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Case Report

Tracheobronchopathia Osteochondroplastica: an uncommon aetiology for recurrent lower respiratory tract infection

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ABSTRACT

Tracheobronchopathia Osteochondroplastica is a rare and benign condition characterised by development of osteocartilaginous nodules within the submuscosal layer of the trachea with sparing of the posterior wall. Majority of the patients remain asymptomatic throughout their lives unless severe airway obstruction develops; in which case patients present with symptoms such as dyspnoea, hoarseness of voice, persistent and often productive cough, haemoptysis, and pneumonia. This is the case of a middle-aged lady who presented with complaints of recurrent lower respiratory tract infection and was diagnosed to have tracheobronchopathia osteochondroplastica on computed tomography imaging, bronchoscopy and histopathological examination.

Keywords: Pneumonia, Tracheobronchopathia osteochondroplastica, Trachea

INTRODUCTION

Tracheobronchopathia Osteochondroplastica (TO) is a rare and benign condition, involving the airways mainly the trachea. It was first described by Wilks in the year 1857.[1] The incidence is between 0.01- 4.2%, and is usually diagnosed in persons over 50 years of age.[2-4]

The incidence is found to be more among males. The condition does not have any predisposing factors and is not associated with any systemic disorders. These patients present with a wide spectrum of clinical manifestations which include dry or productive cough, hemoptysis, dyspnoea, dry throat and atelectasis. Recurrent lower respiratory tract infection is a major complication of TO.[5] These symptoms and complications are the result of narrowing and thickening of the airway walls. To some extent, the rarity of the condition may be attributed to its unawareness. Our patient presented with recurrent lower respiratory tract infection and was found to have TO as its cause.

CASE REPORT

A 42-year-old female presented with history of recurrent episodes respiratory infections for the past 1 year, which were associated with dyspnoea and dry cough. She has been taking formoterol and budesonide combination inhaler along with antibiotics during these episodes. There was no history of hemoptysis.

She is an office worker (no dust exposure). She was not on any regular medications. On examination, she was febrile (temperature 100^0F). Other vitals and general physical examinations were normal. Systemic examinations revealed right sided rhonchi and fine basal crepitations in the right mammary and infraaxillary regions. Her blood investigations showed leucocytosis (12,200 cells/cmm) with neutrophilia (N93 L4 E3). Other investigations like renal and liver parameters, electrolytes, HbA1c and ECG were normal. Chest Xray showed mild haziness in the right lower zone. She was started on intravenous ceftriaxone and oral

clarithromycin. However, she continued to be febrile despite 3 days of antibiotic therapy, with no improvement in total counts. Mantoux test and throat swab for H1N1 were negative.

Her CT thorax showed calcifications in the trachea and hyperinflation of the right middle lobe with narrowing of the right proximal middle lobe bronchus (Figure 1).

