A challenging case of Tracheobronchopathia osteochondroplastica (TPO) in elderly

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Abstract

Tracheobronchopathia osteochondroplastica is a rare, idiopathic benign tracheobronchial abnormality containing osseous or cartilaginous nodules that can be a focal or diffuse disease process arising mainly in the proximal large airways and sparing the posterior membranous portion of the trachea. Majority of cases are symptom free, but sometimes patients can present with haemoptysis as in our case which could have resulted from ulceration of nodule or an acute infection, and in this case the symptoms was exacerbating with using of anticoagulantion.

Introduction

Tracheobronchopathia osteochondroplastica (TPO) is a rare pulmonarydiseasebelieved to arise from multiple sessile cartilaginous or bony submucous nodules that project into the lumen of the tracheobronchial tree, with spare of the membranes portion of the posterior tracheal wall. Gold standards for diagnosis is a bronchoscopy, without the need for a histopathological conformation.

While most patients are asymptomatic,clinical presentation of the disease entity carries a rainbow spectrum of chest symptoms from a great mimicker of obstructive diseases to, a local compressive symptoms to haemoptysis.

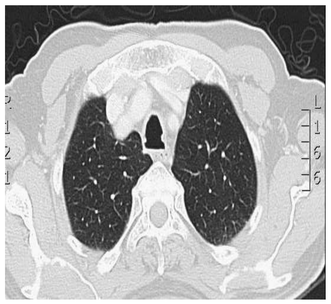

Figure 1 Axial contrast-enhanced CT image of trachea on lung windows showing nodular thickening of the tracheal wall

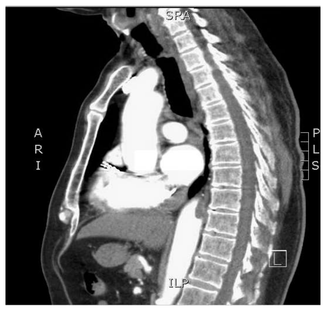

Figure 2 Sagittalcontrast-enhanced CT image of trachea on lung windows showing nodular thickening of the anterior tracheal wall with sparing of the posterior wall.

Methods

We describe the first case of TPO in Kingdom of Saudi Arabia presenting with unusual presentation.

Results

Case ReportA 82 years old with the medical background of Ischemic cardiomyopathy and history of colon cancer underwent right hemicolectomy 2 years ago, referred to the pulmonary service for haemoptysis evaluation.

The haemoptysis was of 6 months duration which was small in amount, associated with occasional cough with absence of constitutional symptoms. No other sites of bleeding from other orifices were present.

The patient is lifelong non-smoker with otherwise no history of any chronic chest disease before or significant exposure asbestos or silica and he has no symptoms suggestive of connective tissue disease. Travel history: non-contributory to potential endemic areas ofmycobacterium or mycotic area. Medication history: Rivaroxaban for atrial fibrillation CHA_2DS_2-VASc score of 5 including heart failure with reduced ejection fraction (HFrEF) of 45%.The local exam of oral, and nose did not show evidence of bleeding or nasal deformity.

The patient is euvolemic. No lymphadenopathy was noted, and peripheral extremity examination did not show evidence of finger clubbing, ecchymoses and/or petechiae, synovitis.

In the Investigations the patient has normal haemoglobin, platelet and his INR was checked several time during the haemoptysis episodes which was therapeutic in the range between 2 and 2.5. CT abdomen was ordered before as follow-up for any metastasis after right hemicolectomy and did not show any mass in the abdomen. CT chest showed irregular thickening and nodularity of the tracheal cartilage, sparing the posterior membranous tracheal wall. (Figure 1 and 2)

The patient underwent bronchoscopy (figure 3 and 4) with results of BAL showed no evidence of malignancy or any infection and biopsy showed: minute detached fragment of unremarkable respiratory epithelium and unremarkable cartilaginous tissue.

Conclusion

The diagnosis of TPO is often incidental, as it is an underrecognized disease entity. In The absences of the common etiologies of hemoptysis TPO should be considered.
Extra-pulmonary manifestations in addition given special attention to the characteristic nodular patterns arsing within the bronchopulmonary tree can help to differentiate disease mimickers. While Tracheobronchopathia osteochondroplastica is famous for sparing the posterior trachea, tracheobronchial amyloidosis has a predominate posterior portion. In addition, relapsing polychondritis share a lot of similarity with TPO but involvement of the ear, nose and tracheomalacia are distinguishing features.
The disease remains to hold excellent prognosis and stable natural history with some patients morbidity/death from sever pulmonary infection.

Treatment of the disease is supportive aiming at elevating symptoms namely antitussives, bronchodilators, and antibiotics. If medical therapy failed to controlled symptoms, then surgical intervention options.

The patient was followed over two years period as the case has atrial fibrillation with highCHA_2DS_2-VASc score at the same time is having recurrent haemoptysis, decision was to observe him and weight risk benefit ratio of continuing anticoagulation. Fortunately, patient did not have bad squeal that mandated the discontinuation of anticoagulation.

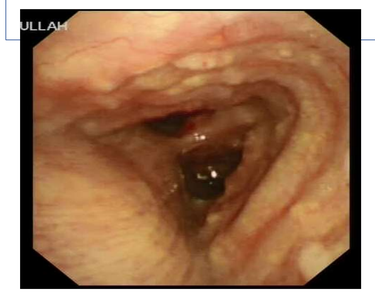

Figure 3 Trachea and bronchial tree showed diffuse nodular changes, with no discrete mass or obstruction

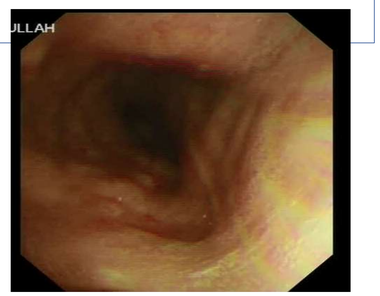

Figure 4 The changes were more prominent in the trachea and proximal bronchial tree and fade away towards the distal bronchi of second-order these areas are easily irritable and likely to bleed.

References