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Large Carinal Chondrosarcoma: Resection of the Main Carina, Distal Trachea, and Orifices of the Mainstem Bronchi and Construction of a New Carina

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Data Interpretation D
Manuscript Preparation E
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Patient: Male, 23-year-old
Final Diagnosis: Carinal chondrosarcoma
Symptoms: Cough • dyspnea • fatigue
Clinical Procedure: —
Specialty: Oncology • Surgery

Objective: Rare disease

Background: Chondrosarcoma of the trachea is an extremely rare malignancy, and tumors located at the main carina are even rarer. Carinal resection and subsequent reconstruction of a new carina constitute a demanding surgical procedure that requires extensive expertise and close collaboration between the anesthetic and surgical teams. In this case report, we aim to highlight the rarity of chondrosarcoma of the main carina and emphasize the critical role of surgical intervention despite the complexity of the procedure. Furthermore, we aim to share our clinical experience and contribute an additional case to the limited literature.

Case Report: A 23-year-old man presented with dyspnea, fatigue, and cough. Chest computed tomography showed a lobulated soft-tissue mass anterior to the distal trachea and main carina. Fiberoptic bronchoscopy revealed widening of the main carina, clinically significant stenosis of the mainstem bronchi, and mucosal infiltration. Biopsy specimens obtained via mediastinoscopy demonstrated well-differentiated carinal chondrosarcoma. Through a right posterolateral thoracotomy, the distal trachea, main carina, and orifices of the mainstem bronchi were resected. A new carina was constructed and anastomosed to the distal trachea. Gross examination of the surgical specimen showed a well-circumscribed mass with a lobulated external surface measuring 7 × 6.5 × 5 cm. The patient remains well 17 years after tumor resection, without signs of recurrence.

Conclusions: Chondrosarcoma involving the distal trachea and main carina is an extremely rare tumor. Given the limited efficacy of chemotherapy and radiotherapy, surgical resection with reconstruction of a new carina is the primary treatment.

Keywords: Chondrosarcoma • Thoracotomy • Tracheal Diseases

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/953333

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Introduction

Chondrosarcoma of the trachea is an extremely rare malignancy, and tumors located at the main carina are even rarer. Chondrosarcomas are malignant cartilage-producing tumors that typically arise in the pelvis, femur, and ribs. To date, only 2 cases of chondrosarcoma of the main carina have been reported in the English-language literature. These tumors exhibit a lobulated architecture with increased cellularity, nuclear atypia, binucleation, and a myxoid or hyaline matrix, often infiltrating pre-existing bone trabeculae. Although carinal resection and the creation of a new carina constitute a complex surgical procedure, this approach is the recommended treatment, provided that complete resection with negative margins is feasible.

Case Report

A 23-year-old man presented to our outpatient clinic with a 6-month history of dyspnea on exertion, fatigue, and cough. His symptoms had worsened over the previous 2 weeks. Clinical examination and chest auscultation revealed decreased breath sounds in both hemithoraces, accompanied by inspiratory and expiratory wheezing. Chest X-ray showed widening of the mediastinum with a shadow protruding into the right hemithorax (**Figure 1**). Chest computed tomography (CT) demonstrated a lobulated soft-tissue mass within the middle mediastinum, anterior to the trachea and main bronchi, most likely originating from the tracheobronchial wall (**Figure 2**). The mass caused substantial narrowing of the distal trachea and both mainstem bronchi, without any lung parenchymal abnormalities. Fiberoptic bronchoscopy performed under neuroleptanalgesia showed widening of the main carina, clinically significant stenosis of the mainstem bronchi, and mucosal infiltration (**Figure 3**). Bronchoscopic biopsy results were nondiagnostic. Histological examination of biopsy specimens from the mediastinal mass obtained via standard mediastinoscopy confirmed the diagnosis of well-differentiated chondrosarcoma. Pulmonary function tests included spirometry, diffusing capacity of the lungs for carbon monoxide, and a flow-volume curve. Spirometry and diffusing capacity measurements indicated obstructive pulmonary disease, whereas the flow-volume curve revealed a fixed intrathoracic obstruction. These results indicated that the planned operation could be performed with acceptable perioperative risk. Conventional CT scans of the brain and upper and lower abdomen showed no evidence of metastatic disease; positron emission tomography-CT was not performed. Electrocardiogram and echocardiogram findings were normal. Complete blood count parameters (white blood cell count, hematocrit, and platelet count), as well as renal and liver function test results, were within normal limits.

