnel, giving different radiological appearances. Parotid masses extending medially through the stylomandibular tunnel have a characteristic 'dumbbell' shape on imaging. In contrast, parotid masses that originate medial to this tunnel have a round appearance, and may grow to a much larger size before diagnosis.[4] Both routes tend to cause posteromedial displacement of the great vessels, whereas schwannomas tend to displace the internal carotid artery anteriorly.[4]

If CT scanning demonstrates parapharyngeal fat between the deep lobe of the parotid and the mass, the lesion can be diagnosed as extraparotid. In this way, CT can correctly diagnose 80–90 per cent of such tumours pre-operatively.[6] However, magnetic resonance imaging (MRI) is needed to more accurately distinguish between an extraparotid salivary gland tumour and a schwannoma, as it has better soft tissue definition. The use of MRI has also reduced the need for angiography, with its inherent risks.

Management of parapharyngeal space tumours is dictated by the type and exact site of the lesion, as well as the patient's general condition. Conservative management may be acceptable in cases of slow-growing benign tumours, as well as for patients unsuitable for surgery. If surgical treatment is undertaken there are various possible approaches,[4] depending on the nature of the tumour, its anatomical site and extent, and its relation to the important neurovascular structures. In the case of parotid deep lobe tumours extending into the parapharyngeal space, the route of spread is also a factor. The cases described above were both excised with external parotid approaches; small benign lesions may however be removed transorally.[7]

Conclusion

This is the first report of pleomorphic adenomata arising in the deep lobe of the parotid gland causing proven obstructive sleep apnoea. Both cases were treated successfully by surgical excision, resulting in a reduction in snoring and apnoeic episodes, and in Case 2 a reduction in the apnoea/hypopnoea index. This highlights the importance of adequate investigation when patients present with symptoms of upper airway compromise, as there are some causes of OSA which are surgically correctable.

The parapharyngeal space is an area of complex anatomy, and is difficult to assess clinically. Radiological imaging of parapharyngeal space tumours is therefore essential for both diagnosis and treatment planning, as the surgical approach is governed by the site and size of the lesion and its route of extension.

External compression of the upper airway secondary to deep lobe parotid tumours has not previously been described, but patients presenting with such masses should be assessed clinically for compression of the upper airway and considered for sleep study prior to surgery.

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Address for correspondence:

Charles Giddings,
74 Walpole House,
126 Westminster Bridge Road,
London, UK.

E-mail: charliegiddings@doctors.org.uk

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