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## Rapid response to radiotherapy in unresectable tracheal adenoid cystic carcinoma: A case report

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### Abstract

#### BACKGROUND

Adenoid cystic carcinoma (ACC) occurs mainly in the head and neck. Tracheal ACC (TACC) is uncommon. Primary resection is recommended as the main treatment of choice, and radiotherapy is considered for residual disease in the postoperative setting. Definitive radiotherapy is an alternative approach to cure unresectable TACC. As the status of radiosensitivity in TACC is uncertain, the evidence for radiotherapy in unresectable TACC is not well established, especially in terms of the optimal dosage and its response evaluation. Herein, we report a case of locally advanced TACC.

#### CASE SUMMARY

A 49-year-old woman was diagnosed with TACC, which included a range of lesions arising in the upper trachea extending caudally 2 cm to 7 cm of the glottis. She was treated with definitive radiotherapy, given the low likelihood of complete resection of the disease. Due to the indolent growth and the propensity for infiltration along the airways, the scheduled radiation dose of 76 Gy in 38 fractions with 6-MV X-ray delivered by intensity-modulated radiotherapy was conducted to the primary tumor volume. After irradiation of 40 Gy, the patient's dyspnea on exertion was dramatically relieved and bronchoscopy revealed that the previous large polypoid intra-luminal mass was significantly eliminated, with near-complete response. The patient completed two phases of scheduled radiotherapy, and acute reactions to treatment included subjective chest tightness and grade 2 esophagitis, managed medically. After 5 years of treatment, the patient is alive without recurrent disease, and there were no serious late radiation esophagus and lung damage, with only slight dysphagia without perforation and fistula.

#### CONCLUSION

Taken together, TACC is uncommon and the treatment of unresectable TACC is challenging. This case indicated that patients with unresectable TACC who

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rapidly respond to radiation may benefit from primary radical radiotherapy. Radiotherapy may be considered an effective alternative treatment modality.

**Key Words:** Adenoid cystic carcinoma; Tracheal cancer; Radiotherapy; Tracheal adenoid cystic carcinoma; Case report

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**Core Tip:** Tracheal adenoid cystic carcinoma (TACC) is uncommon. Primary resection is recommended as the main treatment of choice, and radiotherapy is considered for residual disease in the postoperative setting. Definitive radiotherapy is an alternative approach to cure unresectable TACC. As the status of radiosensitivity in TACC is uncertain, the evidence for radiotherapy in unresectable TACC is not well established, especially in terms of the optimal dosage and its response evaluation. Herein, we report a case of locally advanced TACC who rapidly responded to radiotherapy.

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## INTRODUCTION

Adenoid cystic carcinoma (ACC) is a rare tumor, which is most commonly found in salivary glands of the head and neck[1]. Tracheal ACC (TACC), originating from the submucosal glands of the trachea, is much less common, accounting for approximately 10%-20% of malignant tracheal tumors[2]. The standard treatment of TACC includes primary resection and postoperative radiotherapy for residual disease. Most patients, however, present with locally advanced disease, due to the slow and indolent growth of the tumor and the propensity for invasion into adjacent structures[3]. Primary radiotherapy is the main approach for unresectable TACC. As ACC is prone to indolent growth and the radiosensitivity status is uncertain, most tumors respond poorly to radiotherapy and local progression often occurs shortly after completion of treatment. Herein, we present a case of inoperable TACC, who rapidly responded to radiotherapy. This study was approved by the ethical review committee of West China Hospital of Sichuan University.

## CASE PRESENTATION

### Chief complaints

A 49-year-old woman presented with increasing dyspnea on exertion for 3 wk, occasionally accompanied by chest pain and cough.

### History of past illness

No special history of past illness.

### Physical examination

Physical examination was notable for stridor at rest.

### Imaging examinations

After she was admitted to the local hospital in March 2015, a chest computed tomography (CT) scan showed a soft tissue mass surrounding the inner surface of the upper trachea, leading to irregular lumen stenosis of the trachea (Figure 1A and B). Thus, she was referred to our hospital, in order to initially undergo resection, which is considered the most effective treatment for tracheal tumor. The patient experienced stridor at rest. Bronchoscopy revealed that a large polypoid intra-luminal mass, with its surface presenting rich vascular networks and nodular protrusions, was present in



**Figure 1 Imaging of the case.** A and B: Chest computed tomography pre-treatment revealed a soft tissue mass surrounding the inner surface of the upper trachea; C and D: Chest computed tomography after 60 Gy indicated dramatic regression of the tumor.

the upper trachea extending caudally 2 cm to 7 cm of the glottis (Figure 2A).

