

## Pleomorphic adenoma of tongue base causing dysphagia and dysphasia

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### Abstract

A case of an 87-year-old female with pleomorphic adenoma of the tongue base is reported, with a review of the literature. The tumour had enlarged gradually over a period of three years, causing dysphagia and dysphasia. Computed tomography and magnetic resonance imaging showed that the tumour was exophytic and occupy the oropharynx with little extension into the muscle tissue. The tumour was resected by CO<sub>2</sub> laser. Histological examination revealed a benign pleomorphic adenoma that originated from the minor salivary gland of the tongue base.

**Key words:** Adenoma, Pleomorphic; Tongue

### Introduction

It is well known that pleomorphic adenoma is the most common benign tumour arising in the major and minor salivary glands. According to the AFIP (Armed Forces Institute of Pathology) out of data on 6880 cases of pleomorphic adenoma, 5 115 cases arose in the major salivary glands (63.4 per cent in the parotid gland and 9.5 per cent in the submandibular gland), and the most frequent site in the minor salivary gland was the palate (10.3 per cent), while occurrence in the tongue was extremely rare.<sup>1</sup> We report here our experience of a case of pleomorphic adenoma of the tongue base causing dysphagia and dysphasia, together with a review of the literature.

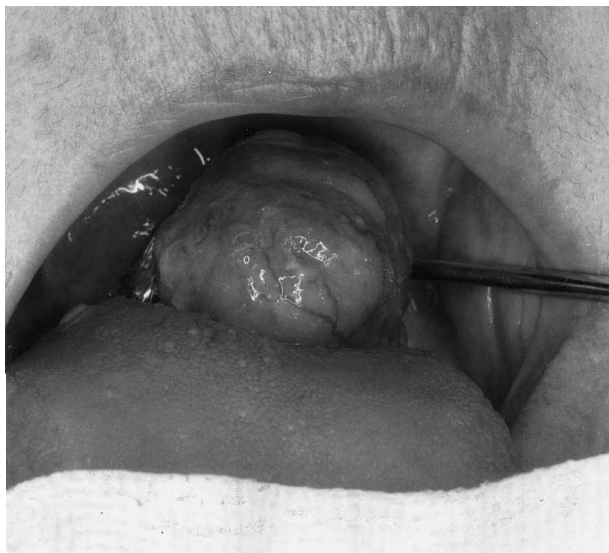


FIG. 1

Large tumour located at the base of the tongue.

### Case report

An 87-year-old female was introduced to our hospital from another hospital on January 20, 1999 because of a three-year history of a tumour at the tongue base causing dysphagia and dysphasia. Physical examination showed that a greyish-white and firm mass was located at the tongue base occupying the oropharynx (Figure 1). No cervical lymph nodes were palpable in the neck. A CT scan showed that the tumour was homogeneous and moderately enhanced (Figure 2). MRI also represented a large round

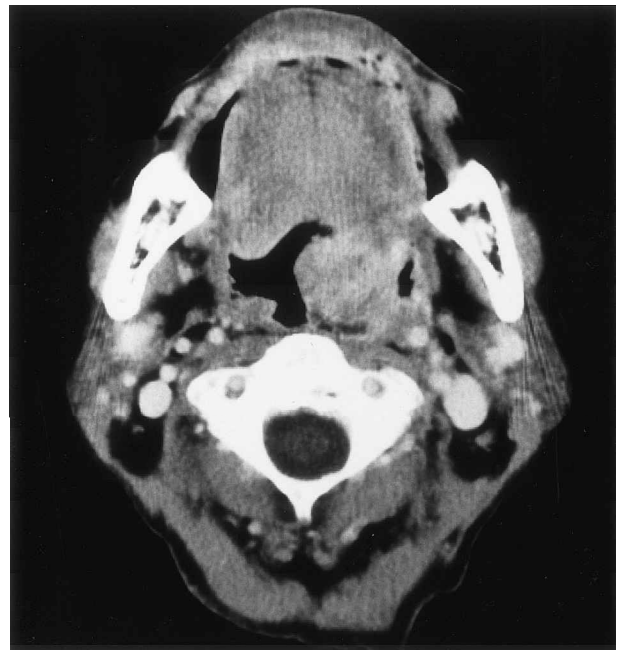


FIG. 2

Computed tomograph scan showed a homogeneously enhanced tumour at the left aspect of the tongue base.

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Accepted for publication: 16 May 2000.

