Case Report

Tracheopathia Osteoplastica Presenting with Haemoptysis in a Young Male

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ABSTRACT

A young man presented with infrequent haemoptysis spanning over 10 years. Chest radiograph was normal. However, the computed tomography (CT) of the chest had shown endotracheal wall changes. The diagnosis of tracheopathia osteoplastica was suggested on fiberoptic bronchoscopy and confirmed histologically. **[Indian J Chest Dis Allied Sci 2010;52:119-121]**

Key words: Tracheopathia osteoplastica, Haemoptysis.

INTRODUCTION

Tracheobronchopathia osteochondroplastica is a rare chronic degenerative disorder seen predominantly in the elderly. It is often asymptomatic, and therefore, may be underdiagnosed. Less than 400 cases have been reported in the literature.[1] It may present with clinical features related to major airways obstruction, such as recurrent cough and stridor. We report this rare disorder in a young patient with an unusual symptom of infrequent haemoptysis.

CASE REPORT

A 28-year-old male, non-smoker, presented with a history of mild haemoptysis that started after a preceding upper respiratory infection, lasted for three days and subsided after treatment with antibiotics. There was no fever or dyspnoea. He had a similar episode of haemoptysis 10 years ago and again an year ago lasting for a few days and was treated conservatively. Chest radiograph and upper gastrointestinal endoscopy done one year ago were not remarkable. There was no past history of tuberculosis or bronchial asthma.

General physicial, cardiaovascular and respiratory system examination were normal. Haemogram, creatinine and liver function tests were also normal. No abnormalities were noted in the coagulation profile. The chest radiograph was normal. Computed tomography of the chest did not show any parenchymal abnormality or mediastinal nodes. However, the tracheal lumen revealed irregular wall with areas of calcification within the tracheal inner lining (Figure 1).

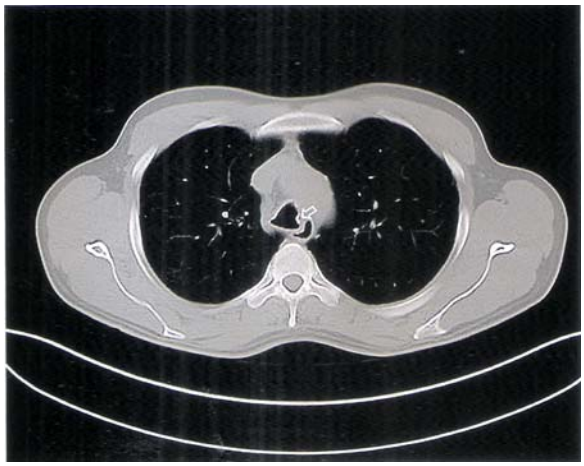

Figure 1. Computerised tomography of chest in the bone window, showing calcified and irregular tracheal inner lining (arrow).

Fiberoptic bronchoscopy revealed white nodular mucosal irregularity all over the anterolateral aspects of the tracheal wall and also extending into both the main bronchi (Figure 2). These nodules were hard to bite with the biopsy forceps, and the secured endobronchial biopsies were sent for histopathology along with bronchial brushings and washings for cytology and microbiology.

Histopathology revealed mucosa lined by squamous epithelium and stroma showing lymphocytic infiltrate and bony spicules without any evidence of granuloma or malignancy (Figure 3).

The endobronchial brushings and cytology were suggestive of similar mild inflammatory infiltration without any evidence of malignancy. Bronchial washings did not reveal any specific pathogenic organisms including fungi or acid-fast bacilli. Serum

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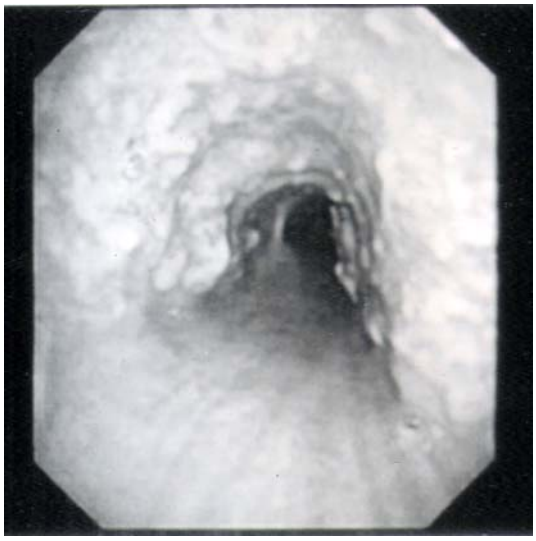

Figure 2. Fiberoptic bronchoscopic view of the trachea, showing multiple, irregular nodular protusions from the mucosa in the typical 'Rock garden' pattern.

