

Tracheobronchopathia osteochondroplastica: a case associated with coal processing and asthma

Erdoğan Kunter (*), Turgut Işıtmangil (**), Zafer Kartaloğlu (*), Rauf Görür (**), Şükrü Yıldırım (***)

SUMMARY

A 41-year-old man admitted with the complaint of chronic cough. His complaint had begun while working at a coal processing facility, but continued after changing his job. Previously he had been treated with the diagnosis of acute bronchitis and/or asthma for several occasions. Initial evaluation tests and examinations confirmed the diagnosis of asthma but response to the treatment was unsatisfactory. Further investigation with computed tomography of the thorax, and bronchoscopy and histopathological examination confirmed the diagnosis of tracheobronchopathia osteochondroplastica. To our knowledge this is the first case associated with both coal dust exposure without interstitial involvement, and asthma. Tracheobronchopathia osteochondroplastica should be considered in asthma cases with chronic cough resistant to therapy, especially when there is a history of inorganic dust exposure.

Key words: *Asthma, coal, tracheobronchopathia osteochondroplastica*

ÖZET

Tracheobronchopathia osteochondroplastica: kömür işleme işi ve astma ile ilişkili bir olgu

Kırk bir yaşında erkek hasta kronik öksürük şikayetiyle başvurdu. Şikayeti bir kömür işleme tesisinde çalışırken başlamış, ancak iş değişikliğine rağmen devam etmişti. Daha önce birçok kez akut bronşit ve/veya astma tanılarıyla tedavi görmüştü. Yapılan ön tetkik ve muayeneler sonucu astma tanısı doğrulandı, ancak tedaviye yeterli cevap alınmadı. Bilgisayarlı toraks tomografisi, bronkoskopi ve histopatolojik incelemeyle yapılan ileri incelemeler sonucu, tracheobronchopathia osteochondroplastica tanısı doğrulandı. Bildiğimiz kadanyla olgumuz interstisyel tutulum olmayan kömür tozu maruziyeti ve astmanın birlikte eşlik ettiği ilk tracheobronchopathia osteochondroplastica olgusudur. Tedaviye dirençli öksürük şikayeti olan astma hastalarında özellikle inorganik toz maruziyeti mevcutsa, tracheobronchopathia osteochondroplastica akla getirilmelidir.

Anahtar kelimeler: *Astma, kömür, tracheobronchopathia osteochondroplastica*

Introduction

Tracheobronchopathia osteochondroplastica (TO) is a rare disease of unknown etiology, which was first described by Wilks in 1857. TO is seen with a frequency of 0.4 percent at bronchoscopy (1-6). The disease is characterized by cartilaginous or bony nodular projections into the tracheobronchial lumen, with sparing of the posterior membranous portion of the tracheobronchial tree. The nodules may be either focal or diffuse. There is a 3:1 male predilection, and the disease typically manifests in patients in their mid-50s. However, recently a 9-year-old girl with TO has been reported (7). The diagnosis is usually established years after the initiation of the process because of the chronic and asymptomatic nature of the condition. Most patients are asymptomatic, but presentation may include persistent cough, hoarseness, dyspnea at exertion, recurrent infection, wheezing and hemoptysis. The last symptom occurs when opposing nodules rub against each other, causing erosion of the mucosa and subsequent bleeding (1). Although most patients have a favorable prognosis, respiratory insufficiency may complicate (3,8). Diagnosis may be difficult with a standard chest radiography because of poor imaging technique and an inadequate search. Computed tomography (CT) improves both the detection and characterization of central airway disease. Bronchoscopy, however, remains the primary procedure for the diagnostic work-up of this disease entity. There is currently no specific treatment to remove the abnormal tissue growth or prevent the development of new nodules. Linear tracheoplasty may be required in patients with symptomatic airway obstruction (9).

Case Report

A 41-year-old male patient admitted with the complaint of chronic cough. He had never smoked. His complaint had lasted for 5 years, beginning after a 10-year period of working at a coal processing facil-

* Department of Thoracic Diseases, Gülhane Military Medical Faculty Haydarpaşa Teaching Hospital

** Department of Thoracic Surgery, Gülhane Military Medical Faculty Haydarpaşa Teaching Hospital

*** Department of Pathology, Gülhane Military Medical Faculty Haydarpaşa Teaching Hospital

This case was presented as a poster presentation at the 15th World Bronchology Congress

Reprint request: Dr. Erdoğan Kunter, Department of Thoracic Diseases, Gülhane Military Medical Faculty Haydarpaşa Teaching Hospital, Acıbadem Cad. Üsküdar-34660, Istanbul, Turkey
E-mail: erkunter@hotmail.com