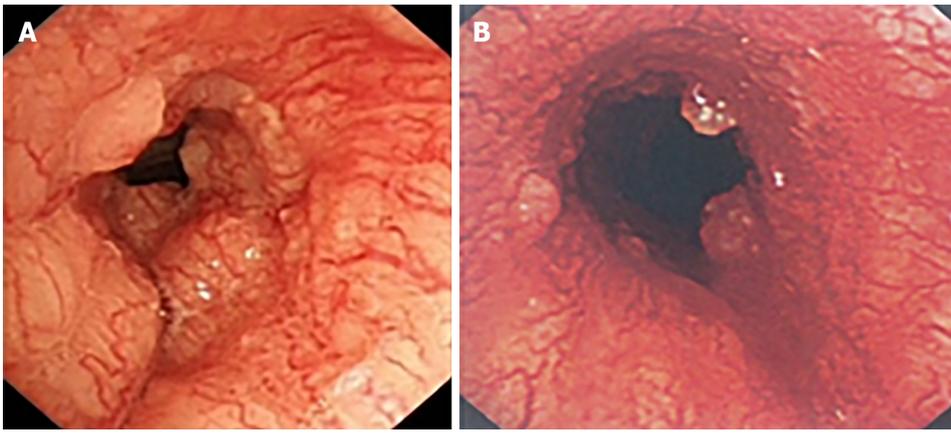
## FINAL DIAGNOSIS

Bronchoscopic biopsy confirmed an ACC, with immunohistochemistry of AE1/AE3 (+), CEA (-), GFAP (-), S-100 (+).

## TREATMENT

Radioisotope bone scans, brain magnetic resonance imaging, and abdominal CT did not reveal enlarged lymph nodes or metastatic disease. After a discussion of this case in our daily multidisciplinary tumor board, she was treated with definitive radiotherapy given the low likelihood of complete resection of disease.

According to the suggestion from the QUANTEC (quantitative analysis of normal tissue effects in the clinic) report[4], the total irradiated dose in our case did not exceed 80 Gy in order to reduce the risk of central airway stenosis. In view of large volume lesions, the therapeutic regimen, which was planned to deliver 76 Gy to the gross tumor volume (GTV) with an additional margin to account for microscopic extension of disease, respiratory motion, and daily variance in patient positioning, was divided into two phases. In the first treatment phase, she received 60 Gy in 30 fractions, using 6-MV X-ray delivered by an intensity-modulated radiotherapy (IMRT) technique. Astonishingly, the patient's dyspnea on exertion was dramatically relieved after 40 Gy of irradiation. Bronchoscopy was conducted which revealed that the previous large polypoid intra-luminal mass was significantly eliminated and the narrow trachea was freed (Figure 2B). However, considering the probable low inherent radiosensitivity of ACC, a prophylactic dose of 60 Gy to the low-risk clinical target volume (CTV<sub>low</sub>), referring to the region for low-risk lymphatic metastasis, was delivered as planned. In the second CT scan of radiotherapy simulation, there was evident regression of the



**Figure 2 Bronchoscopic findings.** A: Pre-treatment revealed that a large polypoid intra-luminal mass, with its surface presenting rich vascular networks and nodular protrusions; B: After receiving 40 Gy, bronchoscopy indicated that the previous large polypoid intra-luminal mass was significantly eliminated.

tumor with increased luminal opening of the trachea (Figure 1C and D). The patient received an additional 10 Gy for the high-risk clinical target volume (CTV<sub>high</sub>), referring to the region for high-risk lymphatic metastasis, and another 6 Gy for the GTV at 2 Gy/d in the second phase of radiotherapy with 6-MV X-ray delivered by IMRT (Figure 3). Thus, the GTV received the prescribed dosage of 76 Gy per 2 Gy in total. The radiotherapy plan beam arrangement consisted of anterior, left, and right anterior oblique, posterior, and left and right posterior oblique fields. The weights of the three posterior fields were reduced compared to the others, in order to minimize the irradiated dose and volume to the esophagus and spinal cord. Due to longitudinal invasion along the trachea, margins of 4-5 cm in the craniocaudal axis were included as suggested by Kaminski[5]. Considering the high-risk of local-regional recurrence, the CTV<sub>high</sub> encompassed the entire GTV with a 2-cm margin of adjacent superior and inferior structures, and with a 0.5-cm margin of adjacent anterior, posterior, and bilateral structures. Given the tumor's propensity for invasion into local nerves, the CTV<sub>low</sub> covered the entire CTV<sub>high</sub> with a 3-cm margin of adjacent superior and inferior structures, and with a 0.5-cm margin of adjacent anterior, posterior and bilateral structures (Figure 3).

