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Liver resection for metastases of tracheal adenoid cystic carcinoma: Report of two cases

Shintaro Hashimoto [a,*], Yorihisa Sumida [a], Shuichi Tobinaga [a], Hideo Wada [a], Kouki Wakata [a], Takashi Nonaka [a], Masaki Kunizaki [a], Shigekazu Hidaka [a], Naoe Kinoshita [b], Terumitsu Sawai [c], Takeshi Nagayasu [a]

[a] Department of Surgery, Division of Surgical Oncology, Nagasaki University Graduate School of Biological Sciences, Japan
[b] Department of Pathology, Division of Surgical Oncology, Nagasaki University Graduate School of Biological Sciences, Japan
[c] Cardiopulmonary Rehabilitation Science, Division of Surgical Oncology, Nagasaki University Graduate School of Biological Sciences, Japan

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ABSTRACT

INTRODUCTION: Tracheal adenoid cystic carcinoma (ACC) is rare and accounts for <1% of all lung cancers. Although ACC is classified as a low-grade tumor, metastases are frequently identified in the late period. Extrapulmonary metastases are rare, and their resection has rarely been reported.
PRESENTATION OF CASE: Case 1: A 77-year-old man underwent tracheal resection for ACC with postoperative radiation (60 Gy) 14 years before (at the age of 63). He underwent two subsequent pulmonary resections for metastases. Fourteen years after the first operation, he underwent extended right posterior segmentectomy with resection of segment IV and radiofrequency ablation for metastases of ACC to the liver. He was diagnosed with metastases to the kidney with peritoneal dissemination 4 years after the liver resection and died of pneumonia 2 years later. Case 2: A 53-year-old woman underwent a two-stage operation involving tracheal resection for ACC and partial resection of liver segments II and V for metastases of ACC to the liver. The tracheal margin was histopathologically positive. Postoperative radiation was performed, and she was tumor-free for 10 months after the liver resection.
DISCUSSION: Complete resection of tracheal ACC provides better survival. Radiotherapy is also recommended. However, the optimal treatment for metastases of ACC is unclear, especially because liver resection for metastases of tracheal ACC is rarely reported. Our two cases of metastases of tracheal ACC were surgically managed with good outcomes.
CONCLUSION: Liver resection for metastases of tracheal ACC may contribute to long survival.

1. Introduction

Tracheal adenoid cystic carcinoma (ACC) is rare and accounts for <1% of all lung cancers [1]. Although ACC is characterized by slow growth and classified as a low-grade tumor [2], metastasis or local recurrence is frequently identified in the late period [3]. Maziak et al. [3] reported that 45% of patients with tracheal ACC were diagnosed with metastases during follow-up in their study, and the mean period from the first treatment to diagnosis was 100 months. Sites of metastasis of tracheal ACC include the lung, liver, bone, brain, kidney, heart, skin, and abdomen. The optimal treatment for metastases of ACC has not been established.

Reports of hepatectomy for liver metastases of ACC, especially tracheal ACC, are rare [4–7]. We herein describe two rare cases of liver metastases of tracheal ACC that were managed surgically. It is reported in line with the PROCESS criteria [8].

2. Presentation of case

2.1. Case 1

A 77-year-old man underwent tracheal resection and reconstruction for tracheal ACC 14 years before (at the age of 63).

* Corresponding author. Present address: Division of Surgical Oncology, Department of Surgery, Nagasaki University Graduate School of Biological Sciences, 1-7-1 Sakamoto, Nagasaki 852-8501, Japan. Tel.: +0958197304.
E-mail addresses: shintahashimoto-ncd@umin.ac.jp (S. Hashimoto), y-sumida@nagasaki-u.ac.jp (Y. Sumida), sh.toby577@gmail.com (S. Tobinaga), gooddream44@gmail.com (H. Wada), kou-waka@hotmail.co.jp (K. Wakata), tnonaka@nagasaki-mc.com (T. Nonaka), kunisakimasaki2002@yahoo.co.jp (M. Kunizaki), hidaka-s@nagasaki-u.ac.jp (S. Hidaka), daimonji@nagasaki-u.ac.jp (N. Kinoshita), sawai@nagasaki-u.ac.jp (T. Sawai), nagayasu@nagasaki-u.ac.jp (T. Nagayasu).

