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A case of myoepithelioma arising in an accessory parotid gland

Yoshiyuki Kawashima, Daisuke Kobayashi*, Norihiko Ishikawa†, Seiji Kishimoto†

Abstract
The present paper describes the first reported case, to our knowledge, of a myoepithelioma arising in an accessory parotid gland. Because pre-operative fine-needle aspiration cytological findings and operative findings suggested that this tumour was malignant, the decision was made to remove the tumour surgically along with the buccal branches of the facial nerve. The resected nerve was reconstructed by nerve transplantation, using the great auricular nerve. Subsequently the tumour was found to be benign. One year after surgery, the patient had excellent facial nerve function and so far there has been no evidence of recurrence.

Key words: Parotid Gland; Salivary Gland Neoplasms; Myoepithelioma; Surgical Procedures, Operative

Introduction

Myoepitheliomas of salivary glands are extremely rare, comprising approximately only one per cent of all salivary gland tumours.[1–3] Additionally, clinical behaviour of accessory parotid gland tumours cannot be predicted, because of the limited number of reported cases and the lack of follow-up information. Here, we present a case of myoepithelioma arising in an accessory parotid gland.

Case report

A 34-year-old woman presented to our university hospital with a four-year history of a slow-growing mass on her right cheek (Figure 1). She suffered from occasional pain. Physical examination revealed a 32 × 32 mm rubbery hard mass in the area, with no associated lymphadenopathy. Facial sensation and facial nerve function were normal. No unusual results were obtained from either a complete blood count, routine blood chemistry or chest X-ray. Ultrasonic examination showed an oval, non-homogeneous hypoechoic mass with well-defined borders. Computed tomography (CT) with contrast medium showed a subcutaneous solid mass with a 20 mm long axis and a slightly enhanced edge, located anterior to the masseter muscle. Magnetic resonance imaging (MRI) revealed that the well-defined tumour had detached from the main parotid gland (Figure 2). Results of cytological examination after fine-needle aspiration cytology (FNAC) suggested that this tumour was an adenoid cystic carcinoma of the right accessory parotid gland. The cytological features were aggregates of small, relatively uniform, round to ovoid cells that surround scattered spheres of hyaline. In some cases isolated cores of the hyaline material were surrounded by rows of adherent cells, whereas in others multiple closely-associated cores simulated the cribriform structures from which they were derived (Figure 3).

Under general anaesthesia, the patient underwent tumour resection using the same approach as that used for superficial parotidectomy (Figure 4(a)). It was noted that the tumour was noncontinuous with the superficial lobe of the prarotid gland. We sacrificed the buccal branches of the facial nerve, the Stensen's duct and the superficial lobe of the main parotid gland, because of

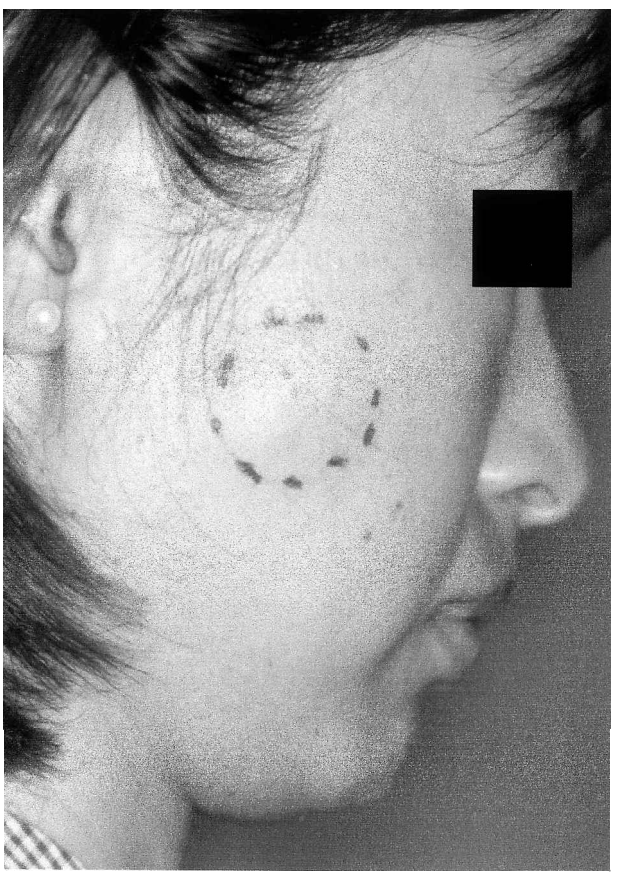

Fig. 1
Slight swelling in the middle area of the right cheek (dotted circle).

