The Journal of Laryngology & Otology
September 2004, Vol. **118,** pp. 724–726

Metastasizing mixed tumour of the parotid gland presenting as multiple lung metastases

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Abstract
Metastasis is a feature of malignant neoplasms. Although rare, metastasizing pathologically benign mixed tumour has been reported. This report describes a 33-year-old woman with multiple lung metastases from mixed tumour. A small preauricular mass had been removed 23 years earlier. Six years later, the patient had the same symptom; the recurrent mass was resected. She was not aware of the exact pathological diagnosis of the previous tumours. One of the most common features of metastasizing mixed tumour is recurrence at the primary site. Inadequate resection of the primary tumour is often associated with metastasis. Thus, it is important for the head and neck surgeon to be aware of the concept of pathologically benign metastasizing mixed tumour and the importance of adequate surgical procedure.

Key Words: Adenoma, Pleomorphic; Neoplasm, Metastasis; Lung

Introduction

Mixed tumours, being synonymous with pleomorphic adenomas, are the most common tumours, accounting for 40 to 70 per cent of neoplasms arising from both the major and minor salivary glands. The diagnoses of malignant mixed tumour; carcinoma *ex* mixed tumour and carcinosarcoma have been well defined and diagnostic criteria have been established. Rarely, metastasizing mixed tumours showing the pathological features of benign mixed tumour are reported.[1] Here, we present a case of benign metastatic mixed tumour detected as a lung mass and discuss the diagnostic difficulty presented by such a tumour.

Case report

A 33-year-old woman was referred to the otolaryngology clinic for examination of a salivary gland tumour. The patient did not have any subjective symptoms. A medical examination with chest X-ray detected a pulmonary shadow (Figure 1), and a computed tomography (CT) scan of the lung demonstrated multiple nodular lesions. A diagnostic pulmonary resection was performed. The patient was diagnosed as having a metastatic lung tumour, and histological assessment of the biopsy specimen suggested a mixed tumour of salivary gland origin.

The patient was referred to our clinic. Ultrasound scan and CT scan showed a multiple nodular parotid tumour (Figure 2). The patient in fact had a history of preauricular subcutaneous mass resected at another hospital when she was 10 years old and again at 16 years old. However, she was only told that the resected mass was a somewhat atheroma-like tumour. Thus, she did not know whether she had undergone parotid gland tumour resection previously.

Multinodular tumour resection with total parotidectomy was performed. The histological diagnosis of the resected tumour was a typical benign mixed tumour, identical to the metastatic lung tumour. Consequently, the patient was given a diagnosis of metastasizing mixed tumour.

Subsequently, there was no recurrence of the parotid gland tumour and no progression of metastatic lung nodules (for 21 months).

Pathological findings

The biopsy specimens of the lung and parotid gland showed quite similar findings. The tumour was composed of epithelial proliferation with focal squamoid features and glandular formation, clear myoepithelioid cells, and myxochondromatous stroma. Neither necrosis, prominent mitotic activity nor cytological atypia was apparent (Figure 3). These were considered typical features of pleomorphic adenoma or benign mixed tumour. The parotid gland tumour showed multinodular growth, strongly suggesting recurrent mixed tumour in the gland. In the lung, the tumour was well circumscribed, but not encapsulated.

Discussion

The existence and nomenclature of metastasizing mixed tumour has been controversial. Metastasizing mixed tumour is histologically indistinguishable from benign mixed tumour, and the histopathological features of the primary tumour and metastatic tumour are also identical.[1–4] One of the most important features of metastasizing mixed tumour is the association with

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Accepted for publication: 27 April 2004.

