CRCP Case Reports in Clinical Practice

Case Report

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Tracheobronchopathia Osteochondroplastica

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Citation Sanaei E, Aghaei Meybodi F, Vaghefi M. Tracheobronchopathia Osteochondroplastica. Case Reports in Clinical Practice. 2019; 4(1):23-26.

Running Title: Tracheobronchopathia Osteochondroplastica (TPO)

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Article info:
Received: 17 January 2019
Revised: 09 February 2019
Accepted: 22 February 2019

Keywords:
Tracheobronchopathia Osteochondroplastica; breathlessness

ABSTRACT

Tracheobronchopathia Osteochondroplastica (TPO) is a rare benign disorder of the lower part of the trachea and the upper part of the main bronchi. A 55-year-old female patient had been referring to our pulmonary and critical care center suffering from chronic dry cough and breathlessness on exertion and retrosternal pain for several months who was admitted to our center when she was complaining about breathlessness on exertion and chronic dry cough and retrosternal pain.

Introduction

Tracheobronchopathia Osteochondroplastica (TPO) is a rare benign disorder of the lower part of the trachea and the upper part of the main bronchi [1-3]. It was first described in the mid-19th century, and approximately 300 cases have been reported since then.

A higher incidence of TPO was seen in northern Europe countries, especially in Finland [4]. Because many cases are asymptomatic, TPO is mainly diagnosed post-mortem. Symptoms can range from productive or non-productive cough, hemoptysis, dyspnea, dryness of the throat, and recurrent pulmonary infections (e.g. retention pneumonia) [4].

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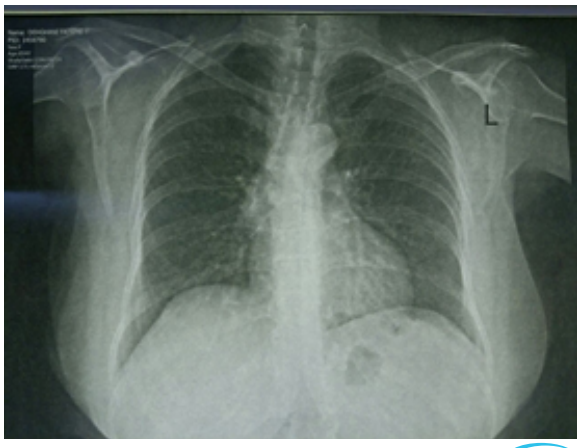

Figure 1. Increasing thickness of the trachea

In severe cases, the diagnosis is made during a difficult intubation. The characteristic bronchoscopic findings are reported as beaded, speculated, rock garden, cobble stoned, or stalactite grotto appearance. The typical histological appearance confirms the diagnosis.

Case Presentation

A 55-year-old female was referred to our pulmonary and critical care center suffering from chronic dry cough and breathlessness on exertion and retrosternal pain since several months ago. She was finally admitted to our center with the complaint of breathlessness on exertion, chronic dry cough, and retrosternal pain. However, her endoscopy and cardiac examination, echocardiography, and coronary artery angiography were normal. She was not a smoker with no history of allergy. Her lung auscultation, lung function tests, and other physical examination were normal. Regarding her laboratory report, her Erythrocyte Sedimentation Rate (ESR) and C-Reactive Protein (CRP), Angiotensin-Converting Enzyme (ACE) level, cytoplasmic-ANCA and perinuclear-ANCA were within normal range. Increasing the thickness of the trachea in the chest radiography was seen (Figure 1).

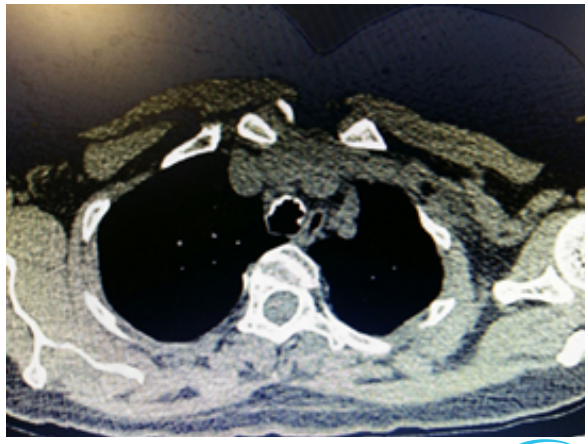

Figure 2. Increasing thickness of the trachea in the form of nodular and calcification

In computed tomography of the chest, the increased thickness of the trachea in the form of nodular and calcification was seen (Figure 2 and 3). So, flexible fiberoptic bronchoscopy was done that revealed diffuse calcified nodularity of the trachea and main and lobar bronchus (Figure 4). Then, multiple biopsies were taken. Histological examination revealed bronchial cartilage and lamellar bone with little marrow (Figure 5). Smears and cultures for Mycobacterium tuberculosis were all negative.