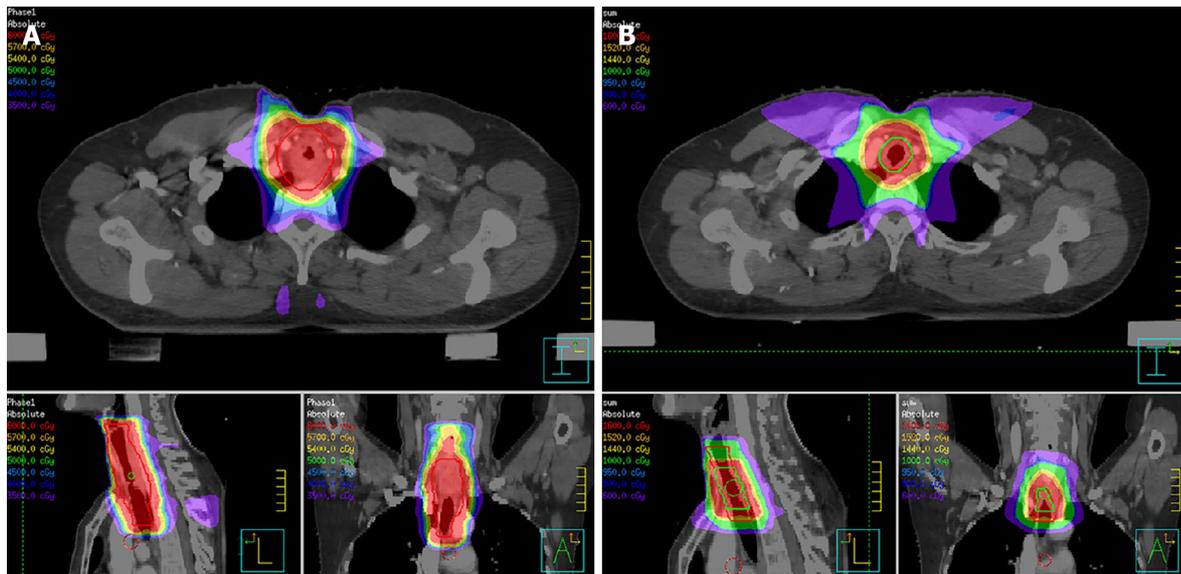
## OUTCOME AND FOLLOW-UP

The patient successfully completed the scheduled two phases of radiotherapy, and acute reactions to treatment included subjective chest tightness and grade 2 esophagitis, managed medically. After 5 years of treatment, the patient is alive without recurrent disease, and no serious late radiation esophagus and lung damage, only slight dysphagia without perforation and fistula.

## DISCUSSION

ACC, formerly known as "cylindroma" and "adenocystic carcinoma," occurs mainly in the head and neck. However, its presence in the pulmonary system is uncommon. Most tumors occur in the trachea and mainstem bronchi, but peripheral tumors from glands of small bronchi have occasionally been reported[6-8]. Women are more often affected than men, and the peak age of incidence occurs between the 4th and 5th decades of life, which is earlier than lung cancer[9,10]. Smoking does not affect the incidence. ACC is well known for its prolonged clinical course, its propensity for peritracheal soft tissue and perineural invasion, and delayed onset of distant metastases[3]. The most common site of distant metastasis is the lung, followed by bone, liver, and brain[11].

Owing to its slow and indolent growth, the clinical course of TACC is prolonged and asymptomatic. When the tumor invades local nerves and structures, patients will present varying symptoms, which are related to airway obstruction. Cough, dyspnea, and hemoptysis are the most common complaints. However, those symptoms often lead to a misdiagnosis of asthma or bronchitis. Chest CT and bronchoscopy are the



**Figure 3** Dose distribution of the first and second treatment plan using intensity-modulated radiotherapy. A: The first radiotherapy plan (30 × 2 Gy) with an irradiated high dose zone for CTV<sub>low</sub>, surrounded by a red line; B: The second radiotherapy plan, where CTV<sub>high</sub> (red line) received an additional 10 Gy in 5 fractions and the GTV (green line) received another 6 Gy in 3 fractions. IMRT: Intensity modulated radiation therapy; CTV: Clinical target volume; GTV: Gross tumor volume.

main diagnostic methods for the diagnosis of TACC. On CT, this tumor usually has a marked tendency toward submucosal extension that manifests as an intraluminal mass with longitudinal extension[12], as seen in our case.