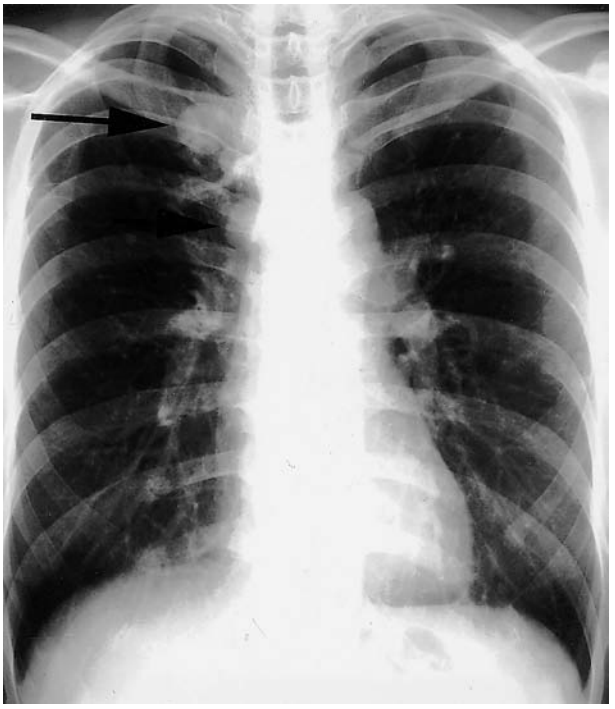


FIG. 1
Chest X-ray at initial diagnosis demonstrating multiple metastatic nodules in the lung (black arrow).

recurrence; metastatic spread of the tumour is usually preceded by recurrent mixed tumour.[1–7]

- **Case report of a pleomorphic adenoma of the parotid, presenting with metastatic disease in the lung**
- **The patient had had preauricular masses excised 17 and 23 years previously, pathology unknown**
- **Previous cases have been reported; this case confirms the potential malignant nature of a disease generally considered to have a benign course**

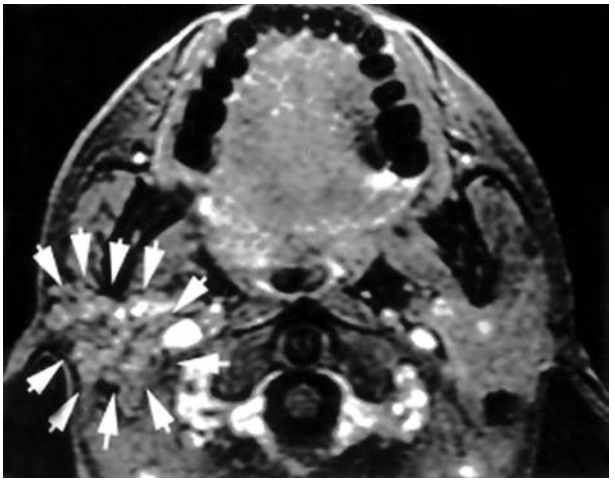
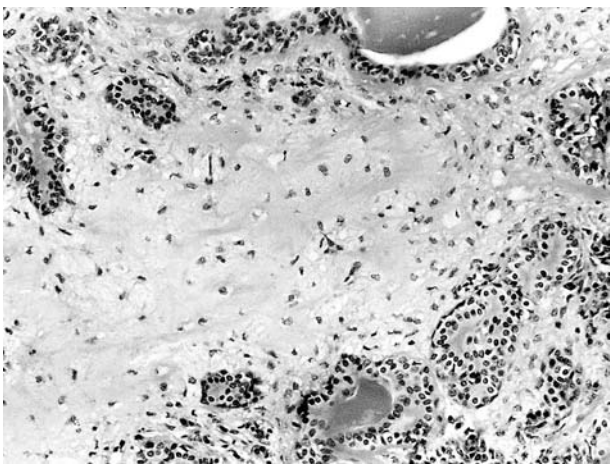


FIG. 2
Gadolinium-enhanced magnetic resonance imaging scan of the primary parotid gland tumour (white arrows indicate the multinodular tumour).

Compared with malignant mixed tumours such as carcinoma arising from benign mixed tumour and carcinosarcoma, the typical clinical course of metastasizing mixed tumour is slowly to moderately progressive.[1] In our case, the growth of metastatic nodules was slow. Common sites of metastasis are lung, bone, and sometimes liver and lymph nodes, suggesting that the tumour tends to metastasize haematogenously rather that lymphatically.[2–9]

Although this type of tumour is considered pathologically benign, metastatic ability is generally a characteristic of malignant tumour. Moreover, relevance of molecular factors such as DNA ploidy, oncogene and tumour suppressor gene with the mechanism of metastatic potential of the tumour has not been elucidated.[8,9] Wenig *et al*. studied their own cases and the relevant literature which contained the information about outcome of the patients after adequate follow-up period. They reported that seven patients died as a consequence of metastatic mixed tumour, resulting in a 22 per cent overall mortality rate.[7] Thus, patients with metastasizing mixed tumour should be treated as having malignant tumour.

