CASE REPORT

Endobronchial hamartoma: an unusual cause of focal bronchiectasis

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SUMMARY

Bronchiectasis, a chronic and debilitating disease, is becoming increasingly prevalent worldwide and causing a growing burden on healthcare systems. This condition can be categorised as diffuse or focal, depending on the underlying cause. Accurate and early diagnosis is essential for effective management and prognosis. Endobronchial hamartomas are a potential cause of recurrent post-obstructive pneumonia with focal bronchiectasis. In this report, we present a case of an elderly gentleman with recurrent pneumonia and right middle lobe bronchiectasis despite receiving courses of antimicrobial therapy. CT thorax showed an oval-shaped endoluminal lesion with popcorn calcifications and fat within the right middle lobe bronchus associated with right middle lobe bronchiectasis. Subsequent flexible bronchoscopy revealed a round, smooth, yellowish intraluminal lesion obstructing the airway of the right middle lobe. We are able to recanalise the airway by removing the endobronchial lesion completely, and the HPE was consistent with endobronchial hamartoma. However, the distal airway was already bronchiectatic. This case emphasises the importance of having a strong clinical suspicion for intraluminal obstruction when focal bronchiectasis is encountered. Bronchoscopy plays a crucial role in obtaining tissue diagnosis and is essential for its therapeutic benefits.

INTRODUCTION

Bronchiectasis is a progressive respiratory disease characterised by permanent bronchial dilatation, cough, sputum production and recurrent respiratory infections.[1] A careful history, review of radiological features and laboratory testing are essential to identify the underlying aetiology. Focal bronchiectasis should raise suspicion of endobronchial obstruction due to recurrent post-obstructive pneumonia, while bilateral diffuse bronchiectasis may suggest systemic illness (i.e. immunodeficiency or previous childhood infection).[2] Identifying the underlying cause of bronchiectasis is critical for treatment and prognostication. Endobronchial obstruction can be caused by malignant or benign diseases including hamartomas.[3] Pulmonary hamartomas within the lung parenchymal are often asymptomatic and were frequently an incidental finding on imaging. In contrast, endobronchial hamartomas commonly present with symptoms of airway obstruction and frequently require bronchoscopy to differentiate malignancy from other benign causes.[4] We present a case of endobronchial hamartomas as the cause of focal right middle lobe bronchiectasis due to recurrent post-obstructive pneumonia.

CASE PRESENTATION

A 66-year-old Iban gentleman who is an ex-chronic smoker with underlying hypertension and chronic obstructive pulmonary airway disease presented with chronic productive cough and recurrent chest infection. Physical examination revealed a healthy man with normal vital parameters. Coarse crepitations were auscultated over the lower half of the right hemithorax, and there was no clubbing or cervical lymphadenopathy.

Initial plain chest radiograph revealed right middle lobe consolidation and bronchiectasis. His sputum smear for acid-fast bacilli and mycobacterium culture were negative. However, despite multiple courses of oral antibiotics, there was no associated radiological resolution. Hence, contrast-enhanced computed tomography (CT) thorax (Figure 1a–c) was arranged which demonstrated an oval-shaped hypoattenuating endoluminal lesion with popcorn calcifications and fat within the right middle lobe bronchus associated with right middle lobe bronchiectasis. Three-dimensional airway reconstruction revealed an endobronchial lesion arising from the posterior wall of the right middle lobe bronchus (Figure 2a, b). Flexible bronchoscopy under conscious sedation then confirmed a round and yellowish intraluminal lesion with smooth surface obstructing the lumen of right middle lobe bronchus (Figure 2c, d); using flexible forceps, the growth was removed en bloc, exposing an erythematous and oedematous right middle lobe sub-segmental bronchus with purulent secretion distally. Minimal post-biopsy bleeding was secured with argon plasma coagulation. Bronchial washing was then performed at the right middle lobe bronchus.

Histopathological examination of the debulked endobronchial lesion revealed bland spindle cells in myxoid stroma with some fibroadipose tissue, smooth muscle, cartilage and bone with overall features suggestive of endobronchial hamartoma. Patient was continued to be followed up regularly and was enrolled in pulmonary rehabilitation with assistance in airway clearance. Patient is

