EJCRIM
European Journal
of Case Reports
in Internal Medicine

LEMIERRE'S SYNDROME: AN UNLIKELY DIAGNOSIS IN A PATIENT PRESENTING WITH DIARRHOEA AND VOMITING, OR MAYBE NOT?

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Received: 24/11/2024
Accepted: 17/03/2025
Published: 23/04/2025

Conflicts of Interests: The Authors declare that there are no competing interests.
Patient Consent: Written informed consent to publish his clinical data was obtained from the patient.
Acknowledgments: We thank the wider senior clinical team for discussions at the time which allowed us to progress this patient's care and further our understanding of this uncommon syndrome. We also thank the patient for allowing us to write up his case and present it.

How to cite this article: Papaleontiou G, Young A. Lemierre's syndrome: an unlikely diagnosis in a patient presenting with diarrhoea and vomiting, or maybe not? *EJCRIM* 2025;**12**:doi:10.12890/2025_005063

ABSTRACT

Lemierre's syndrome is an infectious thrombophlebitis of the internal jugular vein (IJV) where the commonest causative agent is a *Fusobacterium* from a tonsillar or peritonsillar abscess. This is a case presentation of a patient whose primary complaint was diarrhoea. The blood cultures of this patient grew a *Fusobacterium necrophorum* which prompted imaging. The imaging demonstrated a right peritonsillar abscess and IJV thrombosis with septic emboli in the lungs. Key points highlighted are: 1) blood cultures remain a key investigation in making a diagnosis in the septic patient; 2) abdominal symptoms can be part of Lemierre's syndrome; 3) abdominal symptoms (abdominal pain, vomiting and diarrhoea) have been characterised as the third, fourth and fifth most common feature after fever and dyspnoea. This case is presented because it shows how Lemierre's syndrome can present with abdominal features, and this may be incongruent with clinicians' understanding and teaching.

KEYWORDS

Lemierre's syndrome, fusobacterium, gastrointestinal, diarrhoea

LEARNING POINTS

- Lemierre's syndrome is an infectious thrombophlebitis of the internal jugular vein most commonly due to *Fusobacterium necrophorum* originating from a tonsillar or peritonsillar abscess, which may cause septic emboli in the lungs.
- Gastrointestinal symptoms, including diarrhoea, are commoner than appreciated or classically taught in this syndrome.
- Blood cultures are a key investigation in the septic patient and in those that grow a Gram-negative rod, *Fusobacterium* should be considered, and suspicions of Lemierre's syndrome should be raised.

INTRODUCTION

This is a case of a male university student whose most prominent symptom at presentation was diarrhoea. When their blood cultures grew *Fusobacterium necrophorum*. This prompted computed tomography (CT) imaging which demonstrated a key finding of an internal jugular vein

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smc media srl

(IJV) thrombus in keeping with Lemierre's syndrome, an infectious thrombophlebitis of the IJV associated with *F. necrophorum*[1-3]. This gastrointestinal presentation was felt to be atypical. This case serves as a useful reminder of the utility of blood cultures, this syndrome, and as an example of its atypical (or not so atypical) presentation. This case will may contribute to expanding clinicians' knowledge on this topic.

CASE DESCRIPTION

An 18-year-old male student at the local university was admitted to Acute Internal Medicine having presented to the emergency department. He had a 7-day history of diarrhoea and intermittent haematochezia and reported constant central abdominal pain and recurrent epigastric burning sensation triggered by eating. He also had a 3-day history of mild coughing with yellow sputum anda1-day history of vomiting and cold sweats. At presentation he was experiencing light headedness on standing.

Eight days prior to admission he had experienced right sided neck pain and swelling of the tonsils which had been uncomplicated and resolved spontaneously. His medical history was otherwise unremarkable, except for eczema and a longstanding right knee pain.

There was no history of, rashes, urinary symptoms, blood in the stool prior to this presentation or mucous in the stool. The patient had not had any recent ill contacts, had not travelled abroad, did not use recreational drugs, did not smoke and did not regularly drink alcohol. He was not sexually active and there was not a family history of bowel disease.

The positive examination findings were reduced breath sounds and reduced vocal resonance in the right lung base and right submandibular lymphadenopathy and tenderness. Intra-orally, an enlarged right tonsil with mild peritonsillar erythema was seen.

