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Surgical outcomes and long-term prognosis of patients with tracheobronchial adenoid cystic carcinoma

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ABSTRACT

Tracheobronchial adenoid cystic carcinoma (ACC) is a rare malignant disease of the central airways. Complete resection is the primary goal for this tumor; however, it remains challenging due to the complexity of reconstruction, tumor submucosal spread, and the risk of dehiscence. Conversely, tracheobronchial ACC has relatively favorable long-term outcomes due to its indolent growth. Due to the rarity of this tumor, a clear treatment guideline has yet to be established. This study aimed to evaluate treatment strategies, perioperative outcomes, and long-term prognosis of tracheobronchial ACC cases at our institution to identify the optimal therapeutic strategy for this challenging disease. We retrospectively analyzed 11 patients who underwent surgical intervention for tracheobronchial ACC at our institution from 2000 to 2024. Clinical, surgical, and oncologic outcomes were assessed, including the impact of surgical margins and adjuvant therapy. Complete resection was achieved in two cases, while nine patients had positive margins requiring postoperative radiotherapy. No local recurrences were observed, but distant metastases occurred in 54.5% of cases. Oligometastatic disease was effectively managed with local therapy. The 5-year and 10-year overall survival rates were 90% and 75%, respectively. Surgical resection remains the cornerstone of treatment for tracheobronchial ACC, however achieving negative surgical margins is not always feasible. In such cases, postoperative radiotherapy is considered an effective strategy to mitigate the risk of local recurrence. Local treatment of oligometastatic disease may provide a survival. These results indicate the importance of a multimodal treatment approach tailored to the specific challenges of tracheobronchial ACC.

Keywords: adenoid cystic carcinoma, carinal resection, tracheobronchial tumor

Abbreviations:
ACC: adenoid cystic carcinoma
OS: overall survival
RFS: recurrence-free survival

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INTRODUCTION

Tracheobronchial adenoid cystic carcinoma (ACC) is a rare malignant disease of the central airways. The incidence of tracheobronchial malignancies ranges from 0.1 to 0.85 cases per 100,000 population, with ACC accounting for 7%–60% of these tumors, emphasizing its rarity.[1-4] No definitive risk factors for tracheobronchial ACC have been identified, and the disease exhibits no significant sex predilection,[5] frequently affecting relatively younger individuals.[2,3,6] Common symptoms include dyspnea, coughing, and asthma-like manifestations due to airway obstruction, occasionally requiring emergency interventions.[7] Despite acute presentations, tracheobronchial ACC exhibits indolent growth and, unlike high-grade lung cancers, has relatively favorable long-term outcomes.[8,9]

Complete surgical resection is the primary treatment goal for tracheobronchial ACC; however, submucosal spread often causes a broader tumor distribution than expected. The limited elasticity of the trachea restricts resection and reconstruction, sometimes requiring positive surgical margins. Resections exceeding 4 cm increase anastomotic tension,[10] heightening the risk of dehiscence. Surgery should prioritize the postoperative quality of life considering the younger age of the affected patients and the low-grade malignancy of ACC. Tracheobronchial ACC surgery is technically demanding due to its high-risk nature and the need for specialized techniques such as cross-field intubation and extracorporeal membrane oxygenation (ECMO).

Due to the rarity of tracheobronchial ACC, various reports have been published; however, a clear treatment guideline has yet to be established. This study investigated the treatment methods, perioperative outcomes, and long-term prognoses of ACC cases at our institution to identify the optimal therapeutic strategy for this challenging disease.

MATERIALS AND METHODS

Ethical consideration

All studies involving human participants were conducted under the ethical standards outlined by Nagoya University Hospital and national research committees and the 1964 Declaration of Helsinki, along with its subsequent amendments or equivalent ethical standards. The institutional review board of the hospital approved this retrospective study (no. 2017-0034).

Patients

We collected information from patients who underwent surgical intervention for ACC of the central airway from January 2000 to May 2024 at the Nagoya University Hospital. The study included cases in which a macroscopic curative resection was considered feasible based on preoperative imaging and bronchoscopic findings. Patients with laryngeal cancer were excluded. This study excluded cases that did not undergo surgery. Finally, this study examined 11 patients.

