

## Pleomorphic adenoma of parotid gland with cystic degeneration

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### Abstract

Pleomorphic adenoma is the most common tumour to arise in the parotid gland. Diagnosis becomes difficult when such a tumour undergoes degeneration and presents with unusual findings. This can lead to erroneous decisions concerning treatment. We present two patients with pleomorphic adenoma of the parotid gland with cystic degeneration, describe the pathological findings and discuss some pitfalls in diagnosis and treatment. Both patients recovered uneventfully after surgery.

**Key words:** Parotid gland neoplasms; Adenoma, pleomorphic

### Introduction

Pleomorphic adenoma is the tumour that commonly arises in the parotid gland (Thackray and Lucas, 1974). Parotidectomy with preservation of the facial nerve is the treatment of choice. This procedure is widely accepted because a malignant transformation of the tumour is likely after a prolonged period, and because enucleation of the tumour increases the risk of recurrence as viable tumour cells remain in the capsule (Johns and Kaplan, 1987).

However, there are diagnostic and therapeutic challenges such as the case in which the tumour has undergone degeneration and presents with an unusual appearance, making diagnosis difficult and leading to erroneous decisions concerning therapy. Also if the capsule of the tumour adheres to the facial nerve or some other vital structure attempts to separate the capsule from these tissues increases the risk of its rupture.

We present two patients with parotid pleomorphic adenoma with cystic degeneration and discuss its clinical and pathological implications.

### Case reports

#### Case 1

An 87-year-old man was admitted to our hospital with a mass in the left upper neck. Although he had noticed the painless mass 10 years earlier, he had not sought medical care. One month before admission, the mass had enlarged rapidly causing dull pain.

The mass was present below the left pinna and extended to the surface of the mandibular ramus and to the undersurface of the sternocleidomastoid muscle. It measured 5 × 5 cm and was fluctuant. Neither facial paresis nor tenderness was observed. Physical examination revealed no other palpable masses in the neck or elsewhere in the body. There was mild anaemia (Haemoglobin, 9.0 mg/dl) and complete right bundle-branch block on ECG. Computed tomography (CT) depicted the mass as completely cystic in appearance on every slice (Figure 1).

The mass was considered to be a lymphoepithelial cyst from the computed tomogram, but the patient's age and clinical history were not compatible with that diagnosis. It was con-

sidered possible that the mass could be a malignant tumour with cystic degeneration.

Surgery was performed under general anaesthesia. A right preauricular incision with a curved cervical extension was made. The cystic lesion was located between the parotid gland and the sternocleidomastoid muscle. The main part of the cyst adhered tightly to the undersurface of the muscle and skin. As the cyst wall was very fragile, we dissected the cyst together with a cuff of the surrounding muscle and a small amount of skin. The antero-superior portion of the tumour adhered to the parotid gland; partial superficial parotidectomy was performed with preservation of the facial nerve.

The operative specimen proved to be completely cystic, with a tiny solid portion that had not been evident on the computed tomogram. The cystic content consisted of bloody necrotic debris. Microscopically, the cyst wall lacked any epithelial lining. The solid portion showed numerous ductal structures with a hyaline stroma: foci of myxochondroid stroma were also present (Figure 2). The cyst wall consisted of fibrous connective tissue that was very thin at the site of the muscle and relatively thick at the site of the parotid gland (Figure 3). There was no evidence of malignancy.

The patient's post-operative course was uneventful and there has been no evidence of recurrence in the following two years.

#### Case 2

A 44-year-old woman was referred to our hospital with a right upper neck mass that was painless and nontender. It was a little fluctuant, had a smooth surface and measured 3 × 3 cm. No other palpable mass was found in the neck. Physical examination revealed no other abnormalities. Because there was no evidence of malignancy on imaging studies, a right superficial parotidectomy was carried out. The medial side of tumour adhered to the buccal branch of the facial nerve. By microscopic dissection, the mass was separated from the facial nerve.