Once diagnosed, the tumor should be assessed for surgical resection. Several relatively large studies showed that surgery can lead to a significantly better survival than non-surgical management. Two large studies[13,14] reported that the overall 5-year and 10-year survival rates of resected TACC, regardless of complete or incomplete resection, were 52%-88.7% and 29%-43.2%, compared with 33% and 10% in the non-resected group, respectively. Compared with incomplete resection, complete resection with negative airway margins resulted in higher survival with 5-year and 10-year survival rates of 75%-90% and 45%-75%, respectively. It was noted that the incomplete resection of TACC was quite high, with 59%-84.4% R<sub>1</sub> resection rates, probably because it tends to infiltrate along the airways and invade peritracheal soft tissue and perineural tissue. However, the 15-year long-term follow-up results showed that surgery still benefits patients with incomplete resected TACC, compared with the unresectable group (15-year overall survival: 14.5% *vs* 0%)[13]. For TACC patients with incomplete resection (R1), postoperative radiotherapy significantly reduces local-regional failure and improves overall survival. This evidence suggests that a pathologically negative margin might not always be required, and adjuvant radiotherapy is needed in the postoperative setting in such cases. The benefit of adjuvant radiotherapy in R0 resection remains uncertain.

If patients have technically or medically inoperable tumors, radiotherapy should be considered as an alternative local treatment. Nevertheless, the optimal dosage of radical radiotherapy for TACC is not well known, due to the uncertain status of radiosensitivity in TACC. ACC is deemed to be a relatively radioresistant tumor, due to its indolent growth and low inherent radiosensitivity. Lee *et al*[2] assessed the efficacy of irradiation in patients with unresectable TACC, after immediately completing primary radiotherapy. The results showed that among 13 inoperable patients treated with primary radiation of 60 Gy, the objective response rate was only 1/13 (one had a partial response), while four had stable disease, and eight had progressive disease. A Chinese study[9] revealed that two patients with locally advanced disease and invasion of the pulmonary artery received primary radiotherapy with 40 Gy of the scheduled dose, and response to radiation was poor. The lesions remained unchanged following radiotherapy. However, our case showed a significant reduction in tumor size after 40 Gy of irradiation. In contrast, some studies have also indicated that TACC seems to have high radiosensitivity. Bhandari *et al*[15] reported a 51-year-old female with unresectable TACC, who received a dose of 54 Gy/30 fractions IMRT. At 8 mo post-radiotherapy, her CT scan showed a near complete response. Kanematsu *et al*[16] reported that 5 inoperable patients treated

with primary radiation with a median dosage of 60 Gy (50 Gy to 70 Gy), showed a remarkable radiosensitivity including 1 complete response and 4 partial responses, and the 5-year survival rate was 40%. Maziak *et al*[11] also reported that 6 inoperable patients treated with primary radiation with a given dosage varying between 50 and 75 Gy, had a mean survival of 6.2 years. However, most of the irradiated patients harbored high rates of local recurrence, compared with patients treated by primary resection. This may indicate that a higher dosage of irradiation is needed for TACC to improve local control. Bonner Millar *et al*[17] reported a patient with inoperable TACC with a 2.2 cm × 1.9 cm soft tissue mass located posterior to the carina, who received 80 Gy of radiation with 6-MV photons. It was observed that the tumor completely responded to radiation as shown by chest CT scan one month after treatment. There was no evidence of recurrent disease over six years follow-up, and some tolerated side-effects of radiotherapy were noted including chest tightness and pain, diminished force of cough, atrophy of chest wall musculature, and gastroesophageal reflux disease, but not tracheal or esophageal ulcer and fistula. Similarly, our case received 76 Gy of radiation and did not suffer a severe adverse radiation reaction. Higher doses may improve tumor control, but there is also the potential for increased complications using photons (dose > 66 Gy)[18]. Compared to photons, high linear energy transfer (LET) radiation, such as neutrons, harbors a unique physical property of the Bragg peak for sharp dose distribution to significantly diminish irradiated dose and volume of normal tissues around the irradiated target. Bittner *et al*[19] described the effectiveness of fast neutron radiotherapy in 19 unresectable ACC patients, and reported a 5-year actuarial locoregional control rate of 54.1%, with tolerable side-effects (only 2 cases experienced grade 3/4 chronic toxicity).

## CONCLUSION

TACC is uncommon and the treatment of unresectable TACC is challenging. This case indicates that patients with unresectable TACC who rapidly respond to radiation may persistently benefit from primary radical radiotherapy. Radiotherapy may be considered an effective alternative treatment modality. Future research in the form of more case reports, case series, or small-scale clinical trials could add evidence on this phenomenon.

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