Adenoid cystic carcinoma of the tongue: Case report and literature review


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Received: 09/10/2007
Accepted: 06/07/2008

Abstract
Adenoid cystic carcinoma is a common malignant neoplasm originated from salivary glands. The clinical and pathological findings typical of this tumor include slow growth, perineural invasion, and potential local recurrence. It accounts for nearly 2% to 4% of head and neck area tumors. In minor salivary glands it usually affects the palate. We described herein the case of a patient who presented a posterior tongue lesion, which was treated with surgery and radiotherapy as additional treatment. We also carried out a brief literature review on the subject.

Key words: Adenoid cystic carcinoma, tongue, treatment.

Introduction
The Adenoid cystic carcinoma is a relatively rare epithelial tumor of the salivary glands. It accounts for about 5% to 10% of all salivary gland neoplasms, representing 2% to 4% of malignant occurrences of the head and neck area. Approximately 31% of lesions affect minor salivary glands, particularly the palate, though they can also be observed in the submandibular and parotid glands (1).

Although it presents a widespread age distribution, peak incidence occurs predominantly among women, between the 5th and 6th decades of life (2). Typical clinical findings include slow growth, local recurrence, perineural invasion and distant metastasis (3). We present a case of adenoid cystic carcinoma of the tongue, and a brief literature review on its clinical, pathological and therapeutic aspects.

Case Report
A 64-year-old systemically healthy female patient presented to the Division of Stomatology, School of Dentistry at the Federal University of Ceará (Brazil), complaining of an asymptomatic volume growth of the tongue, perceived just one week before consultation. The patient reported a long-term smoking habit, and an otherwise insignificant past medical history. Extra-oral examination and palpation revealed cervical lymph node chain with normal size and consistency. On intraoral clinical exam, a firm sessile nodule of about 4 cm in diameter with a similar coloration to that of the buccal mucosa was observed (Figure 1). Based on patient’s history and clinical exams, associated with a rapid growth of the observed lesion, diagnosis of a malignant neoplasm was hypothesized. An
incisional biopsy was carried out and histopathological analysis demonstrated a neoplasm formed by microcystic spaces rounded by atypical hyperchromatic epithelial cells invading the adjacent muscular tissue, compatible with a cribriform variant of adenoid cystic carcinoma (Figure 2). Subsequently, the patient was sent to the Head and Neck Surgery Division of the Cancer Institute of Ceará (Brazil), where hemiglossectomy and cervical lymph node emptying at the third level were carried out (Figure 3A). Post-surgical radiotherapy sessions were also performed. The patient is currently under a post-surgical 16-month follow-up, showing good health and absence of clinically detected metastasis or local alterations (Figure 3B).
Discussion
In 1859, Billroth used the term “cylindroma” to describe the histological standard of four salivary gland tumors. This term was widely used until 1953, when Foote and Frazell renamed the lesion with the expression adenoid cystic carcinoma (4-6). Evesson and Cawson (7) found a discreet predominance of ACC cases in women (F:M 1.2:1) with ages varying from 24 to 78 years. However, most occurrences took place in the seventh decade of life, average age of 66.3 (+13.1). The presently described case agrees with the observations made by these authors regarding age and gender.

The adenoid cystic carcinoma constitutes approximately 29.6% of minor salivary gland tumors. Although the palate is the most commonly involved site (8), nearly 4.4% of cases are located on the tongue (table 1). In a report by Spiro et al. (9) of 242 salivary gland ACC cases, 171 patients presented lesions involving accessory glands, while 64 patients (26%) presented the palate as the affected site, being the tongue the second most affected area. Isacsson and Shear (10) observed neoplasm occurrences in the palate, floor of the mouth, tongue and gingiva, in this order. Goldbatt, Ellis (11) analyzed 55 salivary gland neoplasms of the tongue, and found that 10% of cases were histologically compatible with ACC. De Vries et al. (12) analyzed 178 cases of salivary gland tumors, out of which only 6 cases were located on the tongue. In these cases differential diagnosis must include benign and malignant tumors related to salivary glands, such as pleomorphic adenoma and adenocarcinoma (8,13).

Treatment of ACC patients has generated great interest and debate among surgeons and pathologists, due to slow clinical development (14). Although it is locally aggressive, with high recurrence levels and late metastasis, commonly leading to patient death between 10 and 20 years after initial treatment (1), compared to other malignant neoplasms, ACC is more difficult to fully remove, with frequently identified positive surgical margins (2).

Table 1. Series of cases of adenoid cystic carcinoma in minor salivary gland tumors.

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Minor salivary gland tumours</th>
<th>Adenoid Cystic Carcinoma</th>
</tr>
</thead>
<tbody>
<tr>
<td>Fine et al.</td>
<td>1960</td>
<td>79 13 (16.5)</td>
<td>2 2.5</td>
</tr>
<tr>
<td>Leafstedt et al.</td>
<td>1971</td>
<td>56 56 (100)</td>
<td>11 19.6</td>
</tr>
<tr>
<td>Spiro et al.</td>
<td>1974</td>
<td>171 171 (100)</td>
<td>26 15.2</td>
</tr>
<tr>
<td>Main et al.</td>
<td>1976</td>
<td>112 28 (25)</td>
<td>3 2.7</td>
</tr>
<tr>
<td>Isacsson, Shear</td>
<td>1983</td>
<td>201 21 (10.5)</td>
<td>2 1</td>
</tr>
<tr>
<td>Evesson, Cawson</td>
<td>1985</td>
<td>336 44 (13.1)</td>
<td>3 0.9</td>
</tr>
<tr>
<td>Huang et al.</td>
<td>1997</td>
<td>48 48 (100)</td>
<td>10 20.8</td>
</tr>
<tr>
<td>Jaber</td>
<td>2006</td>
<td>75 13 (17.3)</td>
<td>3 4</td>
</tr>
<tr>
<td>Wang et al.</td>
<td>2007</td>
<td>737 143 (19.4)</td>
<td>20 2.7</td>
</tr>
</tbody>
</table>

ACC: adenoid cystic carcinoma
Conclusion
The primary treatment objective in adenoid cystic carcinoma patients is local control, normal functionality and distant metastasis prevention. For this purpose, early detection by the surgeon is a requirement, in order to enable a more favorable prognosis and better quality of life. The therapy involving associated radiotherapy remains the modality of choice for most cases.

References