The gross appearance of the operative specimen was of a well encapsulated tumour with cystic degeneration. The cystic cavity was filled with bloody necrotic debris surrounded by normal gland tissue. Microscopically, the solid portion consisted of

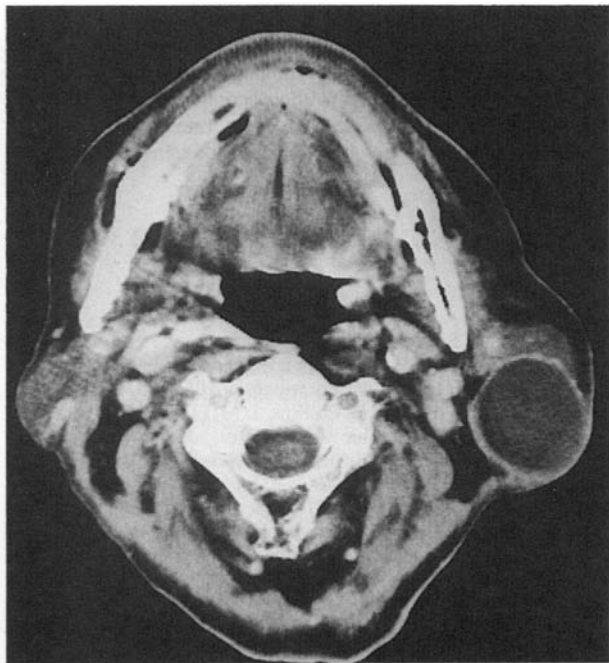


FIG. 1

Axial contrast-enhancement CT scan depicting the mass as completely cystic and lacking any solid portion (*Case 1*).

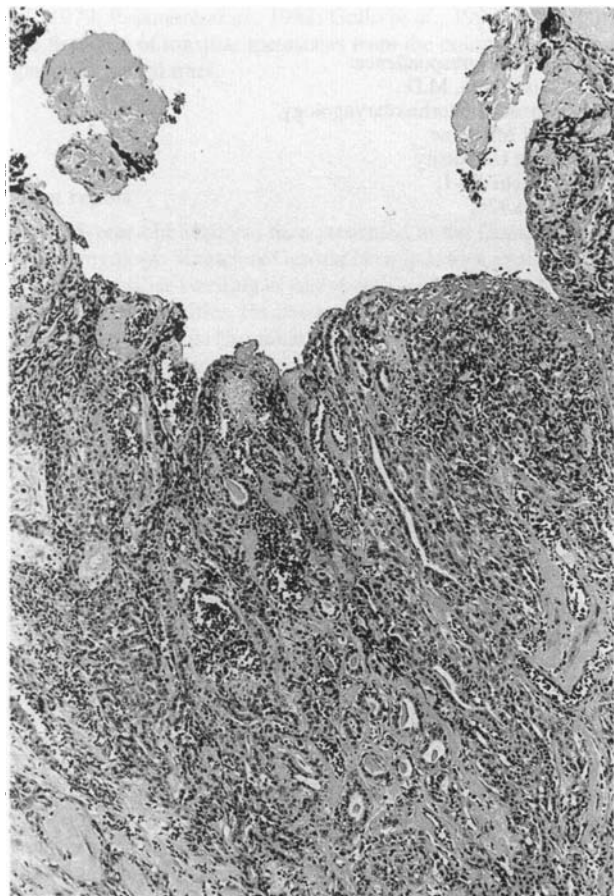


FIG. 2

Low power photomicrograph of the solid portion of the tumour showing the ductal structures arranged in double cell layers and myxoid stroma indicating a pleomorphic adenoma (*Case 1*). (H & E).

tubular structures with a hyaline stroma and foci of myxochondroid pattern. The cystic wall lacked an epithelial lining (Figure 4). Some of the solid portion protruded into the cystic cavity. There was no evidence of malignancy.

The post-operative course was uneventful. There was no evidence of recurrence in the following 10 years.

### Discussion

Although the histogenesis of pleomorphic adenoma is unclear (Dardick *et al.*, 1982), the treatment of parotid pleomorphic adenomas seems well established. But there are some problems in making the pre-operative diagnosis (Johns and Kaplan, 1987).

Modern imaging modalities give us information about the size, location and growth pattern, permitting us to estimate the nature of the lesion, but because many types of tumour can arise in the parotid gland, one cannot make inferences as to the histological type (Som, 1991). Fine-needle aspiration cytology, aided by ultrasonography, can provide accurate information about the biological nature of the tumour and its histology. However fine-needle aspiration carries some risks, because of histological alteration and dissemination of tumour cells (Batsakis *et al.*, 1992).

Gross cystic degeneration of the parotid gland tumour compounds the diagnostic problem. Such a tumour cannot be distinguished from a lymphoepithelial cyst or an epidermoid cyst, if it completely lacks the features of a solid tumour. In addition cystic lesions are not always 'true cysts' but may be pseudocysts from neoplastic lesions. For instance, Warthin's tumour can undergo gross cystic degeneration, as can basal cell adenomas, mucoepidermoid carcinomas and adenocarcinomas (Thackray and Lucas, 1974).

