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FUSOBACTERIUM SEPTICEMIA COMPLICATED BY CEREBRAL SUBDURAL AND EPIDURAL EMPYEMAS: A RARE CASE OF LEMIERRE SYNDROME

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□ **Abstract—Background:** Lemierre syndrome is characterized by postanginal septicemia and internal jugular vein thrombophlebitis with secondary septic emboli, typically to the lungs. The central nervous system (CNS) is rarely involved. **Objective:** To present a case of Lemierre syndrome featuring cerebral subdural and epidural empyemas. **Case Report:** This case report describes the case of a 17-year-old youth with cerebral subdural and epidural empyemas. The findings of chest computed tomography of the neck and the blood cultures were compatible with Lemierre syndrome. The patient recovered well after antibiotic treatment and surgical debridement. **Conclusion:** Lemierre syndrome can result in infection spreading to the CNS, including cerebral subdural and epidural empyemas. This disease entity should be included in the differential diagnoses of CNS bacterial infections. © 2010 Elsevier Inc.

□ **Keywords—**Lemierre syndrome; subdural empyema; epidural empyema; internal jugular vein thrombophlebitis; central nervous system

INTRODUCTION

Lemierre syndrome, or postanginal septicemia, is characterized by anaerobic septicemia, internal jugular vein (IJV) thrombophlebitis, and septic emboli secondary to acute pharyngeal infection (1). *Fusobacterium* species

are the most common cause. Primary infections are usually in the tonsils and oropharynx, but otogenic, odontogenic, and sinogenic sources have been reported (2,3). The lungs represent the most common site for embolic infection and are affected in 79.8% of cases (3). In contrast, infections that spread to the central nervous system (CNS) are rare; when affected, CNS involvement may include cerebral sinus thrombosis, meningitis, cerebral infarcts, cerebral abscess, or occasionally, lower cranial nerves or sympathetic trunk (3–7).

CASE REPORT

A 17-year-old, previously healthy youth presented to the Emergency Department (ED) with a 5-day history of fever, sore throat, and headache. On arrival, his consciousness was clear, body temperature was 37.9°C, and physical examination showed enlarged tonsils. The patient's white blood cell count was 42,290/mm³ with 95% neutrophils, and chest X-ray study and urinalysis were normal. After blood cultures were obtained, the patient was begun on intravenous levofloxacin, 750 mg daily, and was managed in the ED because no inpatient bed was available. On the following day, the patient developed nuchal rigidity and drowsiness. Computed tomography (CT) scan of the brain was unremarkable. Analysis of the cerebrospinal fluid showed a white blood

cell count of $1255/\text{mm}^3$ with 88% polymorphonuclear cells, protein level of 166.2 mg/dL, and glucose level of 53 mg/dL (compared with a serum glucose of 132 mg/dL). He received intravenous cefpirome (2 g every 8 h) and vancomycin (1 g every 12 h) for suspected bacterial meningitis and was admitted to the intensive care unit for further management.

On the third day, the patient began to experience left hemiplegia. Brain magnetic resonance imaging (MRI) with gadolinium enhancement (Figure 1) showed a right frontoparietal subdural empyema, a left medial-frontal epidural empyema, and meningeal enhancement. On craniotomy, pus was found in the subdural and subarachnoid spaces (Figure 2). Surgical debridement was performed, and pathological evaluation of the tissue revealed necrotic debris and fibrinopurulent exudate. Bacterial cultures were negative.

Despite surgical debridement and continued antibiotic treatment, the patient's fever persisted. Physical examination revealed swelling of the right aspect of the neck. Head and neck CT scans showed evidence of right internal jugular vein thrombosis (Figure 3). Meanwhile, the original blood cultures yielded *Fusobacterium nucleatum*.

After treatment with intravenous meropenem (1 g every 8 h) and metronidazole (500 mg every 6 h) for 1 week, his fever subsided and the level of consciousness gradually returned to normal. A brain MRI scan 4 weeks after admission showed resolution of the right-sided subdural empyema and shrinkage of the anterior medial-frontal epidural

