Acinic cell tumour of the maxillary sinus: An unusual case initially diagnosed as parotid cancer

Toshio Yoshihara, Akio Shino, Miwa Shino, Tetsuo Ishii

Department of Otolaryngology, Tokyo Women's Medical College, Tokyo, Japan

SUMMARY
An unusual case of acinic cell tumour of the maxillary sinus is presented. The patient, a 41-year-old male who had undergone incomplete excision of the tumour in the left parotid region previously, was referred to our department for further treatment. The initial pathological diagnosis was adenocarcinoma of the parotid gland. CT-scan not only revealed tumours in the left pre-auricular and upper neck region, but also an enhanced mass in the left maxillary sinus. Although there were neither nasal symptoms nor destruction of the maxillary bone, aspiration biopsy of the maxillary sinus revealed class V. Total maxillectomy, radiotherapy and systemic chemotherapy were performed just after total parotidectomy and radical neck dissection at the left side. The clinical and histopathological findings are discussed.

Key words: acinic cell tumour, maxillary sinus, parotid gland, metastasis

INTRODUCTION
Acinic cell tumour is one of the malignant neoplasms arising from the parotid gland or the other (both major and minor) salivary glands. It has been reported that although metastases of this tumour are rare, regional lymph nodes, the lungs, and bone are affected when they do occur (Thackray and Lucas, 1974). The most frequently-encountered histological type in maxillary cancer is squamous cell carcinoma. On the other hand, maxillary tumours showing glandular structures (such as adenocarcinoma, acinic cell tumour, adenoid cystic carcinoma, and malignant oncocytoma) are extremely rare. To date, there have been very few reports on acinic cell tumour of the maxillary sinus (Manace and Goldman, 1971; Dimitrakopoulos et al., 1992). It is important for a definite diagnosis to differentiate these tumours from metastatic maxillary cancer from the lung, bronchus, breast and colon (Friedmann and Osborne, 1965; Bernstein, 1966). In this report we present a case of acinic cell tumour of the maxillary sinus which was initially diagnosed as parotid gland cancer.

CASE REPORT
The patient was a 41-year-old male. In 1990, he noticed a small painless mass in the left pre-auricular region, and the tumour slowly enlarged. On November 5, 1992, he underwent incomplete extirpation of this tumour under local anaesthesia at a general surgery clinic. Pathological diagnosis was adenocarcinoma originating from the parotid gland. The patient was immediately referred to our department for further treatment. On admission, the tumours, 1.5 × 2 cm in size in the left pre-auricular region and 2.5 × 4 cm in size in the left upper neck, were palpable. Ultrasonography showed an irregularly shaped and hypo-echoic residual tumour in the subcutaneous region and superficial lobe of the parotid gland (Figure 1a). CT-scan not only revealed the parotid and neck tumours, but also an enhanced mass in the left maxillary sinus. There were neither destruction of the maxillary bone (Figure 1b) nor nasal symptoms.

On November 26, 1992, total parotidectomy and left radical neck dissection were performed. In addition, aspiration biopsy of the left maxillary sinus was done post-operatively, and cytological examination revealed class V. After total maxillectomy on December 11, 1992, a total of 50-Gy irradiation and a combined chemotherapy with carboplatin and 5-fluorouracil were performed.

Pathological examination of all specimen revealed that the tumour was composed of variously-sized and oxyphilic cells, and they were arranged in irregular sheats or nests, and often formed duct-like structures (Figures 2a-c). The tumour of the parotid region showed lymphocyte accumulation as well as tumour cells (Figure 2c). Ultrastructurally, the tumour cells
Figure 1. Ultrasonography (A) of the residual tumour of the pre-auricular region. Irregularly shaped and hypo-echoic mass is found from the subcutaneous region to the superficial lobe of the parotid gland. CT scan (B) reveals an enhanced mass in the left maxillary sinus. Destruction of the maxillary bone is not seen.

