The Journal of Laryngology and Otology
March 1995, Vol. **109**, pp. 240–243

Carcinoma ex pleomorphic adenoma of the soft palate

Toshio Yoshihara, Masayo Tanaka, Mitsuko Itoh*, Tetsuo Ishii

Abstract

A case of carcinoma ex pleomorphic adenoma arising in the soft palate is reported. The tumour presented had enlarged gradually over 10 years and finally occupied the oral cavity. The patient was admitted to our hospital due to disturbance of her speech and swallowing, and a sudden haemorrhage from the tumour. The initial pathological diagnosis by open biopsy was benign pleomorphic adenoma. After total resection, histological examination revealed that the tumour was composed partly of benign pleomorphic adenoma and partly of an adenocarcinomatous component. The carcinoma cells with prominent nucleoli were spheroid or polygonal in shape, and frequently formed ductal structures with areas of necrosis. Mitoses were also found. These findings showed that this tumour was a secondary carcinoma which had developed in a pre-existing pleomorphic adenoma.

Key words: Palate, soft; Salivary gland neoplasms; Adenocarcinoma; Pleomorphic adenoma

Introduction

Carcinoma ex pleomorphic adenoma is one of the uncommon neoplasms of a salivary gland. This tumour is also referred to as 'malignant mixed tumour', carcinoma arising in a mixed tumour, or malignant pleomorphic adenoma. Carcinoma ex pleomorphic adenoma accounts for 3.6 per cent of all salivary gland tumours and 11.7 per cent of all malignant salivary gland tumours (Gnepp and Wenig, 1991). It has been reported that the most frequently affected site is the parotid gland (Bears *et al.*, 1957; Eneroth *et al.*, 1968; Gerughty *et al.*, 1969), while the incidence rates in minor salivary gland are much lower. Gerughty *et al.* (1969) found 73 per cent in the parotid gland, 23 per cent in the submandibular gland and one case in a minor salivary gland of the palate. In this report we present a case of carcinoma ex pleomorphic adenoma arising in the soft palate which had gradually enlarged in size to occupy the oral cavity.

Case report

A 71-year-old female presented with a 10-year history of a tumour, having a smooth greyish surface, in the soft palate. The tumour had gradually increased in size. Although the patient had experienced difficulty in speaking and swallowing, she had been afraid to come to a hospital. She was admitted to our hospital after sudden bleeding from the tip of the tumour. Physical examination showed a hard, large, tumour with necrosis occupying the oral cavity (Figure 1). The pathological diagnosis of the initial biopsy was pleomorphic adenoma.

CT examination showed a 5×6 cm tumour mass in the oral cavity compressing the tongue (Figure 2). MRI examination (T_1-weighted) revealed an inhomogeneous high intensity tumour attached to the soft palate (Figure 3). The tumour was totally resected under general anesthesia after tracheostomy. The tumour had not invaded adjacent tissues nor the neck, and no distant metastases were found. The operated region, with exposed muscle tissues, was covered by fresh epithelium after several weeks.

One year after surgery, no functional problems nor local or distant metastases were found.

Pathological findings

Grossly, the surface of the excised tumour (6×5 by 5×4 cm in size) was solid and greyish-white. The tumour was encapsulated except for the area of haemorrhage (Figure 4). Microscopically, the majority of the tumour consisted of typical pleomorphic adenoma, while malignant areas showed poorly differentiated ade-

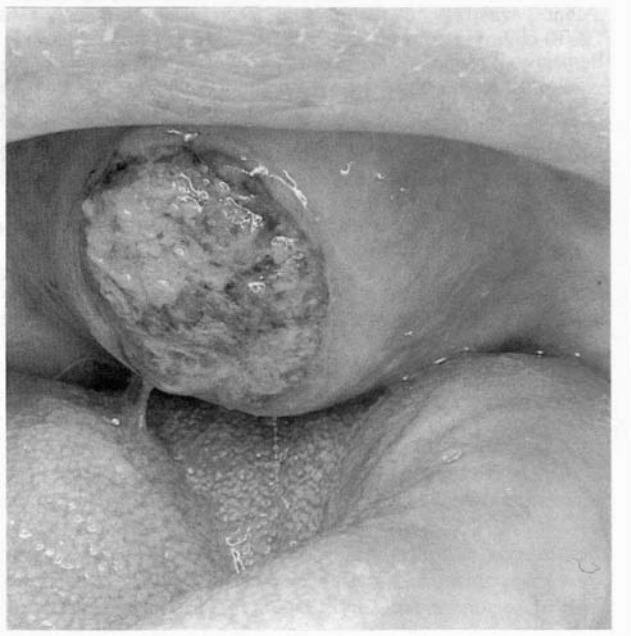

Fig. 1
Large tumour with necrosis occupying the oral cavity.