Database and patient follow-up

The following clinical and pathological characteristics were recorded in our database: age, sex, chief complaint, Eastern Cooperative Oncology Group–performance status (ECOG–PS), tumor location, bronchoscopic intervention types, preoperative vital capacity (%VC), forced expiratory volume in one second (%FEV1), general anesthesia-related issues, innovations during surgery, surgical procedure type, postoperative complications based on the Clavien–Dindo classification,[11] pathological margin diagnosis, postoperative radiation therapy details, recurrence site, recurrence treatment, and survival period. The pathological tumor diagnosis was identified using the World

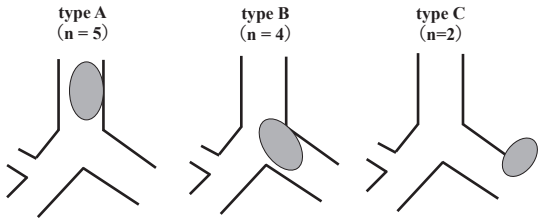

Fig. 1 Classified cases based on tumor location as the trachea (type A), the carina (type B) and distal to the main bronchus (type C)

Health Organization classification of tumors.[12] In this study, we categorized cases based on the primary tumor location as the trachea (type A), the carina (type B), and distal to the main bronchus (type C) (Fig. 1).

Patients were followed up at 3-month intervals for the initial 2 years postoperatively and at 6-month intervals at an outpatient clinic. Recurrence or survival data were collected, whenever possible, for up to 17 years. Recurrence was diagnosed based on compatible physical examination and/or diagnostic imaging findings, and the diagnosis was histologically validated when clinically feasible.

Definition of overall survival and recurrence-free survival

Overall survival (OS) was the interval between the surgery date and all-cause death. Recurrence-free survival (RFS) was the interval between the date of surgery and the date of recurrence detection, all-cause death, or last follow-up. Observations were censored at the last follow-up when the patient was alive or lost to follow-up. The date of the data cut-off was December 2024.

Statistical analysis

The Kaplan–Meier method was used for survival curve estimation. We categorized the participants into margin-negative and margin-positive groups and assessed the observation period, mortality, and recurrence status to calculate the OS and RFS rates. IBM Statistical Package for the Social Sciences version 29.0.1.0 was used for data analyses.

RESULTS

Patient characteristics

Table 1 shows the characteristics, surgical approaches, and postoperative outcomes of the cohort including 11 patients. The median age of the participants was 49 years (range, 21–75), consisting of eight females (73%), and all patients had ECOG–PS of 0. The main presenting complaints included cough (n = 9, 82%), dyspnea (n = 6, 55%), asthma-like symptoms (n = 2, 18%), and hemoptysis (n = 1, 9%). The median tumor diameter was 3.6 cm (range, 1.9–5.0 cm). Of the nine cases involving tracheal or carinal tumors, seven received preoperative bronchoscopic interventions. In most cases, these interventions successfully restored adequate ventilation and achieved favorable pulmonary function, with %VC of 99% (range, 84–133) and %FEV1.0 of 85% (range, 18–132). The tumor localization included the trachea (type A; n = 5), carina (type B; n = 4), and bronchus (type C; n = 2; Fig. 1). No complications were encountered during general anesthesia induction.

Table 1 Patient characteristics

Case	Age	Sex	Chief complaint symptoms	Tumor location	Tumor diameter (cm)	Bronchoscopic intervention (preoperative)	%VC (%)	%FEV1.0 (%)
1	32	F	Cough, Asthma	B	3.6	–	122	74
2	59	M	Cough, Dyspnea	A	3.5	–	99	18
3	61	F	Cough, Dyspnea	A	2.1	Core out	101*	85*
4	75	M	Hemo-sputum	C	5.0	–	133	132
5	61	F	Cough, Dyspnea	A	3.5	Stenting	100*	92*
6	70	F	Pneumonia	C	1.9	–	100	103
7	51	F	Cough, Dyspnea	A	3.6	Stenting	98*	92*
8	37	F	Cough, Dyspnea	B	4.0	Core out, Stenting	84*	71*
9	51	F	Cough, Dyspnea	A	3.6	Core out, Stenting	91*	77*
10	21	F	Cough, Asthma	B	3.0	Core out, Stenting	91*	103*
11	30	M	Cough	B	3.7	Core out, Stenting	86*	70*