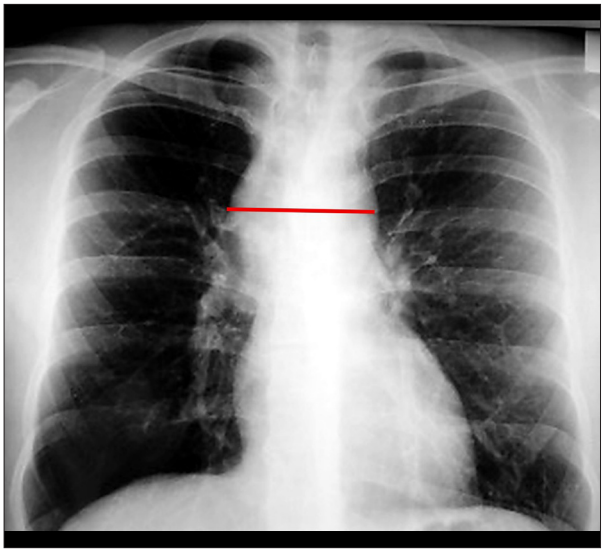

Figure 1. Posteroanterior chest X-ray showing widening of the mediastinum, with a shadow protruding into the right hemithorax. The line indicates the site of mediastinal widening.

After discussion with the patient regarding perioperative risks and emphasizing that surgery was the only effective treatment, the decision was made to proceed with the indicated procedure, and informed consent was obtained.

A right posterolateral thoracotomy was performed. After mobilization of the distal trachea, main carina, and both mainstem bronchi, a circumferential incision was made in the left main bronchus near the visible margins of the tumor. The transoral endotracheal tube was then removed, and a new tube was advanced into the left main bronchus through the operative field. Special attention was given to avoiding obstruction of the left upper lobe bronchus.

The distal trachea (one cartilaginous ring), main carina, and orifices of the mainstem bronchi were resected, and the resected tumor mass was sent for histopathological examination. Continuity of the tracheobronchial tree was restored by constructing a new carina through a side-to-side (laterolateral) anastomosis of the 2 mainstem bronchi. This newly created structure was then anastomosed end-to-end to the distal trachea. A continuous single-layer suturing technique using nonabsorbable polypropylene sutures (3 sutures) was utilized. A pedicled intercostal muscle flap was mobilized to reinforce the anastomotic line.

The patient was extubated in the operating room and transferred to the intensive care unit for continuous monitoring. Postoperative chest X-ray showed complete lung expansion without pneumothorax or pleural effusion (**Figure 4**).

