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Adenoid cystic carcinoma of the maxillary sinus: a clinical–pathological report of 10 years of experience from a single institution

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Abstract. Adenoid cystic carcinoma (ACC) is the most common malignant salivary gland tumour of the maxillary sinus. The present study describes 24 cases seen over a period of 10 years at the Brazilian National Cancer Institute. Socio-demographic, clinical, pathological, and follow-up data were retrieved from the medical files for the period 1997–2006. The mean age of the patients was 51.1 years. Twenty-one (87.5%) presented advanced tumours. The main signs and symptoms found were a tumour mass (87.5%), pain (50%), nasal obstruction (25%), and epistaxis (20.8%). Most cases (62.5%) were treated with surgery and radiation therapy. Follow-up data showed two patients (8.3%) with residual disease, local recurrences in four (16.7%) patients, and distant metastasis in five (20.8%). The overall 5- and 10-year survival rates were 72.61% and 62.11%, respectively. Maxillary sinus ACC has an aggressive but indolent behaviour, typically presenting at an advanced T stage that reflects a poor prognosis for patients.

Key words: maxillary sinus; adenoid cystic carcinoma; salivary gland tumours; head and neck cancer; sino-nasal tract.

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Adenoid cystic carcinoma (ACC) is the most common malignant salivary gland tumour in the maxillary sinus and sino-nasal tract in general.[1–5] It also constitutes the second most common non-squamous malignancy affecting the maxillary sinus, accounting for 45–60% of all cases of non-squamous malignancy at this location.[6–10]

Clinically, maxillary sinus ACC (MSACC) has a slow but not indolent behaviour and is associated with local recurrences, late metastasis, and long-term death. Pain, nasal obstruction, and epistaxis are the most frequent complaints and symptoms become worse as the tumour grows.[4,7,10–17]

MSACC, as with ACC of the minor salivary glands, tends to behave more aggressively than its counterparts in the major salivary glands or minor salivary

glands in other locations. Therefore, treatment usually relies on a combination of different modalities, especially surgery and radiation therapy, and even so the prognosis remains poor.[6,8,10,18,19]

Only a few series of MSACC have been described over the past 40 years[3,4,6,10,11,17,20,21] and conclusive information regarding the clinical and behavioural characteristics of these tumours are relatively lacking in the literature. This is reinforced by the fact that MSACC has been studied as part of a major sample of sino-nasal carcinomas, making the interpretation of its individual characteristics difficult.[4,12,13] The present study aimed to describe 24 cases of ACC affecting the maxillary sinuses, all diagnosed during a 10-year period at a single institution in Rio de Janeiro, Brazil.

Patients and methods

All cases of maxillary sinus malignancy occurring during the period 1997 through 2006 were retrieved from the files of the head and neck surgery department of a single institution. Only cases in which the maxillary sinus was shown to be the epicentre of the tumour, as confirmed in the surgical registers, on imaging examination, and/or on pathological analysis, were included in this study.[21,22] Cases in which an antral origin could not be established completely, as well as those that were not diagnosed at the study institution, had incomplete files, or were identified as metastatic to the maxillary sinus, were excluded from the study.

All cases were confirmed histologically in accordance with the latest World Health Organization (WHO) classification of salivary gland tumours, based on routinely stained tissue sections.[9] The histological diagnoses of the cases included in this study were revised by three of the authors (MFA, ALAE, AML), two of whom are general pathologists and the other an oral pathologist. Tumours were clinically restaged in accordance with Sobin and Wittekind's UICC TNM Classification of Malignant Tumours, for nasal and paranasal sinus tumours.[23]

The following data were collected and reviewed: demographic (age, sex, smoking, and alcoholism), clinical (site of origin, size of the tumour, complaints, signs and symptoms, and TNM classification), pathological (histological diagnoses and classification, presence of residual disease, and local recurrence or distant metastasis), imaging (computed tomography (CT) or magnetic resonance imaging (MRI)), and follow-up; the results were described and analyzed. Follow-up was calculated as the time from the first appointment at the institution for the primary sinus tumour to the date of either the last contact (lost patient) or death. All cases were followed up for a minimum of 5 years (patients diagnosed in 2006) or until recurrence or death; the maximum period of follow-up was 16 years.

Overall survival curves for different clinico-pathological factors were constructed using the Kaplan–Meier method and compared by the log-rank test and Cox proportional hazard model. A *P*-value of less than 0.05 was considered significant. The statistical analysis was conducted using Stata statistical software, version 10 (StataCorp LP, College Station, TX, USA).

This study was approved by the institutional committee on human research and was conducted and developed in accordance with international rules for ethics in research.

Table 1. Clinical parameters of the 24 cases of maxillary sinus adenoid cystic carcinoma.

Age, years	Mean	51.1
	Range	30–74
Sex, *n* (%)	Male	12 (50)
	Female	12 (50)
Race, *n* (%)	White	17 (70.8)
	Black	7 (29.2)
Tobacco use, *n* (%)	Yes	6 (25)
	No	18 (75)
Alcohol use, *n* (%)	Yes	5 (20.8)
	No	19 (79.2)
Evolution time, months	Mean	13.5
	Range	1–84
Complaint (%)	Facial swelling	33.3
	Mouth swelling	29.2
	Nasal obstruction	12.5
Signs and symptoms (%)	Tumour mass	87.5
	Pain	50
	Nasal obstruction	25
	Epistaxis	20.8
	Ocular proptosis	20.8
Stage (%)	Initial	12.5
	Advanced	87.5

Results

A total of 38 salivary gland-type malignant tumours of the maxillary sinuses were identified. Histopathology of surgical specimens revealed 24 cases (63.1%) of ACC over an interval of 10 years (1997–2006).

The mean age of the patients was 51.1 years (range 30–74 years). Most patients (17; 70.8%) were Caucasian and the sexes were equally represented. A minority of the patients were smokers (25%) and drinkers (20.8%). The right and left maxillary sinuses were equally affected by ACC in our study.

All patients had complaints at their first appointment related to some sign or symptom and were seeking medical attention for these complaints. The mean duration of complaints was 13.5 months (range 1–84 months). The most common complaint was 'facial swelling' (33.3%), followed by 'mouth swelling' (29.2%) and 'nasal obstruction' (12.5%). The signs and symptoms most frequently recorded were tumour mass (87.5%), pain (50%), nasal obstruction (25%), and epistaxis (20.8%). Also, all patients had more than one sign and/or symptom, with the combination of facial swelling and pain present in most of the cases (41.7%), all of them being cases of advanced disease (Table 1).

Twenty-one (87.5%) patients were diagnosed with MSACC at an advanced stage of disease and only three (12.5%) were diagnosed in the initial stages of disease. Of those patients with advanced stage MSACC, 14 (58.3%) were T4N0M0 and seven (29.2%) were T3N0M0; all three patients with initial lesions (12.5%) were T2N0M0.

On the basis of the extent of the phenotype, the cribriform histopathology subtype was confirmed in 14 (58.3%) cases; six (25%) cases were of the solid subtype, two (8.3%) cases were of the tubular subtype, and in two (8.3%) cases it was not possible to define the histological subtype that was being presented. Of the