JOURNAL OF
CONTEMPORARY MEDICINE

DOI: 10.16899/jcm.590811
J Contemp Med 2020;10(2):287-289

Case Report / Olgu sunumu

Tracheobronchopathia Osteochondroplastica: A Case Report

Trakeobronchopathia Osteochondroplastica: Bir Olgu Sunumu

Aysel Sunnetcioglu[1], Maşuk Celikel[1], Remzi Erten[2], Ufuk Cobanoglu [3]

[1]Department of Chest Diseases, Yuzuncu Yil University Medical Faculty, Van, Turkey
[2]Department of Pathology, Yuzuncu Yil University Medical Faculty, Van, Turkey
[3]Department of Thoracic Surgery, Yuzuncu Yil University Medical Faculty, Van, Turkey

Abstract

Tracheobronchopathia osteochondroplastica is a rare, benign condition characterized by the presence of multiple cartilaginous and / or bony submucosal nodules protruding into the tracheobronchial lümen. In general, Tracheobronchopathia osteochondroplastica is diagnosed incidentally during bronchoscopy or autopsy and is not associated with a specific disease. It can be asymptomatic or present with non-specific respiratory symptoms. We present a case of a 62 year-old female patient who was diagnosed with Tracheobronchopathia osteochondroplastica by Flexible bronchoscopy and thorax CT performed to investigate chronic cough.

Keywords: Tracheobronchopathia osteochondroplastica, Flexible bronchoscopy, cough

Öz

Trakeobronkopati Osteokondroplastika trakeobronşial lümen içine uzanarak kemik ve/veya kıkırdak submukozal nodüllerle karakterize, nadir, benign bir durumdur. Genel olarak, Trakeobronkopati Osteokondroplastika bronkoskopi veya otopsi sırasında tesadüfen teşhis edilir ve spesifik bir hastalıkla ilişkili değildir. Trakeobronkopati Osteokondroplastika, asemptomatik olabilir veya spesifik olmayan solunum semptomları ile mevcut olabilir. Kronik öksürüğü araştırmak için yapılan Fleksibl Bronkoskopi ve toraks BT ile tanı konulan 62 yaşında kadın hasta sunuldu.

Anahtar Kelimeler: Trakeobronkopati osteochondroplastica (TO), Fleksibl bronkoskopi, öksürük

INTRODUCTION

Tracheobronchopathia osteochondroplastica (TO) is an uncommon benign condition affecting the lumen of the tracheobronchial tree and characterized by abnormal chondrification and ossification of cartilages.[1] Because of the absence of cartilage in this region of the airway, these nodules involve the anterior and lateral walls of the trachea and the bronchus, sparing the posterior membranous wall.[2] The nodular lesions are sessile, calcified and vary in diameter between 1-10mm.

Tracheobronchopathia osteochondroplastica is a chronic disease, with male prevalence (male;female=3;1), and predominantly manifesting between the fifth and seventh decades of life.[3,4] In general, TO is diagnosed incidentally during bronchoscopy or autopsy and is not associated with a specific disease. Chronic cough is the most common complaint and about 54% of patients with TO complain.[5] In this study, we present a case of TO which was detected incidentally in a CT scan performed to investigate chronic cough.

Corresponding *(İletişim)*: Aysel Sunnetcioglu, Department of Chest diseases, Yuzuncu Yil University School of Medicine, Van, Turkey **E-mail *(E-posta)*:** izciaysel@mynet.com
Received *(Geliş Tarihi)*: 11.07.2019 **Accepted *(Kabul Tarihi)*:** 26.04.2020

CASE PRESENTATION

62-year-old female patient was admitted to hospital with the dry cough and occasional shortness of breath for 4 years. He had been treated with the diagnoses of acute bronchitis and/or asthma for several occasions and numerous antitussive prescriptions were given as well. She had never smoked. The patient's respiratory voices were normal and there was no other feature in the resume.

Laboratory studies, complete blood count, biochemical results, and erythrocyte sedimentation Arterial blood gases, pulmonary function tests and current-volume curve was normal. Current and previous posteroanterior chest radiographs were not suggesting any abnormality.

To investigate the underlying causes of chronic dry cough, a thorax CT was performed, which demonstrated multiple nodules lesions at the different levels of the trachea, and also in the right main bronchus. No significant luminal narrowing was observed, and typically the posterior membranous wall of the trachea was normally observed (**Figure 1**).