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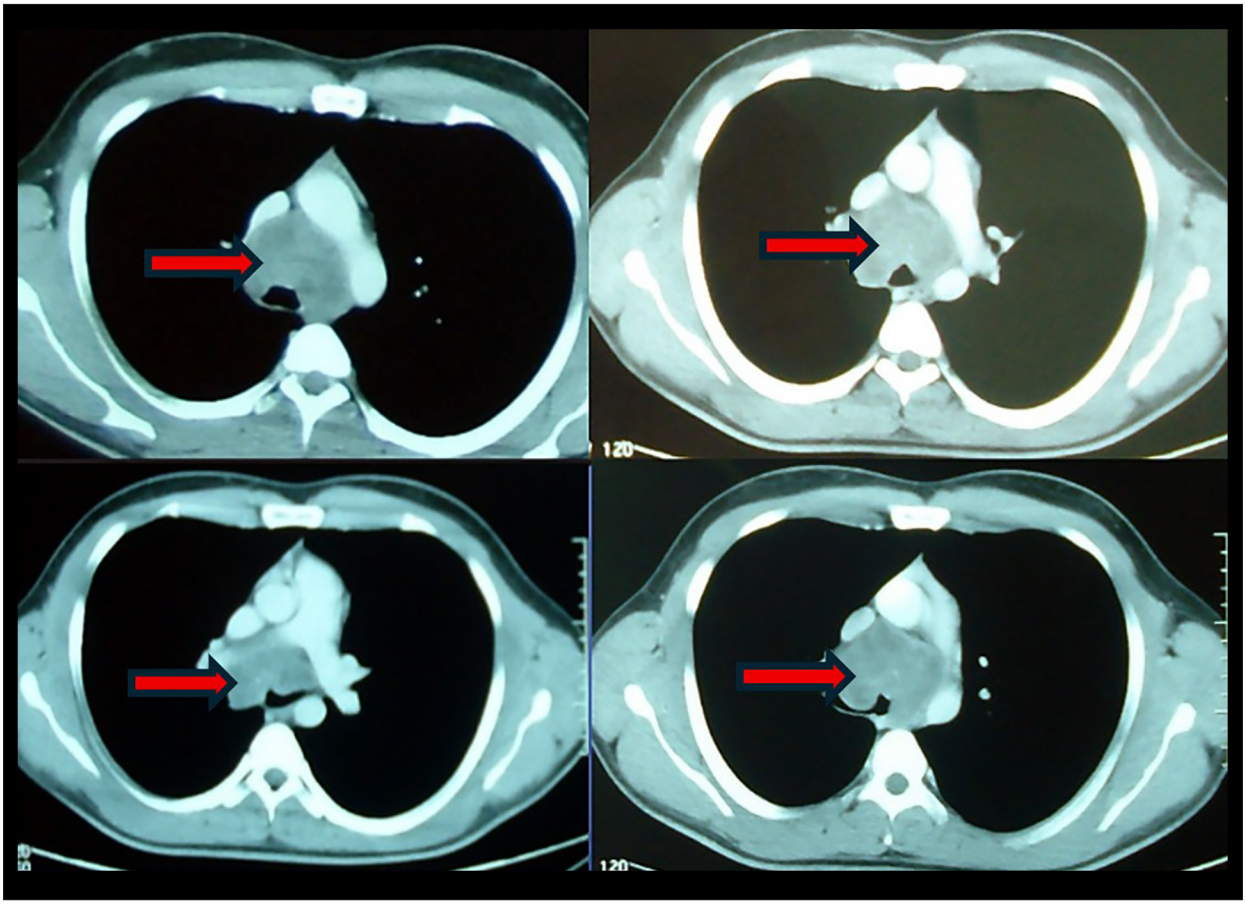

Figure 2. Chest computed tomography images at consecutive levels showing a large lobulated tumor mass causing narrowing of the distal tracheal lumen and the orifices of both mainstem bronchi. Arrows indicate these findings.

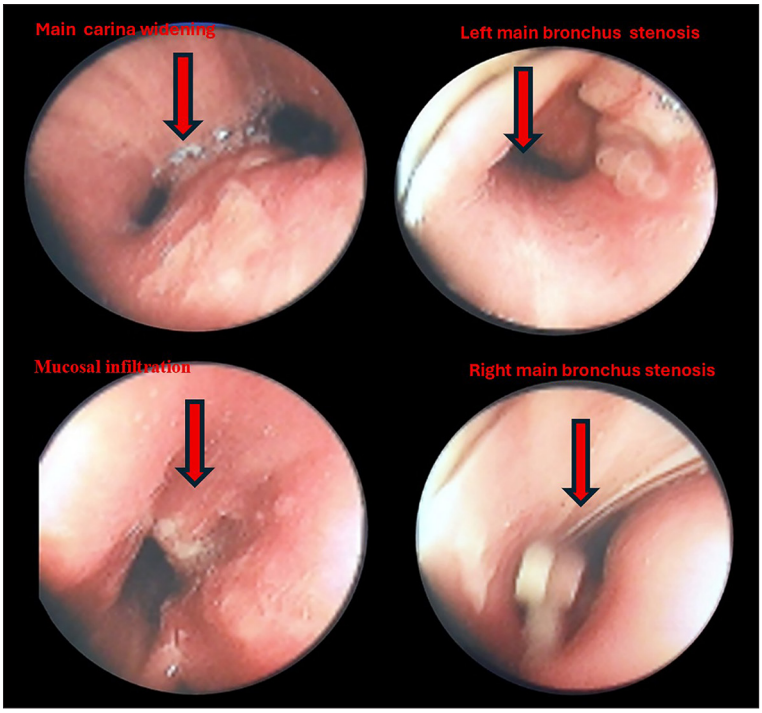

Figure 3. Fiberoptic bronchoscopic images showing widening of the main carina, stenosis of the mainstem bronchi, and mucosal infiltration. Arrows indicate these findings.

