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Clinical Record

Lemierre's syndrome presenting with peritonsillar abscess and VIth cranial nerve palsy

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Abstract

Lemierre's syndrome is characterized by acute oropharyngeal infection complicated by internal jugular venous thrombosis secondary to septic thrombophlebitis, and metastatic abscesses. We report a case of Lemierre's syndrome in an 18-year-old Caucasian woman presenting with a peritonsillar abscess and ipsilateral VIth cranial nerve palsy.

Key words: Peritonsillar Abscess; Abducens Nerve Diseases; Venous Thrombosis; Jugular Veins

Introduction

Lemierre's syndrome is extremely rare and is characterized by a triad of pharyngotonsillitis, internal jugular venous thrombosis secondary to septic thrombophlebitis, and metastatic abscesses. There has been a recent increase in the incidence of Lemierre's syndrome, which may reflect an increasing reluctance to prescribe antibiotics for sore throats. The authors report the case of a young Caucasian woman with Lemierre's syndrome who presented to a district general hospital ENT department with a VIth cranial nerve palsy.

Case report

An 18-year-old Caucasian woman presented to our ear, nose and throat department with a one-week history of sore throat. She had recently returned from a holiday in Greece, where she had been prescribed a seven-day course of Co-amoxiclav (amoxicillin and clavulanic acid mixture) 375 mg tds for tonsillitis. Her symptoms of sore throat, fever, rigors and odynophagia had worsened during the week. She also complained of a two-day history of neck pain, headache and blurred vision.

Examination revealed a pyrexia of 39.2°C, asymmetrical grade III tonsillar enlargement, trismus and a left peritonsillar collection. Palpable lymphadenopathy, which was tender to palpation, was noted in the left anterior cervical chain. Left malar and orbital oedema was also noted. Examination of the cranial nerves demonstrated a left VIth nerve palsy (Figure 1). Fundoscopy revealed mild bilateral papilloedema. There was a significant degree of neck stiffness but Kernig's sign was negative.

Laboratory investigations demonstrated a white cell count of 18.3×10^9/l, a platelet count of 47×10^9/l and a C-reactive protein concentration of 273 mg/l. The Paul Bunnell test was negative. A computed tomography (CT) scan of the patient's head revealed no abnormalities. At lumbar puncture, the opening pressure was 41 cms of water and the cerebrospinal fluid (CSF) was turbid. Analysis of the CSF demonstrated a neutrophil count of $125/mm^3$, 95 per cent of which were polymorphs. A chest X-ray revealed an opacity behind the left heart border, suggesting consolidation.

The patient was admitted to the department and commenced on intravenous benzyl penicillin 1 g qds, metronidazole 500 mg tds and cefotaxime 2 g bd. Polymerase chain reaction of the CSF was negative for meningococcus. Blood and CSF cultures failed to yield any organisms. A presumptive diagnosis of Lemierre's syndrome was made and a magnetic resonance venogram (MRV) was performed, which revealed extensive ipsilateral jugular venous thrombosis (Figure 2).

The patient was treated with intravenous metronidazole and cefotaxime and subcutaneous tinzaparin for two weeks, prior to discharge from hospital on a four-week course of oral amoxicillin, metronidazole and warfarin.

At review one week after discharge, the patient remained apyrexial and well. The left VIth nerve palsy and tonsillar enlargement persisted; however, the peritonsillar collection appeared to have resolved completely. The patient complained of difficulties with articulation of speech and mastication. Examination revealed a partial left hypoglossal nerve palsy.

At review two weeks after discharge, the problems with articulation and mastication persisted. There was some recovery of VIth nerve function. The patient was referred to the speech and language therapy unit and achieved complete recovery of the XIIth nerve palsy after several weeks.

Discussion

Lemierre's syndrome typically involves a triad of pharyngotonsillitis, internal jugular venous thrombosis and septic emboli, as characterized by A Lemierre in his 1936 study of 20 cases, published in the *Lancet*.[1] The pathogenesis most commonly involves the entry of the gram-negative, pleomorphic, obligate anaerobic bacillus

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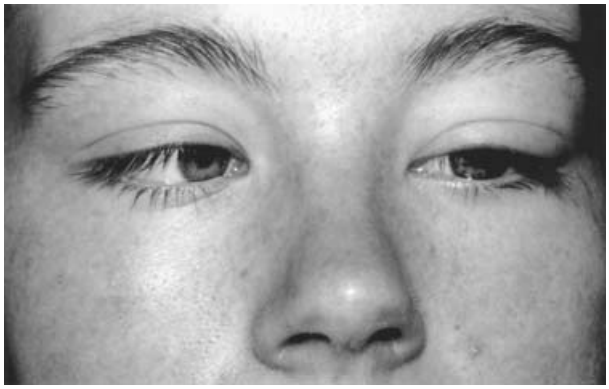

FIG. 1
Patient attempting left lateral gaze, demonstrating left abducens nerve palsy.