From the Department of Otolaryngology, Tokyo Women's Medical College, Tokyo, and the Department of Otolaryngology*, Shiseikai Daini Hospital, Tokyo, Japan.
Accepted for publication: 4 December 1994.

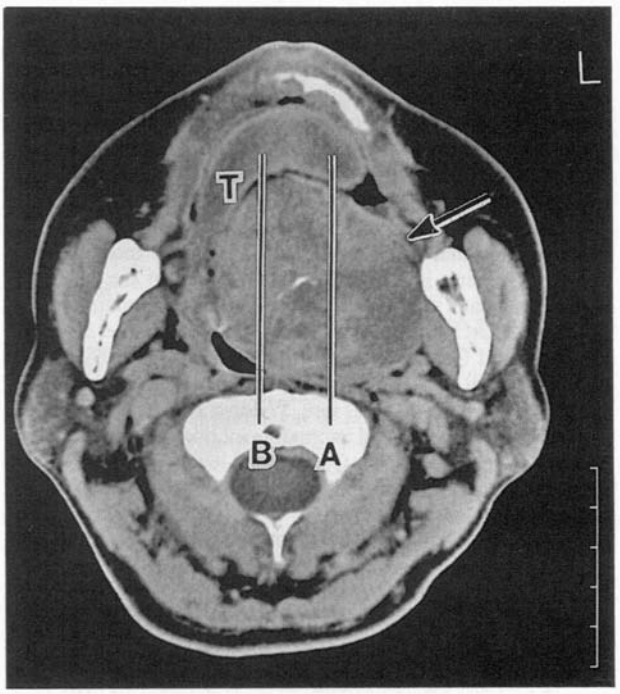

FIG. 2

CT scan showing a 5×6 cm tumour mass (arrowed) in the oral cavity (T: tongue). Lines A and B show areas where cross sections of the specimen were made.

nocarcinoma (Figure 5). The latter was composed of spheroid or polygonal cells with prominent nucleoli and mitoses, and showed ductal structures with comedo-like necrosis (Figure 6). Figure 7 demonstrated small areas of adenocarcinoma in pleomorphic adenoma at two cross-sectional views of the whole specimen (shown by lines A and B in Figure 2). Capsular invasions were found in several areas, but there was no infiltration into neighbouring tissues.

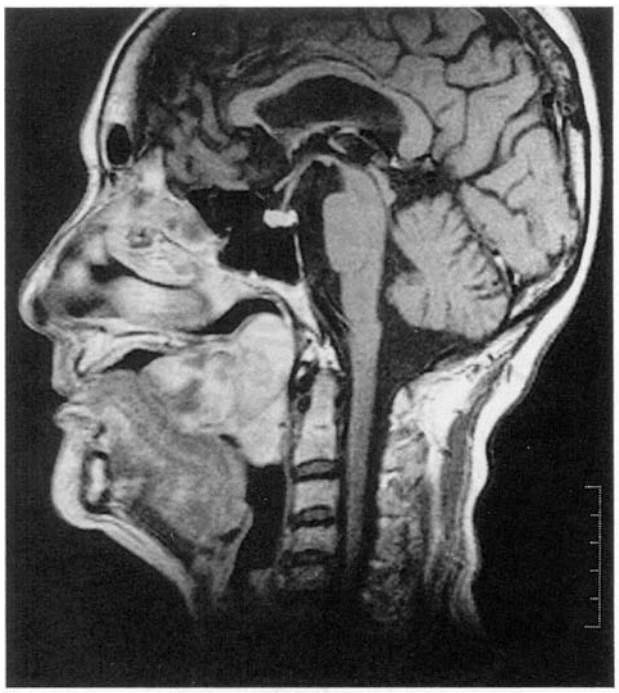

FIG. 3

MRI (T_1-weighted) showing, in sagittal section, an inhomogeneous high density tumour attached to the soft palate.