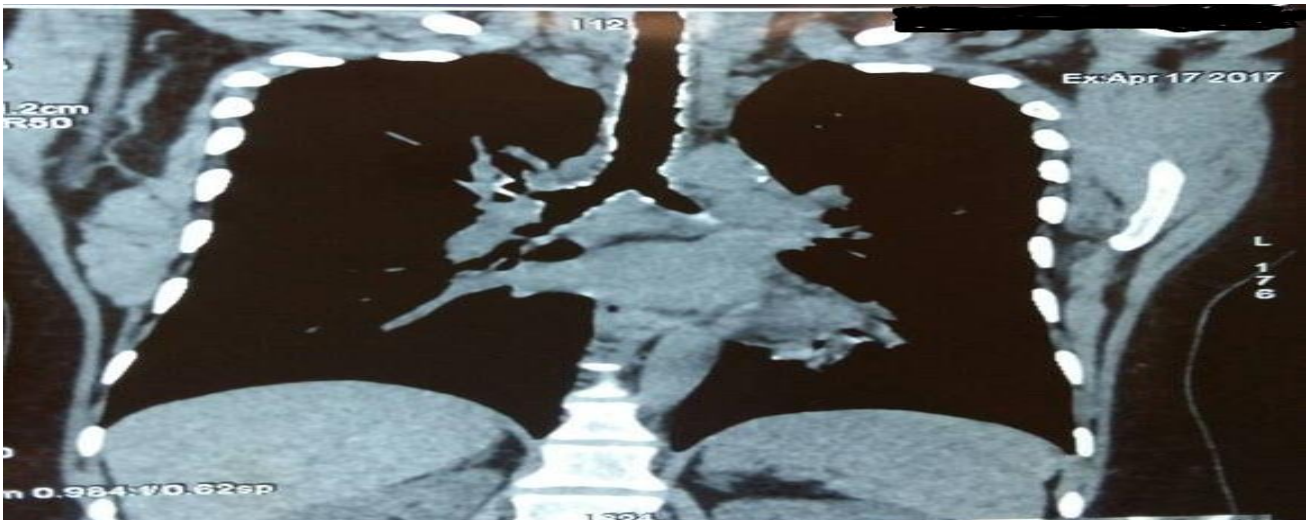

Figure 1: CT Thorax showing tracheal calcifications.

Bronchoscopic evaluation revealed submucosal nodules protruding into the tracheal airway, giving it a cobble stone appearance; with sparing of the posterior wall; suggestive of TO (Figure 2).

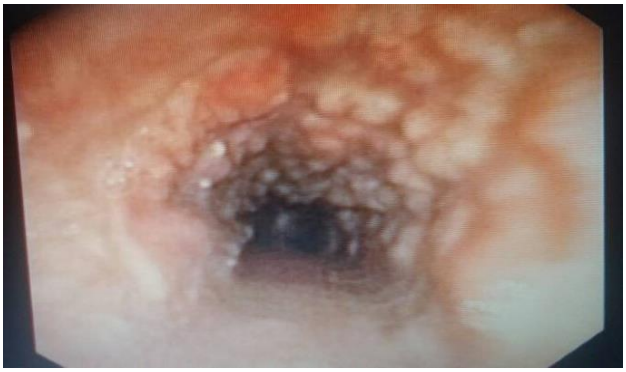

Figure 2: Bronchoscopic image showing submucosal nodules protruding into the trachea.

Biopsy was taken and histopathological examination showed submucosal ossification and cartilaginous tissue. Bronchoalveolar lavage culture grew Pseudomonas aeruginosa and her antibiotics were changed to Piperacillin-Tazobactam (as per culture and sensitivity report); following which she improved. Her PFT showed mild obstructive pattern. She was discharged after 7 days of antibiotics in stable condition and advised to continue formoterol and budesonide inhalers.

DISCUSSION

TO is a rare and benign airway disease of unknown aetiology, characterized by multiple submucosal nodules which project into the lumen of airway. These nodules can be ossified and/or cartilaginous.[3] Chest Xray is usually normal and CT thorax may demonstrate multiple submucosal calcified nodules involving the anterior and lateral wall of tracheobronchus with sparing of the posterior wall.[6]

These findings can be confirmed on bronchoscopy, which reveal sessile submucosal nodules protruding into the lumen of trachea and main bronchi, without disturbing the posterior wall.[7] Histopathological examination of the biopsy shows cartilage and ossification in the submucosa, calcification and mucosal squamous metaplasia.[5,8] Pulmonary function test findings can vary based on the severity of the condition. The test results may be normal or can show obstructive or restrictive pattern, reduced peak flow and abnormal flow-volume loops.[5] The treatment is usually symptomatic. Modalities like surgical resection, mechanical debulking and laser ablation (with Nd:YAG laser) have been tried for symptomatic relief. Patients have shown positive response to inhaled corticosteroids.[5,8]

All treatment modalities are aimed at symptomatic management rather than curing of the condition. Endobronchial sarcoidosis, tuberculosis, tracheobronchial calcinosis, bronchial carcinoma, amyloidosis and papilomatosis are some of the differential diagnosis for TO.[9]

CONCLUSION

As mentioned earlier, TO is a rare and benign airway disease. The manifestations may be either asymptomatic or nonspecific respiratory complaints. The condition is usually discovered incidentally and seldom requires any treatment. Therefore, the awareness of TO, as a

differential diagnosis of neoplasms, is essential to avoid unnecessary chemotherapy or surgery.

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