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Resected tumours of the sublingual gland: 15 years' experience

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Abstract

Sublingual gland tumours are rare, and we have evaluated the clinical features and prognosis of patients treated at a tertiary medical centre in eastern Taiwan. We retrospectively reviewed the cases of nine patients with sublingual gland tumours that were resected from December 1993 to November 2008, four of whom were men and five women. The median (range) age at diagnosis was 52 (39-63) years. Seven had malignant tumours, of which adenoid cystic carcinoma was the most common. All patients with malignant tumours had neck dissections, and four had cervical lymph node metastases. The incidence of lymph node metastases was much higher in patients with advanced primary tumours (T1/2 compared with T3/4: one out of three compared with three out of four). All patients with malignant tumours were given adjuvant radiotherapy. There were no local failures. One patient had regional recurrence in the neck and had a successful further resection. Three patients developed distant metastases, and two died during the follow-up period. Our results suggest that radical resection with postoperative radiotherapy offers adequate local and regional control for malignant sublingual gland tumours. Neck dissection is beneficial, especially for T3/4 disease.

Keywords: Sublingual gland; Sublingual gland tumour; Salivary gland tumour; Adenoid cystic carcinoma

Introduction

Seventy percent of all tumours of the salivary gland are located in the parotid.[1] Sublingual gland tumours are rare, and comprise only 0.5% ∼ 1% of all epithelial salivary tumours and about 1.5% of all carcinomas of the major salivary glands.[2,3] Despite their rarity, most sublingual tumours (80%∼90%) are malignant, with adenoid cystic carcinoma and mucoepidermoid carcinoma being the most common histopathological types.[2,4] Pleomorphic adenoma is the most common benign tumour.[5] Resection is the treatment of choice, but the type of intervention depends on the extent and histopathological type of the primary tumour.[2,4,6,7] Adjuvant radiotherapy is generally recommended for clinically advanced disease, high-grade tumours, or if there are inadequate margins of resection.[2,4]

We have analysed the clinical features and outcomes of treatment in nine tumours of the sublingual gland treated at a tertiary medical centre in eastern Taiwan.

Material and Methods

We made a retrospective review of the nine patients who were newly-diagnosed with tumours of the sublingual gland and

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Table 1
Clinical data, histopathological diagnosis, and staging in nine patients with tumours of the sublingual gland.

Case No	Age (years)	Sex	Size of tumour (cm)	Histopathological diagnosis	TNM stage
1	52	M	2.7	Pleomorphic adenoma	-
2	42	F	3.1	Pleomorphic adenoma	-
3	57	M	2.5	Cribriform adenoid cystic carcinoma	cT2N0M0/pT2N0M0
4	63	F	3.5	Cribriform adenoid cystic carcinoma	cT2N1M0/pT2N2M0
5	43	M	4.2	Cribriform adenoid cystic carcinoma	cT3N0M0/pT3N0M0
6	48	F	3.3	Cribriform adenoid cystic carcinoma	cT2N0M0/pT2N0M0
7	39	F	4.1	Tubular + cribriform adenoid cystic carcinoma	cT3N0M0/pT3N1M0
8	54	M	4.0	Solid adenoid cystic carcinoma	cT4N1M0/pT4N2M0
9	61	F	4.4	High-grade mucoepidermoid carcinoma	cT3N1M0/pT3N1M0

were treated at the Hualien Tzu Chi Hospital, Taiwan from December 1993 to November 2008.

Clinical data were obtained from the patients' medical records. The pretreatment evaluation included chest radiograph, and computed tomography (CT) or magnetic resonance imaging (MRI) of the head and neck. Once malignant neoplasm was confirmed after histopathological examination of the specimen, abdominal sonography and radionuclide whole-body bone scan were also arranged to exclude distant metastases. Patients were restaged according to the 2010 American Joint Committee on Cancer Staging System.

All patients were operated on by a single surgeon (PRC). Intraoperative frozen section biopsy through the floor of the mouth was used to confirm the diagnosis and decide on the extent of resection. If the tumour had invaded the mucosa of the floor of the mouth, we took a punch or incisional biopsy before curative resection. In patients with benign tumours only the sublingual gland, together with the tumour, was removed through an oral approach. In patients with malignant tumours, adjacent structures were also removed to achieve adequate margins according to the extent of the disease (≧1 cm macroscopic soft tissue margin). In general, part of the mandible was removed only when it was attached to or invaded by tumour. Patients with advanced disease, high-grade tumours, close or invaded surgical margins (distance from the tumour to the resection margin less than 4 mm or tumour present at the resection margin), or cervical lymph node metastases, were treated with postoperative radiotherapy. Follow-up data were obtained until December 2014.

Results

There were four men and five women, median age at diagnosis 52 (range 39-63) years. The clinical presentations included swellings in the floor of the mouth (n = 9), pain (n = 3), swelling of the neck (n = 2), numbness of the tongue (n = 2), and oral bleeding (n = 1). The patients with any of the latter four presentations all had malignant tumours. The size of tumour ranged from 2.5-4.4 (median 3.5) cm. The clinical features, histopathological types, and staging are summarised in Table 1.

Two patients had benign tumours and they were treated by resection of the sublingual gland through an oral approach; their wounds were closed primarily. There was no evidence of recurrence after 36 and 48 months.

Seven patients had malignant tumours. The clinical staging was stage II (n = 2), stage III (n = 4), and stage IVa (n = 1) as a result of mandibular invasion based on the imaging study. All patients were classified as M0 at the time of initial management. After final histopathological examination, one patient was upstaged from stage III to stage IVa as the result of the number of lymph node metastases (case 4) (Table 1).

The treatment, histopathological risk factors, and outcome are shown in Table 2. Radical resection was the treatment of choice for malignant tumours, five by combined transcervical and peroral pull-through (cases 3∼7), and two by composite procedures (cases 8 and 9), which included segmental mandibulectomy in one and marginal mandibulectomy in the other. The lingual nerve was sacrificed in six of seven patients and the hypoglossal nerve in two. After resection, the defects of the floor of the mouth were closed primarily in one patient (case 3), reconstructed with a local tongue flap in four (cases 4∼7), and reconstructed with a free flap in two (case 8 = fibular flap and case 9 = radial forearm flap).

All patients with malignant tumours had neck dissection at the time of the primary resection of the tumour, and four had cervical lymph node metastases. The incidence of nodal metastases was much higher in patients with advanced primary tumours (T1/2 compared with T3/4: one out of three compared with three out of four). The histopathological results showed free margins in all patients, with close margins (<4 mm) in two and adequate margins (4 mm or more) in five.

All patients with malignant tumours had postoperative radiotherapy (60∼66 Gy for the primary tumour area, 50∼60 Gy for ipsilateral N0 neck levels I∼V, and 66 Gy for nodes invaded at levels I∼V). The median and mean duration of follow-up from the day of completion of treatment were 84 and 78 months (range 32∼137 months). There were no local failures. One patient (case 4) with a pT2N2 tumour had regional recurrence in the neck at level IIb 24 months after radiotherapy and had a successful further resection. Three patients developed distant metastases, and only supportive care was given. Two patients died during the follow-up