

Tracheobronchopathia osteochondroplastica complicated with a pulmonary adenocarcinoma: A rare case report

*Akciğer adenokarsinomu ile komplike trakeobronkopatia osteokondroplastika:
Nadir bir olgu sunumu*

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ABSTRACT

A 67-year-old female patient presented with a lung nodule one month ago which was detected by physical examination. She was diagnosed with tracheobronchopathia osteochondroplastica complicated with right lower lobe nodules by chest computed tomography, bronchoscopy, and pathological examinations of the biopsy specimens. We performed lobectomy and lymphadenectomy by video-assisted thoracoscopic surgery and small pulmonary lesions were diagnosed as pulmonary adenocarcinomas. At 36 months of follow-up, the patient was free from recurrence. In conclusion, tracheobronchopathia osteochondroplastica complicated with a pulmonary adenocarcinoma is rare, and clinicians should be aware of this possibility, particularly in patients with pulmonary malignancy and tracheal irregularities on chest imaging. Otherwise, tracheobronchopathia osteochondroplastica can be easily misdiagnosed as malignant invasion of the trachea.

Keywords: Pulmonary adenocarcinoma, tracheobronchopathia osteochondroplastica, video-assisted thoracoscopic surgery.

Tracheobronchopathia osteochondroplastica (TO) is a rare benign airway disease characterized by multiple submucosal osseous and cartilaginous nodules in the anterior and lateral walls of the trachea.^[1] The etiology and pathophysiology of TO remains obscure; however, neoplastic disorders has been attributed to the etiology.^[2] Tracheobronchopathia osteochondroplastica with a pulmonary adenocarcinoma (PA) is extremely rare with 20 cases reported since the first case described by Kato et al.^[3] in 1975.

In this article, we report a rare case of TO with PA in the light of literature data.

ÖZ

Altmış yedi yaşında kadın hasta, bir ay önce fizik muayene ile tespit edilen bir akciğer nodülü ile başvurdu. Hastaya göğüs bilgisayarlı tomografi, bronkoskopi ve biyopsi örneklerinin patolojik incelemesi ile sağ alt lob nodülleri ile komplike trakeobronkopatia osteokondroplastika tanısı kondu. Video yardımlı torakoskopik cerrahi ile lobektomi ve lenfadenektomi yapıldı ve küçük akciğer lezyonları akciğer adenokarsinomu olarak tanımlandı. Otuz altı aylık takip süresince, hastada nüks görülmedi. Sonuç olarak, akciğer adenokarsinomu ile komplike trakeobronkopatia osteokondroplastika nadir görülmekte olup, özellikle göğüs görüntülerinde akciğer malignitesi ve trakea düzensizlikleri olan hastalarda hekimler bu olasılığın farkında olmalıdır. Aksi halde, trakeobronkopatia osteokondroplastika kolayca trakeanın malign invazyonu ile karıştırılabilir.

Anahtar sözcükler: Akciğer adenokarsinomu, trakeobronkopatia osteokondroplastika, video yardımlı torakoskopik cerrahi.

CASE REPORT

A 67-year-old female patient was referred to our hospital in October 2018 due to a lung nodule founded by the chest radiography during the physical examination. The patient only complained of chronic cough. She was a non-smoker and had no history of tuberculosis, bronchial asthma or oncology. Chest computed tomography (CT) showed a nodule about 2.2 cm in size in the right lower lobe (Figure 1a, b) with diffuse, irregular nodules and calcification in the tracheal wall (Figure 2a-d). Bronchoscopy was performed and revealed multiple protruding sessile nodules covered with the anterior and lateral walls

