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Lung: Case Report

Endobronchial Inflammatory Myoblastic Tumor (IMT)—A Sleeve to Solve the Issue

Nida S. Bham, MD,[1] and
Jess D. Schwartz, MD, FACS[2]

Inflammatory myoblastic tumors of the lung are rare benign lesions. Here we present the case of a 15-year-old boy with postobstructive pneumonia. Computed tomography of the chest revealed a mass in the left mainstem bronchus that was confirmed on bronchoscopy. A rigid bronchoscopy with core resection was performed. Findings on pathologic examination were consistent with an inflammatory myoblastic tumor. The initial core resection relieved his postobstructive pneumonia; however, repeat bronchoscopy a month later demonstrated recurrence of the lesion. The tumor was removed with a parenchymal-sparing sleeve resection. At 80 months of follow-up, the patient is without evidence of recurrence or stenosis.

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Inflammatory myoblastic tumors (IMTs) of the lung are poorly defined pulmonary lesions that are essentially benign nonneoplastic masses of inflammatory origin. First described in 1973,[1] these "pseudotumors" are rare; however, they are the most common primary lung neoplasm in the pediatric population.[2] Most of these tumors are located in the lung parenchyma, but a small subset are endobronchial in origin. IMTs are difficult to diagnose clinically, and tissue diagnosis with fine-needle aspiration or frozen section is often unreliable. Here we present an unusual case of an endobronchial IMT of the distal left mainstem bronchus in a young male.

A 15-year-old Hispanic male presented with a 3-week history of fever, shortness of breath, cough, and left-sided pleuritic chest pain while being treated for a pneumonia. His past medical history was unremarkable. A computed tomography scan of the chest demonstrated a lesion in the distal left mainstem bronchus (Figure 1). The patient was taken to the operating room for a planned flexible and rigid bronchoscopic biopsy and possible thoracotomy for definitive resection (Figure 2A). Intraoperative frozen section biopsy was suggestive of an IMT, and a decision was made to complete the core resection from within the left mainstem bronchus and to forego definitive resection until a final diagnosis could be ascertained.

The excised tissue was reviewed in-house and by external experts to confirm the diagnosis of an IMT. A review of available literature and the discussion at our tumor board raised the question of the optimal treatment, and a decision was made to repeat bronchoscopy a month later. The repeated bronchoscopy demonstrated a 50% restenosis of the left mainstem bronchus with tumor (Figure 2B). Given the tumor's location at the distal left mainstem, it was thought that the patient's condition could be addressed with a left mainstem bronchial sleeve resection, sparing the entire left lung parenchyma, and the patient was taken to the operating room.

Exploration revealed a chronically atelectatic left lung and multiple, enlarged lymph nodes at stations 5, 6, 7, 8, and 9. Intraoperative bronchoscopy was performed, and the position of the mass was localized with a 25-gauge needle inserted through the left mainstem bronchus. The left main bronchus was incised proximal and distal to the tumor, and the specimen was sent for frozen section evaluation. Pathologic analysis of the specimen demonstrated a negative proximal margin, but microscopic tumor was present at the distal end. To ensure clear margins, the decision was made to perform a left upper lobe sleeve resection.

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[1]Division of Thoracic Surgery, Department of Surgery, Yale School of Medicine, New Haven, Connecticut; and [2]Division of Surgery, Department of Surgery, Oklahoma University Health, Oklahoma City, Oklahoma

Address correspondence to Dr Nida S. Bham, 330 Cedar St, New Haven, CT 06510; email: nida.bham@yale.edu.