%FEV1: percent predicted forced expiratory volume in one second
%VC: vital capacity as percent of predicted
* Pulmonary function following bronchoscopic intervention

Table 2 shows the perioperative outcomes. The surgical procedures included tracheal resection in five cases, sleeve pneumonectomy in three cases, carinal resection with reconstruction in one case, right middle and lower lobectomy in one case, and sleeve left upper lobectomy in one case. A median sternotomy was applied in three cases, whereas a right posterolateral thoracotomy was used in six cases. The approach in the two cases that required sleeve left pneumonectomy began with a right posterolateral thoracotomy, followed by repositioning for either an anterior axillary thoracotomy or video-assisted thoracoscopic surgery (VATS).

Intraoperative field intubation was required in all nine cases requiring tracheal resection and reconstruction (types A and B). One patient treated with left sleeve pneumonectomy (case 10) underwent left main bronchus resection in a left lateral decubitus position, followed by anastomosis between the trachea and the right main bronchus. However, due to the worsening oxygenation caused by the ventilation–perfusion imbalance accompanied by the left lateral decubitus position with right lung ventilation, veno–arterial (V-A) ECMO was employed to maintain oxygenation until the patient was repositioned to the right lateral decubitus position. Strategies in subsequent cases of left sleeve pneumonectomy (case 11) were implemented to mitigate the previously encountered oxygenation issues associated with ventilation–perfusion imbalance. Performing dual-lung ventilation with a surgical field tube inserted into the transected left main bronchus, connecting it to the ventilator through the edge of the incision, and chest closure and repositioning were accomplished without causing hypoxemia from the ventilation–perfusion mismatch. Oxygenation improved after patient repositioning, and ECMO support was avoided. The left pneumonectomy was completed through VATS.

The mean operative time was 333 min (range, 208–568 min), with a mean blood loss of 242 mL (range, 48–950 mL). Complications categorized as grade ≥3 based on the Clavien–Dindo classification included surgical site infection (SSI), chylothorax, and pneumonia in one case each. The case complicated with SSI underwent debridement and irrigation of the posterolateral

Table 2 Perioperative outcomes

Case	Approach	Surgical procedures	Additional treatments for oxygenation	Length of airway resected (cm)	Post operative complication
1	Posterolateral thoracotomy	Right sleeve pneumonectomy		3	
2	Posterolateral thoracotomy	Tracheal resection		3.5	
3	Posterolateral thoracotomy	Tracheal resection		2	
4	Posterolateral thoracotomy	Right middle lower lobectomy+bronchoplasty		–*	Chylothorax
5	Median sternotomy	Tracheal resection		3.5	
6	Posterolateral thoracotomy	Left upper sleeve lobectomy		–*	
7	Median sternotomy	Tracheal resection		4.5	
8	Posterolateral thoracotomy	Modified double-barrel method		4	Surgical site infection
9	Median sternotomy	Tracheal resection		5	
10	Posterolateral thoracotomy anterior axillary incision	Left sleeve pneumonectomy	V-A ECMO	2	
11	Posterolateral thoracotomy →VATS (left)	Left sleeve pneumonectomy	Dual-lung ventilation	3	Pneumonia

V-A ECMO: veno–arterial extracorporeal membrane oxygenation
VATS: video assisted thoracic surgery
%FEV1: percent predicted forced expiratory volume in one second
%VC: vital capacity as percent of predicted
* Since the procedure was a lobectomy with bronchoplasty, there was no shortening of the trachea.

incision site alongside thoracoscopic lavage and chest drainage, which successfully resolved the infection. The case of chylothorax received treatment including fasting, total parenteral nutrition, and pleurodesis, which facilitated chest drain removal on postoperative day 17. Aspiration pneumonia developed in a patient who had undergone left sleeve pneumonectomy, requiring mechanical ventilation. However, the condition improved with appropriate medical management. No cases reported treatment-related death.

Macroscopic-positive margins were identified in three cases, microscopic-positive margins in six cases, and negative margins in two cases. All nine patients with positive surgical margins received postoperative radiation therapy, with a median radiation dose of 60 Gy (range, 50–66 Gy). A summary of intraoperative and postoperative variables is presented in Table 3.