This case is quite educational regarding the basic approach to the mixed tumour as well as metastasizing mixed tumour and emphasizes the importance of



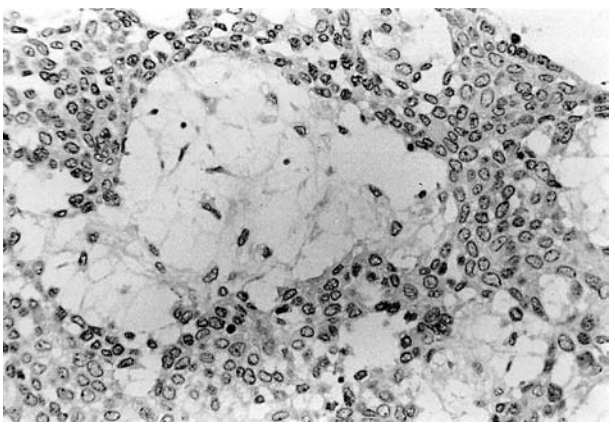


FIG. 3
Section of (a) parotid gland tumour (H&E; –200) and (b) lung tumour (H&E; –400).

https://doi.org/10.1258/0022215042244750 Published online by Cambridge University Press

obtaining an accurate medical history. Lack of accurate diagnostic information about the preauricular mass delayed the patient noticing the metastasizing mixed tumour. Avoiding recurrence of the primary mixed tumour may be the best preventive strategy for metastasizing mixed tumour. The development of metastatic disease should be carefully monitored for a long period, and timely and adequate surgical treatment is required to improve the prognosis and quality of life of the patient.

References

1 Gnepp D, Wenig BM. Malignant mixed tumor. In: Ellis G, Auclair PL, Gnepp G, eds. *Surgical Pathology of the Salivary Glands*. Philadelphia: WB Saunders, 1991:350–68
2 Raja V, China C, Masaki KH, Nakano G. Unusual presentations of uncommon tumors: case 1. Benign metastasizing pleomorphic adenoma. *J Clin Oncol* 2002; **20**:2400–3
3 Hay MA, Witterick IJ, Mock D. Recurrent pleomorphic adenoma of the parotid gland with cervical metastasis. *J Otolaryngol* 2001;**30**:361–5
4 Collina G, Eusebi V, Carasoli PT. Pleomorphic adenoma with lymph-node metastases: report of two cases. *Path Res Pract* 1989;**184**:188–93
5 Czader MC, Eberhart CG, Bhatti N, Cummings C, Westra WH. Metastasizing mixed tumor of the parotid. *Am J Surg Pathol* 2000;**24**:1159–64
6 Wl-Nagger A, Batsakis JG, Kessler S. Benign metastatic mixed tumours or unrecognized salivary carcinoma? *J Laryngol Otol* 1988;**102**:810–12
7 Wenig BM, Hitchcock CL, Ellis GL, Gnepp DR. Metastasizing mixed tumor of salivary glands. *Am J Surg Pathol* 1992;**16**:845–58
8 Jin Y, Jin C, Arheden A, Larsson O, Bauer H, Mandahl N, *et al*. Unbalanced chromosomal rearrangements in a metastasizing salivary gland tumor with benign history. *Cancer Genet Cytogenet* 1998;**102**:59–64
9 Takeuchi E, Shimizu E, Sano N, Yamaguchi T, Yanagawa H, Sone S. A case of pleomorphic adenoma of the lung with multiple distant metastases – observations on its oncogene and tumor suppressor gene expression. *Anticancer Res* 1998;**18**:2015–20

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Dr T. Yoshizaki takes responsibility for the integrity of the content of the paper.
Competing interests: None declared