

Clinical analysis of sinonasal adenoid cystic carcinoma

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ABSTRACT

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Adenoid cystic carcinoma is a rare sinonasal malignancy. Thus, many cases of adenoid cystic carcinoma have been integrated and reported with other adenoid cystic carcinomas of head and neck origin. General features of adenoid cystic carcinoma are slow progression, frequent multiple recurrences after treatment, and frequent local recurrence and distant metastasis after a long time. Although adenoid cystic carcinoma tends to metastasize to distant organs, it is known to have relatively long survival period. However, studies focusing on sinonasal adenoid cystic carcinoma are lacking owing to its low incidence. We therefore retrospectively reviewed 30 cases of sinonasal adenoid cystic carcinoma

to identify its clinical features and prognostic factors. The median age of the patients from this study was 54.1 years. The 5-year overall survival rate and disease-free survival rate were 75.3% and 37.2%, respectively. The maxillary sinus (63.3%) and nasal cavity (23.3%) were the most common sites of primary tumor. The majority of patients were diagnosed with advanced disease (stage III/IV, 80.0%). The most common histopathological subtype was cribriform type adenoid cystic carcinoma (40.9%). Almost all patients (70.0%) underwent surgery and postoperative radiotherapy. The total recurrence rate was 53.3%, and the local and distant recurrence rates were 26.7% and 23.3%, respectively. The mean duration until recurrence from diagnosis was 44.5 months.

Advanced clinical stage and T stage had the worst survival. Positive resection margin and skull base invasion seemed to be poor prognostic factors, but not statistically proven. Local recurrence had worse overall survival than distant recurrence, and distant metastasis did not affect the overall survival even with frequent distant recurrences. This demonstrates that sinonasal adenoid cystic carcinoma has a long-term survival rate after distant metastasis, and effective local treatment and early diagnosis seem to be most important for the long-term survival.

Key words : adenoid cystic carcinoma, sinonasal tract, prognostic factors

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I. INTRODUCTION

Sinonasal malignancy is relatively rare and squamous cell carcinoma accounts for most of sinonasal malignancy, about 80%. The incidence of sinonasal cancer is reported to be one in 200,000 per year.¹ Adenoid cystic carcinoma (ACC) is the second most common sinonasal cancer and makes up for 10% of sinonasal malignancy. It occurs more than 60% more often in the minor salivary glands distributed in the oral cavity, nasal cavity, and paranasal sinuses than in the major salivary glands.²

Thus, many cases of sinonasal malignancy have been integrated and reported with other ACCs of head and neck origin.

General features of ACC are slow progression, frequent multiple recurrences after treatment, and frequent local recurrence and distant metastasis after a long time. Although ACC tends to metastasize to distant organs, it is known to have relatively long survival period.³

However, sinonasal ACC has higher local recurrence and distant metastasis rate than other ACCs of the head and neck . This is because it is difficult to secure enough resection margins for sinonasal ACC as sinonasal ACC occurs in close proximity to the eyes and brain. Therefore, sinonasal ACC has poorer prognosis compared to other ACCs.⁴

The present study aimed to identify the clinical characteristics, treatment outcomes, and prognostic factors of sinonasal ACC by the retrospective research method.

II. MATERIALS AND METHODS

From 1990 to 2010, 30 patients with histopathological diagnosis of sinonasal ACC treated at Shinchon and Gangnam Severance Hospitals were included in this study. A retrospective chart review was conducted concerning sex, age, major symptoms, epicenter, clinical staging, initial treatment modalities, postoperative safety margin, recurrence and its site, the time to first recurrence, survival duration, and final status. All patients were categorized into three groups according to their final 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 (MRI) results, and medical records were used to determine the epicenter of the tumor. Tumors were staged according to the 2010 American Joint Committee on Cancer (AJCC) staging system.

Statistical analyses were performed using SPSS ver. 19.0 for Windows. The outcomes that were measured included disease-specific survival and overall survival. 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. The association

between the final status of patients and postoperative resection safety margin were statistically analyzed by the Fischer's exact test. A *P* value less than .05 was considered statistically significant.

