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CASE REPORT

Lemierre's syndrome in a patient with habitual toothpick usage

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Lemierre's syndrome is characterized by septic thrombophlebitis of the internal jugular vein that is complicated by metastatic infections. The disease usually presents after oropharyngeal infection. In rare cases, odontogenic infection has been implicated as culprit. Here, we report a case of Lemierre's syndrome that most likely developed secondary to toothpick usage. The patient had an uneventful recovery after the timely administration of the appropriate antibiotics.

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Introduction

Lemierre's syndrome was first described in 1936 by Andre Lemierre, a French physician and bacteriologist. Initially referred to as "postanginal septicemia," the disease usually present after oropharyngeal infection, is characterized by thrombophlebitis of the internal jugular vein, and is frequently followed by metastatic infections. The clinical entity became known as "Lemierre's syndrome" in the late 1980s. In the era before antibiotics, the disease had been

largely lethal. However, as the use of antimicrobials for oropharyngeal infections increased, the frequency of Lemierre's syndrome declined and it became known as "the forgotten disease." In the present report, we describe a case of Lemierre's syndrome in a 17-year-old man, whose infectious origin may be linked to toothpick usage.

Case report

A 17-year-old man presented at the emergency department and complained of a 10-day history of fever, chills, productive cough with purulent sputum, and right neck pain when swallowing food. He also indicated generalized malaise, muscle soreness, and headache. One day prior to

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admission, the patient presented at a local hospital due to a fainting episode when getting up from bed. He was diagnosed with hypotension and severe leukocytosis at that hospital. A rapid test for influenza A was positive. The patient was treated with oseltamivir and levofloxacin, then referred to Mackay Memorial Hospital. Review of systems was negative for sore throat, hoarseness, nausea, and vomiting. The patient's past medical history indicated ketamine abuse. The patient had no history of recent travel or animal bites. His medical history indicated significant and habitual use of toothpicks after meals, sometimes to the point of causing bleeding gums.

Upon presentation, the patient appeared very ill. His temperature was 37 °C with a regular pulse rate of 112 beats per minute, a respiratory rate of 20 breaths per minute, and a blood pressure of 73/36 mmHg. Oral examination revealed large cavities in the first molars of the upper and lower left teeth. Swelling and tenderness of the right lateral neck was also found. The patient's cardiac exam was significant for tachycardia. The rest of the physical examination was unremarkable.

The initial laboratory investigations were significant for the following findings: leukocyte count, $46,800 \times 10^3/\mu\text{L}$ (95% neutrophils, 1.0% lymphocytes); hemoglobin, 11.8 g/dL; hematocrit, 35.0%; C-reactive protein, 25.64 mg/dL; aspartate aminotransferase (AST), 51 IU/L; alanine aminotransferase (ALT), 72 IU/L; and creatinine, 2.2 mg/dL. Sodium and potassium levels were within the reference range, and blood gas was compatible with respiratory alkalosis. Chest radiographs showed slightly prominent peribronchial markings over the bilateral parahilar region, which are suggestive of inflammation. Electrocardiogram showed sinus tachycardia. A drug screen was positive for acetaminophen, benzodiazepines, opiates, and cannabis. Blood tests for HIV antibodies, serum *Mycoplasma pneumoniae* IgG and IgM antibodies, and serum antistreptolysin O (ASLO) were all negative. Urine *Legionella* antigen test was also negative.

The patient was admitted to pulmonology services under the initial diagnosis of influenza A that was possibly complicated by pneumonia. Intravenous levofloxacin treatment was continued. The following day, both sets of blood samples that were inoculated into the culture media at the time of admission showed gram-negative bacilli of unknown identity. Meropenem (1 g every 8 hours) was added to the treatment regimen, and levofloxacin was discontinued the following day. Follow-up chest radiograph revealed the progression of right lung pneumonia with parapneumonic effusion. Arterial blood gas levels indicated hypoxemia. The patient was transferred to the intensive care unit due to the possibility of septic shock. A series of studies performed at the ICU, including abdominal ultrasound, chest echogram and echocardiogram, revealed no significant abnormalities. On day 4 in the hospital, the patient's hypotensive status resolved and he was transferred to general ward. Because the organism in the blood culture was found to be anaerobic, metronidazole was empirically added. Two days later, the patient complained of increasing pain on the right side of his neck. Contrast-enhanced computed tomography (CT) revealed an irregular rim-enhanced lesion beneath the right sternocleidomastoid (SCM) muscle along levels II–IV of the internal jugular chain to the supraclavicular region. Lymphadenitis with abscess

formation or septic thrombosis of the internal jugular vein were suspected (Fig. 1).