Date submitted: April 17, 2008 • **Date accepted:** August 10, 2008

ity with remissions and exacerbations but becoming more persistent over time. He had changed his work with the hope that he would feel better. He had been treated with the diagnoses of acute bronchitis and/or asthma for several occasions and numerous antitussive prescriptions were given as well. Current and previous posteroanterior chest radiographs were not suggesting any abnormality. Minimal and scattered rhonchi were heard at chest examination while other organ systems were normal at physical examination. His medical history was unremarkable except that his complaint of coughing worsened when exposed to cigarette smoke, perfumes and some other odors. His routine biochemical and hematologic tests were within normal limits and he was not on any medication at presentation. Pulmonary function tests including carbon monoxide diffusion test were within normal limits but the early reversibility test with inhaled salbutamol was positive. He had a normal peripheral eosinophil count and slightly elevated serum total IgE level (140 IU/mL) but negative prick test with common antigens including common aeroallergens. The patient was put on inhaled long acting beta-2 agonist plus corticosteroid (formoterol and budesonide) along with n-acetyl cysteine as a mucolytic agent for 2 weeks. Inappropriate and insufficient clinical response, i.e. persistent coughing despite improved auscultation findings, to this treatment led us to investigate further, and a chest CT was performed, which demonstrated irregular calcification and projections from the tracheal wall, sparing the posterior wall, into the lumen and bilateral minimal cylindrical bronchiectatic changes (Figure 1). Although this appearance was strongly suggesting TO, bronchoscopy was performed to confirm the diagnosis and investigate any coexisting condition. With bronchoscopy multiple nodules with varying



Figure 1. Computed tomographic image of the affected trachea

dimensions (3-5 mm) were distributed over the anterolateral walls of the trachea. The lower two-third of the trachea and the proximal portions of the primary bronchi seemed to be the most affected sites. The number, dimension and frequency of the nodules were decreasing while going down through the main bronchi (Figure 2). These lesions were difficult to make a biopsy. Biopsy samples from the lesions and adjacent tissues and bronchial lavage samples were examined histopathologically and microbiologically. Disseminated submucosal ossifications were the only abnormality found with bronchoscopic procedures. Taking together the CT, bronchoscopy and biopsy findings, the diagnosis of TO was established.

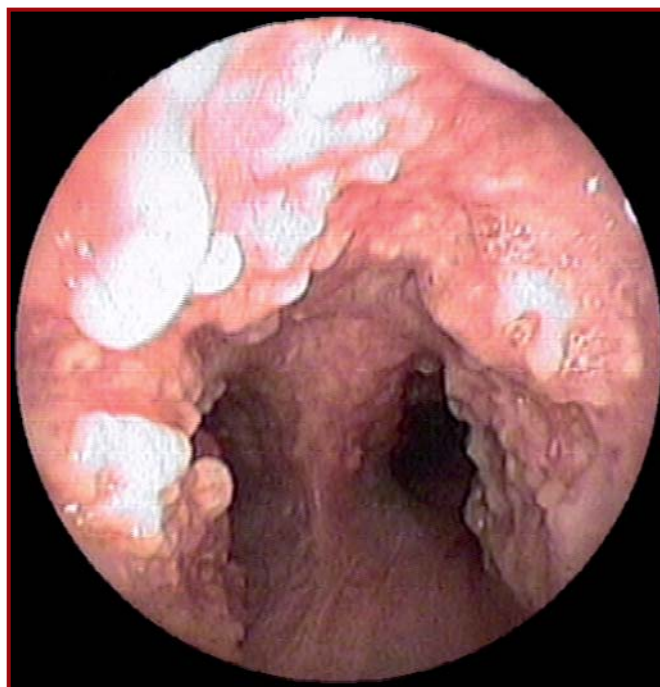


Figure 2. Bronchoscopic image of the lower trachea

Discussion

Although TO has a pathognomonic CT image, sometimes the radiological appearance may mimic other entities such as tracheobronchial amyloidosis, papillomatosis, rhinoscleroma, relapsing polychondritis, etc. Radiologically distinguishing tracheobronchial amyloidosis and papillomatosis from TO might be even more difficult. The classic radiographic appearance of tracheobronchial amyloidosis is nodular and irregular narrowing of the tracheal lumen. Lobar or segmental collapse may be seen with endobronchial obstruction due to amyloid deposition. In certain cases of diffuse involvement, there is a significant component of calcification and ossification of the lesions. In such circumstances, differentiation is made on the basis of posterior membrane sparing in

TO. On the other hand, papillomatosis results from infection of the upper respiratory tract by the human papillomavirus and may show malignant degeneration. Multifocal infection or aspiration of infected tissue from laryngeal papillomas distally may lead to infection of the trachea, the bronchi or even the alveoli. Radiographic findings include nodular narrowing of the airway that may be either focal or diffuse. TO has also been reported as a focal disease but in papillomatosis white and polypoid appearance at bronchoscopy and lack of ossification may suffice to diagnose (1,10). However, malignancies, including lung cancer, have been reported to be associated with TO and needs to be excluded (5,11,12).