III. RESULTS

1. Clinical Characteristics of sinonasal ACC

The mean patient age at presentation was 54.1 years (range, 33-71 years). Among 30 patients, 19 were female and 11 were male patients. Similar to previous studies, the number of female patients was almost twice the number of male patients (Table 1).

Table 1. Demographic Data

Parameter	<i>n</i> (%)
Gender	
Male	11 (36.7)
Female	19 (63.3)
Age, y	
Mean	54.1
Range	33-71

Male mean-age: 51.8, Female mean-age: 55.4

More than half of all patients complained of nasal obstruction and oral swelling. Other symptoms included facial swelling, facial pain, toothache, rhinorrhea, epiphora, and diplopia (Table 2).

Table 2. Chief Complaints of sinonasal adenoid cystic carcinoma

Chief Complaint	<i>n</i> (%)
Nasal obstruction	11 (30.6)
Oral swelling	10 (27.8)
Facial swelling	2 (5.6)
Facial pain	4 (11.1)
Epistaxis	3 (8.3)
Toothache	3 (8.3)
Rhinorrhea	1 (2.8)
Epiphora	1 (2.8)
Diplopia	1 (2.8)

In this study, 86.7% of patients were initially diagnosed at Severance Hospitals, whereas 3.3% presented with residual disease after prior treatment at other hospitals, and 10.0% presented with recurrent disease (Table 3).

The major primary tumor sites were the maxillary sinus (63.3% of patients) and the nasal cavity (23.3% of patients). Other primary tumor sites included the ethmoid sinus (10.0%, 3 cases) and the sphenoid sinus (3.3%, 1 case). At the time of initial diagnosis, 13.3% (4 cases) of tumors invaded the skull base (Table 3).

Most patients presented with tumors that were classified as T4 (43.4%) and T3 (33.3%), followed by T2 (10%) and T1 (10%). At presentation to

the Severance Hospitals, there were one case of cervical lymph node metastatic disease and three cases of distant metastatic disease. As a result, 46.7% of patients had stage IV disease and 53.3% of patients had stage I,II, or III disease (Table 3).

Table 3. Disease Characteristics

Parameter	<i>n</i> (%)
Disease presentation at YUMC	
Initial	26 (86.7)
Persistent	1 (3.3)
Recurrent	3 (10.0)
Tumor epicenter	
Maxillary sinus	19 (63.3)
Nasal cavity	7 (23.3)
Ethmoid sinus	3 (10.0)
Sphenoid sinus	1 (3.3)
Extends to skull base	4 (13.3)
Primary tumor 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)

Histopathological types of ACC were classified into tubular, cribriform, cribriform & tubular, or solid subtype. In this study, 40.9% of patients had cribriform type, followed by cribriform & tubular type (31.8%), solid type (18.2%), and tubular type (9.1%) (Table 4).

Table 4. Histopathology of Sinonasal Adenoid Cystic Carcinoma

Parameter	<i>n</i> (%)
Histologic type	
Tubular	2 (9.1)
Cribriform	9 (40.9)
Cribriform&Tubular	7 (31.8)
Solid	4 (18.2)
Margins	
Negative	5 (18.5)
Positive	22 (81.5)

As an initial treatment, most patients (70%) underwent radical surgery and postoperative radiation therapy, followed by surgery only (16.7%). Besides the initial treatment, each patient underwent palliative debulking surgery, concurrent chemoradiotherapy, and observation only after refusal of all the treatment (Table 5).

Table 5. Initial Treatment Modality of Sinonasal Adenoid Cystic Carcinoma

Treatment Modality	<i>n</i> (%)
OP + RTx	21 (70.0)
OP	5 (16.7)
Palliative OP	1 (3.3)
CCRTx	1 (3.3)
GKS	1 (3.3)
Tx refuse	1 (3.3)

Surgical resection margins were evaluated on the basis of the histopathological extent and a majority of resection specimens contained positive disease (22 cases) with minor negative disease (5 cases) (Table 4).

