

Treatment Outcome of Resected and Nonresected Primary Adenoid Cystic Carcinoma of the Lung

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Background and Objective: The incidence of primary adenoid cystic carcinoma of the lung is relatively rare and the optimal treatment strategy is still unclear.

Methods: Sixteen adenoid cystic carcinoma patients were treated at our institute from 1972 to 1998 and their clinical features, treatments and survivals were reviewed.

Results: Half of all patients were female and the median age was 46 ranging from 30 to 64. All primary lesions were located in the central bronchial tree and 80% of the patients had some symptoms. Eleven patients underwent a resection of the tumor with/without plasty of the trachea or bronchus. Although 6 (55%) of 11 patients had a microscopic residual tumor after resection, 5 patients who received postoperative radiotherapy survived without recurrence from 3 to 17 years. Five patients received radiotherapy as their initial treatment and all tumors responded well to the treatment. The 5-year and 10-year survival rates were 91 and 76% without local recurrence in the resected group and 40 and 0% in the nonresected group, respectively.

Conclusion: These observations suggest that surgical resection should be selected first whenever possible, and, in addition, adenoid cystic carcinoma is sensitive to radiotherapy. (*Ann Thorac Cardiovasc Surg* 2002; 8: 74–7)

Key words: adenoid cystic carcinoma of the lung, resection, radiotherapy

Introduction

Primary adenoid cystic carcinoma of the lung is a relatively rare salivary gland-type malignant neoplasm which originates in the bronchial gland.¹⁾ The speed of growth is slow and the clinical course is relatively long, so it is generally considered to be a low grade-malignancy.

To our knowledge, there have so far been few reports discussing the treatment modalities and its outcome in both resectable and unresectable patients. Therefore, the optimal treatment for patients with marginal lesions that are considered to be between resectable and unresectable tumors remains unclear.

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In the present study, we investigated the clinical features, treatment modalities and outcomes of 16 patients treated at our hospital to determine the optimal treatment strategy for adenoid cystic carcinoma.

Patients and Methods

Since 1972, we have treated 2,846 primary pulmonary carcinomas histologically and/or cytologically diagnosed in our institute. They include 1,402 adenocarcinomas, 832 squamous carcinomas, 305 small cell carcinomas, 206 large cell carcinomas, 65 adenosquamous carcinomas, 16 adenoid cystic carcinomas (0.56%), 10 mucoepidermoids, 6 carcinoids, and 4 carcinosarcomas. We retrospectively reviewed the files of 16 patients with adenoid cystic carcinoma. In addition to these 16 patients, we also found three patients diagnosed to have adenoid cystic carcinoma of the peripheral lung. However, these three patients were excluded from the present analysis for primary pulmonary adenoid cystic carcinoma because adenoid cystic

Table 1. Characteristics of adenoid cystic carcinoma patients

Characteristics	No.
Gender: male/female	8/8
Age: median (range), years	46 (30-64)
PS: 0-1/2/3	15/0/1
Smoking: +/-	7/9
Symptoms	
None	3 (19%)
Stridor/wheeze	4 (25%)
Fever	2 (13%)
Chest pain	2 (13%)
Hemosputum	2 (13%)
Others	3 (19%)
Locations	
Trachea	5 (31%)
Main bronchus	2 (13%)
Truncus intermedius	1 (6%)
Lobar bronchus	3 (19%)
Segmental bronchus	5 (31%)
Periphery	0

PS=performance status

carcinoma in the salivary gland was found in them after a resection of the pulmonary lesion.

The 16 patients were divided into two groups consisting of a resected group (n=11) and a nonresected group (n=5). The clinical features, recurrent pattern and survival were all investigated in each group.

Survival was calculated by the Kaplan-Meier method, and a comparison of survival curves was made by the log-rank test. The data were considered to be significant when the p value did not exceed 0.05.

Results

As shown in Table 1, eight patients were males and eight were females ranging in age from 30 to 64 years (median; 46 years). Fifteen patients (94%) were determined to be Eastern Cooperative Oncology Group Performance Status (PS) 0-1, and only one patient with PS 3 suffered from severe pneumonia. Seven patients (44%) were smokers with a mean of 37 pack years ranging from 11 to 80. Thirteen patients (81%) were symptomatic. Four patients who complained of stridor/wheezing were first treated as having bronchial asthma. The median duration from the appearance of symptoms to diagnosis was 15 months ranging from one week to 5 years. All the primary lesions were located in the trachea and central bronchi. The median maximum diameter of the primary tumor was 40 mm ranging from 20 to 80 mm.

Table 2. Stages of the resected and nonresected groups

	Resected group (n=11)	Nonresected group (n=5)	Total (n=16)
Clinical stage			
I	3	0	3
II	3	0	3
IIIA	1	1	2
IIIB	4 (36%)	4 (80%)	8 (50%)
Pathological stage			
I	2		
II	1		
IIIA	1		
IIIB	7 (64%)		

The histological subtypes [2, 3] of the adenoid cystic carcinoma were retrospectively examined. Adenoid cystic carcinoma with a cribriform pattern and with a mixture of cribriform and tubular patterns was observed in 10 and 6 patients, respectively. No solid pattern was observed in our series.

The clinical and pathological stages of the resected and nonresected group are shown in Table 2. Eight patients in clinical stage IIIB showed tracheal invasion of the primary tumor. None of the patients were stage IV at the time of diagnosis.