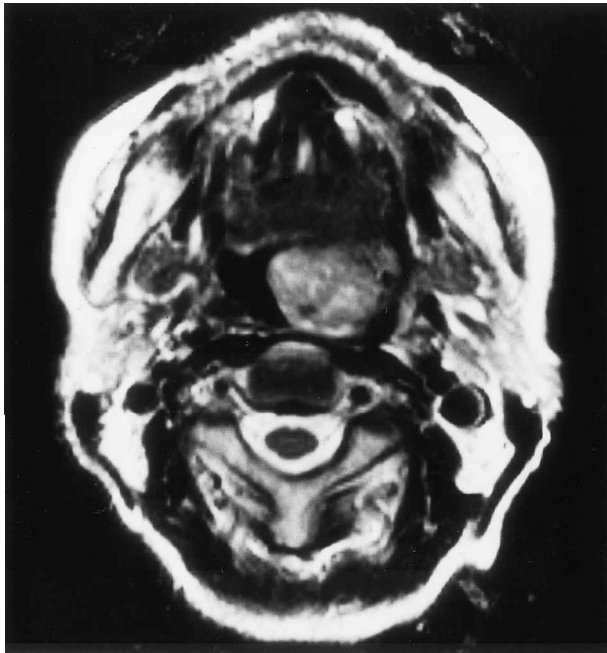
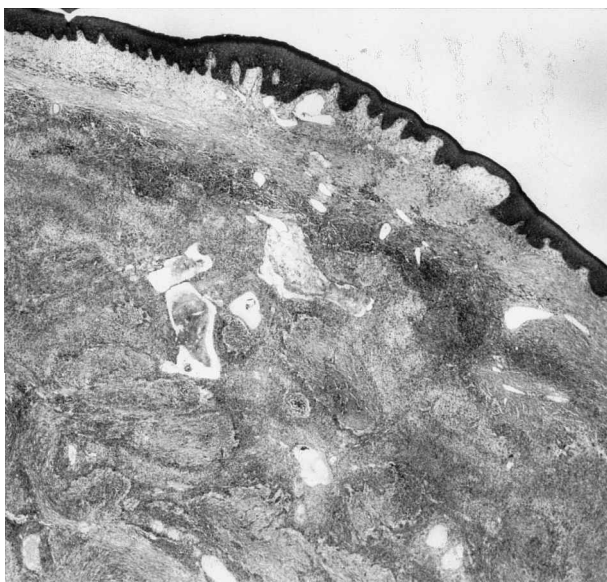


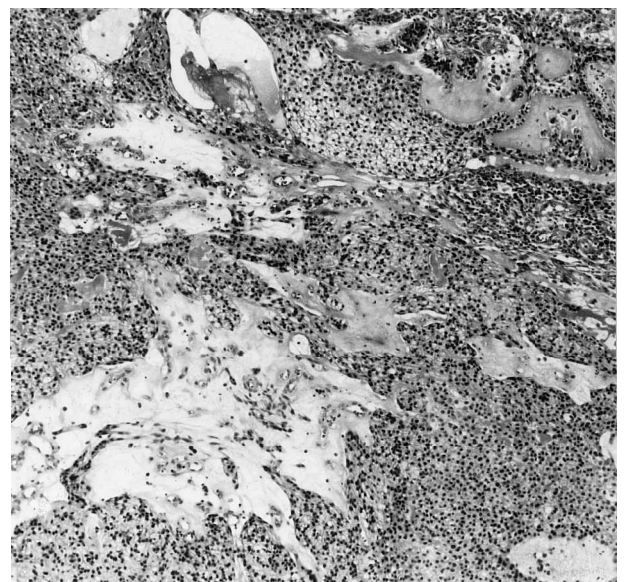
FIG. 3

Magnetic resonance imaging (T2-weighted) showed a tumour with high intensity.

mass on the left side of the oropharynx, which showed a high intensity on the T2-weighted image (Figure 3). Tumour invasion into the surrounding tissues was not observed. Nor was it possible to detect any lymph node metastasis in the neck. The thyroid gland was normally located in the anterior neck. The rest of ENT examinations were normal. On February 26, the tumour was completely resected with the adjacent muscle tissues, approximately 5 mm in width, by CO<sub>2</sub> laser under general anaesthesia by nasal intubation. There was little bleeding during the operation and a tracheostomy was not performed. The excised tumour was well circumscribed and the cut surface was yellowish-white. The tumour was composed of cellular



(a)



(b)

FIG. 4

(a) The tumour was covered with squamous epithelium, and was composed of variously sized cellular nests and solid sheets of cells. (b) The tumour was predominantly cellular with eosinophilic hyaline stroma. Myxoid area was rarely seen. [H&E; (a)  $\times 20$ , (b)  $\times 80$ ].

nests and sheets of polygonal or spindle-shaped cells. Mitotic figures were not seen. Eosinophilic hyaline material was often seen. Immunohistochemically, the cells were positive for cytokeratin and epithelial membrane antigen (EMA), whereas they were negative for actin, desmin, CD31, CD34 and MIB-1. The pathological diagnosis was benign pleomorphic adenoma (cellular type) (Figure 4). The post-operative course was good and there has been neither functional disturbance, nor any signs of recurrence to date.