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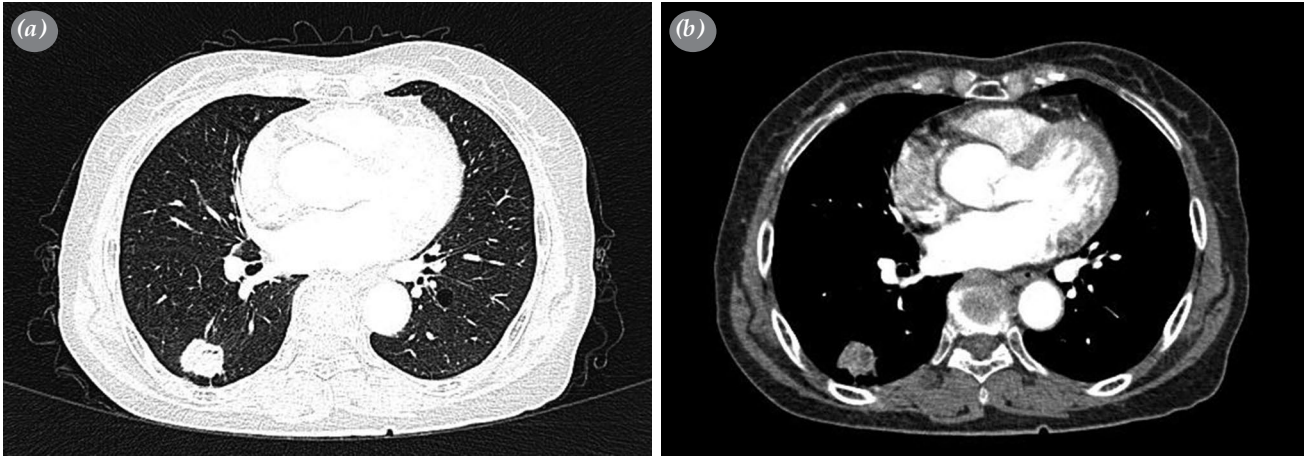


Figure 1. Computed tomography images showing a nodule in about 2.2 cm size in the right lower lobe, presented as burr and pleural traction sign. (a) Lung window, (b) mediastinal window.