The total recurrence rate was 53.3% (16 cases), and the average time from the initial treatment to first recurrence was 44.5 months, about 4 years. Most of the initial recurrent types were local recurrence in 8 cases and distant metastasis in 7 cases, and all the initial distant recurrence occurred in the lung. Neck lymph node recurrence was very rare in this study (1 case). The lung was the most common site of distant metastasis, followed by the liver (3 cases) and bones (2 cases) (Table 6).

Table 6. Recurrence Features of Sinonasal Adenoid Cystic Carcinoma

Parameter	n (%)
Recurrence rate	16 (53.3)
Mean duration until recurrence from diagnosis	44.5 mo.
Type of 1 st recurrence	
Local	8 (26.7)
Regional	1 (3.3)
Distant	7 (23.3)
Sites of distant metastasis (all)	
Lung	6
Liver	3
Bone	2

Abbreviations: mo., Months

2. Prognostic factors affecting the survival of sinonasal ACC

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

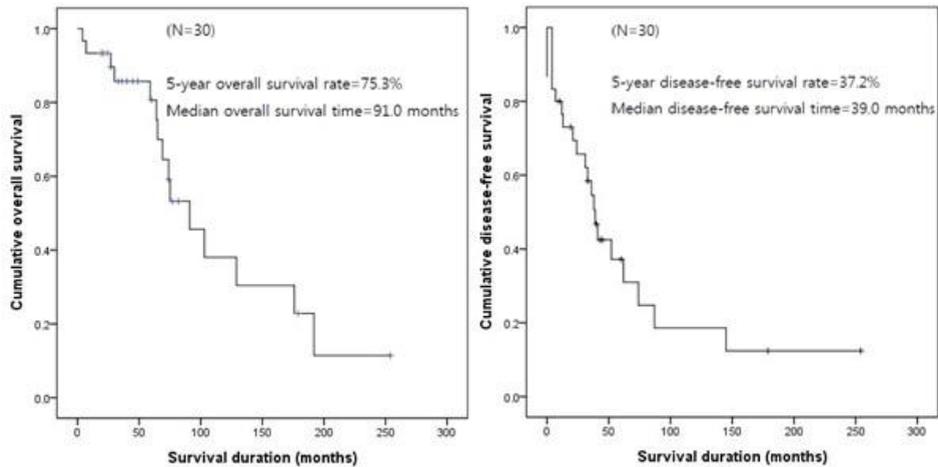


Fig. 1. Overall and disease-free survival rates for all patients with sinonasal ACC. 5-year overall survival rate was 75.3% and 5-year disease-free survival rate was 37.2%.

The time difference between disease-free survival time and overall survival time was about 4 years, and the overall survival rate at 5 years after surgery was relatively higher than 70% although the disease-free survival rates at 5 years after surgery was very low. In other words, patients with sinonasal ACC had the tendency to survive for a long time even after recurrence.

Patients with stage IV disease had worse overall survival and disease-free survival than patients with stage I, II, or III disease ($p=0.027$ and $p=0.001$, respectively) (Fig. 2).