Discussion

The term 'malignant mixed tumour' has often been used for salivary gland tumours showing both a carcinomatous area and a pleomorphic adenoma. However, the term 'malignant mixed tumour' should be divided into three different clinical and histological entities: (1) carcinoma ex pleomorphic adenoma (carcinoma in pre-existing pleomorphic adenoma); (2) true malignant mixed tumour (carcinosarcoma) (Batsakis, 1974; Livolsi and Perzin, 1977); and (3) so-called benign metastasizing pleomorphic adenoma) (Chen, 1978). Most cases of 'malignant mixed tumour' are carcinoma ex pleomorphic adenoma, while the latter two types are extremely unusual. Thackray and Lucas (1974) showed that the carcinomatous component in carcinoma ex pleomorphic adenoma is most often an adenocarcinoma or undifferentiated carcinoma. Evans and Cruickshank (1970) reported that squamous cell carcinoma is the most frequent type. In our case, the components of poorly differentiated adenocarcinoma were found in several small areas of every section including those demonstrated is Figure 7.

Histological evidence for carcinomatous transformation in pleomorphic adenoma is as follows: (a) unusual destructive and infiltrative growth; (b) abnormal nuclear changes with mitoses; (c) necrosis; (d) haemorrhage; (e) dystrophic calcification; (f) vascular or neural invasion, or both; and (g) local and distant metastases (Gerughty *et al.*, 1969; Rauch *et al.*, 1970; Batsakis, 1974). Nagao *et al.* (1981) also proposed the following criteria for carcinomatous transformation in the parotid gland: (i) capsular invasion; (ii) infiltration into adjacent organs; (iii) proliferation of atypical cells within fibrous tissues and chondroid matrix in the area of pleomorphic adenoma; (iv) vascular involvement; and (v) mitotic figures. The present case also showed necrosis, haemorrhage, capsular invasion and mitoses.

There is no definite sex incidence for this tumour. Foote and Frazell (1953) and Evans and Cruickshank (1970) showed that carcinoma ex pleomorphic adenoma are more common in females than in males, whereas Bears *et al.* (1957) and Eneroth *et al.* (1968) reported the predominance of males. It has been reported that the size of carcinoma ex pleomorphic adenoma is larger than that of other benign tumours (Foote and Frazell, 1953). Nagao *et al.* (1981) showed that 50 per cent of the carcinoma ex pleomorphic adenoma of the parotid gland were greater than 5 cm in dimension. On the other hand, Livolsi and

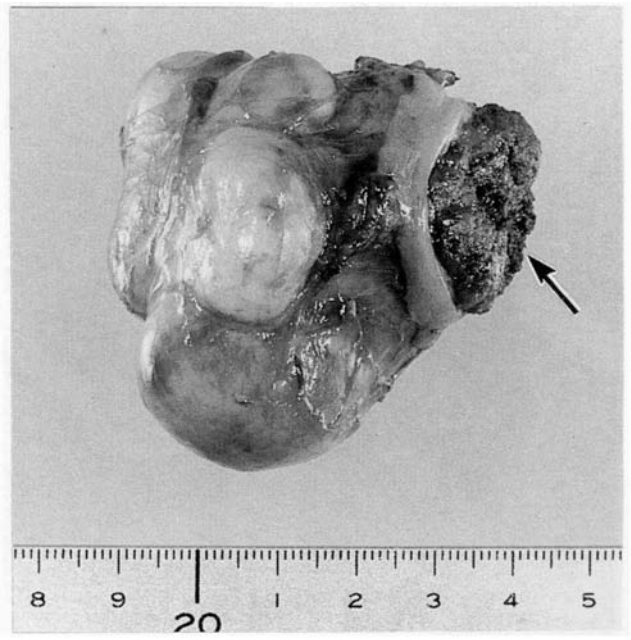

FIG. 4

Excised tumour which is greyish-white with a necrotic region (arrowed).

https://doi.org/10.1017/S0022215100129809 Published online by Cambridge University Press