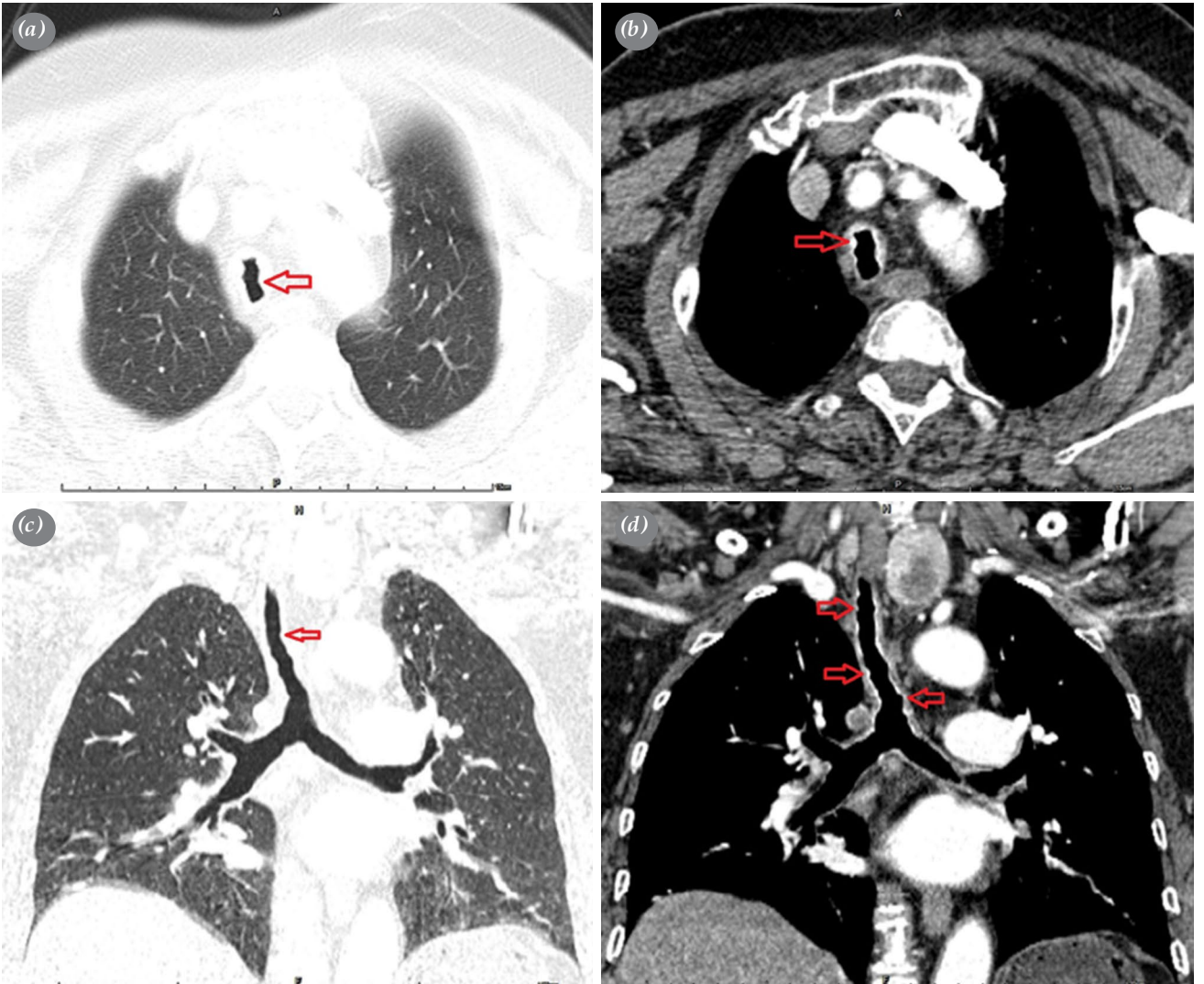


Figure 2. Computed tomography images showing calcifications on the anterior and lateral walls of the trachea that extended from the origin of the trachea to the carina (arrow). (a and c) Lung window, (b and d) Mediastinal window.

of the trachea without any obvious endobronchial lesions (Figure 3a, b). Histopathological examination showed cartilaginous and osseous submucosal nodules consistent with the diagnosis of TO (Figure 4a). Tumor markers (CEA, CA19-9, CYFRA21-1, proGRP, NSE, and SCC) were all negative. Sputum was also negative for acid-fast bacilli in three occasions. After rapid anesthesia induction, the bronchoscope passed through

the stenosed region, and a 6.5-mm endotracheal tube was inserted with bronchoscopic assistance, and one lung ventilation with bronchial blocker. We performed right lower lobectomy and systematic lymphadenectomy by video-assisted thoracoscopic surgery (VATS), and wedge resection was performed and the biopsy was confirmed as an PA in the operations firstly (Figure 4b). The pathological stage of this patient

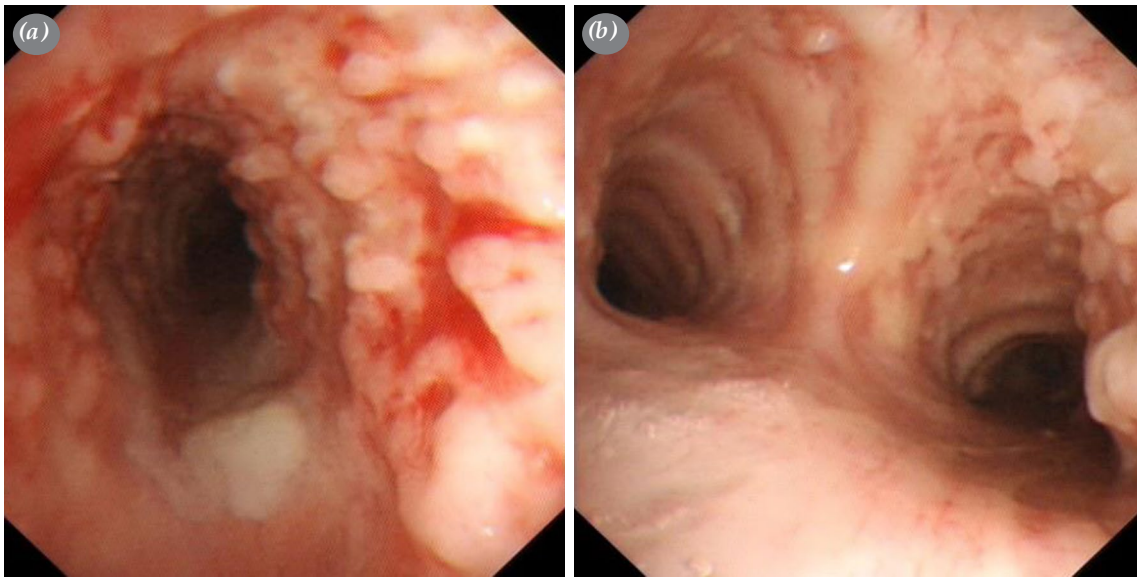


Figure 3. (a, b) Bronchoscopy showing multiple protruding sessile nodules covered with the anterior and lateral walls of trachea without any obvious endobronchial lesions.

