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Synchronous bilateral mucoepidermoid carcinoma of the parotid gland

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Abstract
It is rare for a parotid gland tumour to arise bilaterally, the most common example being Warthin's tumour. Furthermore, it is rare for a parotid gland cancer to occur bilaterally. Here, we describe a case of synchronous bilateral mucoepidermoid carcinoma arising in the parotid gland. A case of bilateral facial nerve dysfunction is presented in which aggressive surgery failed to save the life of a 48-year-old man. This is the first such case reported in the available English literature.

Key words: Parotid Neoplasms; Carcinoma, Mucoepidermoid

Case report

Clinical course

A 48-year-old man had noticed a slowly growing mass causing dull pain in the left pre-auricular area and had been treated with antibiotics for three months at a local clinic. However, left facial nerve dysfunction occurred, so he visited our clinic. When first examined, he had a 6×4 cm minimally tender firm tumour in the left pre-auricular area that adhered strongly to the skin. There was also a 1×1 cm firm movable nodule in the right pre-auricular area of which he had been unaware. His left facial nerve dysfunction was very severe (House-Brackman Grade-V), but his right facial nerve function was intact. Magnetic resonance imaging (MRI) showed a $3 \times 4 \times 5$ cm enhanced mass in the left parotid gland and a 1 cm diameter enhanced cystic mass in the right parotid gland, with associated cervical lymphadenopathy on the left side only (Figure 1). Chest X-ray, full blood count and liver function tests were normal. Lung computed tomography, hepatic echogram, and systematic gallium scintillation count excluded the existence of another primary mucoepidermoid carcinoma. An open biopsy of the left parotid gland was performed under general anaesthesia, and showed mucoepidermoid carcinoma. On 22 December 1999, a left total parotidectomy and inferior external auditory canal dissection were performed with radical left neck dissection. Fourteen days after the first operation, however, right nerve dysfunction gradually appeared. On 8 February 2000 a right total parotidectomy and radical right neck dissection were performed with radical post-operative radiotherapy to both sides of the parotid gland and neck. Both facial nerves were dissected and a nerve transplant was performed on the right side using the cervical nerve. On pathologic examination, both tumours were typical high-grade mucoepidermoid carcinomas. In follow-up no local recurrence was observed, but multiple bone and brain metastases were detected by a gallium scintigram. Four months after the second operation, the patient died of respiratory suppression resulting from the brain metastases.

Pathologic description

The histology of the specimen revealed a high-grade mucoepidermoid carcinoma. Most of the tumour was a differentiated malignant neoplasia with an admixture of

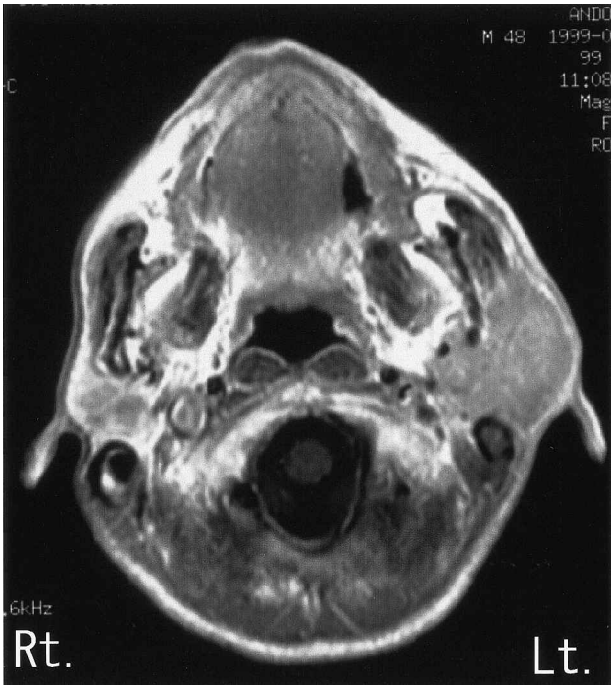

Fig. 1
Magnetic resonance imaging (MRI) showed a $3 \times 4 \times 5$ cm enhanced mass in the left parotid gland and a 1 cm diameter enhanced cystic mass in the right parotid gland.

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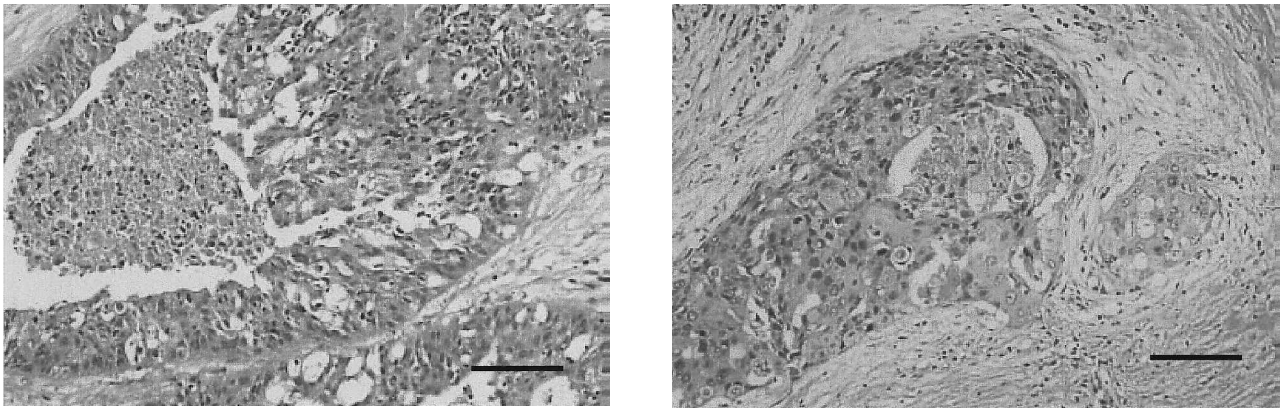

