



Adenoid cystic carcinoma of the minor salivary glands: a retrospective series of 29 cases and review of the literature

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Objective. Approximately 78% of minor salivary gland tumors are malignant, of which adenoid cystic carcinoma (ACC) represents 15% in our series. ACC is an uncommon tumor characterized by slow growth and a high potential for recurrence. This series of 29 consecutive patients examines clinicopathologic features, management, and survival outcomes.

Materials and Methods. This study is a retrospective chart review of 29 patients with ACC of the minor salivary glands in a period of 23 years (1989 and 2012).

Results. The mean age was 61.2 years (16-89 years), with no gender predilection. The majority occurred in the palate/maxilla (66%) and initial presentation was stage IV. Mean follow-up was 42.6 months. Recurrence rate was 10% local, 14% distant over the observation period.

Conclusions. The palate/maxilla is the preferred location for occurrence, and initial presentation at stage IV is common. Postoperative radiation remains a common strategy to prevent local recurrence in lesions with adverse features. (Oral Surg Oral Med Oral Pathol Oral Radiol 2016;121:210-214)

Adenoid cystic carcinoma (ACC) of the minor salivary glands is rare and presents as a unique subset of minor salivary gland tumors (MSGTs) that carry with it different characteristics, principles for treatment, and prognosis than ACC of the major salivary glands. The purpose of this study was to review the clinical and pathologic features, management, and survival outcome of ACC of the minor salivary glands in the maxillofacial region.

PATIENTS AND METHODS

This is a retrospective study/chart review of all adenoid cystic carcinomas occurring within a minor salivary gland identified within the University of Maryland Department of Oral and Maxillofacial Surgery Oncology Database in a 23-year period between 1989 and 2012. Inclusion criteria were positive ACC on

histopathologic examination, initial treatment at the University of Maryland, and sufficient data for review. This study was approved by the University of Maryland Institutional Review Board for ethical human research.

RESULTS

A total of 243 minor salivary gland tumors (MSGTs) were identified. Malignant tumors were identified in 189 (78%) patients of this group and 29 (15%) were ACC. The mean age was 61.2 years (range 16-89 years), with no gender predilection. The majority occurred in the palate/maxilla, 66% of cases, followed by the upper lip, lower lip, and buccal mucosa. Of note, lesions identified in the palate were T3-T4 (58.6%), whereas T1-T2 (27.6%) lesions presented in the lips and buccal mucosa (Table I). The most common presenting symptom was intractable pain, of which 67% of patients had perineural invasion (PNI). The incidence of nodal and metastatic disease was uncommon, 3.6% and 7.1% respectively, both with a preference to develop in the presence of a T4 lesion.

All patients underwent surgery as initial treatment, with 61% of patients treated by surgery alone and 38%

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Statement of Clinical Relevance

Adenoid cystic carcinoma of the minor salivary glands is an uncommon tumor with a distinct presentation. Lesions that occur in the palate present at a late stage, T3-T4. Postoperative radiation therapy is a standard treatment strategy for reducing local recurrence.

Table I. Association between initial presentation location and T stage classification

Location	T stage at presentation		Fisher's exact test (P = .05)
	T1-T2	T3-T4	
Upper lip	5	0	.0057
Retromolar fossa	1	0	.4074
Buccal mucosa	2	0	.1567
Palate	2	17	.0001
Lower lip	1	1	.6581

treated with surgery and adjuvant radiation therapy. Resection margins were designed greater than 1.5 cm from the clinical margin of the tumor. Indications for radiation therapy were for positive margins, perineural involvement, T3-T4 size or bone involvement, and evidence of cervical lymphadenopathy. Reconstruction of the defect was completed via local tissue closure/secondary intention in 43.5%, maxillary obturator fabrication 39.1%, and microvascular free flap reconstruction in 17.4%. The radial forearm fasciocutaneous free flap or the fibula osteocutaneous free flap were the reconstruction methods of choice when a free flap was used.

Notable features from pathologic evaluation revealed 60% with positive margins and the presence of perineural invasion in 67% of patients. The incidence of recurrence among patients with a positive margin was 29%, compared with 10% of patients with a clear margin. The presence of PNI reflected a 23% incidence of recurrence, compared with 20% in patients without identified PNI.

Mean follow up was 42.6 months (5-220 months), and no recurrence was observed in 76.3%, local recurrence in 10%, and distant metastasis in 14%. After the acute postoperative phase, with or without adjuvant radiation, patients were followed regularly and received annual imaging in the form of magnetic resonance imaging, computed tomography, and/or positron emission tomography scans to assist in surveillance of the head, neck, and chest. Of note, patients who have undergone microvascular reconstruction have required more frequent imaging studies to assist in the monitoring for local regional recurrence as opposed to patients who have undergone obturation with a removable prosthesis.

Patient survival dependence on tumor recurrence, treatment, margins, location, stage, and T level was tested by χ^2 or Fisher's exact test (P = .05). No statistically significant dependence was found in any relationship; however, this sample size may be too small to be statistically powered. A significant relationship was noted between tumor site and stage of disease with lesions located in the palate presenting at an advanced stage versus other sites of the oral cavity.

DISCUSSION

Malignant MSGTs are rare and account for an estimated 2%-4% of all head and neck cancers.¹ Among minor salivary gland tumors, the proportion that are malignant have been generally described to be 50%, with studies reporting a range from 33% to 90%. The incidence has been described to be 0.9 cases per 100,000 populations for malignant MSGT.² They can occur throughout the upper aerodigestive tract, with the majority (70%) presenting in the oral cavity; other locations include the nasal cavity/sinuses/nasopharynx (25%) and the larynx (3%).¹ Within the oral cavity, the likely areas of presentation are the palate, buccal mucosa, and lip, with approximately 50% presenting on the palate. The highest concentration of minor salivary glands has been described to be located at the junction of the hard and soft palate.^{3,4} Of malignant MSGTs, the most common entities are the adenoid cystic carcinoma and the mucoepidermoid carcinoma, contributing 40%-50% and 30%-40%, respectively.⁵

Originally described in 1859 by Theodor Billroth and referred to as a "cylindroma," the nomenclature was redefined in 1953 as the adenoid cystic carcinoma. It is a malignant salivary gland tumor arising from ductal and myoepithelial cells that have 3 distinct histologic patterns.⁶ These variations include the cribriform, tubular, and solid pattern; tumors that exhibit more than 30% of the solid subtype carry a worse prognosis.^{7,8} The cribriform pattern is the most commonly occurring (46.3%), followed by the solid (38.3%) and tubular (14.9%) variants.^{9,10}

Among cases of ACC, 48%-65% occur in the minor salivary glands.¹¹ The palate is the most common site affected by minor salivary gland ACC and comprises 8.3% of all palatal tumors and 17.7% palatal malignant MSGT.^{9,11} ACC is characterized to have aggressive behavior with a tendency for neurotropism and recurrence. Initial clinical presentation within a minor salivary gland is generally of a slow, firm growing mass with ill-defined margins as a result of an infiltrative submucosal growth pattern. Dysesthesia is common and reflects this tendency for perineural invasion. Compared with this study, we note comparable demographic similarities to the current literature. Common is a wide age distribution, from 16 to 89 years, with a peak incidence in the fifth to sixth decade and no gender predilection.^{12,13} Minor salivary gland ACC of the palate comprised 66% of the patients studied, followed by the lip at 21% (Table I). Current literature would confirm this observation, with the majority of ACC occurring in minor salivary glands and with more than 50% occurring in the palate.^{7,9,14,15} It was noted that T1-T2 lesions were located in the lip, whereas T3-T4 lesions presented

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