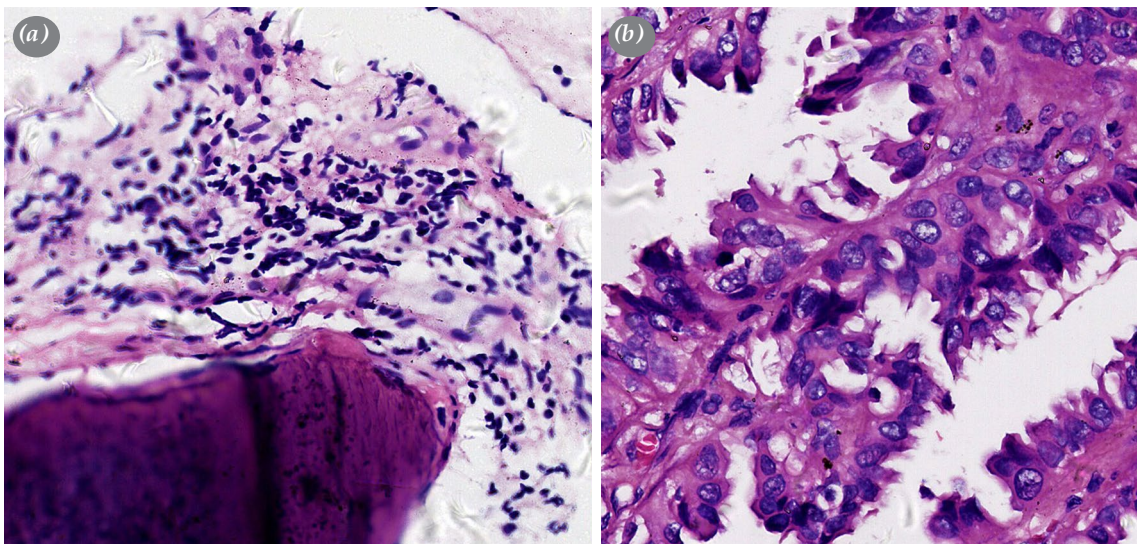


Figure 4. Histopathological findings. (a) Tracheobronchopathia osteochondroplastica illustrate submucosal ossification and cartilage formation, accompanied by a number of inflammatory cells (H&E, $\times 400$). (b) The lower lobe nodules of the right were invasive adenocarcinomas with 90% of acini and 10% micropapillary (H&E, $\times 400$).

Table 1. Summary of reported cases of tracheobronchopathia osteochondroplastica complicated with pulmonary adenocarcinoma

Case	Age (year)	Sex	Chief complaint	TO location	Tumor location	Treatment	Author	Year
1	62	F	Cough	T (-), B (+)	RLL	Conservative	Kato et al. ^[3]	1975
2	62	F	--	--	--	--	Ozawa et al. ^{[15]a}	1980
3	60	F	Cough	T (-), B (+)	LLL	Chemotherapy	Wagai et al. ^[16]	1986
4	58	F	Asymptomatic	T (+), B (+)	RUL	Surgery	Ikeda et al. ^[17]	1993
5	80	F	--	--	--	--	Yoshimoto et al. ^{[18]a}	1993
6	63	F	--	--	--	--	Araki et al. ^{[19]a}	1994
7	70	F	--	--	--	--	Antaku et al. ^{[20]a}	1994
8	67	F	--	--	--	--	Kikumoto et al. ^{[21]a}	1996
9	86	M	Back pain	T (+), B (+)	RUL	Conservative	Yokoyama et al. ^[7]	1996
10	75	M	Asymptomatic	T (+), B (+)	RUL	Surgery	Ueshima et al. ^[4]	2000
11	77	F	Dyspnea	T (+), B (-)	RL	Pleural biopsy	Yasuhiro et al. ^[22]	2007
12	65	F	Cough and hemoptysis	T (+), B (-)	LLL	Chemotherapy	Wu et al. ^[23]	2014
13	56	M	Cough and hemoptysis	T (+), B (-)	RUL	--	İlvan et al. ^[8]	2014
14	-	-	--	--	--	--	Luo et al. ^{[6]b}	2017
15	69	F	Cough	T (+), B (-)	LUL	Chemotherapy	Sneha et al. ^[24]	2021
16	67	F	Cough	T (+), B (-)	RLL	Surgery	<i>Present case</i>	2022

TO: Tracheobronchopathia osteochondroplastica; T: Trachea; B: Bronchi; RLL: Right lower lobe; LLL: Left lower lobe; RUL: Right upper lobe; RL: Right lobe; LUL: Left upper lobe; a: Only the abstract was available; b: Six cases only the abstract was available.

was IA3 according to the American Joint Committee on Cancer (AJCC) staging system. After 36 months of follow-up, CT showed diffuse, irregular nodules and calcification in the tracheal wall, and the patient was free from symptoms and without any evidence of recurrence.