Figure 2. The tumour cells are granular and oxyphilic. They are arranged in irregular sheets or nests, and often form duct-like structures. Figure 2a shows the maxillary tumour (×340); figure 2b illustrates the upper neck tumour (×170). In Figure 2c the pre-auricular tumour is shown to contain numerous lymphocytes (arrowheads) as well as tumour cells (P: parotid gland tissue; ×170).

It is known that the primary organs that most frequently metastasize to the nose and paranasal sinuses are the kidney, breast, uterus, and digestive systems (Friedmann and Osborne, 1965; Bernstein, 1966). This case was diagnosed as acinic cell tumour of the maxillary sinus with subsequent pre-auricular and upperneck lymph-node metastasis, because of the following findings: (1) electron-microscopical examinations of the specimens showed typical features of acinic cell tumour; (2) the pre-auricular tumour was located from the subcutaneous region to the superficial lobe of the parotid gland and contained many lymphocytes and tumour cells, indicating the tumour to be a lymph node metastasis; and (3) periodical examinations of the contained numerous secretory granules with low electron density (Figure 3). From these findings, the final diagnosis was acinic cell tumour of the maxillary sinus. Periodical examinations of the lung, kidney, stomach, colon and the other digestive systems have been carried out. To date, neither local recurrence nor distant metastasis have been found.

DISCUSSION
The present case was referred to our hospital for treatment of incomplete extirpation of parotid gland cancer and regional lymph node metastasis. Lightmicroscopically, the initial pathological diagnosis was adenocarcinoma arising from the superficial lobe of the parotid gland. Without cytological examination, it could not be confirmed if the enhanced area of the left maxillary sinus was due to an inflammatory or cancerous lesion. Concerning the relation between the tumours of the maxillary sinus and the parotid gland, the following speculations were discussed with the pathologists: (1) the primary lesion was the maxillary sinus; (2) parotid gland cancer was the primary lesion and then metastasized to the maxillary sinus and the regional lymph nodes; or (3) the primary lesion was another organ and then metastasized to both the parotid gland and maxillary sinus.
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lungs, kidneys, and digestive organs have shown no abnormalities to date.

Acinic cell tumour accounts for approximately 1% of all salivary gland tumours, and 17-19% of all malignant tumours of the parotid gland (Batsakis, 1974). This tumour is often called "acinic cell carcinoma" or "acinic adenocarcinoma", because it behaves in a malignant nature. Eneroth et al. (1966) have described that the 5-year survival rate is 90% and that the tumour metastasizes in 20% of the cases. Rauch et al. (1970) have reported a 5-year mortality of 50%. Frequent metastatic sites are the regional lymph nodes, lungs, vertebrae, brain, and bone (Kleinsasser, 1969; Evans and Cruickshank, 1970; Batsakis, 1974; Thackray and Lucas, 1974).

There have been a number of reports of acinic cell tumour of the parotid gland, while studies on acinic cell tumour of the maxillary sinus have been very few (Manage and Goldman, 1971; Dimitrakopoulos et al., 1992). Acinic cell tumour must be distinguished from the clear-cell variant of mucoepidermoid tumour, clear cell adenoma, metastatic renal-cell adenocarcinoma, and malignant oncocytoma. It is well known that acinic cell tumour is characterized by typical ultrastructural features, i.e. the cells contain numerous secretory granules which are similar to those of the normal parotid gland (Erlandson and Tandler, 1972). Judged solely from light-microscopical studies, several cases of acinic cell tumour of the maxillary sinuses might have been diagnosed as adenocarcinoma. In our case, ultrastructural study was very useful for a definite differential diagnosis.

Figure 3. Electron-microscopically, the tumour cells contain numerous secretory granules (x20,000).

ACKNOWLEDGEMENTS
The authors are grateful to Professor M. Kawakami (Department of Pathology, Tokyo Women's Medical College) for valuable advice. We also thank Miss N. Abo for technical assistance.

REFERENCES

Toshio Yoshihara, MD
Department of Otolaryngology
Tokyo Women's Medical College
8-1 Kawada-cho
Shinjuku-ku
Tokyo 162
Japan