An ultrasound of the neck was performed, as shown in Fig. 2. On the left side, smooth blood flow was seen in the internal jugular vein and carotid artery. On the right side, however, circulation was absent in the internal jugular vein, indicating total obstruction. A diagnosis of Lemierre's syndrome was made.

On day 10 in the hospital, *Fusobacterium necrophorum* was identified in the initial blood culture. Meropenem was replaced by penicillin (4 MU were intravenously administered every 4 hours), which was then changed to ampicillin/sulbactam (3 g were intravenously administered every 8 hours) on day 15 in the hospital. A Ga-67 scan was performed to identify other septic emboli that might have failed to show increased uptake in regions other than the internal jugular vein.

The patient was administered metronidazole for a total of 19 days. Ampicillin/sulbactam was administered for another 4 weeks. His clinical condition markedly improved. We arranged for teeth extraction during admission. Amoxicillin was prescribed to patient upon discharge. On a followed-up appointment 2 months after disease onset, the patient was doing well and was symptom-free, though his right internal jugular vein remained occluded.

Discussion

Lemierre's syndrome is a rare but potentially lethal disease. The incidence is estimated to be 0.8–1.5 cases per 1 million persons per year.¹ The disease used to be more common before the advent of antibiotics. With antimicrobial usage, the number of cases reported declined, only to



Figure 1. Contrast-enhanced CT showing an irregular rim-enhancing lesion beneath the right SCM along internal jugular chain level I–IV to the supraclavicular region, revealing septic thrombosis of internal jugular vein.

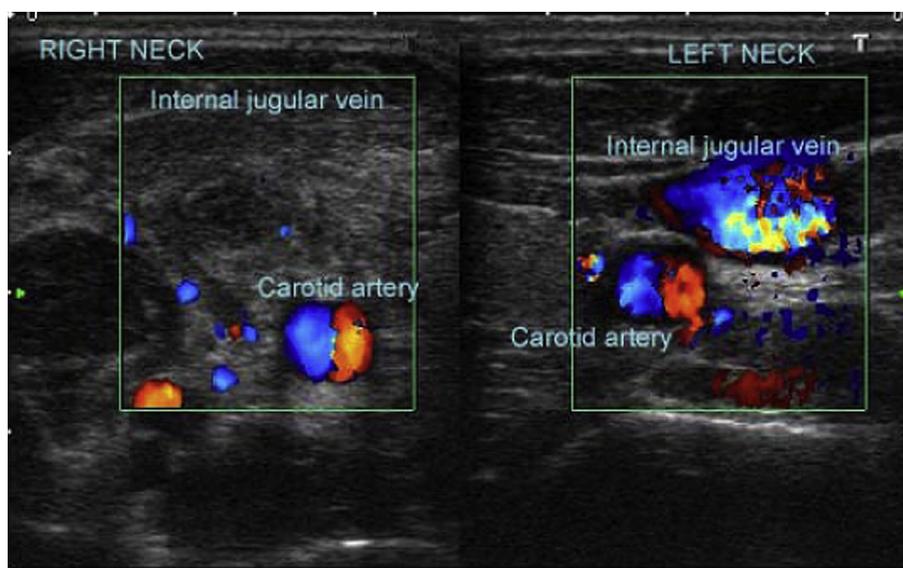


Figure 2. Ultrasound of the neck revealing the absence of blood flow in the right internal jugular vein.