In the resected group, a single lobectomy of the lung was performed in three patients, a pneumonectomy in five, and a tracheal resection in three. All the patients underwent an ordinary lymph node dissection. Of the patients with a lobectomy, one received bronchoplasty (sleeve upper lobectomy). In 5 patients with a pneumonectomy, 3 underwent carinal resection.

There were seven pathological stage IIIB patients with a tracheal invasion of the primary tumor. Ten of these 11 resected patients (91%) had no histological evidence of lymph node metastases. Six patients (55%) had a microscopic malignant tumor at the tracheal or bronchial stump (Table 3). Five of the 6 patients with microscopic residual tumors received locoregional radiotherapy with the dose ranging from 50.0 to 61.2 Gy after operation.

The nonresected group consisted of five patients. The reasons for unresectability were an extensive tracheal invasion of the tumor in four patients and a poor pulmonary function for a pneumonectomy in one patient. Radiotherapy alone was selected as the initial treatment. A median irradiated dose was 60.0 Gy ranging from 50.0 to 70.0 Gy. The tumor of all patients showed a remarkable radiosensitivity including 1 complete response and 4 partial responses.

Table 3. Operative procedure and microscopic residual tumors at the trachea or bronchial stump

Operative procedure	Microscopic residual tumor	
	(+)	(-)
Lobectomy (n=3)*	0	3
Pneumonectomy (n=5)**	4	1
Tracheal resection (n=3)	2	1
Total	6	5

*One patient underwent a sleeve lobectomy.

**Three patients underwent a sleeve pneumonectomy.

The survival curves of the two groups are shown in Fig.1. The 5 year and 10 year survival rates were 91% and 76% in the resected group and 40% and 0% in the nonresected group, respectively. Five patients with microscopic residual tumors who received postoperative radiotherapy survived without recurrence 3, 3, 6, 12, and 17 years after operation, respectively.

In the resected group, three patients died. One patient died of multiple pulmonary metastases which appeared two years after the operation, while another died of old age 17 years after the initial treatment, and the other died of operative complications (pyothorax). On the other hand, all patients died in the nonresected group. The causes of death were uncontrollable local regrowth of the tumor in three patients and multiple brain metastases or small cell carcinoma in each patient. The latter two patients also developed a local regrowth of the primary tumor before death.

Discussion

The incidence of tracheal tumors is less than 0.2 per 100,000 people per year,⁴⁾ and about half of them appear to be adenoid cystic carcinoma. In the present study we found the incidence of adenoid cystic carcinoma of the lung among all primary pulmonary tumors (2,846 cases) to be 0.56% (16 cases). Most of our patients had a good performance status on admission, spending a lengthy time to reach the diagnosis after their symptoms had appeared. The symptoms are usually due to chronic bronchial obstruction or irritation, averaging two years' duration before diagnosis in reported cases.^{4,5)} In the present study, 81% of the patients had symptoms such as stridor or hemoptysis and the median duration before diagnosis was 15 months.

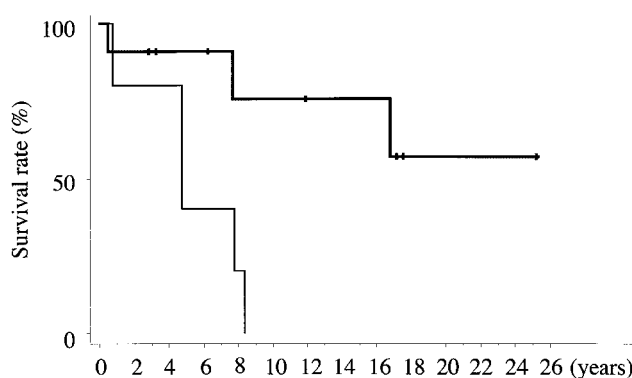


Fig. 1. Survival curves of the resected group and nonresected group.

(—) Resected group (n=11), (---) nonresected group (n=5). Each tick mark represents a patient who is alive.

Adenoid cystic carcinoma seems to possess radiosensitivity. Maziak et al.⁵⁾ reported that patients who initially received radiotherapy alone had a mean survival time of 37 months, ranging 4 months to 7 years. Grillo⁶⁾ also reported the median survival time of irradiated patients to be 28 months. In the present study, all five patients responded to radiotherapy and the median survival time was 5 years. However, all irradiated patients eventually developed a regrowth of the primary tumors which turned out to be a direct cause of death in three patients.

A surgical resection has been thought to be the most favorable procedure to control localized lesions. The median survival time of resected patients was reported to range from 7.5 years to 118 months.⁵⁻⁷⁾ However, adenoid cystic carcinomas, because they tend to infiltrate along the airways, are often incompletely resected and thus are prone to recur locally. The complete resection rates in reported cases ranged from 42 to 57%. Pearson et al.⁷⁾ reported that nine of the 26 adenoid cystic carcinoma patients who underwent an operation resulted in an incomplete resection and seven of the nine died of local failure. In the present study, six (55%) of 11 resected patients had microscopic residual tumors at the surgical margin and five patients with residual tumors received postoperative radiotherapy. So far, there has been no local recurrence and the survival of the resected patients compares favorably with previous reports.

In conclusion, a surgical resection including an extensive resection, such as a carinal resection, should therefore be selected first whenever possible. In addition, radiotherapy should be considered in patients with either an incomplete resection or none at all.

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