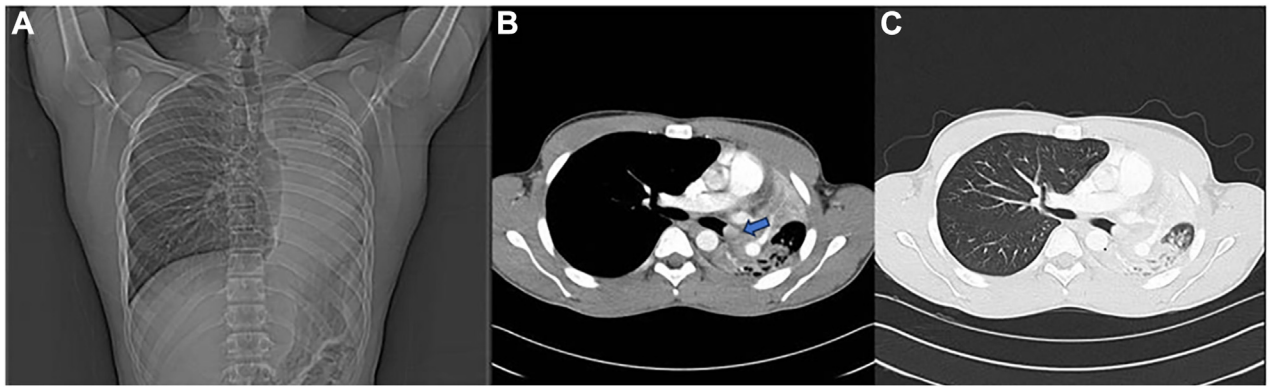

FIGURE 1 (A) Scout film of the patient's chest shows complete collapse of the left lung with mediastinal shift to the left and silhouette of lesion occluding left mainstem bronchus. (B, C) Computed tomography images of the chest show a well-circumscribed mass completely obstructing the lumen of left distal bronchus (arrow).

A standard, left upper lobe sleeve resection in conjunction with a left hilar pericardial release was performed, and subsequent frozen sections were negative. The patient had an uneventful postoperative course, requiring 1 toilet bronchoscopy for plugging on postoperative day 1, and he was discharged on postoperative day 7. Final pathologic examination revealed a spindle cell proliferation consistent with the diagnosis of IMT. All margins and lymph nodes submitted were negative for tumor on final pathologic examination. Postoperative follow-up with bronchoscopy at 48 months (Figure 3A) and computed tomography scan at 80 months (Figures 3B, 3C) demonstrated a well-healed and patent anastomosis, expansion of the left lower lobe, and no evidence of recurrence.

COMMENT

IMTs are exceedingly rare lesions of the lungs, accounting for less than 1% of all pulmonary tumors.[3] First described by Bahadori and Liebow[1] in 1973, these are the most common primary lung neoplasms in children younger than 16 years. No gender or ethnic predominance has been reported.

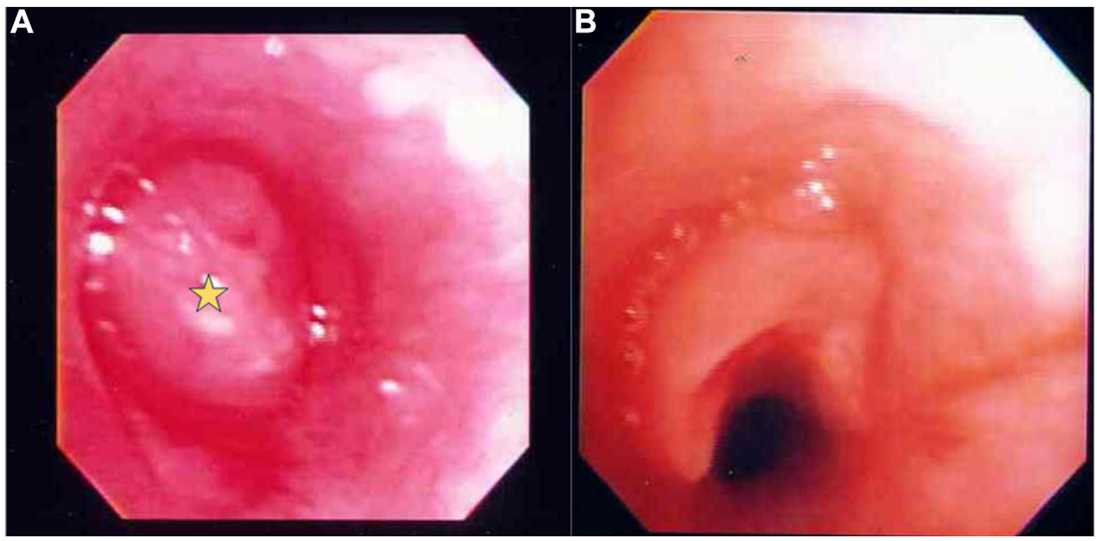

FIGURE 2 (A) Endobronchial view of left mainstem tumor (star). (B) View of left mainstem tumor after rigid bronchoscopic "core" resection biopsy.