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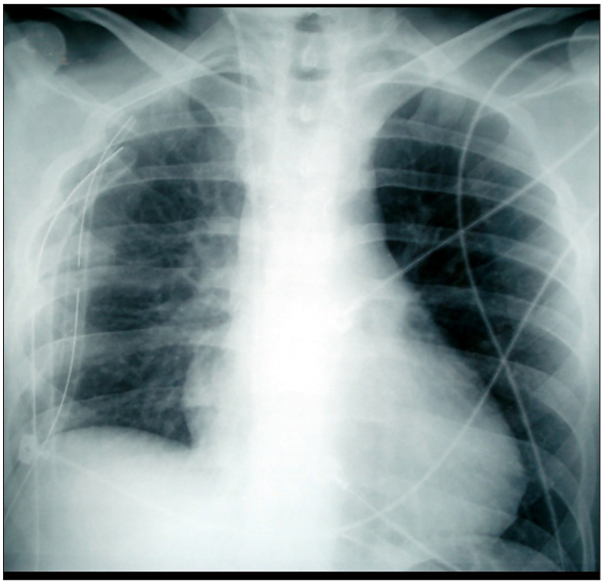

Figure 4. Postoperative chest X-ray showing complete expansion of both lungs without pneumothorax or pleural effusion. Chest drainage tubes are visible.

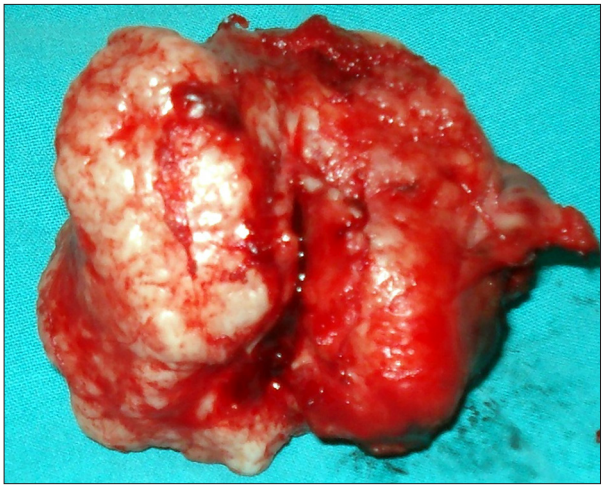

Figure 5. Photograph of the resected tumor, measuring 7 × 6.5 × 5 cm.

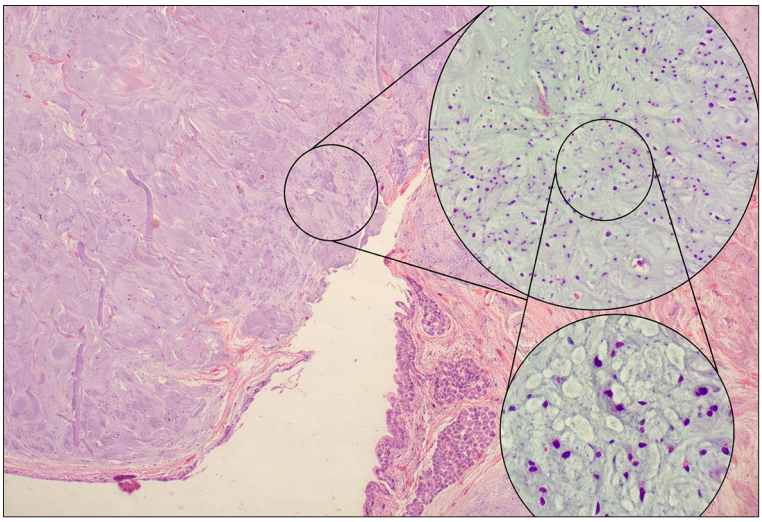

Figure 6. Histopathological image showing well-differentiated grade 1 chondrosarcoma with lobulated hyaline cartilage, increased cellularity, nuclear atypia, and binucleated chondrocytes. Areas of myxoid change and disrupted lacunar architecture further support the diagnosis.