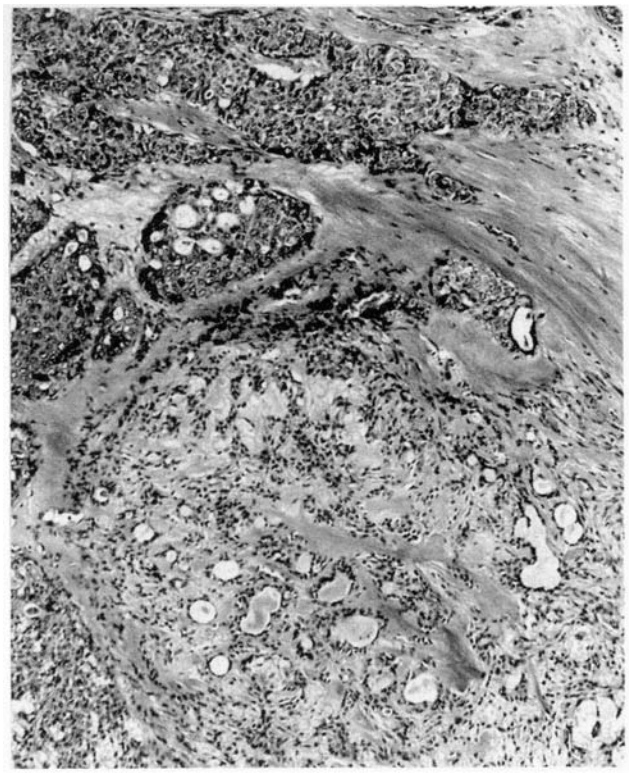

FIG. 5

Tumour consisting of two components: (1) benign pleomorphic adenoma (below); and (2) typical adenocarcinoma (above). (H & E; ×70).

Perzin (1977) stated that this tumour, arising in a minor salivary gland, is smaller than that arising in a major salivary gland.

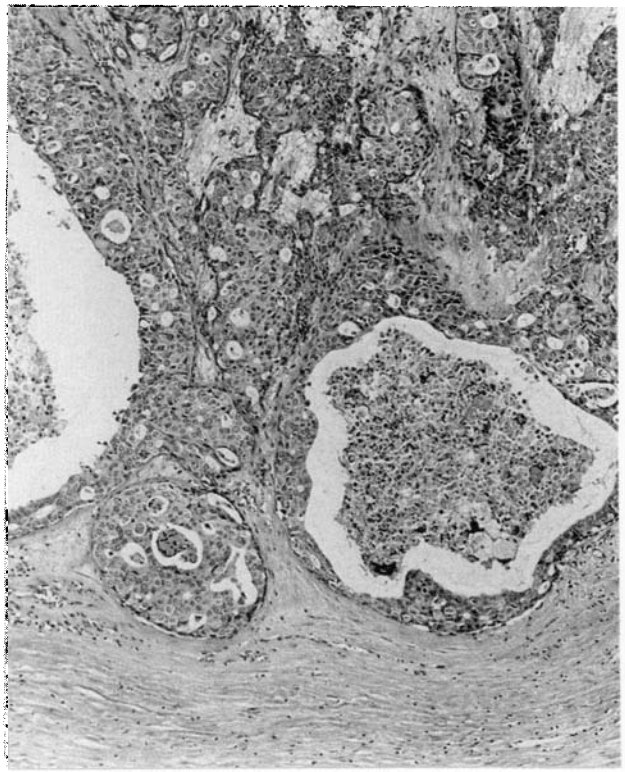

FIG. 6

The adenocarcinomatous component with comedo-like necrosis. (H & E; ×70).

Although our case originated from a minor salivary gland of the soft palate, the tumour had enlarged greatly and finally occupied the oral cavity because of the patient's refusal to consult a doctor until significant haemorrhage appeared. According to the AFIP data of 326 patients with carcinoma ex pleomorphic adenoma, 80 per cent of the tumours occur in the major salivary glands and 17.5 per cent in the minor salivary glands (Gnepp and Wenig, 1991). Furthermore, in the oral cavity, 36 cases (11 per cent of all 326 cases) arise in the palate (Gnepp and Wenig, 1991). It has been reported that pleomorphic adenoma is the most common neoplasm arising in a minor salivary gland of the palate. In malignant palatal tumours, mucoepidermoid carcinoma and adenoid cystic carcinoma are frequently found (Regezi *et al.*, 1985; Da-Quan and Guang-yan, 1987; Waldron *et al.*, 1988). When compared with these tumours, carcinoma ex pleomorphic adenoma arising in the palate is rare. Waldron *et al.* (1988) reported only four cases (described as 'malignant mixed tumour') out of 181 palatal tumours. Da-Quan and Guang-yan (1987) also reported 19 cases out of 160 cases but Regezi *et al.* (1985) did not report any out of 109 cases.

