Bronchoscopic Resection of Inflammatory Myofibroblastic Tumour: A Rare Cause of Recurrent Haemoptysis and Lobar Atelectasis

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ABSTRACT

This case report presents a rare example of a benign inflammatory myofibroblastic tumour (IMT) in the bronchus, highlighting the importance of a thorough diagnostic work-up and successful bronchoscopic resection without surgery. A 25-year female patient presented with low-grade fever, progressive dyspnoea, cough, and intermittent haemoptysis. The diagnostic work-up revealed a tumour in the bronchus intermedius causing right-lower lobe atelectasis. A bronchoscopy and debulking of the tumour were successfully performed, and histopathological examination confirmed the diagnosis of IMT. The patient experienced a seamless recovery and remained asymptomatic during follow-up appointments, with no evidence of tumour recurrence at one year, demonstrating the effectiveness of bronchoscopic resection for selected cases of IMT.

Key Words: *Bronchoscopic resection, Inflammatory myofibroblastic tumour, Pulmonary pseudotumour, Haemoptysis*.

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INTRODUCTION

Inflammatory myofibroblastic tumour (IMT) of the lung is a term that encompasses a range of pulmonary lesions, also previously referred to as plasma cell granuloma, inflammatory pseudotumour, fibrous histiocytoma, fibroxanthoma, and xanthogranuloma.[1] Lesions similar to IMTs can also occur in various other tissues and organs, including the orbit, skull base, thyroid, liver, spine, spleen, and lymph nodes.[2] Two prevailing theories suggest that IMT may be an inflammatory response to either an underlying infection or a low-grade malignancy. Supporting the reactive inflammatory theory, immunohistochemical staining reveals the presence of IgG-predominant, polyclonal plasma cells within IMT lesions.[3] IMTs can occur in any age group of both genders, but over one-half of the patients are <40 years of age. Initial management with bronchoscopic resection can be a viable and effective treatment option.[4]

CASE REPORT

A 25-year female college student presented with a two-month history of intermittent low-grade pyrexia (up to 101°F), progressive exertional dyspnoea, and cough with intermittent haemoptysis.

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There was a gradual progression of dyspnoea from heavy exertion to moderate exertion, such as brisk walking. She reported coughing up bright red blood, approximately 5-10 ml on affected days, without any associated wheezing, orthopnoea, or paroxysmal nocturnal dyspnoea. Her past medical, personal, family, socio-economic, and gynaecological histories were non- contributory. Upon general physical examination, no abnormalities were detected. Vital signs were within normal limits with oxygen saturation of 99% and body mass index (BMI) of 21 kg/m^2. Respiratory system examination revealed reduced chest movements on the right lower side, accompanied by dull percussion and decreased intensity of breath sounds. The remaining systemic examination was unremarkable, with no other abnormalities detected.

Laboratory investigations, including complete blood count, showed anaemia with a haemoglobin level of 9.5 g/dl, accompanied by mild leucocytosis with a white blood cell count of 11.5/cm and normal differential counts. Coagulation studies were normal with prothrombin time (PT) of 13 seconds (control 14 seconds) and an activated partial thromboplastin time (aPTT) of 28 seconds (control 30 seconds). The remaining laboratory investigations, including renal and liver function tests and serum electrolytes, were within normal limits.

Her chest radiograph (PA view) showed a loss of volume in the right hemithorax, accompanied by a triangular opacity in the right lower zone, with its base towards the heart. Further evaluation with High-Resolution Computerised Tomography (HRCT) of the chest revealed a tumour in the bronchus intermedius, resulting in right lower lobe atelectasis (Figure 1).

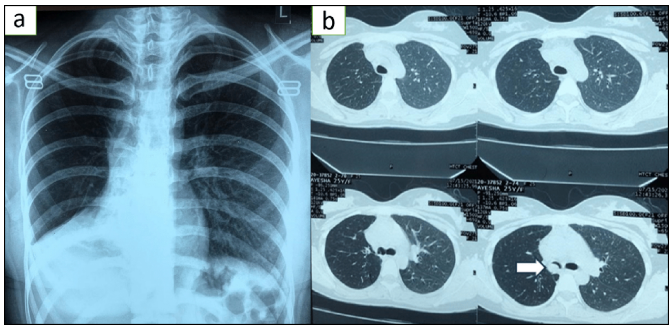

Figure 1: (a) Chest radiograph (PA view) showing volume loss in the right hemithorax with a triangular opacity in the right lower zone, obscuring the right hemidiaphragm; and (b) an HRCT chest image (pulmonary window) revealing a tumour in the bronchus intermedius, causing partial narrowing (indicated by the "wink sign," white arrow).