On postoperative day 6 (POD 6), a small pneumothorax developed at the base of the right hemithorax, requiring retention of the chest tube for an additional 7 days. The chest tube was removed after cessation of the air leak and complete lung expansion. The patient was discharged from the hospital on POD 15.

Gross examination of the surgical specimen showed a well-circumscribed mass with a lobulated external surface measuring 7 × 6.5 × 5 cm. Cut surfaces were relatively homogeneous and whitish, with focal yellowish areas and a firm texture (**Figure 5**). Histopathological examination demonstrated a well-differentiated neoplasm consisting of chondrocytes with mild nuclear atypia and binucleated forms, in the absence of necrotic changes. Immunohistochemistry findings were positive for S100, and the Ki-67 proliferation index was less than 5% in the neoplastic cell nuclei. These findings were compatible with a well-differentiated grade 1 chondrosarcoma with negative resection margins (**Figure 6**).

Four months postoperatively, the patient presented with acute respiratory failure, and chest CT showed complete obstruction of the right main bronchus. Fiberoptic bronchoscopy revealed complete obstruction of the right main bronchus by fleshy necrotic tissue; loose sutures were visible within the necrotic debris (**Figure 7A**). After neodymium-doped yttrium aluminum garnet (Nd: YAG) laser ablation, all necrotic tissue and visible sutures were removed (**Figure 7B**). The procedure was performed over 3 sessions. Two months later, follow-up fiberoptic bronchoscopy

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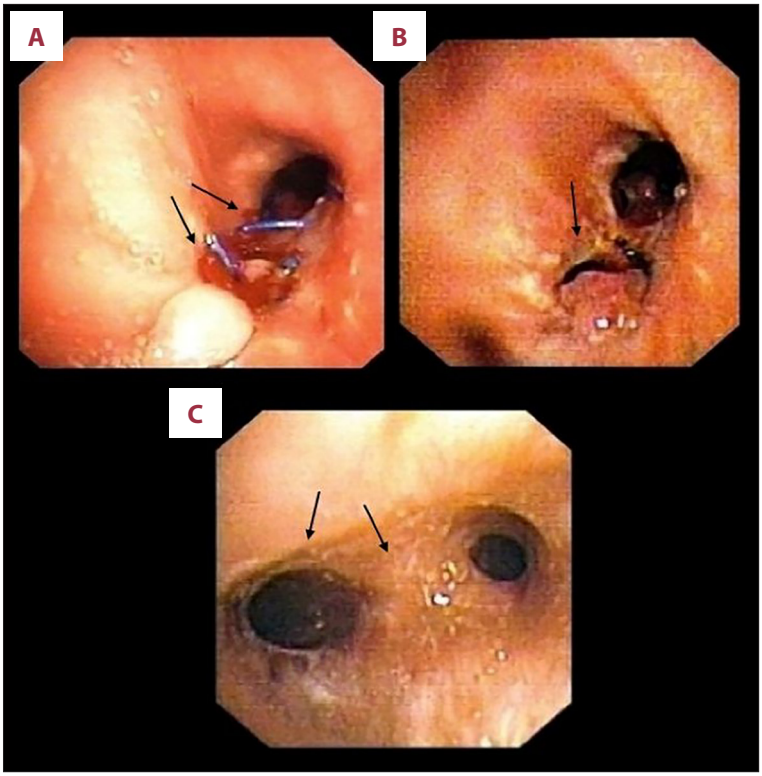

Figure 7. Fiberoptic bronchoscopic images. (**A**) Complete obstruction of the right main bronchus by fleshy necrotic tissue. A polypropylene suture is visible. (**B**) Appearance after the first session of neodymium-doped yttrium aluminum garnet (Nd: YAG) laser treatment. (**C**) Appearance 2 months after the third session of Nd: YAG laser treatment, showing complete patency of the distal trachea, newly constructed carina, and orifices of both mainstem bronchi, without intraluminal foreign tissue. Arrows indicate these findings.

detected no abnormalities (**Figure 7C**). Histological evaluation of the excised necrotic tissue revealed an inflammatory reaction without evidence of recurrence. Because the tumor was completely resected with negative margins, neither chemotherapy nor radiotherapy was administered.