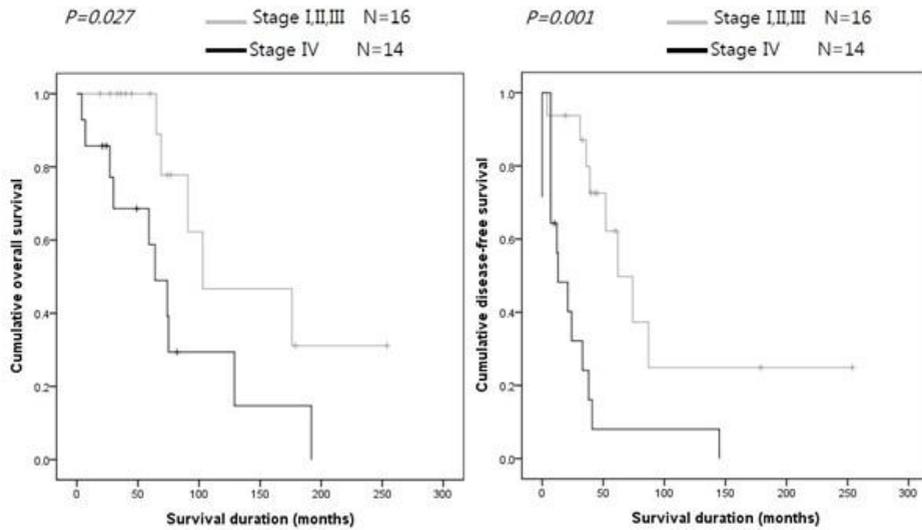


Fig. 2. Overall and disease-free survival rates by clinical stage. Stage IV of sinonasal ACC predicted a poorer prognosis than stage I,II,III ($p=0.027$ and $p=0.001$, respectively).

Also, as a review of the final status of patients by the initial clinical staging, all stage I and II patients except one stage II patient ($n=5$) were in the NED group, and most stage III and IV patients (especially stage IV patients) were in the DOD group and the AWD group (Table 7).

Table 7. Final Status by AJCC Clinical Staging

Final Status	NED				AWD				DOD			
<i>n</i>	9				6				15			
Clinical Stage	I	II	III	IV	I	II	III	IV	I	II	III	IV
<i>n</i>	3	2	3	1	0	0	3	3	0	1	4	10

Similar to the clinical staging, patients with T4 disease had worse overall survival and disease-free survival compared with patients with T1, T2, or T3 disease ($p=0.016$, $p=0.001$, respectively) (Fig. 3).

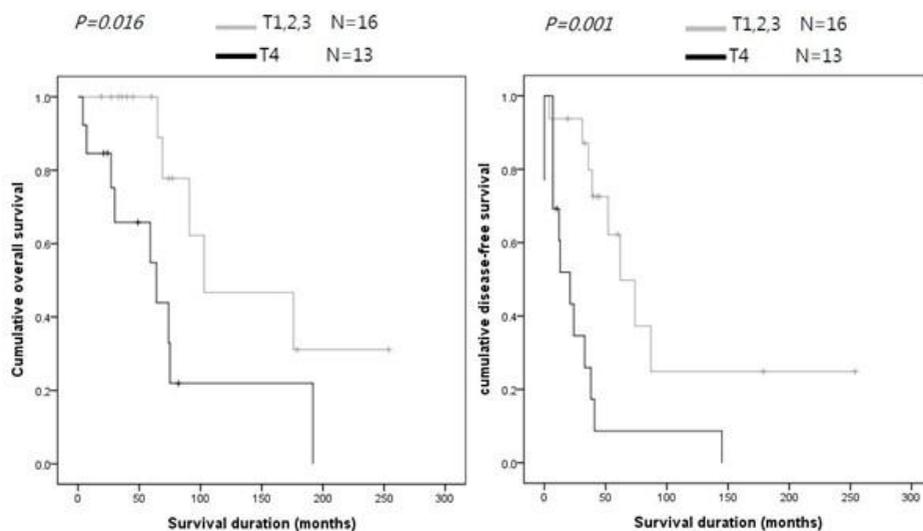


Fig. 3. Overall and disease-free survival rates by T stage. T4 of sinonasal ACC predicted a poorer prognosis than T1,2,3 ($p=0.016$ and $p=0.001$, respectively).

Distant metastasis was not identified as a significant factor that affected the overall survival ($p=0.380$), but as a statistically significant factor that affected the disease-free survival ($p=0.029$) (Fig. 4).