From the Departments of Otolaryngology, Pathology*, and Head and Neck Surgery†, Tokyo Medical and Dental University, School of Medicine, Tokyo, Japan.
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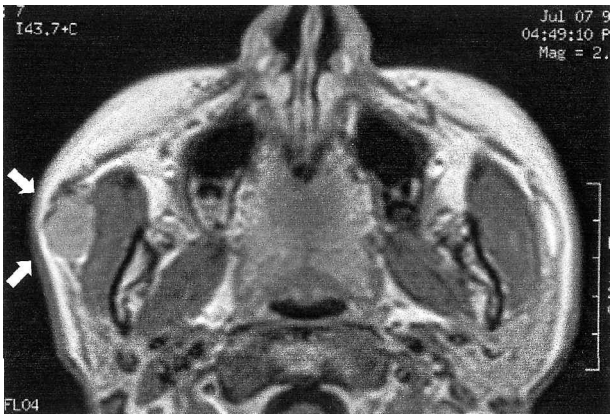


FIG. 2

MRI shows subcutaneous tumour (arrows) outside the masseter muscle and separated from the right main parotid gland.

observed tumour-like adhesions and the findings of pre-operative FNAC. Using the right great auricular nerve, a free facial nerve graft was performed (Figure 4(b)).

Under microscopic examination, the tumour mainly exhibited a reticular pattern composed of polygonal-shaped cells with hyalinized stroma (Figure 5(a)). There were a few focal areas composed of spindle cells (Figure 5(b)). Immunohistochemical examination revealed that both types of tumour cells were positive for α-smooth muscle actin, S-100 protein, vimentin, cytokeratin, and epithelial membrane actigen, and that both were negative for glial fibrillary acidic protein and desmin. Altogether, the pathological findings indicated that the tumour was a myoepithelioma of the accessory parotid gland. The superficial lobe of the main parotid gland was free from tumoural involvement. One year after surgery, the patient had excellent facial nerve function (i.e. House-Brackmann grade I).

Discussion

Accessory parotid glands occur in approximately 21 per cent of humans.[4] Generally, one accessory gland occurs on each side of the mouth, approximately 6 mm anterior to the main parotid gland and usually just above Stensen's duct, and each accessory gland has its own duct.[2] Clinically, they are usually found at the midpoint of an imaginary line extending from the tragus to a point midway between the ala of the nose and the vermilion border of the upper lip.[5] Therefore, in diagnosis of a mass presenting in the mid-anterior cheek, including lymphadenopathy, lipoma, neurofibroma, schwannoma, haemangioma, fibroma, epidermal inclusion cyst, and primary parotid duct tumours, care must be taken to determine whether the tumour has arisen from the main parotid gland or the accessory parotid gland. The locations of the accessory parotid gland and the tumour are important in diagnosis.

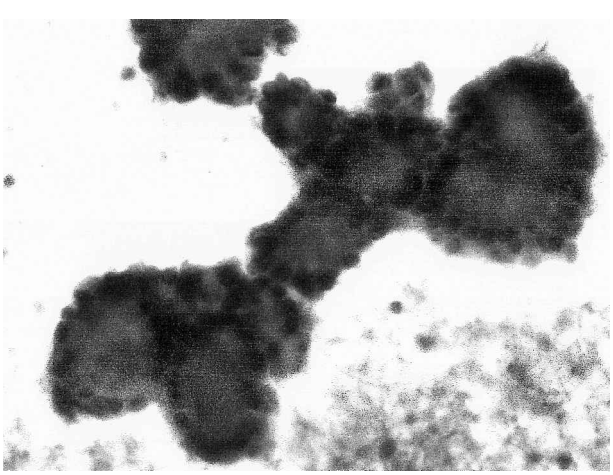
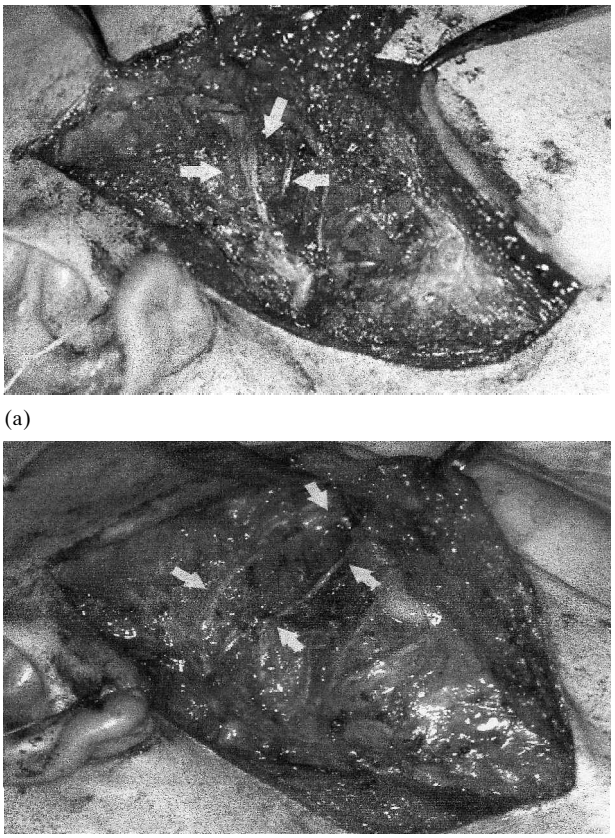


FIG. 3

Relatively uniform, round to ovoid cells that surround scattered spheres of hyaline core (Papanicolaou stain, × 300)



(a)

(b)

FIG. 4

(a) Intra-operative exposure of the main trunk of the facial nerve and its branches. Arrows indicate the tumour. (b) Using the right great auricular nerve, facial nerve grafts were performed (arrows).

