

56세된 여자의 하악 선양낭성암 환자 증례보고

박희용, 조경효, 손창규, 조종관
대전대학교 부속한방병원

Adenoid Cystic Carcinoma of the Submandible in a 56-Year-Old Woman

Hee-yong Park, Jung-hyo Cho, Chang-gue Son, Chong-kwan Cho

Department of Internal Medicine, College of Oriental Medicine, Daejeon University

연구목적 : 선양낭성암은 모든 두경부 종양에서 불과 1% 미만의 희소암으로, 본 논문에서는 하악 선양낭성암 기원의 간에 전이성 거대 종양을 가진 증례를 통해 선양낭성암에서의 한방 치료의 유효성평가와 함께 이에 대한 지속적인 연구를 유도해내는 계기로 삼고자 한다.

연구방법 : 본 연구에서는 한약물치료, 침치료, CT검사 및 혈액학적 검사를 포함한 모든 치료와 검사결과를 추적 조사하였다. 치료로 청간탕과 화침법을 사용하였으며, 청간탕은 1일 3회 복용, 화침은 월 1회 시술하였다. 또한, 혈액학적 검사는 월 1회 하였으며, CT검사는 19개월간의 치료기간 중 2회에 걸쳐 수행되어졌다.

연구결과 : 재검사시의 CT소견과 내원당시의 CT소견을 비교하였을 때, 큰 변화가 없이 잘 유지하였으며, 비록 일부의 혈액학적 검사소견이 약간 악화되었지만, 환자의 주증상은 빠르게 경감되었다.

결론 : 본 증례는 진행성 선양낭성암 환자에게서 한방치료가 삶의 질 개선과 생존을 향상을 위한 고식적 목적의 치료에 유용하게 사용되어질 수 있는 가능성을 제시해 준다.

Key Words: Adenoid cystic carcinoma (ACC), Distant metastasis, Oriental medical treatment

I. Introduction

Adenoid cystic carcinoma(ACC) is accounting for less than 1% of all head and neck carcinomas and 10% of all salivary gland malignancies. ACC is secondly frequent malignant epithelial tumour of salivary glands. It is characterized by slow-growing but aggressive tumour with a propensity for peri-neural invasion. It also frequently recurs and metastasis to distant organs such as lung or bone after treatment, and the prognosis is poor^{1,2}.

In this study, we will report a rapid growing, progressive adenoid cystic carcinoma in a 56-year-old female patient. It is the aim of the present paper to derive further studies evaluating the effectiveness of oriental medical treatment on ACC patients, we present this case of a woman with metastatic huge mass who received oriental medical treatment. However, a review of the literature reveals no previous report of such a case in oriental medicine.

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· 교신저자 : Chong-kwan Cho, East-West Cancer Center,
Dunsan Oriental Hospital of Daejeon University,
1136 Dunsan-dong, Seo-gu, Daejeon
(Tel. 82-42-470-0400, Fax. 82-42-470-9005
E-mail : orimedc@dju.ac.kr)

II. Case Report

A 56-year-old woman with epigastric pain, general weakness and sputum was referred to our hospital

on February 2004. Six years ago, adenoid cystic carcinoma of the right submandibular gland was diagnosed and wide excision of right submandibular and sublingual gland was done at Severance Hospital of Yonsei University. In addition, she also right tonsillectomy was done. Since then she had been in healthy state with mass-free for four years. However, distant metastasis was found in right upper lobe of lung on November, 2003. The medical team of Severance Hospital performed wedge resection of right upper lobe of lung. Histologic examination of the lung specimen showed a metastatic ACC. Unfortunately, before she recovered from the operation, it was relapsed in liver on January, 2004. The medical team performed fine-needle aspiration biopsy before operation. Fine-needle aspiration biopsy revealed the metastatic ACC. They decided to apply resection of liver mass. For the course of operation, they found multiple nodule which was presumed to be cancer seeding in omentum and abdominal wall. On frozen biopsy, it was also revealed the metastatic ACC. Therefore the procedure of operation was not performed. They recommended her chemotherapy as an palliative therapy for adenoid cystic carcinoma, and believed it could offer the best chance for prolon-

ging survival. However, she and her family refused the proposal. She made her choice to receive oriental medical treatment.

On visit our hospital, previous computed tomography(CT) scan(on January, 2004) showed about 14×11cm sized oval shaped low density mass in left and quadrate lobes of liver. There were no abnormalities on chest radiograph except for right phrenic elevation which was due to resection of right upper lobe of lung. Laboratory tests revealed elevation of γ -GTP and ALP. All other blood parameters were in normal range, including carcinoembryogenic antigen and squamous cell carcinoma antigen. She presented with intermittent abdominal pain in the right upper quadrant and general weakness. She had obstructive phlegm in her throat after resection of lung. Though she had several symptoms, her performance status was good relatively. She was restricted in physically strenuous activity but able to carry out work of a light in the house.