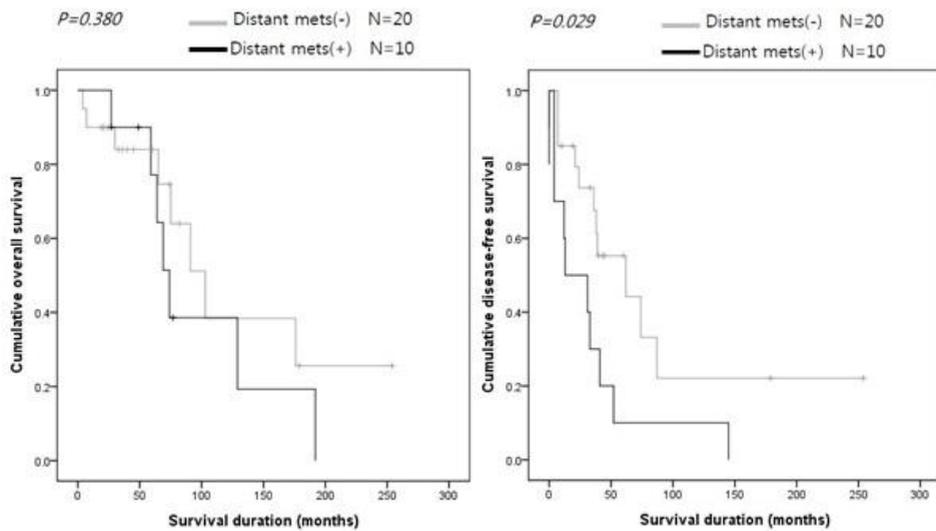


Fig. 4. Overall and disease-free survival rates by distant metastasis. Distant metastasis predicted a poorer prognosis by the disease-free survival rate ($p=0.029$), not by the overall survival rate ($p=0.380$).

When compared, patients with local recurrence had worse overall survival than patients with distant recurrence ($p=0.027$) (Fig. 5).

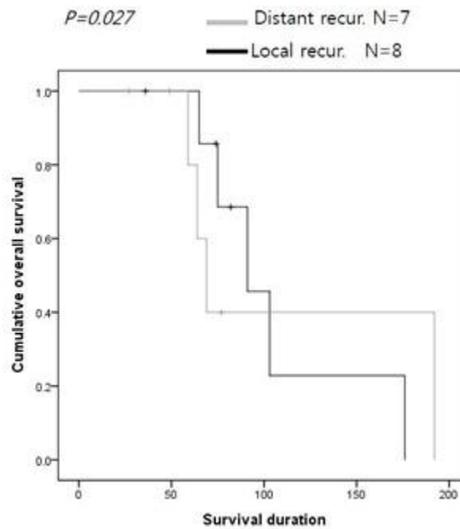


Fig. 5. Overall survival rate by recurrence type. Local recurrences had worse overall survival than distant recurrences ($p=0.027$).

Skull base invasion was not identified as a statistically significant factor for overall survival and disease-free survival compared with no skull base invasion ($p=0.101$ and $p=0.089$, respectively) (Fig. 6). However, if sample size is increased, statistical significance will be expected by comparing with survival curves because this study included only four patients with skull base invasion.

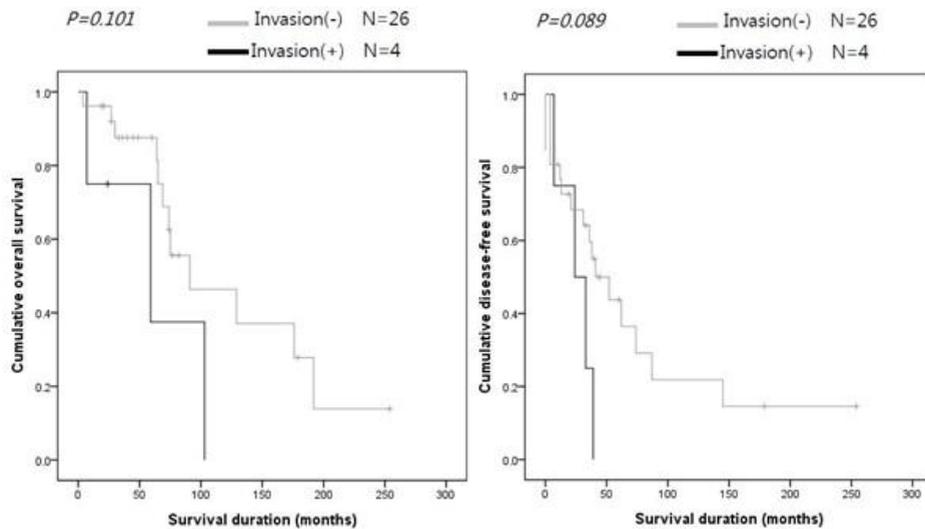


Fig. 6. Overall and disease-free survival rates by skull base invasion. Skull base invasion had no significant worse survival ($p=0.101$ and $p=0.089$, respectively).