Postoperatively, the patient was monitored every 3 months for the first year and every 6 months for the subsequent 4 years. Follow-up included clinical examination, conventional thoracic and upper abdominal CT scans, and comprehensive laboratory evaluations (complete blood count, urea, creatinine, transaminases, alkaline phosphatase, bilirubin, gamma-glutamyltransferase, and lactate dehydrogenase). For the next 5 years, annual chest and upper abdominal CT scans showed no abnormalities. After this initial 10-year period, follow-up care was transferred to his primary care physician. During recent telephone contact, the patient reported undergoing annual low-dose chest CT scans that demonstrated no pathological findings. To date, the patient has been followed for a total of 17 years; he remains fully active and employed as a police officer.

Discussion

Chondrosarcoma of the trachea is an extremely rare malignancy; tumors located at the main carina are even rarer. Chondrosarcomas are malignant cartilage-producing tumors that typically arise in the pelvis, femur, and ribs. They exhibit a lobulated architecture characterized by increased cellularity, nuclear atypia, binucleation, and a myxoid or hyaline matrix, often infiltrating pre-existing bone trabeculae. The Ki-67 proliferation index is low in grade 1 tumors and progressively higher in grade 3 lesions. Tumor grading is based on cellularity, nuclear atypia, and mitotic activity; higher-grade lesions demonstrate more aggressive behavior and a substantially worse prognosis. Tracheal chondrosarcomas tend to be low-grade, slow-growing tumors with a low risk of metastasis, resulting in an overall 10-year survival rate of approximately 95% [1,2]. According to the 2020 World Health Organization classification, chondrosarcomas are divided into 8 subtypes and may occur throughout the body; most arise in the extremities (femur and humerus) and axial skeleton (iliac bone and spine) [3,4]. Other sites include the head, neck, and ribs [5,6]. Extraskeletal chondrosarcomas are uncommon, representing fewer than 2% of all chondrosarcomas [7,8].

Chondrosarcomas generally have an equal incidence in both sexes, whereas tracheal chondrosarcoma occurs more frequently in men (91%) [9]. These tumors can be classified as primary when arising de novo from normal hyaline cartilage or secondary when resulting from malignant transformation of benign precursors, such as enchondromas. In the present case, histopathological examination demonstrated a well-differentiated neoplasm consisting of chondrocytes with mild nuclear atypia and binucleated forms; necrotic changes were not detected. Immunohistochemistry findings were positive for S100,

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and the Ki-67 proliferation index was below 5% in the neoplastic cell nuclei. The final diagnosis was a well-differentiated, grade 1 chondrosarcoma. These findings, combined with negative surgical margins, may explain the patient's long-term recurrence-free survival.

The first case of tracheal chondrosarcoma was published by Jackson and Jackson in 1959 [10]. A 2022 review identified only 35 cases reported in the English-language literature between 1959 and 2020 [11]. A subsequent PubMed search by our group—covering 2020 to 2025—identified 3 additional cases [12-14]. We also identified a clinical video presentation from the 2024 annual meeting of the American Association for Thoracic Surgery [15], bringing the total number of reported cases to 39. Among these cases, 2 young patients were diagnosed with Maffucci syndrome [16,17]. This syndrome is associated with mutations in the isocitrate dehydrogenase genes *IDH1* and *IDH2*; it is clinically characterized by multiple enchondromas, which carry an increased risk of malignant transformation [18]. Other pathological conditions associated with the development of chondrosarcoma include Ollier disease and Paget disease [11].

Most tracheal chondrosarcomas are diagnosed in older men, with a median age of 68 years. The tumor is generally asymptomatic initially; symptoms become apparent only when the mass occupies most of the tracheal lumen (≥75%) [11]. Dyspnea is the predominant symptom; cough and wheezing are also present in approximately one-third of patients. Almost half of patients are initially misdiagnosed, primarily with asthma [2,3]. Most tumors are low-grade and well-differentiated (grade 1), with a median size of 3.0 cm, and predominantly involve the proximal trachea [2,11]. In some cases, chondrosarcoma has been diagnosed in the setting of a previously known tracheal enchondroma [19]. Most cases involve the proximal trachea (54%), followed by the distal (29%) and middle (17%) trachea [11]. To date, only 2 cases of chondrosarcoma of the main carina have been described, both of which were treated via resection and reconstruction. The first case of carinal chondrosarcoma resection was reported by Voltolini et al [20]; the second was reported by Gauthier et al, who performed resection via median sternotomy, with reconstruction requiring the creation of a new carina [15].