### Discussion

Pleomorphic adenoma is the most common lesion in the minor salivary gland, however, approximately 50 per cent of all minor salivary gland tumours are malignant.<sup>2</sup> Common sites of minor salivary gland tumours are the palate, followed by the maxillary sinus, lip, cheek, tongue and others.<sup>1-5</sup> In the tongue, malignant neoplasms of salivary gland origin such as adenoid cystic carcinoma, adenocarcinoma, mucoepidermoid carcinoma were predominant over benign tumours.<sup>2,4,5</sup> On the other hand, Chaudhary *et al.*<sup>6</sup> described that pleomorphic adenoma accounted for nearly 56 per cent of all intra-oral minor salivary gland tumours. AFIP data showed the anatomical distribution of 1227 cases of pleomorphic adenoma in the minor salivary gland as follows: 711 palate, 297 lip, 126 cheek, 38 tonsil/oropharynx, 16 tongue and 89 others.<sup>1</sup> Pleomorphic adenoma arising in the base of the tongue is extremely rare. Bardwil *et al.*<sup>4</sup> described only two cases in their review of 100 tumours of minor salivary gland origin. Furthermore, Goepfert *et al.*<sup>7</sup> reported only one case of pleomorphic adenoma in 21 cases with salivary gland tumours of the tongue base. To our knowledge there have been only five reports in the literature (Table I).<sup>7-10</sup> Although some other cases were included in the old series by Frable and Elzay<sup>2</sup> and Bardwil<sup>4</sup> the patients' age, sex and other clinical findings are not described. According to these five reports, most patients were not aware of the tumours until difficulty in swallowing appeared, otherwise they were detected on routine physical examinations by general practitioners.

TABLE I  
REPORTS OF PLEOMORPHIC ADENOMA OF TONGUE BASE

Authors	Year	Age	Sex	Size	Treatment	Symptoms
Goepfert <i>et al.</i> <sup>7</sup>	1976	39	F	?	RTCo60, Surgery	?
Grewal <i>et al.</i> <sup>8</sup>	1984	35	M	4 cm	Surgery	Dysphagia
Deitmer and Stoll <sup>9</sup>	1985	29	M	2 × 3 cm	Surgery	Dysphagia, pain
Banerjee <sup>10</sup>	1987	32	F	2 × 3 cm	Surgery	No symptoms
Magliulo <i>et al.</i> <sup>11</sup>	1996	82	F	3 × 4 cm	Surgery	Dysphagia
Present case		87	F	2 × 3 cm	Surgery	Dysphagia

M = male; F = female.

Patients' ages ranged from 29 to 87 years (average being 50.7 years), and the male-to-female ratio was 4:2. MRI and/or CT scan are necessary for understanding the nature and the submucosal extension of the tumour, but all previous reports except for one<sup>10</sup> showed none of these examinations. The surgical approaches to the base of the tongue varied according to the site and size of the tumour. Generally, the following approaches were recommended: transoral resection, midline transhyoid approach and lateral pharyngotomy. In our case, the tumour could be completely resected transorally because there was little tumour extension into the normal muscle tissue and vallecular area. When the tumour is malignant with extensive invasions into the surrounding tissues, the latter two approaches are recommended. General anaesthesia by nasal intubation is appropriate for this surgery, and tracheotomy is required after the resection when bleeding is marked.

Although the present case showed poor development of the characteristic myxoid and chondroid stroma as compared with that of major salivary gland, pleomorphic adenoma of the minor salivary gland is known to be more cellular with fewer mesenchymal components.<sup>1</sup> In cases of the aged, malignant change in benign tumours of the minor salivary gland, that is, carcinoma ex pleomorphic adenoma must be taken into consideration. According to the AFIP data on carcinoma ex pleomorphic adenoma, 17.5 per cent of the tumour occurred in the minor salivary gland.<sup>12</sup> In our case, the histological examinations showed neither carcinomatous component nor mitotic activity. Immunostaining of MIB-1 as a proliferation marker was also negative. The other immunostainings such as actin, desmin, CD31 and CD34 were performed in order to determine whether the tumour was of epithelial origin, and to differentiate pleomorphic adenoma from glomus tumour. Thyroglossal duct cyst and ectopic thyroid tissue are also important as a differential diagnosis. CT and MRI examinations were available for a pre-operative diagnosis of these diseases. To date, the operated region has been covered by normal epithelium and the patient has no complaint of dysphagia or dysphasia.

#### Acknowledgement

We are grateful to Dr N. Shibata, Department of Pathology, Tokyo Women's Medical University for his valuable advice for this presentation.

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Dr T Yoshihara takes responsibility for the integrity of the content of the paper.

Competing interests: None declared