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Case Report

Endotracheal involvement from atypical mycobacterium causing respiratory difficulty: Case report and literature review☆

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ABSTRACT

Atypical mycobacterium infections often present as cervicofacial lymphadenitis in pediatric patients. Endotracheal involvement, however, is rare, and has not been previously described with imaging and photographs.

An infant with natural killer T-cell deficiency was admitted with cough, rhinorrhea, and cervical lymphadenopathy. Laryngotracheobronchitis-type symptoms persisted and imaging revealed an intraluminal abnormality of the trachea. Endoscopy confirmed a mediastinal lymph node with intrusion into the tracheal lumen. Intraluminal biopsy was deferred due to concerns of airway loss. Biopsy of the associated cervical lymph node confirmed Mycobacterium avium-intracellulare infection. The patient was managed with antibiotics and steroids with clinical resolution of his respiratory symptoms.

1. Introduction

Atypical, or non-tuberculous, mycobacterium infection is a common source of chronic cervicofacial lymphadenitis in the immunocompetent pediatric patient [1–4]. Patients typically present with a unilateral enlarged lymph node that is not responsive to standard antibiotic treatment. As surgical excision is favored over long term antibiotic treatment, these patients represent an important group to the otolaryngologist [2].

These infections are generally of low virulence. However, in the setting of immunodeficiency, atypical mycobacterial infection can result in regional or systemic spread [5]. This spread may result in further complications depending on the organ system involved. Natural killer T-cell deficiency is a rare disorder, but causes impairment of innate and adaptive immunity [6,7].

We present a pediatric patient with natural killer T-cell deficiency with atypical mycobacterial involvement of the trachea, leading to persistent respiratory symptoms masked as laryngotracheobronchitis. Further work-up with imaging and bronchoscopy was performed to characterize his disease. We describe the presentation, clinical course, work-up, management, and follow-up of this patient's disease process. Imaging and photographs, which have not been previously utilized in the literature, are provided. In addition, a review of the literature is performed. IRB approval was not required for this case report per our institution's guidelines.

2. Case report

Our patient is a 24-month old male with natural killer T-cell deficiency. He had a complex infectious disease history starting with diffuse varicella infection after receiving the varicella vaccine. He also developed multiple other viral infections including HHV-6 viremia and pneumonitis, adenovirus viremia and pneumonitis, CMV pneumonitis, and EBV pneumonitis. Work-up prior to admission revealed his natural killer T-cell deficiency.

As an outpatient, he was evaluated for a recent fever with rhinorrhea, cough, and emesis. Due to his history, he was admitted to All Children's Hospital for further work-up and treatment, as well as eventual plan to proceed with bone marrow transplant. Four weeks into his admission, the patient continued to have noisy breathing. There was concern by the primary and pulmonary teams for possible adenoidal contribution to his respiratory issues and the pediatric otolaryngology service was consulted. He also had cervical lymphadenopathy, primarily an enlarged right cervical lymph node. Due to his symptoms and presentation, recommendation was made for microlaryngoscopy and rigid bronchoscopy to further

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evaluate the patient's airway. However, at that time, he was not yet cleared for general anesthesia. He continued to be observed and treated for presumed laryngotracheobronchitis.

Initial chest x-ray imaging was unremarkable beyond a mild degree of prominence of the perihilar lung markings. Due to persistent respiratory symptoms, a CT neck and thorax were then obtained by the primary team. This revealed a 2.3 × 1.8 cm mediastinal mass with intrusion into the trachea causing >50% obstruction (Fig. 1). At this point, the pediatric otolaryngology service was re-consulted. He was cleared for general anesthesia and underwent microlaryngoscopy with rigid bronchoscopy. The laryngoscopy exam was clear of concerning findings, but the bronchoscopy revealed a posteriorly based intraluminal mass with smooth mucosa, located within the distal trachea (Fig. 2). The remaining trachea was clear to the carina. Any manipulation of the mass caused bleeding.

The decision was made to observe the intraluminal mass for multiple reasons. At the time, despite his mild stridor at rest, the patient was clinically comfortable without major respiratory distress. A needle or excisional biopsy could potentially compromise the airway due to bleeding or spillage of contents. In addition, the etiology was unclear at the time, but he also had mediastinal and cervical lymphadenopathy. It was deemed safer to perform a biopsy of his cervical lymphadenopathy in order to obtain a diagnosis.