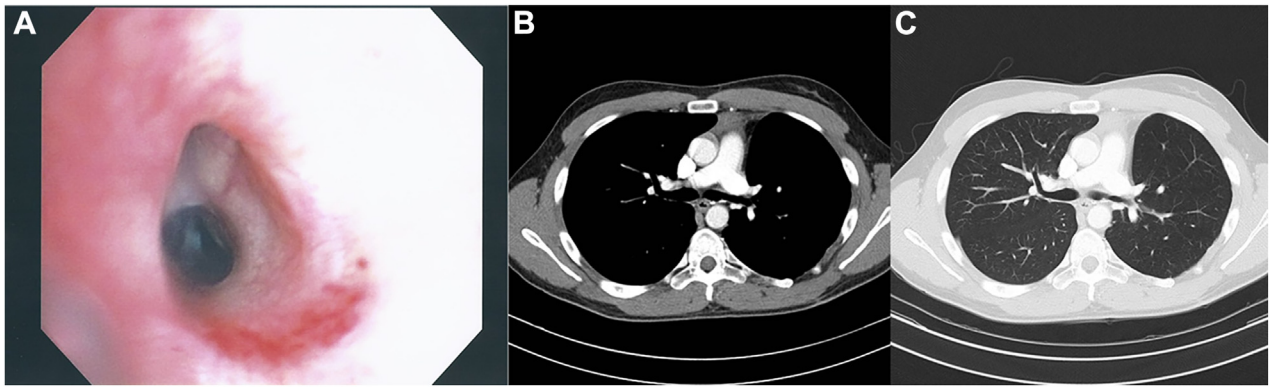

FIGURE 3 (A) Surveillance flexible bronchoscopy 48 months after resection. (B, C) Surveillance computed tomography 80 months after resection.

These tumors have been reported in the literature with variable nomenclature (pseudotumor, xanthoma, fibroxanthoma, histiocytoma xanthogranuloma, plasma cell tumor) owing to their difficult pathologic characteristics and unknown cause. The most current widely accepted description is a benign pulmonary neoplasm, usually parenchymal in origin, typically composed of plasma and spindle-shaped mesenchymal cells.[4] Although the lesion is histologically benign, it may have features such as angioinvasion, local recurrence, distant metastases, and cytogenetic clonal changes.[1,2]

These lesions have been classified in the literature into 2 types, noninvasive and invasive. The noninvasive subtype is more predominant and is usually asymptomatic. These are frequently diagnosed on routine chest radiographs as "coin" lesions.[2] The invasive subtype usually occurs in pediatric patients and is associated with systemic symptoms such as fever, fatigue, and weight loss. These are larger in size with a high risk of extension into local mediastinal structures or the chest wall.[4]

When these tumors are intraparenchymal, they may have long, protracted courses before diagnosis. However, in the small subset of patients who have endobronchial lesions, the presentation may be more acute and clinically impressive because of the postobstructive pneumonia. The treatment of choice is unequivocally an operation, although historically nonoperative management with radiotherapy and corticosteroids has been described, although it is not popular.[5]

Intraluminal bronchoscopic removal as definitive therapy has most recently garnered some interest owing to the low metastatic potential of these lesions; however, current data are lacking on the long-term success of these techniques.[6] The techniques generally involve use of the following therapies, either as standalone treatments or more commonly in combination: thermocoagulation, cryotherapy, neodymium laser, photodynamic therapy, and argon plasma coagulation. However, in the pediatric population, the recurrence rate after endobronchial therapy is estimated to be 46% compared with 6% after surgical resection.[7] Frontline interventional bronchoscopy is mandatory in cases of life-threatening obstructions and can serve as the sole initial treatment for polypoid tumors with a base area limited to 10 mm^2.[8]

Complementary cryotherapy may be applied to the base to prevent local recurrence. These interventions are often reserved for unresectable, multifocal, or relapsing cases.

Delays in diagnosis and treatment can significantly increase the extent of the required surgical intervention, which was a major concern in our case. Our initial rigid bronchoscopy indicated the potential for a complete parenchyma-sparing operation by performing a left mainstem bronchial sleeve resection. Although it cannot be proven, the month-long delay and subsequent tumor regrowth may have compromised our distal resection margin, necessitating a left upper lobe sleeve resection instead. Therefore, in cases in which a parenchyma-sparing sleeve resection is feasible, we highly recommend early and expedited surgical intervention.

In conclusion, IMTs are rare lesions and are the most common benign pulmonary tumors in children. They are usually asymptomatic, but owing

to their large size, they can behave like a malignant tumor with potential invasion into mediastinal structures or the chest wall. Frozen sections performed at the time of the index operation can often be indeterminate, which can pose a dilemma to the surgeon at the time of the operation. Overall, the consensus remains that early diagnosis and complete resection not only decrease the rate of recurrence but also prevent the escalation of the type of surgical resection required with the added advantage of excluding underlying malignant disease.

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PATIENT CONSENT
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