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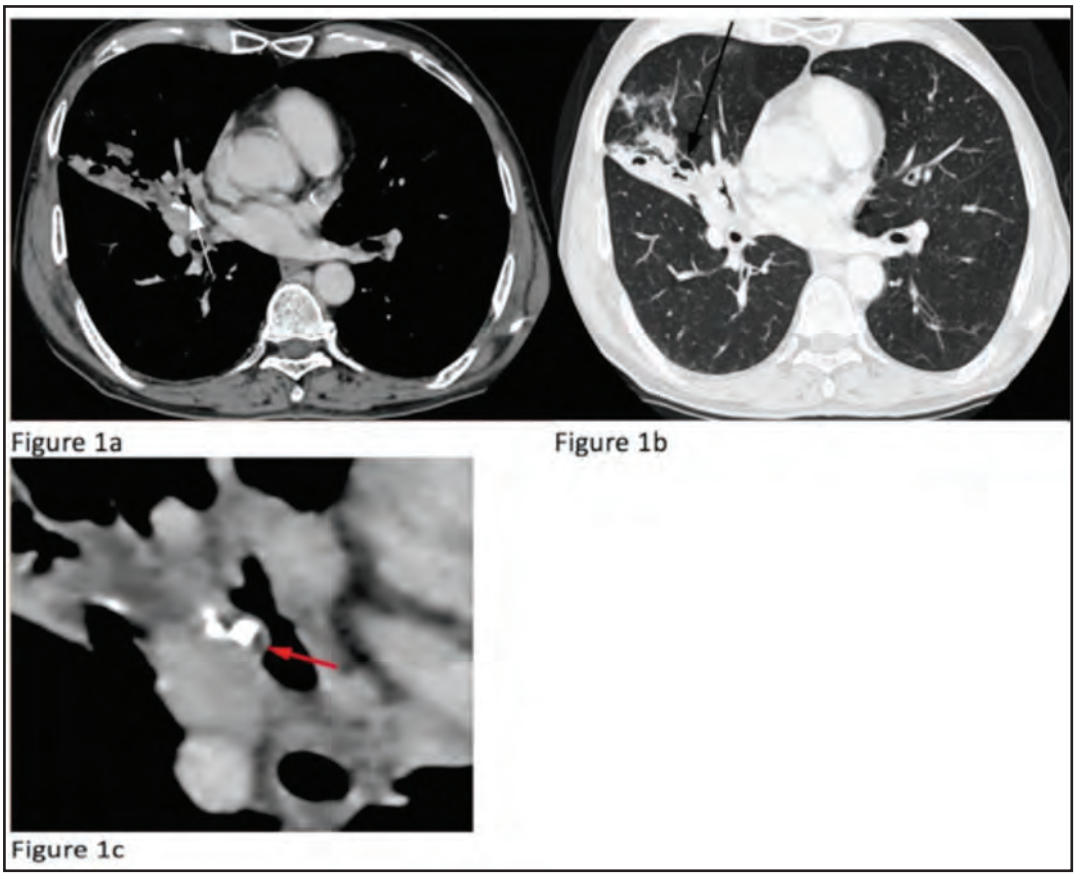

Fig. 1: Contrast-enhanced CT Thorax in mediastinal window (Figure 1a) and lung window (Figure 1b) axial views show an oval-shaped hypoattenuating endoluminal lesion with popcorn calcifications (white arrow in Figure 1a) as well as macroscopic fat with an attenuation value of -40HU (red arrow in close-up image in Figure 1c) within the right middle lobe bronchus. It is associated with bronchiectatic airways of the right middle lobe (black arrow in Figure 1b).

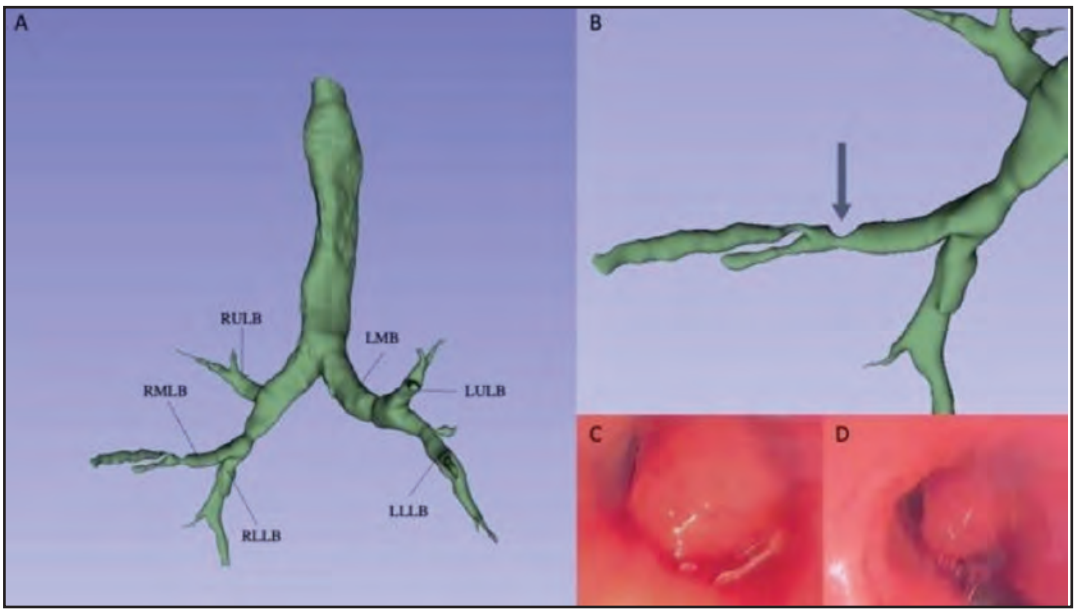

Fig. 2: Three-dimensional airway reconstruction (A) revealed a stenotic right middle lobe bronchus with bronchiectatic dilatation of distal airways (B) which was confirmed by flexible bronchoscopy with an intraluminal mass (C) and was removed en bloc (D) via flexible forceps.