be followed by a surge in the last two decades, which is speculated as related to the reduction in outpatient usage of antibiotics.^{1,2} Most affected patients are between 12–28 years of age and male.¹ *Fusobacterium necrophorum* is the organism most commonly implicated.^{1–3} Believed to be a commensal organism that lives in the oropharynx of human beings, *Fusobacterium* manifest as opportunistic infections after the disruption of the oropharyngeal mucosa by dental caries. While virulence factors such as capsular polysaccharides and pili have been observed in the organism, possibly playing a role in its adherence, extracellular enzymes including proteases, collagenase, DNase, neuraminidase, and hyaluronidase enable bacteria to break through the epithelium.⁴ Lemierre's syndrome arises when infection of the oropharynx spreads to the lateral pharyngeal space, which contains the internal jugular vein. From the neck, metastatic infections spread mostly commonly to the lungs, followed by joint, muscle, bone, liver, skin, spleen, or heart. Meningitis, epidural/subdural abscess, cavernous/sigmoid/transverse/lateral sinus thrombosis, and stroke have also been reported.²

The clinical manifestations of internal jugular vein thrombosis may include cervical swelling and pain. A cord may be palpated beneath the SCM muscle. In a large series of patients with internal jugular vein thrombosis reported by Tovi et al, the most common clinical manifestation was fever (83%), followed by leukocytosis (78%), neck swelling (72%) and cervical pain (66%). Less commonly observed signs included cord sign (39%) and sepsis syndrome (30%).⁵

The mainstay of treatment is the timely administration of antibiotics. *F. necrophorum* is usually susceptible to penicillin, clindamycin, metronidazole, and chloroamphenicol.^{1,6,7} Jacinto et al was able to isolate *F. necrophorum* from primary endodontic infections in a Brazilian population. The clinical isolates were 100% susceptible to amoxicillin, amoxicillin/clavulanate, and cephaclo.⁸ Brazier et al found that among 205 isolates of *F. necrophorum* studied in England and Wales from 1990–2000, all of the isolates were susceptible to metronidazole, but 2%

were also resistant to penicillin and 15% were resistant to erythromycin.⁹ Kuriyama et al studied 158 strains of *F. nucleatum*-like bacteria, of which a few may have been *F. necrophorum*. There were only five strains that showed resistance to amoxicillin, and no strain was positive for β -lactamase production.⁶ Aldridge et al studied 22 clinical isolates of *Fusobacterium* and found that 4.5% demonstrated β -lactamase production.⁷ The concern for β -lactamase production by the microorganism has prompted the recommended use of ampicillin/sulbactam.¹⁰ Surgical management is indicated only for patients in whom other medical therapies have failed. The role of anticoagulation in patients with Lemierre's syndrome is controversial.

In a search of the English medical literature for articles published between 2001–2008, Rosado et al. found that for Lemierre's syndrome the primary site of infection is the oropharynx (59.5%), followed by manifesting as mastoiditis (15%). Only 7 of 99 cases surveyed presented with primary odontogenic infection (7%).³ Although our patient could certainly have developed Lemierre's syndrome secondary to *Fusobacterium* harboring in dental caries, one question whether his habitual usage of toothpicks may have contributed to his disease. An instrument that is well known to all cultures, the toothpick has played a major role in the history of dental health for 2 million years.¹¹ Although the modern toothpicks are made of wood or bamboo, in the past, they were made of variety of materials, including gold, bronze, silver, chicken or fish bones, and goose quill.¹¹ Regardless of the type of material used, the use of these objects is possibly the oldest bad habit known to mankind. In minor cases, toothpicks can cause tooth abrasion.¹² In severe cases, they have been found to cause intestinal perforation,^{13–15} liver abscess,¹⁶ pancreatitis,¹⁷ and even cardiac tamponade.¹⁸ Extrinsic mucosal trauma caused by toothpick usage may have induced Lemierre's syndrome in our patient.

A high index of suspicion is required in the diagnosis of Lemierre's syndrome. Early signs may be subtle and the disease may be overlooked as a simple case of pneumonia.

Because the causative organism is often anaerobic, microbiologic confirmation is often delayed. Here, we presented a case of Lemierre's syndrome associated with toothpick use. The importance of this life-threatening disease cannot be overlooked.

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