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Case report

Bacteroides bacteremia complicating otogenic Lemierre's syndrome

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ABSTRACT

Lemierre's syndrome is a complex infection involving oropharyngeal sources leading to septic jugular thrombophlebitis. Otogenic variants may arise from complicated ear disease and involve cranial venous sinuses. We describe a case of a 9 year boy with chronic suppurative otitis media leading to mastoiditis, cranial venous sinus and jugular vein thrombophlebitis, and *Bacteroides* bacteremia. Both the pathogen and the extent of venous involvement were unusual in this case. Otogenic Lemierre's syndrome is a serious infection associated with significant morbidity which should be considered in any child with otogenic findings and septic thrombophlebitis.

1. Introduction

Lemierre's syndrome was first described in the early 1900s by Courmont and Cade and further characterized in 1936 by André Lemierre [1]. The classic form of the disease consists of a primary oropharyngeal infection, followed by thrombophlebitis of the internal jugular vein leading to anaerobic septicemia and metastatic infection [2]. Others have described clinically similar variants of Lemierre's syndrome, such as "Lemierre's-like syndrome," using various alternate criteria [3]. Full characterization of this condition remains elusive due to lack of consensus on the case definition. In a comprehensive review, Riordan identified no fewer than twelve case definitions used in previous case series, and summarizes major points of contention, including the minimum radiographic findings required for diagnosis and restriction to cases with oropharyngeal infection [4]. Disagreement on the requirement for isolation of *Fusobacterium necrophorum,* the most commonly implicated causative organism, also continues to be debated, as Lemierre's syndrome has been shown to involve other pathogens, including *Bacteroides*, *Prevotella*, streptococcal and staphylococcal species [4–6]. We here describe an unusual case of otogenic Lemierre's syndrome with bacteremia in a child secondary to *Bacteroides fragilis*, a less common pathogen.

2. Case report

A 9 year-old male with a history of frequent "acute otitis media" according to the reports of the caregiver was transferred to our facility for jugular venous thrombosis. He had two episodes of suppurative otitis media in the four months prior to presentation. At an outside hospital emergency department (ED) 15 days prior to transfer, he was diagnosed with bilateral suppurative otitis media and treated with azithromycin. Despite this therapy, he developed neck pain and his otorrhea persisted. A specimen of the ear drainage was sent for bacterial culture and his antibiotic was changed to amoxicillin-clavulanate. This aerobic culture grew *Proteus vulgaris*, prompting another antibiotic change to oral trimethoprim-sulfamethoxazole.

He continued to worsen with development of left neck swelling, increased neck and ear pain, torticollis, and headache. He returned to the ED three days prior to transfer and was admitted after non-contrast computed tomography (CT) of the left temporal bone showed left mastoid hypoplasia and opacification, interpreted as

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mastoiditis. On admission to the referring hospital, he was febrile to 39.4° Celsius (C). Complete blood count revealed total white blood cell count (WBC) of 14600/mm^3 with 66% neutrophils and 20% band forms, hemoglobin 9.4 g/dL, and platelets 111000/mm^3. Erythrocyte sedimentation rate was 60 mm/hr. Bacterial cultures of the peripheral blood and urine were obtained and did not yield growth. He was treated with intravenous ceftriaxone and topical ciprofloxacin but remained febrile, and his neck pain and swelling worsened. This prompted repeat CT of the head and neck, which showed diffuse thrombosis of the left jugular vein with extension through the jugular bulb to the proximal transverse sinus (Fig. 1). His antibiotics were changed to vancomycin and piperacillin-tazobactam and he was transferred to our hospital.