A biopsy of his enlarged right supraclavicular lymph node was then performed. Microbiology returned as Mycobacterium avium-intracellulare (MAI). The patient was started on antibiotic treatment for his MAI infection, utilizing a regimen of azithromycin, ciprofloxacin, ethambutol, and rifampin. In addition, he was started on IV methylprednisolone to decrease the degree of lymphadenitis.

Repeat imaging at 3 and 6 months post-procedure showed no significant radiographic change of the intraluminal mass. Clinically, however, the patient continued to improve from a respiratory standpoint until he had resolution of his respiratory symptoms within 10 months. Overall, he had a prolonged admission to treat multiple infections. He was continued on antibiotics and steroids. There was discussion of weaning his steroids, however he was found to be an acceptable candidate for bone marrow transplant and thus the steroids were continued in preparation for his transplant. He received the transplant 1.5 years after admission, with discharge occurring 6 months later. He has continued to follow up as an outpatient, displaying no further respiratory issues. He has been transitioned to oral hydrocortisone and single-agent ciprofloxacin, and will remain on antibiotics until demonstrating immune reconstitution. An outpatient CT thorax obtained for post-transplant monitoring 2.5 years after his bronchoscopy showed radiographic resolution of the intraluminal mass.

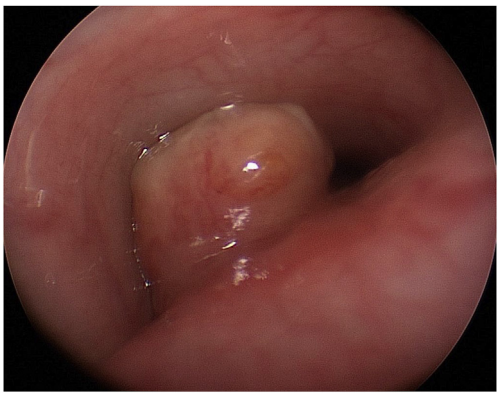

Fig. 2. Intraluminal mass within the distal trachea as visualized with rigid bronchoscopy.

3. Discussion

Atypical, or non-tuberculous, mycobacterium (AMB) are mycobacterium that do not cause tuberculosis or leprosy. They are acid-fast staining, aerobic bacteria that are ubiquitous throughout the environment [1–4]. They are generally of low virulence [1]. However, in healthy children under the age of 5, they are the most frequent cause of chronic lymphadenitis [1,2]. Decreased immunity and the penchant to place objects, which may be contaminated with soil, in their mouths have been theorized as potential causes for the increased incidence among young children and localization to the cervicofacial lymph nodes [4]. The most frequent bacterial isolate is Mycobacterium avium-intracellulare [1–3]. Most commonly, patients present with a unilateral, enlarging, painless cervical mass that does not respond to antibiotics. The mass may progress to cause an overlying violaceous skin discoloration, skin changes and breakdown, and a chronic draining wound. Diagnosis is clinically based as cultures may take weeks to result, or may not result at all. However, PCR can be used to expedite isolation of an organism [1,3]. It is important to rule out tuberculosis as the causative agent. Imaging is primarily utilized to determine extent of disease, or for preoperative planning.

Standard of care for AMB cervical lymphadenitis is to perform total excision with or without limited neck dissection [1,3]. Panesar et al. performed a retrospective study of 79 children over 10 years and found a 1.8% (1/55) recurrence rate in patients treated with complete excision; in comparison, there was a 95.2% (20/21) recurrence rate in patients treated with incision and drainage [1]. In addition, incision and drainage is associated with chronic wounds,

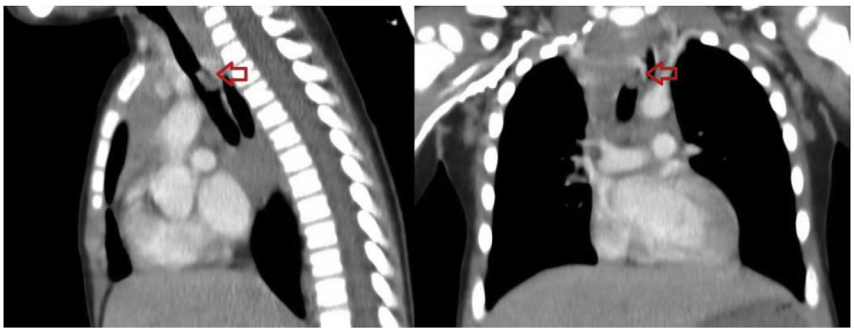

Fig. 1. CT image. Left image is the sagittal view revealing endotracheal obstruction by the mass (arrow). Right image is the coronal view revealing mediastinal mass effect on the airway and endotracheal extension (arrow).