DISCUSSION

The etiology and pathophysiology of TO still remains unclear.^[1] Recently, some studies have shown an association between TO and neoplasm, arguing that this may be a factor in disease progression.^[2,4] To date, approximately 100 cases have been published, since the first one reported that TO was associated with malignant tumors by Narutomi^[5] in 1964. Luo et al.^[6] reported that 18% (13/73) of TO patients were associated with malignancy, and the most common histological type in concomitant cancers was PA, but we could not obtain satisfactory information about these patients. Yokoyama et al.^[7] reported 14 Japanese cases of TO complicated with lung cancer, with the highest proportion of lung adenocarcinoma (8/14). Although some cases of TO were found to be associated with malignancy, significant evidence for the etiological relationship between TO and malignancy is lacking.

Yokoyama et al.^[7] reported the mean age of TO complicated with lung cancer was around 67 years, and the incidence of TO in men was similar to women. It was further found that the majority were women in TO with a PA. Our review of the 15 cases of TO complicated with PA showed more women than men (12:3), with a mean age of 67.8 years (Table 1). Clinical manifestations of TO complicated with lung cancer are quite variable and non-specific, and the most frequent complaint is chronic cough,^[8] which is similar to the case in this report. According to current data in the literature, in six out of 10 TO complicated with PA patients with detailed recorded data, cough is the main symptom, which may be related to TO invasion of the trachea.

Chest CT and bronchoscopy may be useful in demonstrating submucosal calcified nodules.^[1] However, there are still a number of missed diagnosis cases by radiologists and, therefore, a better understanding of the disease is of utmost importance. In this case, the initial X-ray imaging examination did not reveal a diagnosis of TO disease. Therefore, in case of TO complicated with cancer, careful examination is necessary, including bronchoscopy, chest CT, and pathological examination of the biopsy specimens.

Lobectomy requires one-lung ventilation, causing stenosis and deformation of the tracheal lumen due to TO, which may lead to difficult placement of double-lumen tracheal intubation and even damage to the airway.^[9] Myojo et al.^[10] reported that performing bronchial blocker catheter was safe and effective in patients with TO with PA. We evaluated the degree of tracheal stenosis before surgery, selected endotracheal intubation under bronchoscopy guidance, and one-lung ventilation with a bronchial blocker. The anesthesia process was smooth and no airway damage occurred. The literature reveals that using the laryngeal mask airway anesthesia is safe for patients with TO.^[11] As this patient was expected to undergo prolonged thoracoscopic surgery in the lateral position in this report, considering the risk of aspiration, endotracheal intubation was chosen over a laryngeal mask. Nonetheless, further studies are needed to evaluate whether laryngeal mask combined with bronchial occlusion is applicable in patients with TO complicated with lung cancer.

There is no consensus on the treatment of TO complicated with PA, and most TO treatments are palliative, with a major focus on symptoms.^[12] Ueshima et al.^[4] reported a case of TO complicated with PA in whom right upper lobe resection was performed. In our case, the right lower lobectomy was performed by powered linear cutter, and successful postoperative recovery. In general, TO involves the trachea and the main bronchi, rarely involves the lobe and segment bronchi. The bronchial stump in lung lobectomy and segmentectomy is not strengthened. Multiple submucosal osseous and cartilaginous nodules in the tracheal wall may affect surgical stump healing. For pneumonectomy and trachea, sleeve resection patients of the bronchial stump needed to be covered with pleura and intermittent suture reinforcement. Bronchopleural fistulas can occur after pulmonary resections as a complication with high morbidity and mortality.^[13] The prognosis of patients with tumors with TO is usually favorable, with most cases rarely progressing over a few years.^[14] Our patient is under follow-up for 36 months and her condition remains unchanged.

In conclusion, clinicians should be aware of this possibility, particularly in patients with pulmonary malignancy and tracheal irregularities on chest imaging. For patients with a pulmonary adenocarcinoma, tracheobronchopathia osteochondroplastica can be easily misdiagnosed as a malignant tracheal invasion, and chest computed tomography, bronchoscopy and positions of pathological examinations of the

biopsy specimens should be performed in identifying patients with a high suspicion of tracheobronchopathia osteochondroplastica.

Patient Consent for Publication: A written informed consent was obtained from patient.

Data Sharing Statement: The data that support the findings of this study are available from the corresponding author upon reasonable request.

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