Positive safety resection margin was not identified as a statistically significant factor for overall survival and disease-free survival compared with negative safety resection margin ($p=0.082$ and $p=0.128$, respectively) (Fig. 7). The reason might be that the sample size was small and the patient data of negative safety resection margin were almost early-censored. Therefore, by comparison with both survival curves and final status, positive resection margin is assumed to have worse survival than negative resection margin.

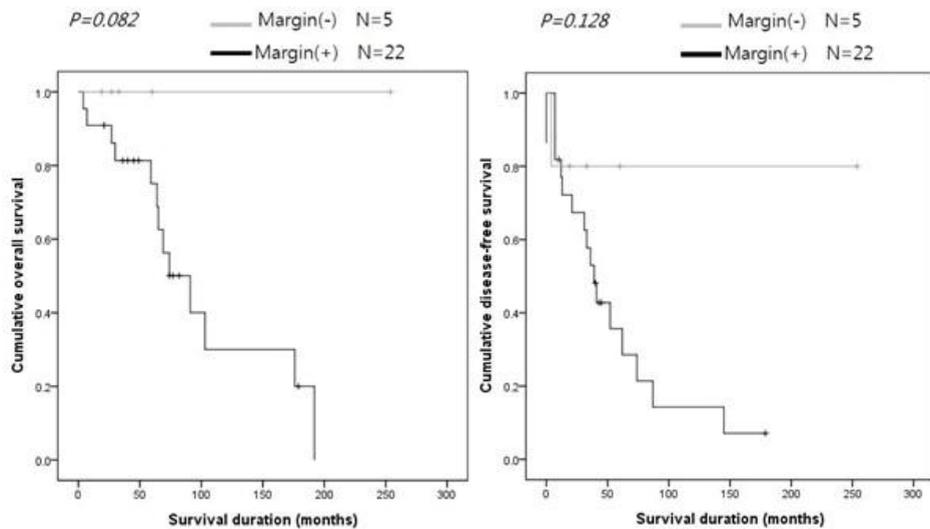


Fig. 7. Overall and disease-free survival rates by safety resection margin. Positive safety resection margin had no significant worse survival ($p=0.082$ and $p=0.128$, respectively).

In comparison to the final status of patients by resection safety margin, all patients with negative safety margins were in NED status, and patients with more than 70% of positive safety margins were in DOD status. Both results were statistically significant ($p=0.014$) (Table 8).

Table 8. Final Status by Safety Margin

		Final Status		Total
		NED	DOD	
Margin	+	5	14	19
	-	4	0	4
Total		9	14	23

Fischer's exact test, $p=0.014$

There was no significant difference in overall or disease-free survival for epicenter, histopathological subtype, and initial treatment modality. Statistical analysis between survival and initial treatment modality was not carried out in this study because almost all patients underwent surgery and postoperative radiation therapy. There was also no significant difference in overall or disease-free survival for sex, age, and recurrence site (local, lymph node, or distant site).

IV. DISCUSSION

Adenoid cystic carcinoma originating from the paranasal sinuses and nasal cavity was first described in 1962 by Tauxe et al.⁵, but not many studies conducted clinical analysis of such ACC and the majority of these studies had a small sample size. Up to now, the incidence of sinonasal ACC was so low that the largest sample size was 105 cases from the United States in 2007, followed by 35 cases from South Korea in 2006.^{6,7} Meanwhile, of all patients with ACC of the head and neck, those whose disease originates from the sinonasal tract had the poorest prognosis.⁸ The reason was that the anatomical structure surrounding the tumor was complex and complete resection of the tumor was difficult because the tumor exhibited frequent local recurrence and early perineural invasion.⁹ The tumor also progressed slowly and had multiple recurrences in organs including its primary site. Distant metastasis was observed even with radical surgery and radiation therapy. Several studies have reported the occurrence of distant metastasis in about 40% of ACC patients.⁶ Nevertheless, many studies reported that patients were alive for more than several years after lung metastasis.^{7,9-11} However, regional lymph node metastasis or recurrence was very uncommon.^{6,7,9}