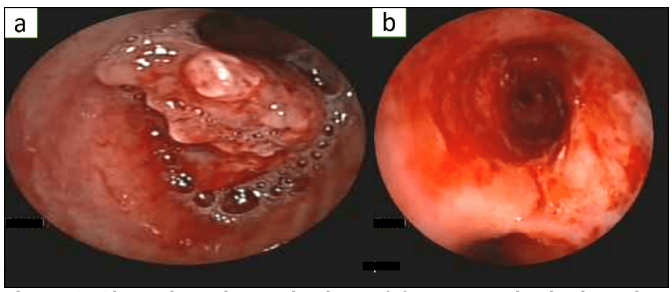

Figure 2: Shows bronchoscopic views: (a) a tumour in the bronchus intermedius partially obstructing the airway, with the middle lobe bronchus orifice visible at 12 o'clock; and (b) the airway after tumour debulking, with restored patency.

No previous bronchoscopy was done to obtain a biopsy from the tumour, and she was referred to our centre for further management. Airways evaluation using both flexible fibre-optic (Fujinon® 270T) and rigid bronchoscopies (Karl Storz® (10318-C) was performed under general anaesthesia to visualise and treat the tumour. A reddish, sessile, and vascular tumour was identified in the bronchus intermedius, causing partial (>70%) obstruction and obliteration of the bronchial lumen (Figure 2). The tumour was successfully excised using a combination of an electrocautery probe, repeated bursts of Argon Plasma Coagulation (APC) beam directed at the tumour, and mechanical debulking using the bevelled edge of the rigid bronchoscope (apple coring). A fleshy tumour, measuring approximately 2 cm in length (28 × 17 × 8 mm), was carefully grasped using rigid biopsy forceps and successfully extracted through the lumen of the rigid bronchoscope. The tumour was retrieved in its entirety, allowing for the complete removal of the obstructing lesion and restoration of the bronchial lumen (Figure 2). The base of the lesion was further coagulated using an APC beam. Post-tumour extraction, bronchoscopic examination revealed normal distal airways in the middle and lower lobes, with no visible tumourlets or nodules on either side of the bronchial tree.

The patient experienced a seamless and uncomplicated recovery, leading to a successful discharge from the hospital. A series of follow-up appointments were conducted to monitor the patient's progress, with the first follow-up occurring at one week after the procedure. At this initial follow-up, she remained asymptomatic, and a chest radiograph confirmed complete radiological clearance of the opacity from the right lower zone, indicating a successful treatment outcome. Histopathological examination of the tumour tissue revealed a characteristic proliferation of myofibroblastic spindle cells, accompanied by a mixed inflammatory infiltrate consisting of lymphocytes, plasma cells, and eosinophils. Immunohistochemical (IHC) staining showed negative reactivity for Desmin, S100, EBER, and CK. However, a positive stain for anaplastic lymphoma kinase (ALK) was observed, which is a distinctive feature of IMTs. The combination of these histopathological and IHC findings led to a conclusive diagnosis of an IMT of the bronchus.

Subsequent follow-up appointments at three and six months revealed consistent findings, with the patient remaining asymptomatic and radiographs showing no signs of recurrence. The final follow-up appointment, conducted at one year post-procedure, included a chest radiograph and surveillance bronchoscopy, both of which confirmed no tumour recurrence in the affected bronchus.

DISCUSSION

IMT of the bronchus is a rare benign neoplasm that can present with non-specific symptoms such as dyspnoea and haemoptysis and is pathologically characterised by a proliferation of myofibroblastic spindle cells with accompanying mixed inflammatory cells.[4] In this case, the patient presented with a two- month history of low-grade fever, progressive exertional dyspnoea, and cough with intermittent haemoptysis. The wink sign, which refers to the appearance of a partially obstructed bronchus or airway, was evident in our patient's chest CT scan.[5] Bronchoscopy and debulking of the tumour were successfully performed, and histopathological examination confirmed the diagnosis of IMT.

Complete surgical resection is usually the treatment of choice for IMT.[6] However, for such benign tumours, initial management should be attempted using bronchoscopic resection or debulking, as was done in this case.[7] This approach allows for tumour removal while preserving lung function. For patients ineligible for surgical resection due to compromised health, multiple lesions, or advanced disease, alternative non-surgical options may be considered, including corticosteroids, chemo-therapy, or radiotherapy. While anecdotal evidence suggests varying degrees of success and failure with these treatments, their efficacy remains uncertain.[8] In such cases, crizotinib, a competitive inhibitor of the ALK tyrosine kinase, has been used with varying success.[9]

This case presents a rare example of a benign IMT in the bronchus, highlighting the importance of a thorough diagnostic work-up and successful bronchoscopic resection without surgery.

PATIENT'S CONSENT:

The patient provided written informed consent.

COMPETING INTEREST:
The authors declared no conflict of interest.

AUTHORS' CONTRIBUTION:
TM: Inception, detailed write-up, data collection, and revision.
SNT: Consent from patient, initial write-up, data collection, and revision.
Both authors approved the final version of the manuscript to be published.

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