At our facility, his temperature was 40.0 °C, pulse 134 beats per minute, respirations 40 per minute, with blood pressures of 88/51 mmHg. He was comfortable without supplemental oxygen with saturations of 96%. Weight was 23.5 kg (10th percentile for age), height was 129 cm (20th percentile for age), and BMI 14.1 kg/m^2 (5th percentile for age). His examination was significant for scarred and opacified tympanic membrane bilaterally with perforation and purulent material in the external canal. He had diffuse left neck tenderness and swelling with decreased range of motion but had absence of swelling, erythema or tenderness over the mastoids. Aerobic and anaerobic blood cultures were obtained from a single peripheral blood specimen, vancomycin and piperacillin-tazobactam were continued, and enoxaparin was started for anti-coagulation. He was taken to the operating room (OR) for bilateral microscopic myringotomy with tympanostomy tube insertion. His initial peripheral blood culture grew *B. fragilis* from the anaerobic bottle, while operative specimens obtained from the middle ear grew rare *P. vulgaris* on aerobic cultures and *B. fragilis* on anaerobic cultures; additional blood cultures obtained 48 and 72 hours after admission were negative. Over the following four days, he remained febrile with persistent left neck swelling and pain. Repeat CT scan with contrast showed progression of thrombosis into the left transverse sinus and proximal left external jugular vein, septic emboli in bilateral lung apices, and bilateral coalescent mastoiditis with left ossicular, septal and cortical erosion (Fig. 2). On his seventh hospital day, he returned to the OR for left microscopic tympanomastoidectomy and otoendoscopy. He was found to have a left middle ear and mastoid cholesteatoma which was excised, a bony defect over the sigmoid sinus with purulence, and absence of the short process of the incus, which was removed. Operative specimens yielded growth of *Proteus* spp. from the mastoid bone from aerobic cultures, and *Prevotella oris* from anaerobic cultures. He was continued on piperacillin-tazobactam, and vancomycin was discontinued on day 11. On day nine the following magnetic resonance (MR) imaging using a 3 T magnet following pulse sequences of the brain were obtained: sagittal T1, axial diffusion, axial T2, MR venogram, and axial fluid-attenuated inversion recovery (FLAIR). These images showed unchanged thromboses in the cranial and jugular venous drainage, and follow-up CT with contrast on day 12 showed no progression of the thromboses. An immune evaluation was performed including quantitative immunoglobulins, T and B cell subsets, neutrophil oxidative burst assay and tetanus, diphtheria, measles, *Haemophilus influenzae* type B and pneumococcal titers, all of which were within normal limits. He was discharged home with a peripherally-inserted central venous catheter to complete six weeks of antibiotic therapy. He returned 9 days after discharge for scheduled right microscopic tympanomastoidectomy with excision of right middle ear and mastoid cholesteatoma. He completed two additional weeks of oral amoxicillin-clavulanate and ciprofloxacin after completion of intravenous therapy. He finished three months of enoxaparin therapy for his thrombosis, however, repeat CT with angiography showed complete loss of the left transverse sinus down to the entry of the left jugular vein into the innominate vein.

3. Discussion

This manuscript describes a case of otogenic Lemierre's syndrome characterized by chronic suppurative otitis media (CSOM) with cholesteatoma, chronic mastoiditis, and cranial sinus and jugular thrombophlebitis with septic pulmonary emboli. CSOM is a term used to describe a patient who has a perforated tympanic membrane with persistent drainage from the middle ear which lasts several weeks. This can occur with or without a cholesteatoma. To our knowledge, this is only the second case of *B. fragilis* bacteremia associated with otogenic Lemierre's syndrome reported in the literature [7]. There are a number of features of this case which are unique. The normal external physical examination of the mastoid bone despite a progressive otogenic source was a complicating factor. This was likely due to undiagnosed cholesteatoma and chronic mastoiditis, which was not fully appreciated

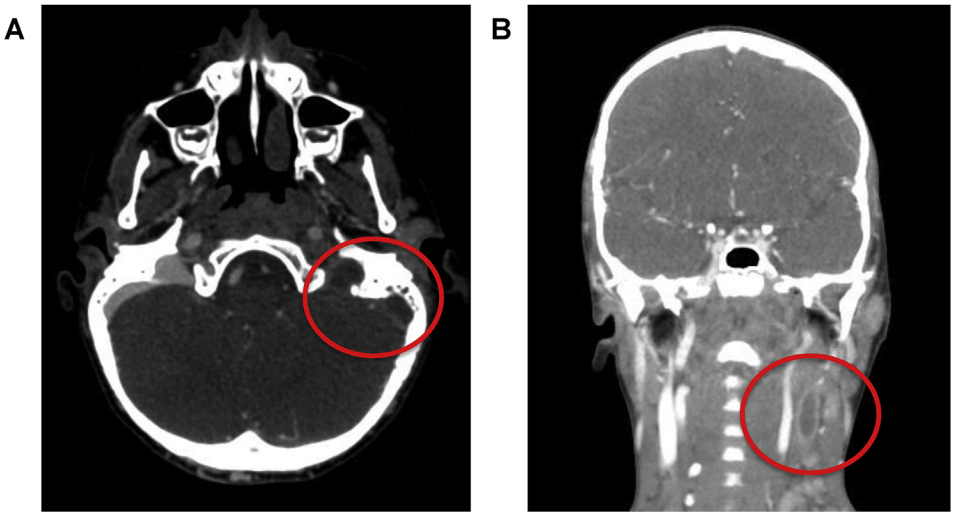

Fig. 1. Computed tomography of the neck. Axial radiograph (A) shows absence of contrast in the transverse sinus and jugular foramen consistent with thrombus. Coronal image (B) demonstrates no contrast in the left jugular vein but thickened walls with associated soft tissue swelling and inflammatory changes involving the left sternocleidomastoid muscle.