ACC of the head and neck had a variety of female/male ratio, but female/male ratio of sinonasal ACC was generally reported to be more than 1 to 1.5.^{6,7,9,10} This study also reported that the number of female patients was predominant, about twice the number of male patients (63.3%). The mean age in this study was a sixth decade, which is in agreement with that of previous studies.^{6,7,9,10}

In this study, more than 70% of patients had advanced clinical staging (stage III, IV) and T staging (T3, T4) at diagnosis, and the patients of clinical stage IV and T4 had statistically significant worse overall survival and disease-free survival. This means that earlier diagnosis results in better survival. However, early diagnosis has been difficult so far because sinonasal ACC has a few early symptoms and the majority of manifestations mimic inflammatory diseases such as chronic rhinitis and rhinosinusitis.

Several studies of ACC of the head and neck reported that patients with distant metastasis had poorer prognosis¹¹⁻¹⁴, but only one study reported that distant metastasis of sinonasal ACC was a poor prognostic factor.⁷ On the other hand, many studies of sinonasal ACC reported that distant metastasis was not a significant prognostic factor, which is in agreement with this study. Nevertheless, distant metastasis did not have a

significantly worse overall survival but a significantly worse disease-free survival. Disease-free survival had significant difference probably because of frequent distant metastases of sinonasal ACC. Meanwhile, we may assume that overall survival did not have significant difference because of the long-term survival after distant metastasis. Furthermore, local recurrence had significant worse overall survival than distant recurrence.

Several studies also reported that radical surgery and postoperative radiotherapy provided the best survival rate compared with other treatment modalities^{6,9,10}, but other studies reported the opposite.^{7,15} The largest scale study proposed new prognostic factors that better survival was observed in the nasal cavity compared to other site, with no skull base invasion, with the cribriform subtype, and with lower clinical and T stages. However, unlike other studies, this study showed no significant difference in survival for distant metastasis and local recurrence.⁶ Our study agreed with the newly proposed prognostic factor of clinical and T stages.

This study was not able to compare statistically significant difference in survival for treatment modalities because almost all patients underwent surgery. Because surgery group was relatively large, we compared the

final status of the patients by resection safety margin. As a result, we confirmed that positive resection margin had statistically significant poorer final status (DOD) than negative resection margin. This result may emphasize that radical surgery with enough negative resection margins has favorable outcomes, also known as the major principle of malignant tumor surgery. On the other hand, this result may also emphasize sinonasal ACC was already advanced that patients were unable to have radical surgery with a wide negative margin. Indeed, in comparison with clinical stages by resection margin, patients with positive resection margins had stage I in 1 case, stage II in 2 cases, stage III in 8 cases, and stage IV in 12 cases, whereas patients with negative resection margins had stage I in 2 cases, stage II in 1 case, and stage III in 2 cases. Patients with negative resection margins had no stage IV disease, but almost all patients with positive resection margins had advanced III or IV disease and all patients of stage IV had positive resection margins. However, to exactly compare the significant survival difference for resection margin, we need to compare the survival difference for resection margin in the same clinical stage. Thus, we can solve the dilemma between the importance of radical surgery with wide resection margin and that of conserving anatomical functions for quality

of life.

In addition, there was no significant difference in survival for epicenter, histopathological subtype, and skull base invasion, but skull base invasion had a tendency to have a poorer prognosis when compared to the survival curves according to skull base invasion.