The cause of the condition is unknown but several theories, including exposure to inhaled noxious substances have been postulated. One of the patients of Ashley has had pneumoconiosis and there are other published cases of TO, in whom silicosis has been accused for the development of the disease (11,13-15). Our patient was exposed to coal dust but radiologically and clinically there was no evidence of pneumoconiosis.

In TO, spirometry frequently shows a mild obstructive pattern, but in spite of marked radiographic changes, patients are only rarely symptomatic since severe airway obstruction is unusual (6,16,17). However, to our knowledge there is only one previous case report of TO that the patient meets the criteria of bronchial asthma as a coexisting disease (16).

TO is a rare condition but considering that most of the cases stay asymptomatic for a long period, the true prevalence of the disease might be higher than the reported numbers. Clinicians should include this disease in the list of differential diagnoses when confronted with symptoms like persistent and often productive cough, hemoptysis, dyspnea and wheeze. We think that thorax CT findings are very helpful to establish a diagnosis, but bronchoscopic evaluation will enable the clinician to prove it and look for the coexisting situations like malignancies. We hope that our case of TO associated with bronchial asthma and coal dust exposure contributes to future investigations, since in diseases of unknown etiology, describing the associated conditions may help to define the etiology and develop prevention and treatment strategies.

References

1. Prince JS, Duhamel DR, Levin DL, Harrell JH, Friedman PJ. Nonneoplastic lesions of the tracheobronchial wall: radiologic findings with bronchoscopic correlation. *Radiographics* 2002; 22: 215-230.
2. Mboti FB, Ninane V, Larsimont D, et al. Acute respiratory failure from tracheopathia osteoplastica. *Acta Chir Belg* 2005; 105: 227-228.
3. Shigematsu Y, Sugio K, Yasuda M, et al. Tracheobronchopathia osteochondroplastica occurring in a subsegmental bronchus and causing obstructive pneumonia. *Ann Thorac Surg* 2005; 80: 1936-1938.
4. Restrepo S, Pandit M, Villamil MA, Rojas IC, Perez JM, Gascue A. Tracheobronchopathia osteochondroplastica: helical CT findings in 4 cases. *J Thorac Imaging* 2004; 19: 112-116.
5. Hantous-Zannad S, Sebai L, Zidi A, et al. Tracheobronchopathia osteochondroplastica presenting as a respiratory insufficiency: diagnosis by bronchoscopy and MRI. *Eur J Radiol* 2003; 45: 113-116.
6. Zack JR, Rozenshtein A. Tracheobronchopathia osteochondroplastica: report of three cases. *J Comput Assist Tomogr* 2002; 26: 33-36.
7. Karlikaya C, Yuksel M, Kilicli S, Candan L. Tracheobronchopathia osteochondroplastica. *Respirology* 2000; 5: 377-380.
8. Nienhuis DM, Prakash UB, Edell ES. Tracheobronchopathia osteochondroplastica. *Ann Otol Rhinol Laryngol* 1990; 99: 689-694.
9. Grillo HC, Wright CD. Airway obstruction owing to tracheopathia osteoplastica: treatment by linear tracheoplasty. *Ann Thorac Surg* 2005; 79: 1676-1681.
10. Tukiainen H, Torkko M, Terho EO. Lung function in patients with tracheobronchopathia osteochondroplastica. *Eur Respir J* 1988; 1: 632-635.
11. Lundgren R, Stjernberg NL. Tracheobronchopathia osteochondroplastica. A clinical bronchoscopic and spirometric study. *Chest* 1981; 80: 706-709.
12. Simsek PO, Ozcelik U, Demirkazik F, et al. Tracheobronchopathia osteochondroplastica in a 9-year-old girl. *Pediatr Pulmonol* 2006; 41: 95-97.
13. Ashley DJB. Bony metaplasia in trachea and bronchi. *J Pathol* 1970; 102: 186-188.
14. Kissler W, Wierich W, Meesem D. Tracheobronchopathia osteochondroplastica und bronchialkarzinom. *Prax Klin Pneumol* 1977; 31: 660-663.
15. Mariotta S, Pallone G, Pedicelli G, Bisetti A. Spiral CT and endoscopic findings in a case of tracheobronchopathia osteochondroplastica. *J Comput Assist Tomogr* 1997; 21: 418-420.
16. Pinheiro GA, Antao VC, Muller NL. Tracheobronchopathia osteochondroplastica in a patient with silicosis: CT, bronchoscopy, and pathology findings. *J Comput Assist Tomogr* 2004; 28: 801-803.
17. Roggenbuck C, Hau T, de Wall N, Buss H. Simultaneous occurrence of tracheobronchopathia osteochondroplastica and mucoepidermoid carcinoma. *Chirurg* 1995; 66: 231-234.