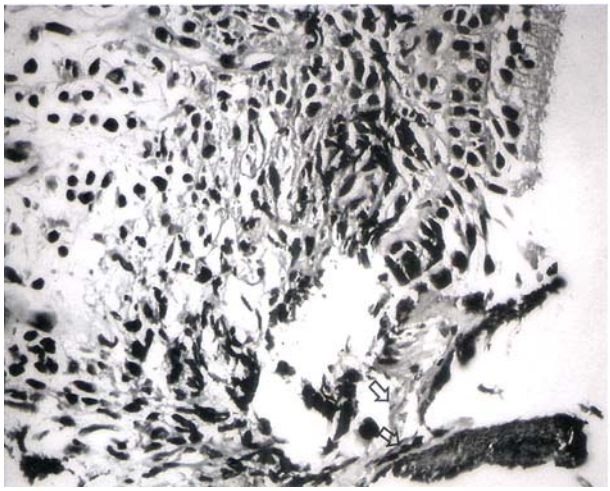

Figure 3. Histological sections of endotracheal biopsy, showing mild inflammation and bone formation (arrows).

calcium levels were found to be normal. The patient did not have any further episode of haemoptysis and was discharged with a diagnosis of tracheobronchopathia osteoplastica.

DISCUSSION

Tracheobronchopathia osteochondroplastica is a rare chronic degenerative disorder of the trachea and is predominantly seen in the elderly.[2] As this disorder is often asymptomatic, estimates of incidence vary between 1/2000 bronchoscopies and 1/400 at autopsy.[3] The cause of this disorder is not well established but may occasionally be associated with chronic inflammation, trauma and chronic chemical or mechanical irritation.[4]

It may manifest clinically with non-specific signs and symptoms, although stridor, cough, and dyspnoea are the usual presenting symptoms.[4] Our patient presented with an unusual history of infrequent haemoptysis each time following upper respiratory infection suggesting an infective trigger.

Radiologic studies may suggest the diagnosis if scalloped nodular calcified opacities are seen in the submucosa, particularly on the CT of the chest as were seen in our patient.[5] The diagnosis is confirmed after endoscopic and pathologic examination. The bony lamellae may protrude into the mucosa, that is the characteristic appearance on bronchoscopy (Rock garden pattern)[6] and is more prominent in the lower two-thirds of the anterolateral tracheal wall often extending to major bronchi as in the present case.

On histopathology, metaplastic cartilage and bone are found in the submucosa, often in continuity with the inner surface of the tracheal cartilage. The overlying mucosa is intact and may appear to be normal or metaplastic. Foci of calcification may be seen and the irregular bony spicules have thin walls surrounding fatty marrow.[2] The presence of mild inflammatory infiltration of the stroma in our patient favours the occurrence of an infective exacerbation resulting in haemoptysis. Otherwise, inflammatory changes are not typical features of the pathology of tracheopathia osteoplastica.

The differential diagnoses include post-intubation granulation tissue, chronic granulomatous inflammation (tuberculosis, sarcoidosis, fungal infection, etc), focal multinodular amyloidosis, papillomatosis and tumours. These were all ruled out on histopathology. Chronic chemical (occupational) or mechanical irritation (endotracheal intubation) and hypercalcemic states were also excluded in our patient.

The treatment of tracheopathia osteoplastica is generally conservative and aims at symptomatic relief. While localised disease may not require treatment, significant narrowing may require laser removal and dilation.[7] Surgical resection of obstructing lesions may be required and extensive involvement may require tracheoplasty.[8]

Being an infrequent condition with few symptoms, tracheopathia osteoplastica is likely to be underdiagnosed or misdiagnosed unless there is an awareness of radiological and bronchoscopic features.

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