Studies up to now reported that the 5-year overall survival rate of sinonasal ACC was usually 50 to 80%.^{7,11} The 5-year overall survival rate of the current study was 73.5%, and the disease-free survival rate was 37.2%. In other words, the time difference between the disease-free survival time and the overall survival time was about four years. Moreover, patients with local recurrence had worse overall survival than distant recurrence ($p=0.027$). On the basis of this result, when the primary local treatment is carried out successfully, it is possible to increase the survival rate and duration of the disease despite the high distant metastatic tendency of sinonasal ACC. Therefore, we must develop a method to diagnose and treat sinonasal ACC at an early stage without complications and functional loss.

V. CONCLUSION

Sinonasal ACC is a rare tumor that usually presents as late-stage disease. Sinonasal ACCs of advanced clinical stage and T stage had worse survival, and positive resection margin and skull base invasion seemed to be poor prognostic factors, but not statistically proven. Local recurrence had worse overall survival than distant recurrence, and distant metastasis did not affect the overall survival even with frequent distant recurrences. This demonstrates that these tumors have a long-term survival after distant metastasis, and effective local treatment and early diagnosis seem to be most important for the survival.

Despite the clinical implications, clinical reports with large sample sizes are lacking owing to the low incidence of sinonasal ACC. Therefore, in the future, large multicenter studies including a long-term follow-up must be carried out to find new prognostic factors, and the new development of diagnostic and therapeutic modalities or methods will help control the confirmed prognostic factors. Further studies will improve the survival and quality of life of patients with rare sinonasal ACC that has poor prognosis.

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ABSTRACT(IN KOREAN)

비부비동 선양낭성암종의 임상적 분석

<지도교수 김창훈>

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선양낭성암종은 비부비동 악성 종양의 약 10%를 차지하며, 주타액선보다 구강 및 비부비동에 분포하는 소타액선에서 60% 정도로 더 많이 발생한다. 비부비동의 선양낭성암종은 흔하지 않기 때문에 다른 두경부에 발생한 선양낭성암종과 통합하여 보고된 경우가 많았다. 선양낭성암종의 일반적 특징은 성장이 완만하고, 치료 후 다발성 재발이 빈번하며, 오랜 시간이 경과한 후에도 국소재발이나 원격전이를 잘하지만 원격전이가 있는 경우에도 오랫동안 생존을 유지할 수 있다는 보고들이 있었다. 하지만 비부비동에 국한된 임상적 연구가 적어서, 이에 따른 종양의 특징 및 예후인자에 대해서는 아직까지 명확히 밝혀진 것이 없었다. 본 연구의 목적은 비부비동 선양낭성암종 환자 30명을 대상으로 후향적 연구를 통해 그 임상적 특징 및

예후인자를 규명하고자 하는 것이다. 환자들의 평균 연령은 54.1세였으며, 5년 생존률 및 무병생존률은 각각 75.3%와 37.2%였다. 상악동(63.3%)과 비강(23.3%)이 가장 흔한 원발 부위였으며, 80.0%가 진행된 병기인 3, 4기로 진단되었다. 조직학적 아형은 사상형(40.9%)이 가장 많았고, 대부분(70.0%)의 환자는 수술 및 술후 방사선 치료를 초치료로 받았다. 재발율은 53.3%였고, 국소 재발이 26.7%, 원격 재발이 23.3%였다. 진행된 임상적 병기 및 T병기일수록 생존률이 낮은 것으로 나타났으며, 통계적 의미는 확인하지 못했지만 절제연 양성일 경우나 두개저 침범이 있을 경우에 생존률이 나쁠 것으로 예측되었다. 국소 재발이 원격 재발보다 생존률이 낮았다. 또한 잦은 원격 전이에도 불구하고 생존률에는 통계적으로 의미있는 영향은 없는 것으로 나타났다. 이러한 사실은 원격전이 이후에도 비교적 오랜 기간 생존을 유지한다는 것을 보여주는 것으로, 결과적으로 생존율을 높이기 위해서는 효과적인 국소 치료와 조기 진단이 중요하다는 사실을 시사하고 있다.

핵심되는 말 : 선양낭성암종, 비부비동, 예후인자