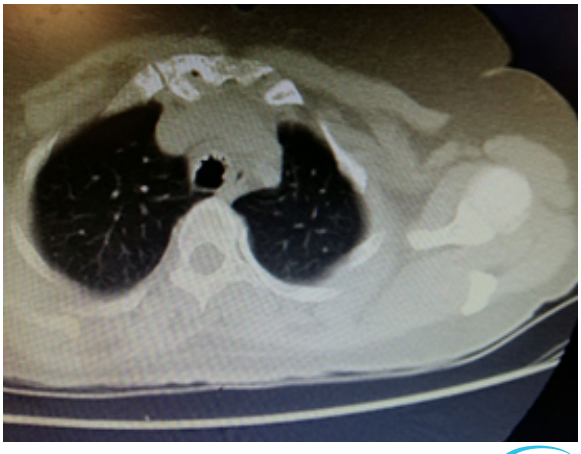

Figure 3. Increasing thickness of the trachea in the form of nodular and calcification (lung view)

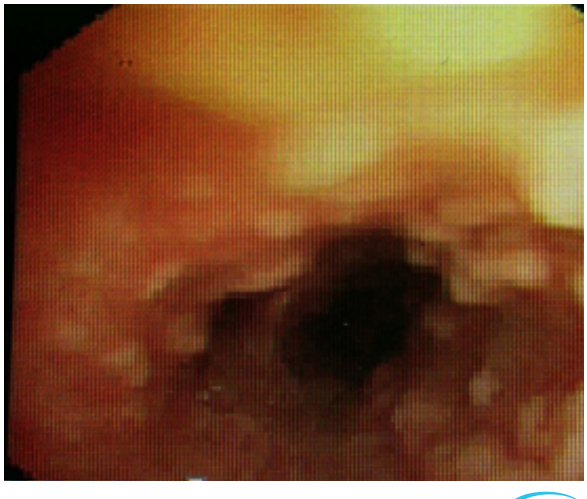

Figure 4. Diffuse calcified nodularity of the trachea

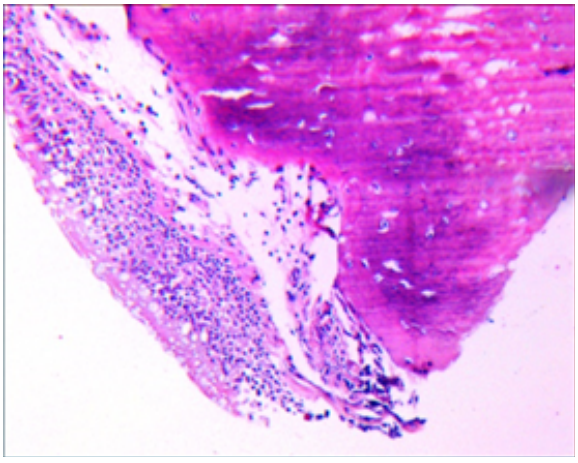

Figure 5. Bronchial cartilage with abnormal and unevenly distributed mineralization supporting the diagnosis of Tracheobronchopathia Osteochondroplastica

Discussion

In the bronchoscopic view, TPO appears as whitish, hard spicules projecting into the tracheal lumen from the anterior and lateral walls, sparing the posterior wall. Also, the larynx and the main bronchi could be involved. The diagnosis of TPO is confirmed by typical histological findings, usually from biopsies or postmortem analysis. Our patient suffered from exertional dyspnea and chronic dry cough.

Moreover, TPO nodules may also be caused by endobronchial sarcoidosis, calcification lesions of tuberculosis, papillomatosis, malignant lesions, and tracheobronchial calcinosis [5-10]. Some patients were initially thought to have asthma [11] or bronchial/trachea tumors.

Because typical symptoms are absent, TPO is most likely underdiagnosed. In this case, the patient was referred to the clinic with breathlessness on exertion, chronic dry cough and retrosternal pain while her endoscopy, cardiac examination, echocardiography, and coronary artery angiography reports were normal. Only severe cases suffer from wheezing and dyspnea caused by the obstruction of the airway lumen. Hemoptysis can occasionally result from the ulceration of a nodule or an acute infection. Sometimes TPO causes difficulties in endotracheal intubation [12, 13]. In most cases, the disease progresses very slowly although it may eventually end in respiratory insufficiency [14].

Because no specific therapy is available, treatment is only symptomatic, which includes antibiotics in case of bacterial infections, mechanical measures to remove obstruction nodules by using cryotherapy, laser excision, external beam irradiation, radiotherapy, stent insertion, interventional bronchoscopy, or surgical resection [15, 16]. Also, successful recovery with nonsteroidal anti-inflammatory drugs and dapsone has been reported in symptomatic cases.

For our patient, the first treatment was prescription of ICS fluticasone, naproxen 500 mg q 12 h, and pregabalin 75 daily respiratory unite of Shahid Sadoughi University of Medical Sciences. After 4 weeks, because of unsuccessful treatment of the symptoms, the patient was referred to another center for interventional bronchoscopy.

Ethical Considerations

Compliance with ethical guidelines

There was no ethical considerations to be considered in this research.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Conflict of interest

The authors declared no conflict of interest.

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