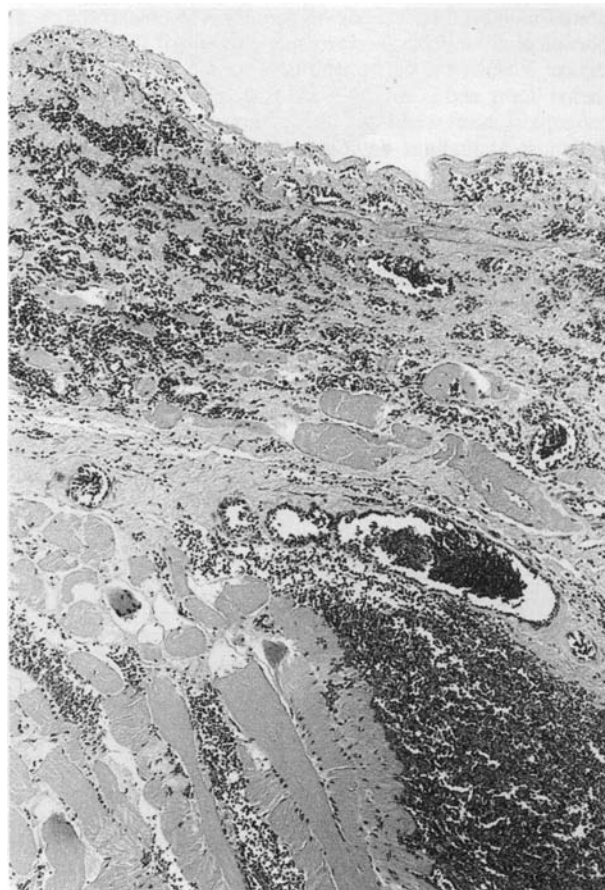


FIG. 3

Low power photomicrograph of the cystic portion of the tumour showing a thin fibrous capsule with blood vessels adhering to the muscle (*Case 1*). (H & E).

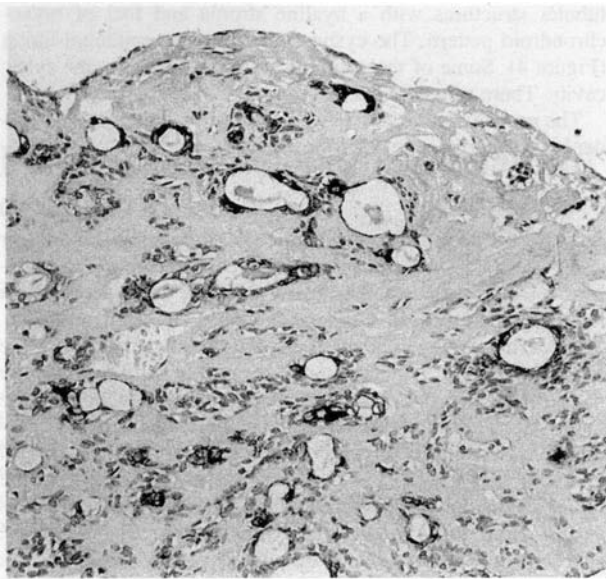


FIG. 4

High power photomicrograph showing immunoreactivity in ductal cells, but not in cells around the cystic cavity (Case 2). (Keratin (56 kd) immunostain; counterstained with haematoxylin).

Cystic lymphoid hyperplasia of the parotid gland has recently been described in patients who are HIV positive. One must pay special attention to the patient's history, particularly regarding sexual habits and drug abuse (Wernig, 1991).

From the point of view of surgery, parotid pleomorphic adenomas with cystic degeneration possess problems. If the cystic portion protrudes into the surrounding tissues, it is impossible to dissect it along the cystic wall because the wall lacks an epithelial lining and is very thin and fragile. Violation of the wall can rupture the cyst and spill its contents on to the operative field. Pathological findings have shown that the cyst may contain viable tumour cells; in which case rupture can lead to a multinodular recurrence. Thus surgeons should avoid injuring the cystic wall. We recommend dissecting the cyst together with a cuff of the surrounding tissue while avoiding injury to the facial nerve. Microscopic dissection is useful in cases involving adhesion to the facial nerve.

### Conclusions

We present two patients with benign pleomorphic adenomas of the parotid gland. Despite cystic degeneration, both tumours were successfully resected without complications, and there have been no recurrences. In such cases, one must avoid direct dissection of the cystic wall in order to prevent rupture of the cyst.

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