Endobronchial Atypical Mycobacteria in an Immunocompetent Child

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Summary. Endobronchial granulomas in children are mainly caused by mycobacterial infections. In addition to Mycobacterium tuberculosis, other organisms such as nontuberculous mycobacteria (NTM) have emerged. These organisms cause a broad spectrum of pulmonary diseases. An isolated endobronchial NTM infection in a child is reported. After bronchoscopic removal, a decision not to add drug treatment was made, with satisfactory results. Treatment options are not well established in children and remain a source of controversy. Different options are discussed.


Key words: endobronchial granuloma; nontuberculous micobacteria; Mycobacterium avium.

INTRODUCTION

Endobronchial granulomas in children are mainly caused by mycobacterial infections. In addition to Mycobacterium tuberculosis, other organisms such as nontuberculous mycobacteria (NTM) have emerged, and at present, Mycobacterium avium is responsible for 80% of all NTM infections.1 Historically, NTM have not caused invasive disease in immunocompetent hosts. However, in recent decades, an increasing number of case reports have documented the spectrum of disease caused by these bacteria in healthy individuals.2 An isolated endobronchial NTM infection in a child is reported.

CASE REPORT

A 14-month-old male, with a history of airway disease and wheezing was admitted to our hospital for pneumonia. One month later, the patient’s existing wheezing disease had worsened and a physical examination revealed asymmetrical breath sounds. A chest X-ray was obtained showing air trapping in the left lung suggestive of endobronchial obstruction.

Therefore, the patient was admitted for evaluation by bronchoscopy (rigid bronchoscope extended length, Karl Storz), where granulation tissue was seen to be obstructing the left mainstem bronchus (Fig. 1); a sample biopsy was taken, revealing necrotizing granulomatous inflammation but no mycobacterial organisms were demonstrated histologically by way of acid-fast stain (Fig. 2). Bacterial, fungal, virus, and mycobacterial cultures were done, all of which were negative. Other tests to rule out tuberculosis infection were negative (morning gastric aspirate, tuberculin skin test, and Quantiferon® test). In order to assess lung extension, a chest CT was performed, showing an endobronchial mass occluding the left mainstem bronchus, but no lymphadenopathy or parenchymal infiltrates were seen (Supplementary Fig. 1).

The granulation tissue was removed with surgical forceps by rigid bronchoscope and sent for new cultures. Fifteen weeks later, M. avium was cultured from the endobronchial mass. Due to scarce growth, no susceptibility tests could be performed.

To confirm immunocompetent status, other tests were performed: white blood cell counts and differentials were normal, testing for human immunodeficiency virus was
negative. Cystic fibrosis was also ruled out. To complete the evaluation, a new chest radiograph was performed, offering no evidence of disease. Consequently, we decided not to add chemotherapy. At 2 years follow-up the patient remains asymptomatic, and chest radiograph is normal (Supplementary Fig. 2). No further bronchoscopy testing has been carried out.

DISCUSSION

NTM is a collective term used to refer to different species of the genus *Mycobacterium* not belonging to the *M. tuberculosis* complex. *M. avium* is the second most common cause of mycobacterial infection after tuberculosis and its incidence has dramatically increased in recent years; however, its true magnitude is difficult to ascertain since is not a reportable disease.

Historically, NTM were commonly believed to be laboratory contaminants or to colonize airways, not causing disease in immunocompetent hosts. Except in patients with cystic fibrosis, children rarely develop pulmonary disease, and lymphadenitis is by far the most common expression of infection with NTM in the immunocompetent pediatric population. Immunocompromised patients, on the other hand, have a much greater tendency to develop disseminated disease. However, there has also been an increasing number of reports of previously healthy pediatric patients afflicted with atypical mycobacterial pulmonary infection, ranging from isolated endobronchial forms as in the case reported to more aggressive involvement like parenchymal infiltrates, cavities, and pleural involvement.

Diagnosis of *M. avium* pulmonary disease is extremely difficult, especially in pediatric patients. Signs and symptoms are variable and nonspecific (chronic cough, sputum production, dyspnea, fever, or hemoptysis). There is also a wide range of radiographic findings including isolated pulmonary nodules, mediastinal lymphadenopathy, parenchymal infiltrates, or bronchial obstruction. In the absence of specific diagnostic features in the history and physical examination, a chest roentgenogram, differential skin testing, and NTM isolation in a culture are essential for diagnosis. However, as these organisms are commonly found in nature, contamination of culture material or transient infection does occur. Thus, a single positive sputum culture, especially containing small numbers of organism, does not always suffice for diagnosis of NTM disease. According to these factors, the American Thoracic Society (ATS) has proposed diagnostic criteria.

Controversy also exists regarding treatment options for the disease. One of the major historical concerns was under what circumstances patients should be given drug therapy. The newer macrolides have had a significant impact on the treatment of this disease, but studies on selection and emergence of drug-resistant bacteria have shown that they should not be given alone, but rather in a three–four drug regimen usually including rifampicin (or rifabutin), ethambutol, or isoniazid. The ATS recommended prolonged antibiotic therapy in adults with pulmonary infection caused by NTM, but no standard therapeutic regimen exists for children.

On the other hand, surgical excision for cervical lymphadenitis in children is nearly always curative for *M. avium* (95% of success rate without chemotherapy). This experience, coupled with the side effects of the antibiotic regimen and the high level of drug resistance in NTM isolates, calls for caution when deciding to treat pediatric patients with multiple antibiotics must be made with caution. The need for and effectiveness of antimicrobial therapy for pulmonary NTM has not been established in immunocompetent children, and some clinicians recommend surgical resection for the lesions. When endobronchial MNT lesions produce symptoms of obstruction, lesions can usually be removed by forceps or thermal laser.

In all pediatric cases of NTM pulmonary disease reported thus far, acid-fast bacilli were present in the
initial biopsy; consequently, all completed a multiple regime of antibiotics.\(^1,\)\(^2\) Conversely, in our case, neither the histological sample nor the acid-fast stain revealed mycobacterial organisms, and therefore the patient did not receive medical therapy following endobronchial lesion removal. We received the results from the culture 15 weeks after the patient was discharged; patient condition and chest X-ray had not worsened, so the decision was made not to add chemotherapy. As previously reported, at 2 years follow-up, the patient remains in good health. Given that cervical lymph nodes in immunocompetent patients undergo spontaneous resolution in 12–18 months, we suggest that endobronchial lesions have a similar resolution. As in any patient, the decision to treat should be made by weighing the anticipated risk and benefits, and close observation is indicated if a decision is made not to treat.\(^2\)

REFERENCES