Myoepithelial cells are present in a number of secretory organs including the breasts, the lacrimal glands, the sweat glands, and the salivary glands.[6,7] Since Sheldon first described a myoepithelioma of the salivary gland in 1943,[8] over 50 additional cases have been reported.[1,9] However, criteria for classifying myoepitheliomas of the salivary glands have never been well-defined or clearly described. The tumours have been classified in the past as a form of pleomorphic adenoma. Undoubtedly, they bear a close relationship to pleomorphic adenoma but differ in the lack of a prominent ductular component and the shape demarcation of the periphery of the cellular cords from the myxo-vascular stroma.[10] The neoplastic cells of myoepitheliomas demonstrate cytokeratin immunoreactivity. There is considerable variation of tumour cell expression of α-smooth muscle actin.[11] Immunoreactivity for S-100 protein is usually strong, whereas it is more variable for vimentin and glial fibrillary acidic protein.[12,13] In this case, the overall microscopic appearances were most consistent with a diagnosis of myoepithelioma. To our knowledge, myoepithelioma of the accessory parotid gland has not previously been reported.

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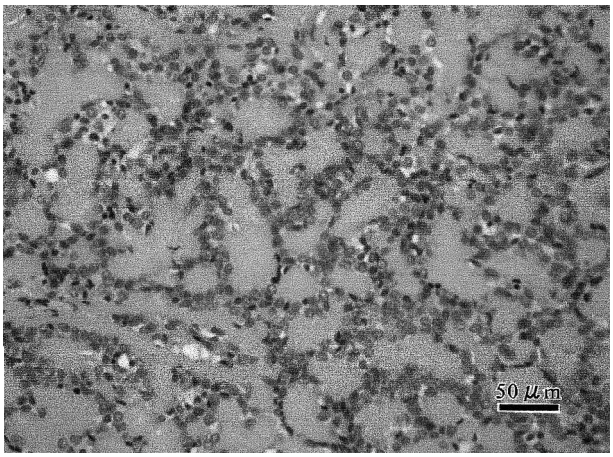
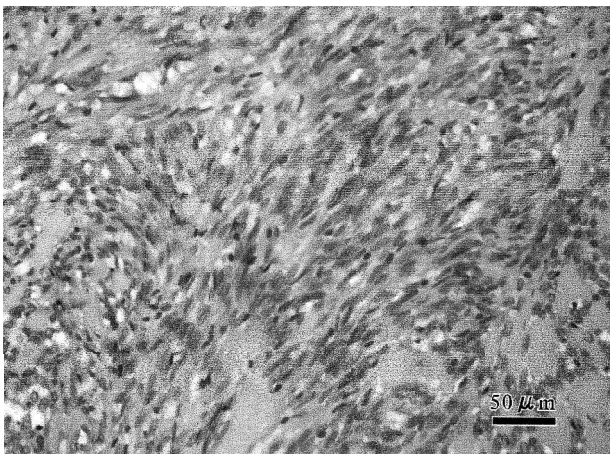


(a)



(b)

FIG. 5

(a) Microscopically, the tumour mainly exhibited a reticular pattern, composed of ramifying cords of polygonal-shaped cells surrounded by ample hyalinized stroma (H & E; × 160; range bar, 50 μm). (b) A few areas of this tumour were composed of spindle-shaped cells (H & E; × 160; range bar, 50 μm)

It is generally accepted that one to 7.7 per cent of all parotid tumours occur in accessory gland tissue, and that 50 per cent of these tumours are histologically malignant.[5,14] Neoplastic characteristics of acessory parotid gland tumours are similar to those of tumours of the main parotid gland. For this reason, surgical treatment should generally be performed, regardless of cytological findings of fine needle aspiration.

In our case, the decision to resect the buccal branches of the facial nerve was based mainly on the operative finding of tumour-like adhesions. FNAC is a well-established tool for investigating many head and neck conditions. Its application in parotid tumours is, however, controversial. Whilst it is an accurate method of distinguishing neoplastic from non-neoplastic lesions it may not be possible always to predict accurately a specific tumour type due to the overlapping spectrum of cytological appearance found in a wide variety of salivary gland neoplasms. This can result in a differential diagnosis in the cytology report for those tumours not characteristic of a specific type. So the extent of excision is a decision that depends on the anatomical findings at the time of surgery. Incomplete excision of salivary gland tumour is to be avoided because of the high recurrence rates. In surgical treatment of patients with a main parotid gland tumour, immediate repair by free facial nerve graft has been possible since Cawthorne introduced routine use of the operating microscope for surgery of the facial nerve in 1938.[15] In our experience, nerve-tumour dissection with free facial nerve grafting is a satisfactory method of surgical treatment of tumours of the accessory parotid gland.

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Address for correspondence:
Yoshiyuki Kawashima, M.D.,
Department of Otolaryngology, School of Medicine,
Tokyo Medical and Dental University,
1-5-45, Yushima, Bunkyo-Ku,
Tokyo 113-8519, Japan.

Fax: +81 3-3813-2134

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