Table 3 Intraoperative and postoperative variables

N=11	Value (%)
Surgical procedure	
Tracheal reconstruction	5 (42)
Pneumonectomy with carinal resection	3 (27)
Carinal resection with reconstruction	1 (9)
Lobectomy with bronchial reconstruction	2 (18)
Approach	
Median sternotomy	3 (27)
Posterolateral thoracotomy	6 (55)
Posterolateral → Anterior axillary incision	1 (9)
Posterolateral thoracotomy → VATS	1 (9)
Additional treatments for oxygenation	
Surgical field intubation	9 (82)
Extracorporeal membrane oxygenation	1 (9)
Dual-lung ventilation	1 (9)
Post operative complications	
Pneumonia	1 (9)
Chylothorax	1 (9)
Surgical site infection	1 (9)
Resected margin	
Negative (R0)	2 (18)
Microscopic positive (R1)	6 (55)
Macroscopic positive (R2)	3 (27)

IQR: interquartile range
VATS: video assisted thoracic surgery
R0: resection for cure or complete remission
R1: microscopic residual tumor
R2: macroscopic residual tumor

No local recurrences were reported; however, distant metastases were determined in six (54.5%) cases. Regarding the distribution of metastatic sites, including overlapping cases, pulmonary metastasis was the most prevalent, occurring in four cases. Further, liver metastases were observed in three cases, subglottic metastasis in one case, and sacral metastasis in one case. Regarding prognosis, five (45%) cases were alive without disease. Among the recurrence cases, four (36%) cases were considered oligorecurrences and received local treatment, leading to survival with the disease.

The two (18%) cases with synchronous multiple metastases died of the disease. One patient with multiple lung metastases experienced recurrence at 11 months postoperatively. The patient was treated with six courses of ociperlimab + tislelizumab; however, the disease progressed. Subsequently, four courses of carboplatin + paclitaxel were administered, but the disease progression was observed, and the patient died 38 months after surgery (Case 10).

Table 4 and Fig. 2 present the recurrent sites, corresponding treatments, and clinical course.

Table 4 Resection margin, clinical course and prognosis

Case	Resected margin	Postoperative radiotherapy (Gy)	Time to recurrence (month)	Recurrent site	Treatment for recurrence	Prognosis
1	R2	50	170	Bone (sacrum)	Radiation	AWD
2	R2	60	94	1. Pulmonary 2. Liver	1, 2. Resection	AWD
3	R1	60				NED
4	R2	60	87	Liver (multiple)	BSC	DOD
5	R1	60	108	1. Pulmonary 2. Subglottic	1. Resection, 2. RT	AWD
6	R1	50	64	1. Pulmonary 2. Chest wall 3. Liver	1. Resection, 2. RT, 3. BSC	AWD
7	R0	–				NED
8	R0	–				NED
9	R1	66				NED
10	R1	60	11	Pulmonary (multiple)	Chemotherapy	DOD
11	R1	60				NED

AWD: alive with disease
BSC: best supportive care
DOD: dead of disease
NED: no evidence of disease
RT: radiotherapy
R0: resection for cure or complete remission
R1: microscopic residual tumor
R2: macroscopic residual tumor

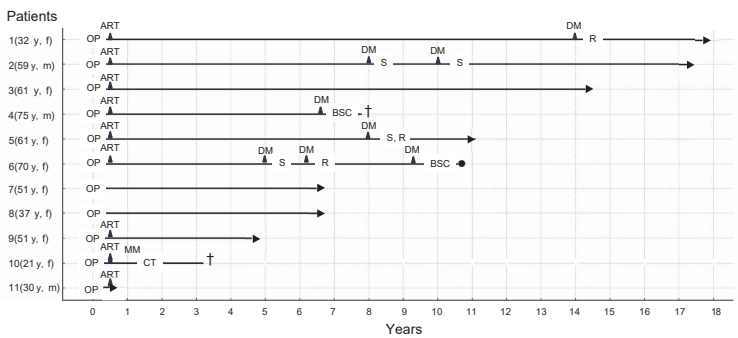

Fig. 2 Individual follow-up progress for each patient
OP: surgery for tracheobronchial adenoid cystic carcinoma
ART: adjuvant radiation therapy
DM: distant metastasis
MM: multiple metastasis
S: surgical resection for metastasis
R: radiation therapy for metastasis
CT: chemotherapy
BSC: best supportive care
†, dead of disease; ▶, no evidence of disease; •, alive with disease.