Given the rarity of tracheal chondrosarcoma, treatment guidelines have not been established, and therapeutic decisions are primarily based on evidence from published case reports [11]. Carinal resection and reconstruction of a new carina are the indicated treatment for chondrosarcoma originating from the main carina. This is a complex and demanding procedure associated with potentially fatal perioperative complications. As noted by Mathisen, the procedure requires familiarity with the principles of airway surgery, preservation of the tracheal blood supply, avoidance of anastomotic tension, and close cooperation with the anesthetic team [21].

The procedure can be performed via right posterolateral thoracotomy or median sternotomy, with or without extracorporeal membrane oxygenation (ECMO). Carinal reconstruction and restoration of tracheobronchial continuity may be achieved using either continuous or interrupted suturing techniques [22]. Ziaian et al concluded that the continuous suturing technique was associated with a lower frequency of restenosis than the interrupted technique [23]. Both absorbable and nonabsorbable sutures may be used, depending on the surgeon's preference. Suturing techniques and materials have been used successfully by many surgeons, with or without ECMO. We performed the procedure through a right posterolateral thoracotomy using nonabsorbable sutures and a continuous suturing technique, without ECMO.

Complications after carinal resection and reconstruction include anastomotic stenosis, tracheopleural fistula, and pulmonary and pleural infections. Our patient developed a small pneumothorax at the base of the right hemithorax that was managed conservatively. The chest tube remained in place for a total of 13 days, after which the patient was discharged with almost normal chest X-ray findings. Four months after surgical resection, the patient presented with acute respiratory failure due to complete obstruction of the right main bronchus by necrotic inflammatory tissue. Using a flexible bronchoscope, all necrotic tissue was removed by Nd: YAG laser ablation. Three bronchoscopy sessions were required to achieve complete patency of the right main bronchus. Histological examination of the removed necrotic tissue revealed no evidence of malignancy.

The Nd: YAG laser has an established role as a key palliative intervention for intraluminal malignancies of the tracheobronchial tree. When a tumor grows intraluminally and surgical resection is contraindicated due to comorbidities or local extension into adjacent anatomical structures, or in cases of intraluminal recurrence after resection, Nd: YAG laser ablation can be performed as palliative treatment [24]. Furthermore, this technique is valuable for reducing clinically significant luminal obstruction in patients scheduled for surgical resection, thereby facilitating safe passage of the endotracheal tube.

Palliative treatment is recommended for patients with metastatic disease, those who cannot tolerate surgical resection because of increased operative risk, and those who decline surgery [25]. Tumors that are inoperable for any of these reasons can be treated by endoscopic resection when they exhibit intraluminal growth. The procedure can be performed using a rigid or flexible bronchoscope. Rigid bronchoscopy enables both diagnostic tissue sampling and therapeutic tumor debulking via mechanical coring or laser resection. For inoperable

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tumors with predominantly extraluminal growth, radiotherapy—with or without chemotherapy—is a potential alternative. Patients with metastatic disease often require local treatment of the primary lesion, as well as systemic treatment for distant disease. However, chemotherapy has limited utility because chondrosarcoma is generally chemoresistant [25].

Conclusions

Surgical resection of the main carina for chondrosarcoma is the treatment of choice whenever feasible and can be performed via median sternotomy or right thoracotomy. Complete tumor resection with negative margins offers the potential for cure. Nonsurgical treatment is generally less effective and is primarily recommended when the tumor is deemed inoperable. Given the rarity of tracheal chondrosarcoma, no treatment guidelines have been established, and therapeutic decisions are based on evidence from published cases.

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Department and Institution Where Work Was Done

Thoracic Surgery Department, Athens Medical Center, Athens, Greece.

Patient Consent

The patient provided informed consent for publication of this report and accompanying images.

Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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