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Treatment outcomes of sinonasal adenoid cystic carcinoma: 30 cases from a single institution

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ABSTRACT

Objective: To establish the clinical features and prognostic factors of sinonasal adenoid cystic carcinoma (ACC).
Material and methods: Thirty patients with histopathological diagnosis of sinonasal ACC who were treated at Severance Hospital between 1990 and 2010 were included in this retrospective chart review study.
Results: The 5-year disease-specific survival and disease-free survival rates were 75.3% and 37.2%, respectively. The maxillary sinus (63.3%) and nasal cavity (23.3%) were the most common sites of primary tumour. Most patients were diagnosed with advanced-stage (III/IV) disease (80.0%) and had undergone surgery and postoperative radiotherapy (70.0%). The most common histopathological subtype was cribriform type (40.9%). Local recurrence rate and distant metastasis rates were 26.7% and 23.3%, respectively. The mean time from primary treatment to recurrence was 44.5 months. Sinonasal ACC patients with stage IV and T4 disease had significantly worse survival than those with low stage and T disease. Patients with local recurrence had worse disease-specific survival than those with distant recurrence. Distant metastasis was associated with disease-free survival but not disease-specific survival.
Conclusion: Despite the frequent occurrence of distant metastasis, early diagnosis and effective local control seemed to be the most important factors influencing the survival of sinonasal ACC.

1. Introduction

Adenoid cystic carcinoma (ACC) is the second most common sinonasal cancer, accounting for 10% of sinonasal malignancies. It occurs more often in the minor salivary glands distributed in the oral cavity, nasal cavity, and paranasal sinuses than in the major salivary glands (Matsuba et al., 1986). ACC is the second most common total salivary gland malignancy and the most common parotid gland malignancy (20.6%) (Luksic et al., 2012). High-grade malignant salivary tumours, including ACC, are associated with significantly worse survival (Mercante et al., 2013). General features of ACC are slow progression and frequent local recurrence with distant metastasis that occur in an unpredictable manner. ACC is known to have relatively long-term survival despite the frequent occurrence of distant metastasis (Seifert et al., 1990).

Because ACC of the sinonasal area is a very rare malignancy, many cases of sinonasal ACC have been integrated and reported with other ACCs of head and neck origin. Maso and Lippi (1985) reported that sinonasal ACC has a poorer prognosis compared with other ACCs because of the close proximity to the eyes and brain and difficulty in securing a sufficient resection margin.

The aim of this retrospective chart review study was to identify the clinical characteristics, treatment outcomes, and prognostic factors of sinonasal ACC.

2. Material and methods

Thirty patients with histopathological diagnosis of sinonasal ACC who were treated at Severance Hospital of Yonsei University College of Medicine between 1990 and 2010 were included in this retrospective chart review study. The following information was

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obtained from chart review: sex, age, major symptoms, tumour epicentre, clinical stage, initial treatment modalities, postoperative resection margin, recurrence, recurrence site, time from primary treatment to recurrence, survival duration, and final disease status. All patients were categorized into three groups according to their final disease status: no evidence of disease (NED) group, alive with disease (AWD) group, and death of disease (DOD) group.

Computed tomography (CT) scans, magnetic resonance imaging results, and medical records were used to determine the epicentre of the tumour. Tumours were staged according to the 2012 American Joint Committee on Cancer staging system.

Statistical analyses were performed using SPSS ver. 21.0 for Windows. Treatment outcomes included disease-free and disease-specific survival, and curves describing these outcomes were generated using the Kaplan–Meier product-limit method. Statistical significance was assessed by the log-rank test for equality of survival. A p-value <0.05 was considered statistically significant.

3. Results

3.1. Clinical characteristics of sinonasal ACC

The mean age of patients at presentation was 54.1 years (range, 33–71 years). Among the 30 patients, 19 were female and 11 were male. Similar to previous studies, the number of female patients was almost twice that of male patients.

More than half of all patients complained of nasal obstruction and an intraoral mass. Other symptoms included facial swelling, facial pain, toothache, rhinorrhoea, epiphora, and diplopia (Table 1).