OS and RFS

The median follow-up period was 107 months (range, 6–208 months). The 5-year and 10-year OS rates for all cases were 90% and 75% and that of RFS rates were 79% and 36%, respectively. The 5-year and 10-year OS rates in patients with negative resection margins were 100% and the corresponding RFS rates were also 100%. The 5-year and 10-year OS rates in the case of positive resection margins were 87% and 75% and that of RFS rates were 87% and 29%, respectively (Fig. 3).

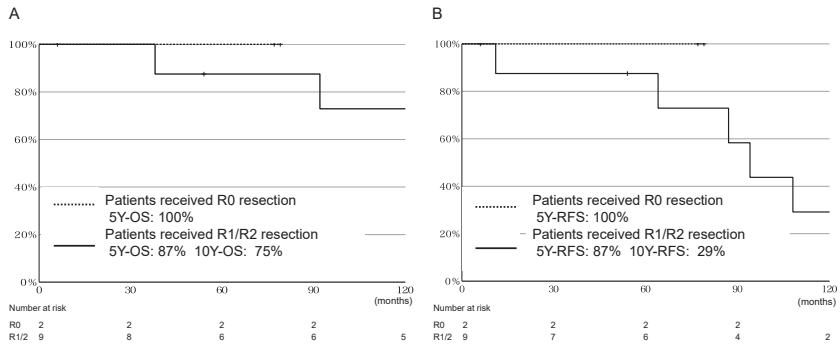

Fig. 3 The prognosis of ACC in our series

Fig. 3A: Kaplan-Meier survival curves comparing the OS between the R0 and R1/R2 resection groups

Fig. 3B: Kaplan-Meier survival curves comparing RFS between the R0 and the R1/R2 resection groups

OS: overall survival

RFS: recurrence-free survival

R0: resection for cure or complete remission

R1: microscopic residual tumor

R2: macroscopic residual tumor

DISCUSSION

We analyzed patients with tracheobronchial ACC who underwent airway reconstruction surgery to identify the optimal treatment strategies for this rare disease which required complex interventions. Our findings reinforce and extend previous evidence on the importance of postoperative radiotherapy and local treatments in tracheobronchial ACC. Wang et al reported that, among 163 surgically treated cases, patients with R1 resection who received postoperative radiotherapy achieved similar RFS as those with R0 resection, while those without radiotherapy had significantly poorer outcomes. Furthermore, they demonstrated that local therapies such as surgery or radiotherapy improved post-recurrence survival compared to chemotherapy or best supportive care.[13] In our study, all nine patients with positive surgical margins received adjuvant radiotherapy, and none experienced local recurrence. Additionally, four patients with oligometastatic recurrence underwent local treatments—surgical resection or radiotherapy—and remained alive at follow-up. These results support the effectiveness of adjuvant radiotherapy in preventing local recurrence after incomplete resection and suggest that local therapies can offer durable disease control in selected patients with limited metastases. Although similar strategies have been proposed in previous large-scale studies, our study adds further validation in a more recent cohort with detailed treatment data and long-term follow-up.

Gaissert et al and Yang et al analyzed over 130 tracheobronchial ACC cases and reported a favorable prognosis with pathological negative margins.[9,14] Similarly, our two patients with negative margins remained recurrence-free. However, submucosal extension frequently causes unexpected invasion, thereby complicating complete resection. Estephen et al analyzed 59 ACC cases with incomplete resection, demonstrating no survival difference between the R0 and R1 groups receiving radiotherapy but exhibiting poor prognosis in margin-positive cases without it.[15] In our study, all R1/R2 cases received adjuvant radiotherapy with no local recurrence, emphasizing its essential role in margin-positive cases.

Surgical treatment for tracheobronchial ACC is challenging because of the complex airway reconstruction and significant anastomotic complications.[16] Wright et al reported 37 anastomotic dehiscence cases among 901 tracheal resections, including 6 fatal cases due to airway obstruction, brachiocephalic artery fistula, or mediastinitis. The determined risk factors included a resection length of >4 cm, preoperative tracheostomy, age of 17 years, reoperation, and laryngotracheal resection.[10] In our cohort, three cases involved resections of ≥4 cm, aligning with the reported risk factors; however, no anastomotic dehiscence or local recurrence occurred.