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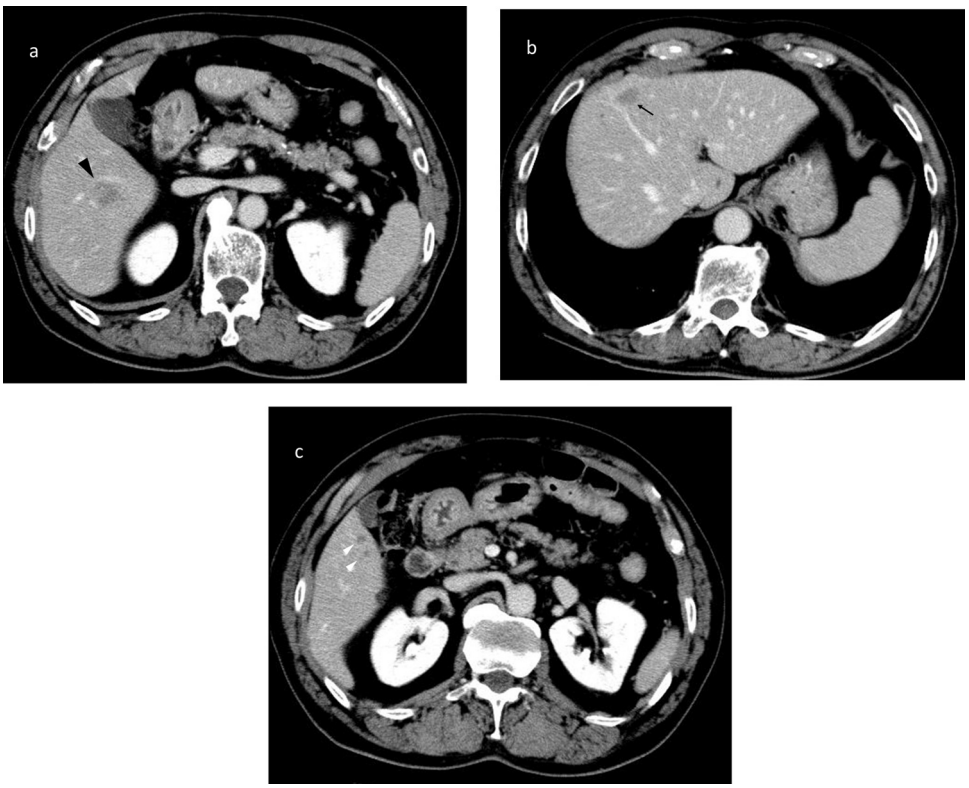

Fig. 1. Abdominal CT showed (a) a peripherally enhanced 3.5-cm mass in liver segments V/VI (black arrowhead) and (b) a peripherally enhanced 1.5-cm mass in segment IV (arrow). (c) CT also showed unenhanced masses in segments I, IV, V, and VII (white arrowhead).

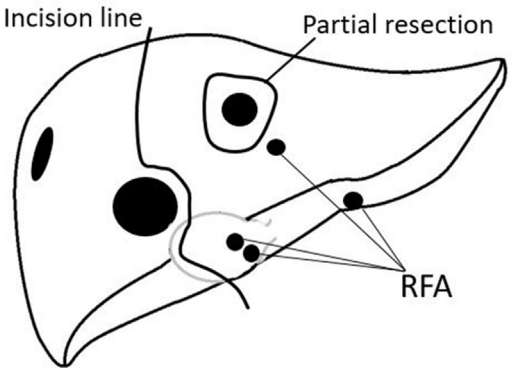

Fig. 2. The patient underwent extended right posterior segmentectomy with resection of segment IV and radiofrequency ablation for the lesions in segments I, IV, V, and VII.

Histopathologically, the margin was negative and no lymph node metastasis was found. Postoperative radiation therapy (60 Gy) was performed. He was diagnosed with solitary pulmonary metastases and underwent pulmonary partial resection 6 and 8 years after the original tracheal resection and reconstruction. Abdominal computed tomography (CT) showed 3.5- and 1.5-cm masses with peripheral enhancement in liver segments V/VI and IV, respectively (Fig. 1a-c). CT also showed unenhanced masses in segments I, IV, V, and VII. The patient's Eastern Cooperative Oncology Group (ECOG) performance status was 1. The result of an indocyanine green test was 12.1%. The laboratory data showed no evidence of organ disorder, including liver dysfunction. CT showed no other metastases or local recurrence. The patient was diagnosed with multiple liver metastases and underwent extended right posterior segmentectomy with resection of segment IV and radiofrequency ablation for the lesions in segments I, IV, V, and VII (Fig. 2). He was observed and diagnosed with metastases to the bilateral kidney with peritoneal dissemination 3 years 8 months after the liver resection. Because of his poor activities of daily living and high age, conservative therapy was performed. He died of pneumonia 6 years 6 months after the liver resection (21 years after the first operation).

2.2. Case 2

A 53-year-old woman presented with complaints of wheezing. Chest CT revealed a tumor on the left posterior side of the upper trachea. Bronchoscopy showed a hypervascular tumor occupying the tracheal airway (Fig. 3). Bronchoscopic examination revealed a tracheal ACC. Abdominal CT revealed 2.5- and 1.0-cm masses in liver segments V and II, respectively. Dynamic magnetic resonance imaging of the liver showed 25- and 8-mm masses with peripheral enhancement in segments V (Fig. 4a) and II (Fig. 4b), respectively. The patient was diagnosed with metastases of tracheal ACC, and a two-stage operation was planned. First, she underwent tracheal resection, and the left thyroid and left recurrent nerve were resected in combination because of the neoplastic invasion. Histopathologically, the tracheal ACC was found to have invaded the thyroid, and the margin was diagnosed as positive due to preservation of the larynx. No lymph node metastasis was found. Postoperative radiation therapy (60 Gy) was performed. Two months after the first operation, a liver operation was planned. The patient's ECOG performance status was 1. The result of an indocyanine green test was 3.0%. The laboratory data showed no evidence of organ disorder, including liver dysfunction. She underwent laparoscopic partial hepatectomy of segments II and V. Histopathologically, the tumor showed a cribriform pattern and the specimens were diagnosed as metastases of ACC (Fig. 5). She remained tumor-free for 1 year after the liver resection (1 year 2 months after the first operation).