In our case, we first decided to apply Chunggantang(CGT) for the purpose of improving of liver function. CGT is a new herbal formula modified from In-Jin-O-Ryoung-San(Korean pronunciation for 茵陳五苓散) which has been a standardized formula for liver injury related jaundice for several

Table 1. Prescription of Chunggantang(CGT)

General Name	Part used	Relative Amount(g)
<i>Artemisia capillaries</i>	Herba	10
<i>Pueraria thunbergiana</i>	Radix	12
<i>Trionyx sinensis</i>	Carapace	10
<i>Raphanus sativus var</i>	Semen	10
<i>Atractylodes macrocephala</i>	Rhizoma	6
<i>Poria cocos</i>		6
<i>Atractylodes japonica</i>	Rhizoma	6
<i>Pueraria thunbergiana</i>	Flos	4
<i>Polyporus umbrellatus</i>		4
<i>Amomum villosum Lorur</i>	Fructus	4
<i>Glycyrrhiza uralensis</i>	Radix	2
Total amount		72

centuries³(Table 1). And then, we applied Hwa Acupuncture Therapy(HAT) as Korean traditional acupuncture. Upon theory of HAT, we diagnosed her the excess of earth and asthenia of wood(土實木虛) by the comparative examination of the palpitation of the pulse⁴. CGT was administered daily by three times a day and acupuncture on the acupoint(Sobu H-8, Kyonggo L-8, Taedon Liv-1 and

Taebaek Sp-3) was performed by once a month. As a rule, a follow-up of laboratory tests in a month was recommended. However, the patient was lost to follow-up several times due to personal reasons.

Although elevation of AST and γ -GTP were aggravated slightly from February, 2004 to August, 2005, her chief complaint was rapidly relieved (Table 2.). After two months of treatment, abdo-

Table 2. Changes of Liver Function Tests for Duration of Treatment

Laboratory Tests	2004.					2005.							
	2. 9.	3. 9.	6. 1.	6. 28.	11. 25.	1. 6.	2. 12.	3. 12.	4. 9.	5. 9.	6. 8.	7. 14.	8. 16.
AST(<40 IU/L)	28	38	58	100	56	42	104	89	118	120	66	105	50
ALT(<40 IU/L)	25	28	63	96	58	43	96	76	66	97	44	99	39
ALP(<120 IU/L)	375	418	453	393	253	231	332	328	252	301	302	364	316
γ -GTP(<32 IU/L)	222	181	296	241	389	425	664	598	548	612	485	673	521

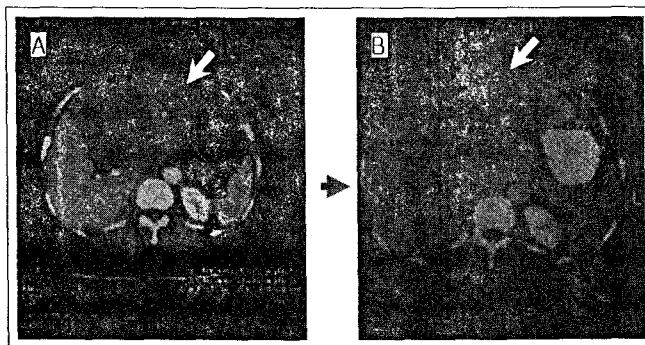


Fig. 1. CT images

A: CT (on May, 2004) showing a huge and low density tumor about 15 × 11 cm sized in left and quadrate lobes of liver
 B: CT (on October, 2004) showing no interval change and no evidence of newly appeared lesion, since May, 2004

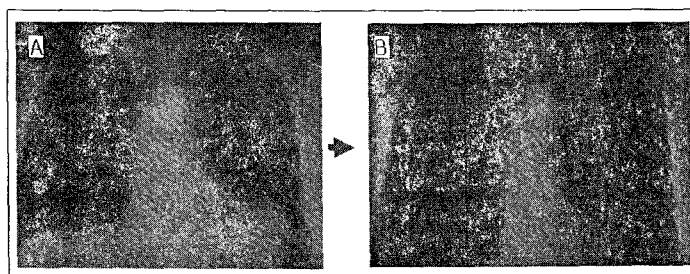


Fig. 2. Chest radiograph images

A: Chest radiograph (on May, 2004) showing no abnormalities except for right phrenic elevation
 B: Chest radiograph (on October, 2005) showing no interval change and no evidence of newly appeared lesion, since May, 2004

minal pain was disappeared. She also recovered in the debility state. Follow-up of CT scan was performed twice for the duration of treatment. CT on May, 2004 showed about 15×11cm sized oval shaped mass in left and quadrate lobes of liver. Since then, CT on October, 2004 showed no interval change and no evidence of newly appeared lesion, since May, 2004(Fig. 1). Chest radiograph on August, 2005 also showed no abnormalities(Fig. 2.). We recommended her a Follow-up of CT scan in 6 months continually. However, she refused further examination of CT scan. Recently, from the aggravation of laboratory tests, we can assume that the tumor is progressive continually. In spite of aggravation in the clinical course, she lives in asymptomatic healthy state on August, 2005.