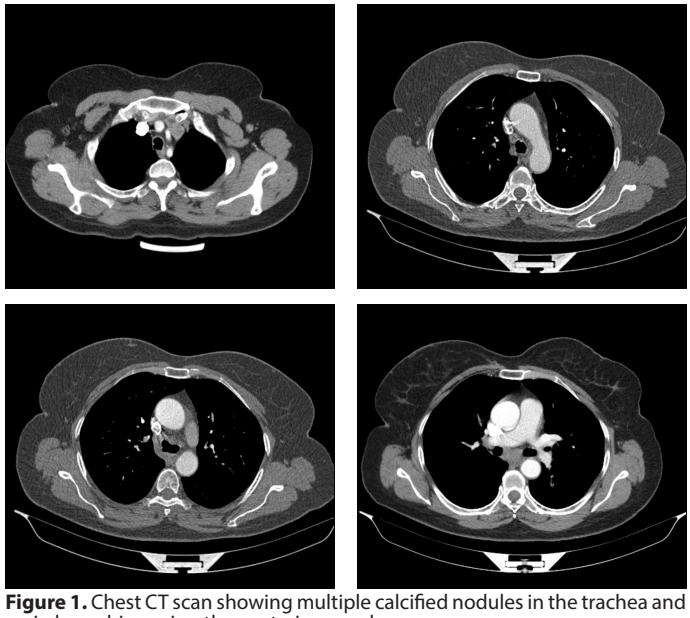


Figure 1. Chest CT scan showing multiple calcified nodules in the trachea and main bronchi, sparing the posterior membrane.

Flexible bronchoscopic (FB) examination of the patient is localized on the anterior and lateral walls of the trachea, a large number of white colored, hard-structured and irregularly appearing nodular lesions were detected. The number of nodules were decreasing while going down through the main bronchi (**Figure 2**). The biopsy was performed difficult because of the nodules lesions were hard-structured. Histopathological examination showed fragments of normal cartilage and bone formation with normal mucos (**Figure 3**).

Taking together the thorax CT, FB and biopsy findings, the diagnosis of TO was established. No specific treatment for TO was offered to patient. The patient remains under clinical follow-up with management of symptoms.

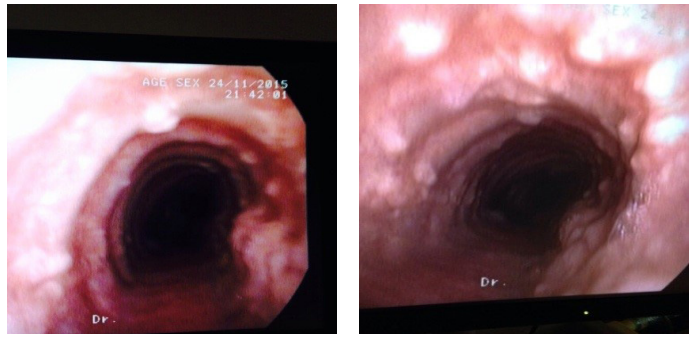
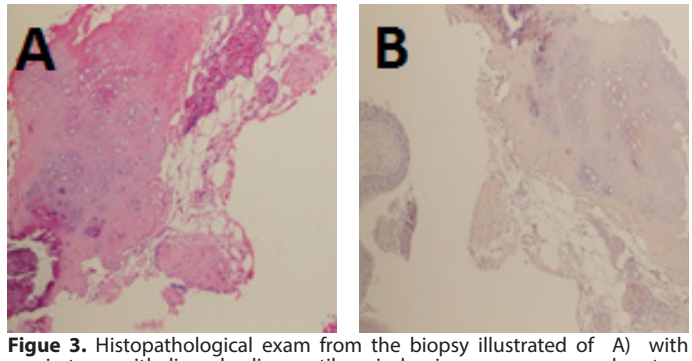


Figure 2. Flexible bronchoscopy showing nodular deformities along multiple nodules from anterolateral wall of trachea and main bronchi protruded into the lumen



Figue 3. Histopathological exam from the biopsy illustrated of A) with respiratory epithelium, hyaline cartilage in benign appearance and mature bone tissue. (Hematoxylin-Eosin x100); B) Amyloid staining was not observed. (Congo-red x100)

DISCUSSION

Tracheobronchopathia osteochondroplastica is a rare and benign disorder with characterization of multiple sub-mucosal osseous and cartilaginous nodules in the anterior and lateral wall of trachea and bronchus It is usually diagnosed in patients aged over 50 years, but one case in a 9-year-old child.[5] Although chronic infection, congenital anomaly, chemical or mechanical irritation, degenerative or metabolic abnormalities, and genetic predisposition are considered factors contributing to development, the cause of the disease is currently unknown.[6] Our case is over 50 years old and there were no etiologically conceivable exposures. In addition, no calcium metabolism disorder was detected.