Previous reports showed that metastases of carcinoma ex pleomorphic adenoma ranged from 30 per cent (Moberge and Eneroth, 1968) to 70 per cent (Gerughty *et al.*, 1969). One-third of patients with this tumour of parotid gland origin showed distant metastases according to Foote and Frazell (1953); however there were none out of 11 patients with palatal tumours according to Livolsi and Perzin (1977). Some authors reported that carcinoma ex pleomorphic adenoma of the palate developed high recurrence rates (Spiro *et al.*, 1977), while others reported low recurrence rates (Livolsi and Perzin, 1977). Because the number of cases are very small, there is no distinct tendency as to the rate of recurrence and rate of formation of metastases.

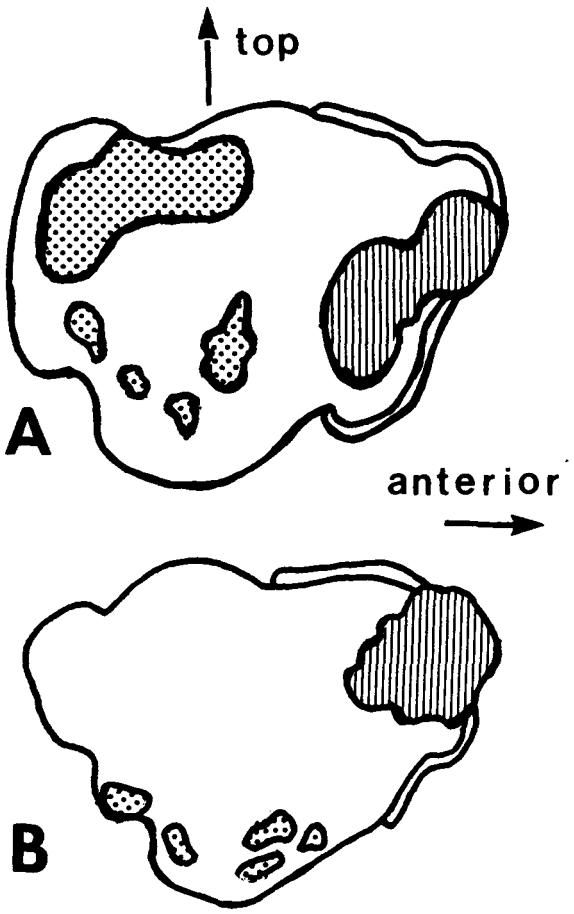

FIG. 7

Cross sections from A and B (see Figure 2) demonstrating the area of adenocarcinoma (dotted area) and pleomorphic adenoma (white area). Stripes demonstrate the area of haemorrhage.

https://doi.org/10.1017/S0022215100129809 Published online by Cambridge University Press

It is generally known that carcinoma ex pleomorphic adenoma shows a poor prognosis in comparison with other malignant tumours of salivary gland origin. Five-year survival rates varied from approximately 25 per cent to 65 per cent (Eneroth *et al.*, 1968; Gerughty *et al.*, 1969; Eneroth and Zetterberg, 1974; Spiro *et al.*, 1977; Boles *et al.*, 1980; Hickman *et al.*, 1984; Spitz and Batsakis, 1984).

To date, there has been neither local recurrence nor neck and distant metastases in our case.

References

Batsakis, J. G. (1974) *Tumors of the head and neck. Clinical and pathological consideration*, Williams & Wilkins Co., Baltimore, pp 1–51.

Bears, O. H., Woolner, L. B., Kirklin, J. W., Devine, K. D. (1957) Carcinomatous transformation of mixed tumors of the parotid gland. *Archives of Surgery* **75:** 605–614.