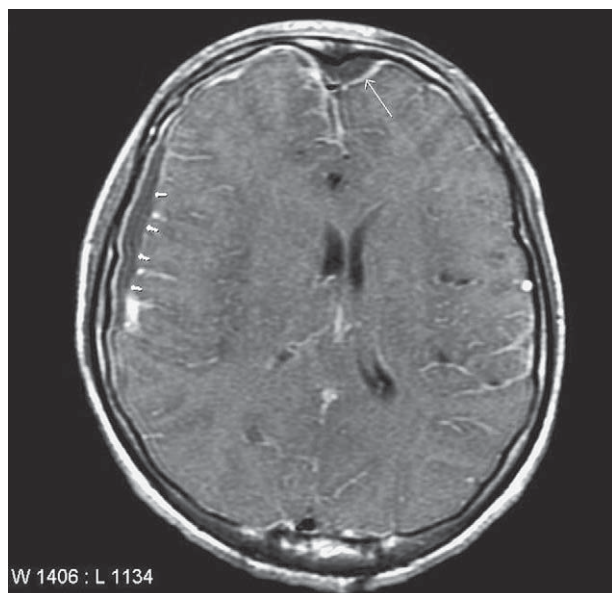


Figure 1. T1-weighted brain magnetic resonance imaging after gadolinium injection shows diffuse leptomeningeal enhancement bilaterally over the cerebral hemispheres with a faint high signal, suggesting a fluid collection with ring enhancement at the right frontoparietal subdural space (small arrows) and left medial-frontal epidural space (large arrow).

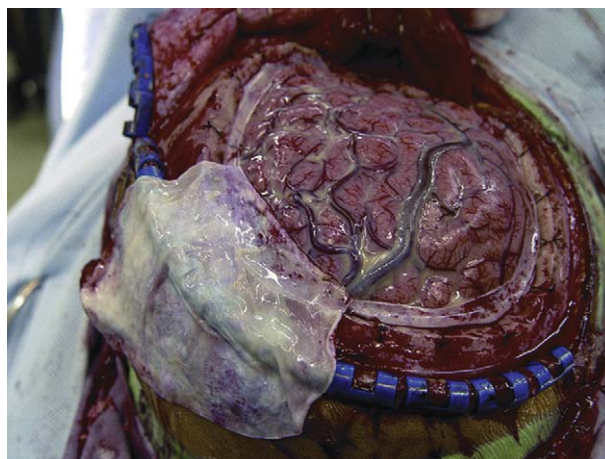


Figure 2. On craniotomy, purulent fluid was found in the subdural and subarachnoid spaces.

empyema. The patient never exhibited evidence of any other embolic infection, and was discharged without any neurologic sequelae after 8 weeks of antibiotic treatment.

DISCUSSION

Fusobacterium species are part of the normal oropharyngeal flora of healthy adults. The first stage of primary infection in Lemierre syndrome is pharyngitis in 87.1% of patients (3). With disease progression, bacteria invade mucosa weakened by the preceding pharyngitis and move to the lateral pharyngeal space, where they can cause IJV septic thrombophlebitis (3). The CNS is rarely involved

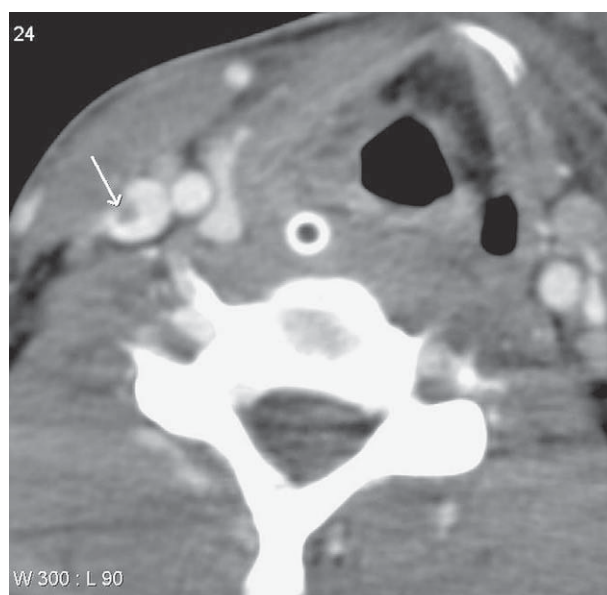


Figure 3. Post-contrast neck CT scan reveals a filling defect (arrow) in the lumen of the right internal jugular vein at the level of the thyroid gland, suggesting thrombus formation.

in embolic infection in patients with Lemierre syndrome, probably because the cerebral veins are upstream from the major sources of septic thrombi in the IJV.

Culturing *F. nucleatum* from our patient's blood, along with observing his evolving signs and symptoms, allowed us to make the appropriate diagnosis and manage his care. Based on the findings of this case and others in the literature, we recommend anaerobic cultures and anaerobic antibiotic coverage in patients who have pharyngitis and neck swelling, which may be preceding events of a CNS infection, until the causative organisms are definitively identified. In the management of patients with CNS infections, a thorough and careful physical examination is essential and may provide important clues to both the underlying pathology and appropriate treatment. Pharyngitis, although a very common disease entity, can cause complications such as Lemierre syndrome. Careful examination looking for signs of deep neck infection should not be omitted in any patient with pharyngitis.

CONCLUSION

Lemierre syndrome can cause embolic infections in the CNS, including cerebral, subdural, and epidural empyemas.

Lemierre syndrome is becoming a forgotten diagnosis, but it should be considered in the differential diagnosis of CNS bacterial infections.

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