Majority of the patients with TO are asymptomatic. Chronic cough and dyspnea are the most frequent presentations. Other presentations include hemoptysis, dry throat, recurrent lower respiratory tract infection.[1] In our case, chronic cough was the most frequent respiratory symptom. Chest radiograph is usually normal. The chest CT imaging may demonstrate multiple submucosal irregular, sessile and calcified nodules involving the anterior and lateral wall of the tracheobronchus, usually sparing the posterior membranous.[7,8]

Flexible bronchoscopy is the most valuable tool for establishing the diagnosis. Diagnosis is ultimately dependent on the bronchoscopic finding of sessile submucosal nodules

protruding into the lumen of the trachea and main bronchi, with a beaded or "rock garden" appearance. Obtaining a bronchoscopic forceps biopsy may prove to be difficult owing to the stony hard nature of the nodules. Although biopsy is not necessary for diagnosis of this disorder, it may be helpful to exclude disorders included in the differential diagnosis of nodularity within the trachea. The differential diagnosis of TO includes amyloidosis, sarcoidosis, calcificating lesions of tuberculosis, papillomatosis, tumors of bronchial and tracheal.[9] The characteristic sparing of the posterior wall may help to differentiate the TO from tracheobronchial amyloidosis. Histopathological analysis from the bronchoscopic biopsy specimens shows heterotopic bone formation with abnormal cartilage proliferation and calcium deposits. Histopathological picture may help to rule out other diseases such as amyloidosis, mucoepidermoid carcinoma, papillomatosis, sarcoidosis. In our case, the diseases included in the differential diagnosis were excluded by histopathological evaluation of bronchoscopic biopsy.

No specific therapy for TO is currently available. Treatment is only symptomatic. Patients with TO could be simply followed for prolonged periods of time. Surgical removal, laser ablation or radiotherapy, or stent implantation should be performed only in cases of the airway narrowing by the lesions.[10]

In conclusion, TO is a rare and benign trachea and bronchi disease, characterizing by submucosal ossified and cartilaginous nodules. Thorax CT and FB should be performed to reveal the rare underlying causes of chronic cough patients

ETHICAL DECLARATIONS

Informed Consent: Written informed consent was obtained from all participants who participated in this study.

Status of Peer-review: Externally peer-reviewed.

Conflict of Interest Statement: The authors have no conflicts of interest to declare.

Financial Disclosure: The authors declared that this study has received no financial support.

Author Contributions: All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

REFERENCES

1. Leske V, Lazor R, Coetmeur D, Crestani B, Chatté G, Cordier JF. Tracheobronchopathia osteoc-hondroplastica: a study of 41 patients. Medicine (Baltimore) 2001;80:378-90.
2. Abu-Hijleh M, Lee D, Braman SS. Tracheobronchopathia osteochondroplastica: a rare large airway disorder. Lung 2008;186:353-9.
3. Lundgren R, Stjernberg NL. Tracheobronchopathia osteochondroplastica. A clinical bronchosco-pic and spirometric study. Chest 1981;80:706-9
4. Baran A, Güngör S, Ünver E, Yılmaz A. Tracheobronchopathia osteochondroplastica: a case re-port. Tuberk Toraks 2004;52:183-5.
5. Simsek PO, Ozcelik U, Demirkazik F, et al. Tracheobroncho-pathia osteochondroplastica in a 9-year-old girl. Pediatr Pulmonol 2006;41(1):95-7.
6. Doshi H, Thankachen R, Philip MA, Kurien S, Shukla V, Korula RJ. Tracheobronchopathia oste-ochondroplastica presenting as an isolated nodule in the right upper lobe bronchus with upper lobe collapse. J Thorac Cardiovasc Surg 2005;130:901–2.
7. Jabbardarjani HR, Radpey B, Kharabian S, Masjedi MR. Tracheobronchopathia osteochondrop-lastica: presentation of ten cases and review of the literature. Lung 2008;186:293–7.
8. Al-Busaidi N, Dhuliya D, Habibullah Z. Tracheobronchopathia osteochondroplastica: case report and literature review. Sultan Qaboos Univ Med J 2012;12:109–12.
9. Meyer CN, Dossing M, Broholm H. Tracheobronchopathia osteochondroplastica. Respir Med 1997;91:499-502.
10. Luo S, Wu L, Zhou J, et al. Tracheobronchopathia osteochond-roplastica: two cases and a review of the literature. Int J Clin Exp Pathol 2015;8(7):8585–90.