In this study, 26 patients (86.7%) were initially diagnosed at our institution, whereas 1 patient (3.3%) presented with residual disease after prior treatment at other hospitals, and 3 patients (10.0%) presented with recurrent disease (Table 2). The major primary tumour sites were the maxillary sinus ($n = 19$; 63.3%) and nasal cavity ($n = 7$; 23.3%). Other primary tumour sites included the ethmoid sinus ($n = 3$; 10.0%) and sphenoid sinus ($n = 1$; 3.3%). In four cases (13.3%), tumours had invaded the skull base at the time of initial diagnosis (Table 2). Most patients presented with tumours that were classified as T4 ($n = 13$; 43.4%) and T3 ($n = 10$; 33.3%), followed by T2 ($n = 3$; 10%) and T1 ($n = 3$; 10%). Cervical lymph node metastasis and distant metastasis at presentation were found in one case and three cases, respectively. Stage IV disease was present in 46.7% of patients, whereas 53.3% of patients had stage I, II, or III disease (Table 2).

Histopathological types of ACC were classified into tubular, cribriform, cribriform and tubular, or solid subtypes. In this study, specific histopathological subtypes were reported in 22 cases. Nine patients (40.9%) had cribriform type, seven patients (31.8%) had cribriform and tubular type, four patients (18.2%) had solid type, and two patients (9.1%) had tubular type.

Twenty-one patients (70%) underwent radical surgery and postoperative radiation therapy as initial treatment and five patients (16.7%) received surgery alone. One patient each (3.3%) underwent palliative debulking surgery, concurrent chemoradiotherapy, and observation only after refusal of all treatment (Table 3).

Among the 27 cases of patients who underwent surgical resection as initial treatment, 22 cases (81.5%) contained specimens with positive margins and 5 cases (18.5%) contained negative margins. Histopathological diagnosis showed that most positive margins were in the posterior portion of the specimens.

The total recurrence rate was 53.3% ($n = 16$), and the average time from initial treatment to first recurrence was 44.5 months. The mode of recurrence was local recurrence in eight cases (26.7%) and distant metastasis in seven cases (23.3%). All initial distant recurrence occurred in the lung, whereas neck lymph node recurrence was detected in only one case (3.3%). The most common site of distant metastasis was the lung ($n = 7$), followed by the liver ($n = 3$) and bone ($n = 2$) (Table 4).

3.2. Prognostic factors affecting survival of sinonasal ACC

The median disease-specific survival time was 91.0 months, and the 5-year disease-specific survival rate after surgery was 75.3%. The median disease-free survival time was 39.0 months, and the 5-year disease-free survival rate after surgery was 37.2% (Fig. 1).

Table 1
Chief complaints of sinonasal ACC.

Chief complaint	n (%)
Nasal obstruction	11 (30.6)
Intraoral mass	10 (27.8)
Facial swelling	2 (5.6)
Facial pain	4 (11.1)
Epistaxis	3 (8.3)
Toothache	3 (8.3)
Rhinorrhoea	1 (2.8)
Epiphora	1 (2.8)
Diplopia	1 (2.8)

Table 2
Disease characteristics of sinonasal ACC.

Parameter	n (%)
Disease presentation	
Initial	26 (86.7)
Persistent	1 (3.3)
Recurrent	3 (10.0)
Tumour epicentre	
Maxillary sinus	19 (63.3)
Nasal cavity	7 (23.3)
Ethmoid sinus	3 (10.0)
Sphenoid sinus	1 (3.3)
Skull base invasion	4 (13.3)
Primary tumour status	
T1	3 (10.0)
T2	3 (10.0)
T3	10 (33.3)
T4	13 (43.4)
Nodal status	
N0	29 (96.7)
N+	1 (3.3)
Distant metastasis	
M0	27 (90.0)
M1	3 (10.0)
Clinical stage	
I	3 (10.0)
II	3 (10.0)
III	10 (33.3)
IV	14 (46.7)

Table 3
Initial treatment modality for sinonasal ACC.

Treatment modality		n (%)
Operation	Alone	5 (16.7)
	+ Postoperative RT	21 (70.0)
	For palliation	1 (3.3)
CCRT		1 (3.3)
GKS		1 (3.3)
Tx refusal		1 (3.3)

Abbreviations: CCRT, concurrent chemoradiotherapy; GKS, gamma-knife surgery; Tx, treatment; RT, radiotherapy.