III. Discussion

ACC was first described by Billroth in 1856 and called cylindroma⁵. It is a malignant tumor arising from the major and minor salivary glands, lacrimal glands, upper respiratory tract, breast, or uterine cervix. It has a biologic behavior characterized by slow growth, multiple local recurrences, and long clinical course, often complicated by development of delayed distant metastasis. Current treatment of this tumor can generally achieve good short-term prognosis, with slow growth that allows prolonged survival. However, the long-term prognosis is poor. The 5-year survival rates are 62-75%, in contrast to 20-year survival rates for patients with these tumors located in head and neck sites are 13%. These rates are due to the invasive nature and to metastases, frequently to the lung and bone, occurring long after initial treatment^{6,7}.

Distant metastasis in ACC may appear at a considerable time from the initial diagnosis and

treatment. The median time between the diagnosis of the primary lesion and the detection of distant metastasis was 60 months(range: 18-120 months)². In the present case, distant metastasis occurred 4 years after treatment of the primary tumor. The distant metastases appear to progress very slowly, remaining stable for a long period. In case of pulmonary metastasis, median freedom from progression in a resection with clear margins group was 30 months⁸. On the contrary, the patient shows a rapid tumor-growing and a progressive clinical course in our case. She was developed the distant metastases in liver and abdominal wall within just 2 months from the lung metastasectomy. And then, the most important factor influencing survival was distant metastasis and influencing quality of life (QOL) was primary or secondary tumor site⁸. Before our initial treatment, we worried about serious complications was caused by peritoneal seeding and huge mass in liver, such as ascites, abdominal pain and bowel obstruction. However, she did not show any symptoms for the duration of our treatment (19 months). Moreover, her chief complaints on visit our hospital were rapidly relieved in spite of aggravation of laboratory parameters. In general, the detection of distant metastases in asymptomatic patients is not an indication for further treatment, as radiotherapy and chemotherapy are ineffective for the management of secondary ACC patients. Chemoradiation or even surgical excision of metastatic tumor have some palliative benefit in cases of symptomatic metastatic disease². Like this, our treatment was useful for promoting of QOL and prolonging of survival in the present case.

So far, we described a case of rapid progressive ACC. The case may give us a possibility of that oriental medical treatment offer potential benefits (from an approach aimed at promoting of QOL and

extending survival to palliative care) for patients with advanced ACC patients.

REFERENCES

1. Qureshi SS, Nadkarni MS, Shrikhande SV, Desai S, Deodhar K, Ramadwar M et al. Hepatic resection for metastasis from adenoid cystic carcinoma of parotid gland. *Indian J Gastroenterol.* 2005;24(1):29-30.
2. Alexander DR, Nikolaos G, Hariklia G, Gregory F, Spyros DS, George AV et al. Adenoid cystic carcinoma of the head and neck. Clinicopathological analysis of 23 patients and review of the literature. *Oral Oncology*, 2005;41(3):328-35.
3. Cho JH, Lee YY, Seo SH, Yoo HS, Choi WJ, Lee YW et al. A Clinical Report about 57 Patients with Chronic Liver Disease. *Korean Journal of Oriental Medicine.* 2001;21(4):112-21.
4. Gam CW, Park DI. Study on Hwa-acupuncture Therapy. *Korean J. of Physiology & Pathology.* 2005;19(2):363-9.
5. Kokemueller H, Eckardt A, Brachvogel P, Hausamen JE. Adenoid cystic carcinoma of the head and neck-a 20 years experience. *International Journal of Oral and Maxillofacial Surgery.* 2004;33(1):25-31.
6. Kazumoto K, Hayase N, Kurosumi M, Kishi K, Uki J, Takeda F. Multiple brain metastases from adenoid cystic carcinoma of the parotid gland: Case report and review of the literature. *Surgical Neurology.* 1998;50(5):475-9.
7. Wahlberg P, Anderson H, Biörklund A, Möller T, Perfekt R. Carcinoma of the parotid and submandibular glands-a study of survival in 2465 patients. *Oral Oncology.* 2002;38(7):706-13.
8. Laura DL, Marco G, Paolo B, Paolo PBM, Barbara C, Elena F et al. Lung metastasectomy in adenoid cystic carcinoma (ACC) of salivary gland. *Oral Oncology.* In Press. 2005.