(a) (b)

FIG. 2

The histology of the specimen (haematoxylin eosin staining. (a): left parotid gland. (b): right parotid gland). A high-grade mucoepidermoid carcinoma with an admixture of mucus, intermediate, and epidermoid cells. A few ducts were present and there was circumscribed squamous metaplasia or focal inclusion of goblet-like cells. (bar = 100 μm)

mucus, intermediate, and epidermoid cells. A few ducts were present within the tumour. In some areas there was circumscribed squamous metaplasia or focal inclusion of goblet-like cells, that were periodic acid-Schiff (PAS)-positive and stained with Astra blue. There were many microcysts filled with mucous substances (Figure 2).

Discussion

Salivary gland tumours are rare, ranging from three to six per cent of all head and neck tumours. Roughly 80 per cent of these present in the parotid gland.[1] Among parotid gland tumours, Byrne and Spector[2] in 1988 reported that 63.2 per cent were benign tumours, 21.6 per cent were malignant neoplasms, and 15.2 per cent were non-neoplastic lesions, while Williams[3] reported in 1980 that 27 per cent of parotid tumours were malignant.

Bilateral parotid gland tumours, either asynchronous or simultaneous, are known, but rare.[2,4] Byrne and Spector[2] reported that 3.5 per cent of 231 cases and Turnbull 1.3 per cent of 1805 cases of parotid gland tumours were bilateral. Turnbull[4] also stated that 86 per cent of bilateral tumours of the parotid gland were Warthin's tumours.

In the English literature, 14 bilateral malignant tumours of the parotid gland have been reported with detailed descriptions. In 13 cases, the tumour was the same histiotype of carcinoma bilaterally, while the histology differed in one case.[5] Ten were acinic cell carcinoma[4,6–12] and two were adenocarcinoma.[13,14] To our knowledge, only one previous case of asynchronous bilateral mucoepidermoid carcinoma has been reported[15] and we have described the first case of synchronous bilateral mucoepidermoid carcinomas arising in the parotid gland.

Mucoepidermoid carcinoma is the most common malignant neoplasm observed in the parotid glands, with incidences ranging from 4.2 to 12 per cent of all parotid gland tumours.[2,8,12] Other malignant tumours of the parotid gland are less common. The incidence of acinic cell tumour ranges from three to five per cent of all parotid gland tumours.[1,8] In the parotid gland, bilateral mucoepidermoid carcinoma is much rarer than bilateral acinic cell carcinoma. The parotid gland tumour on one side was not thought to be a metastasis from the other side, because the lymphatic drainage of the parotid glands occurs via the cervical route on each side, and there is no direct connection. Moreover, Spiro *et al.*[9] reported in 1978 that distant metastasis occurred in only 15 per cent of mucoepidermoid carcinomas of the parotid gland, and most metastases were to the lungs, skeleton, and brain.

Based on the histological appearance, mucoepidermoid carcinoma is classified into 'high-grade malignancy' and 'low-grade malignancy', or the clinical and pathologic findings are used to separate the tumours into low-, medium-, and high-grade malignancy groups.[16] Goode *et al.*[17] reported that the mortality in the high-grade group

TABLE I
BILATERAL PAROTID GLAND CARCINOMA

Author	Age	Sex	Histology	Occurrence
Bauer and Bauer (1953)[6]	65	M	Acinic cell carcinoma	synchronous
Diamant *et al.* (1961)[7]	71	M	Acinic cell carcinoma	synchronous
	55	M	Acinic cell carcinoma	synchronous
Enroth (1964)[8]	27	M	Acinic cell carcinoma	asynchronous
Clarke *et al.* (1969)[9]	57	F	Acinic cell carcinoma	synchronous
Turnbull and Frazell (1969)[4]	36	F	Acinic cell carcinoma	synchronous
	60	M	Acinic cell carcinoma	asynchronous
Levin *et al.* (1975)[10]	57	F	Acinic cell carcinoma	synchronous
Nelson *et al.* (1978)[11]	59	M	Acinic cell carcinoma	asynchronous
Di Palma *et al.* (1999)[12]	35	F	Acinic cell carcinoma	asynchronous
Berkeley (1959)[13]	5	M	Adenocarcinoma	synchronous
Ferlito (1978)[14]	67	M	Adenocarcinoma	synchronous
Seifert (1997)[15]	73	M	Mucoepidermoid carcinoma	asynchronous
Hakuba *et al.* (2003) present case	48	M	Mucoepidermoid carcinoma	synchronous
Assor (1974)[5]	58	M	Adenocarcinoma (lt.) Carcinoma in Warthin's tumour (rt.)	asynchronous

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is high, even with aggressive therapy combining total parotidectomy with radical neck dissection and radiation therapy; and the existence of facial nerve invasion is an aggravating factor in mucoepidermoid carcinoma of the parotid gland. Our patient presented with bilateral mucoepidermoid carcinoma synchronously and complained of facial nerve dysfunction bilaterally. The clinical findings were very dramatic and aggressive surgery and post-operative radiotherapy failed to save his life.

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