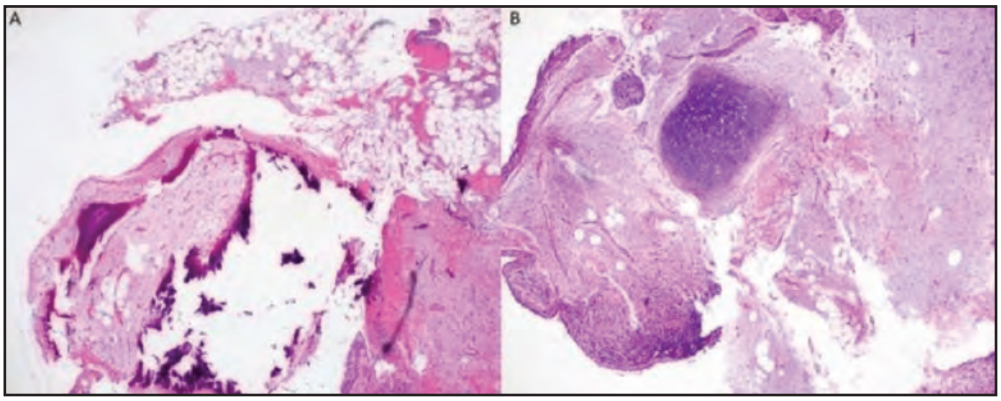

Fig. 3: Histopathological examination of the removed tumour demonstrated bland spindle cells in myxoid stroma with some fibroadipose tissue, smooth muscle, bone (A), and cartilage (B) with overall features suggestive of endobronchial hamartoma. (×40 magnification, haematoxylin and eosin stain).

doing well now with no further recurrent admission for pneumonia. His vaccination status is also up to date as an important preventive healthcare measure for patient with bronchiectasis.

DISCUSSION

Endobronchial hamartomas are rare benign lesions with an incidence of 0.025–0.032% typically affect males in their fifth and sixth decade of life with a higher prevalence in smokers.[5] The presentation varies from asymptomatic to bronchial obstruction, which can cause atelectasis, chronic cough, and recurrent chest infections. Radiologically, they can be difficult to distinguish from malignant lesions, but generally present as well-circumscribed nodules or masses, with typical *"popcorn"* calcifications present in 20–30% and fat seen in up to 60% of lesions.[6] Tissue biopsy via bronchoscopy thus remains the gold standard for diagnosis and exclusion of other causes of endobronchial obstruction. Furthermore, it also offers therapeutic options by enabling airway recanalization through tumour debulking.[7]

Early detection and treatment of bronchiectasis can potentially reverse and stabilise the condition. It is important to determine the underlying cause as a variety of factors can contribute to bronchiectasis including systemic illnesses and severe childhood infections. In regions with a high prevalence of pulmonary tuberculosis, post-tuberculous bronchiectasis remains a common cause of bronchiectasis. These aetiologies frequently presented with typical distribution, i.e., bi-basal symmetrical distribution for systemic diseases and bi-apical involvement for tuberculosis.[8] Hence, when bronchiectasis was focal in distribution which was limited only to a segment or lobe, clinician should consider more localised diseases such as sequestrated lung, congenital bronchial atresia, extrinsic compression and endobronchial obstruction. In this case, we present a patient with recurrent post-obstructive pneumonia resulting in focal right middle lobe bronchiectasis due to endobronchial obstruction from an endobronchial hamartoma. Unfortunately, the distal segment from the endobronchial lesion was already bronchiectatic due to recurrent post-obstructive pneumonia.

CONCLUSION

Although endobronchial hamartomas are benign lesions, it is crucial to make an early and accurate diagnosis as an endobronchial lesion can cause non-resolving obstructive pneumonia and eventually irreversible focal bronchiectasis. Early diagnosis can prevent progression to bronchiectasis and other complications such as corpulmonale and respiratory failure. Therefore, clinicians should have a high index of suspicion for endobronchial lesions in patients with recurrent post-obstructive pneumonia, and prompt investigation with tissue biopsy, ideally via bronchoscopy, is recommended to facilitate early diagnosis and management.

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