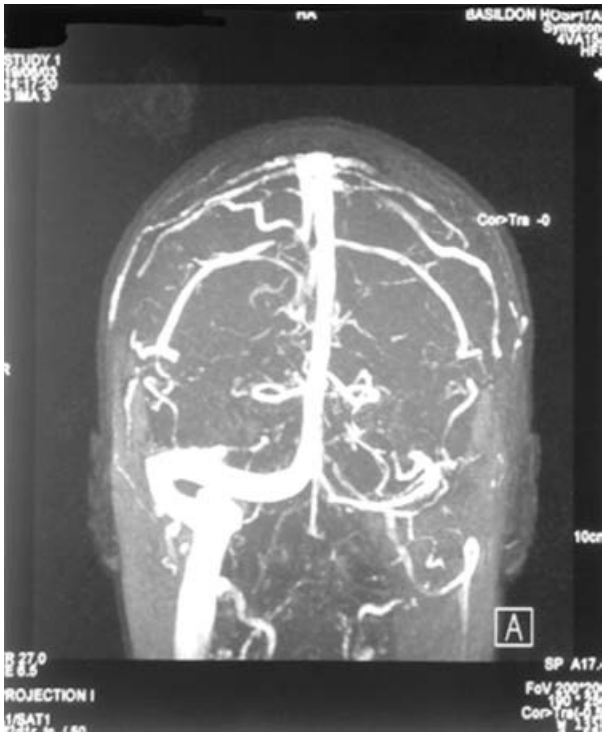

FIG. 2
Magnetic resonance venogram of head and neck, demonstrating left external jugular venous thrombosis.

Fusobacterium across the oropharyngeal mucosa secondary to trauma, inflammation or tissue destruction.[2] Infection by Epstein Barr virus may also contribute additionally by reducing T-cell-mediated immunity.[3] Fusobacterium forms part of the normal flora of the oropharyngeal, gastrointestinal and genito-urinary tracts. *F. nucleatum* and *F. necrophorum* are the most frequently isolated species, of which *F. necrophorum* is the most virulent.[4] *Fusobacterium necrophorum* produces a lipopolysaccharide endotoxin that aggregates platelets in culture, thereby creating the optimal anaerobic conditions required for bacterial propagation.[5] These organisms are generally sensitive to penicillin; however, before the antibiotic era Lemierre's syndrome was invariably fatal. The current mortality associated with Lemierre's syndrome is estimated to be between 4 and 12 per cent.[6] There appears to have been a recent increase in the incidence of Lemierre's syndrome in the United Kingdom, which may reflect an increasing reluctance to prescribe antibiotics for sore throats.[7,8]

The progression from the appearance of oropharyngeal infection to septicaemia is estimated to take approximately seven days.[9] The condition typically affects otherwise healthy adolescents and young adults. Septic emboli most commonly occur in the lungs but have also been reported in the joints, bones, skin, kidneys and meninges.[1,8] In our case, an abducens nerve palsy was also noted at presentation. This may have arisen from involvement of the cavernous sinus in the septic thrombophlebitis, although there appeared to be no extension of the thrombosis to the sinus on the original MRV scan. The patient also demonstrated an ipsilateral hypoglossal nerve palsy. Hypoglossal nerve palsies have been reported to occur secondary to retropharyngeal collections, in tuberculous infections for example, and this may explain the phenomenon in this case. Interestingly, the Epstein Barr virus may cause a similar clinical picture via neuritis affecting individual cranial nerves;[10] however, the Paul Bunnell test in our case was negative. It may be the case that both the VIth and XIIth cranial nerve palsies resulted from a cysterical meningitis; together with nasopharyngeal carcinomas, this is a recognized cause of the so-called clival syndrome.[11]

There is one report in the literature of Lemierre's syndrome involving meningitis and cerebral abscesses. These sequelae are thought to occur more commonly with primary ear infections as opposed to tonsillar infection. However, the current case, to our knowledge, is the first presentation of Lemierre's syndrome with a peritonsillar collection and VIth and XIIth cranial nerve palsies. Fortunately, there was no evidence of any cerebral collections on CT.

Standard therapy for Lemierre's syndrome involves the use of high-dose intravenous antibiotics for several weeks and consideration of drainage of any septic foci. Our patient was successfully managed with intravenous cefotaxime and metronidazole for two weeks, followed by a four-week course of oral amoxicillin and metronidazole. Our patient was also anticoagulated with tinzaparin and warfarin (under the guidance of the haematology department), although the efficacy of anticoagulation in such cases remains controversial.

We postulate that the unusual findings of VIth and XIIth nerve palsies may be sequelae of severe meningeal infection. There was no evidence of cavernous sinus involvement in the thrombosis on MRV. Compression of the hypoglossal nerve by the retropharyngeal abscess or by associated lymph nodes remains within the realms of possibility.

Conclusion

Lemierre's syndrome remains a rare but potentially life-threatening condition typically involving young adults and adolescents, which can be treated with prolonged courses of antibiotics. We have presented the first case, to our knowledge, demonstrating oropharyngeal infection, internal jugular venous thrombosis and cranial nerve palsies.

- **Lemierre's syndrome is characterized by a triad of pharyngotonsillitis, internal jugular vein thrombosis and metastatic abscesses**
- **This paper describes a case of Lemierre's syndrome associated with VIth cranial nerve palsy**
- **Aggressive treatment with high-dose intravenous antibiotics is required. The authors discuss the aetiology and clinical course of the condition in relation to the present case**

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