Boles, R., Raines, J., Lebovits, M., Fu, K. (1980) Malignant tumors of salivary glands: a university experience. *Laryngoscope* **90:** 729–736.

Chen, K. T. K. (1978) Metastasizing pleomorphic adenoma of the salivary gland. *Cancer* **42:** 2407–2411.

Da-Quan, Ma., Guang-yan, Yu (1987) Tumours of the minor salvary glands. *Acta Otolaryngologica (Stockholm)* **103:** 325–331.

Eneroth, C. M., Blanck, C., Jakobsson, P. A. (1968) Carcinoma in pleomorphic adenoma of the parotid gland. *Acta Otolaryngologica (Stockholm)* **66:** 477–492.

Eneroth, C. M., Zetterberg, A. (1974) Malignancy in pleomorphic adenoma: a clinical and microspectrophotometric study. *Acta Otolaryngologica (Stockholm)* **77:** 462–432.

Evans, R. W., Cruickshank, A. H. (1970) Carcinoma ex mixed tumour epithelial tumors of the salivary glands. In *Major Problems in Pathology*. (Bennington, J. L., ed.), W. B. Saunders, Co., Philadelphia, pp 226–241.

Foote, F. W., Frazell, E. L. (1953) Tumors of the major salivary glands. *Cancer* **6:** 1065–1133.

Gerughty, R. M., Scofield, H. H., Brown, F. M., Hennigar, G. R. (1969) Malignant mixed tumors of salivary gland origin. *Cancer* **24:** 471–486.

Gnepp, D. R., Wenig, B. M. (1991) Malignant mixed tumor. In *Surgical Pathology of the Salivary Glands*, (Ellis, G. L., Auclair, P. L., Gnepp, D. R., eds.), W. B. Saunders Co., Philadelphia, pp 350–368.

Hickman, R. E., Cawson, R. A., Duffy, S. W. (1984) The prognosis of specific types of salivary gland tumors. *Cancer* **54:** 1620–1624.

Livolsi, V. A., Perzin, K. H. (1977) Malignant mixed tumors arising in salivary glands: 1: Carcinomas arising in benign mixed tumors: a clinicopathologic study. *Cancer* **39:** 2209–2239.

Moberger, J. G., Eneroth, C. M. (1968) Malignant mixed tumors of the major salivary glands. Special reference to the histologic structure in metastases. *Cancer* **21:** 1198–1211.

Nagao, K., Matsuzaki, O., Saiga, H., Sugano, I., Shigematsu, H., Kaneko, T., Katoh, T., Kitamura, T. (1981) Histopathologic studies on carcinoma in pleomorphic adenoma of the parotid gland. *Cancer* **48:** 113–121.

Rauch, S., Seifert, G., Gorlin, R. J. (1970) Diseases of salivary glands: tumors. In *Thomas' Oral Pathology*, (Gerlin, R. J., Goldman, H. M., eds.), C. V. Mosby Co., St Louis, pp 1003– 1070.

Regezi, J. A., Lloid, R. V., Zarbo, R. J., McClatchey, K. D. (1985) Minor salivary gland tumors. A histologic and immunohistochemical study. *Cancer* **55:** 108–115.

Spiro, R. H., Huvos, A. G., Strong, E. W. (1977) Malignant mixed tumors of salivary origin. A clinicopathologic study of 146 cases. *Cancer* **39:** 388–396.

Spitz, M. R., Batsakis, J. G. (1984) Major salivary gland carcinoma. *Archives of Otolaryngology* **110:** 45–49.

Thackray, A. C., Lucas, R. B. (1974) Tumors of the major salivary glands. In *Atlas of Tumor of Pathology*, Second series, Fascicle, 10, Armed Forces Institute of Pathology, Washington, D.C., pp 107–117.

Waldron, C. A., El-Mofty, S. K., Gnepp, D. R. (1988) Tumors of the intraoral minor salivary glands: a demographic and histologic study 426 cases. *Oral Surgery, Oral Medicine, Oral Pathology* **66:** 323–333.

Address for correspondence:
Dr Toshio Yoshihara,
Department of Otolaryngology,
Tokyo Women's Medical College,
8–1 Kawada-cho,
Shinjuku-ku,
Tokyo 162,
Japan.