Several surgical techniques for resection and reconstruction have been reported in cases of tumors located in the carina. Grillo et al described 13 patterns of resection and reconstruction.[17] Preoperatively evaluating the tumor location and the length of resection is crucial to determine the most appropriate surgical approach. Furthermore, one-lung ventilation via cross-field intubation[18] is often required. Thus, severe hypoxemia due to ventilation–perfusion mismatch can occur in cases of left sleeve pneumonectomy, particularly when the patient is approached in the left lateral decubitus position. Nakamura et al[19] revealed that V-A ECMO after the left main bronchus transection and dual-lung ventilation using the planned resected lung after the carinal transection can be useful for maintaining oxygenation in certain cases. Due to the complexity of the tracheobronchial procedure, such modifications are often required to ensure airway management and safe surgery completion.[20]

Oligometastatic cases in recurrent tracheobronchial ACC may benefit from local control through surgery or radiotherapy. Hashimoto et al reported hepatic metastases that recur 14 years after the initial treatment, treated with hepatectomy and radiofrequency ablation, with the patient surviving six years.[21] Park et al described two hepatic metastases resected 3 years after primary tumor removal, with a 2-year RFS.[22] They recommended metastasectomy for well-controlled primary tumors with sufficient organ function and good PS,[21,22] particularly for oligometastatic recurrence. Our four cases received local treatment, achieving favorable prognoses, consistent with previous results.

Systemic therapy for ACC includes regimens, such as platinum doublets that contain cisplatin or carboplatin, with a reported median progression-free survival of 8.9–9.7 months, Immune checkpoint inhibitors for distant metastases yield a median progression-free survival of 4.5–6.6 months and remained unestablished as standard therapy.[23] Whole-genome-based pharmacotherapy combined with surgical expertise may improve prognosis. Parikh et al revealed that ACC overexpressed MYB/MYBL1 and is related to NOTCH1 mutations, demonstrating similar transcriptional profiles across tissues.[24] NOTCH1 is a poor prognostic factor in ~20% of ACC cases, but the development of NOTCH inhibitors demonstrates potential.[25]

Regarding the long-term prognosis of patients with ACC, current cases demonstrated 5-year and 10-year survival rates of 90% and 75%, respectively, indicating an improvement compared with previously reported rates (5-year, 52%–85%; 10-year, 29%–63%).[9,13,14] This improvement may be associated with the relatively older data in previous large-scale studies and advancements in surgical techniques, radiotherapy, and chemotherapy, which have improved both treatment safety and efficacy.

In addition to resection margins and recurrence patterns, tumor size may also be a prognostic factor in tracheobronchial ACC. A recent large-scale study by Behbahani et al using the US National Cancer Database identified tumor diameter ≥40 mm as an independent negative prognostic factor for OS in patients with primary tracheal ACC.[26] In our series, two patients had tumors exceeding this threshold. One (Case 4) had a 5.0-cm tumor with R2 resection and developed multiple liver metastases 87 months after surgery, ultimately succumbing to the disease. Another patient (Case 8) had a 4.0-cm tumor that was completely resected (R0) and remains recurrence-free at 78 months. These cases support the notion that larger tumors may be more likely to result in incomplete resection and potentially poorer outcomes, consistent with previously reported findings.

The main study limitation is that it is a single-institution report on a rare disease, with a small sample size of only 11 cases. Consequently, statistical analyses aimed at identifying prognostic factors were impossible. Conducting analyses that reflect the current treatment landscape by accumulating cases from multiple institutions is warranted.

In conclusion, surgical resection remains the cornerstone of treatment for tracheobronchial ACC, achieving favorable long-term outcomes through complete pathological resection. However, considering the lack of a viable airway substitute and anatomical constraints on resection length, achieving negative surgical margins is not always feasible, particularly when anastomotic dehiscence prevention is a priority. In such cases, postoperative radiotherapy is considered an effective strategy to mitigate the risk of local recurrence and prevent airway obstruction due to recurrent disease. Furthermore, distant metastases tend to occur in the late postoperative period; however, local treatment of oligometastatic disease may provide a survival benefit. These results indicate the importance of a multimodal treatment approach tailored to the specific challenges of tracheobronchial ACC.

CONFLICTS OF INTEREST

All authors have no conflict of interest to report in relation to this study.

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