In the first 24 hours of admission the patient was persistently hypotensive, tachycardic and febrile, with a temperature up to 40°C, despite broad spectrum antibiotics and fluid resuscitation. His electrocardiogram revealed sinus tachycardia. Viral swab for respiratory viruses (the standard admission testing, at this hospital, at the time, was for COVID-19, Influenza A and B and respiratory syncytial virus) was negative. Blood tests demonstrated leucocytosis (18 $\times 10^9$/l) secondary to neutrophilia (16 $\times 10^9$/l). There was a mild thrombocytopaenia (147 $\times 10^9$/l). The haemoglobin level and prothrombin time were normal. C-reactive protein was markedly raised (274 mg/l). Electrolytes, renal function and liver enzymes were normal.

Initial imaging was a chest X-ray, and this was unremarkable. A CT of the abdomen and pelvis showed consolidation in the left lung base not appreciable on the admission chest X-ray, but no intra-abdominal pathology.

By day 2 of his hospital stay, the patient had become hypoxic. *Legionella* and *Streptococcal* urine antigens, urine culture, stool culture (specifically for *Salmonella*, *Shigella*, *Campylobacter*, *Cryptosporidium* and *Giardia*), *Clostridium difficile*, glutamate dehydrogenase antigen and toxins testing were all negative. HIV testing was negative. The admission blood cultures had grown Gram negative rods.

By day 3, the organism in the positive blood cultures had been identified as *F. necrophorum*. This finding prompted a subsequent CT of the neck and chest which demonstrated a right peritonsillar abscess and an IJV thrombus, as well as small bilateral pleural effusions, bi-basal atelectasis and bilateral septic thoracic emboli. This constellation of findings was consistent with Lemierre's syndrome[1].

We are pleased to report that with a combination of antibiotic therapy, low molecular weight heparin and dexamethasone the patient's condition improved. There was an attempt at drainage of the peritonsillar abscess by otolaryngology colleagues, but this was unsuccessful. The patient had a 3 month follow up CT of the neck and chest which demonstrated resolution of the previous findings, and at 8 months he had elective prophylactic bilateral tonsillectomy.

DISCUSSION

Lemierre's syndrome is characterized by a septicaemia of anaerobic organisms that spreads to the IJV from tonsillar or peritonsillar abscesses[1-3]. This causes thrombophlebitis of the IJV and secondary septic emboli, most frequently to the lungs. It typically affects young adults, and the classical symptoms and signs are thought to be: fever, sore throat, submandibular lymphadenopathy, dyspnoea and haemoptysis[1]. The causative organism is most commonly the Gram negative *F. necrophorum*. Lemierre's syndrome has potentially life-threatening consequences, with a mortality rate of 4-12%[1-3]. Therefore, early diagnosis is crucial to allow early treatment with appropriate intravenous antibiotics and surgery, if required, to address the underlying tonsillar pathology. It is worth noting that anticoagulation and vessel recanalization is not known to be of benefit.

This patient presented due to gastrointestinal symptoms the most prominent of which was diarrhoea. This provided a wide differential diagnosis. It was the blood culture results that prompted further imaging leading to the final diagnosis and onward management. Therefore, this case is educationally useful in several ways. Firstly, it highlights the diagnostic utility of blood cultures in the septic patient, a vital first line test which should not be overlooked and often confirms or clinches a diagnosis. Secondly it prompted exploration of the literature and broadened our personal understanding of this syndrome. There are reports of Lemierre's syndrome presenting with abdominal symptoms. In those patients with abdominal presentations there was typically had a direct link to intra-abdominal septic emboli, thrombi or abscesses[4]. We found no such pathology in our patient (within the limits of the contrast enhanced CT imaging that was undertaken). There are also reports of a suggested gastrointestinal variant of Lemierre's syndrome, but this is associated with *Fusobacterium nucleatum* and portal vein thrombus, neither of which were present in this case[5].

There is a longitudinal retrospective Swedish study of invasive *F. necorphorum* which provides information on characteristics seen in Lemierre's syndrome[3]. The study suggests that gastrointestinal features may be commoner than is generally understood[3]: with abdominal pain, vomiting and diarrhoea present in approximately 44, 38 and 19% of the cases respectively[3]. This makes abdominal pain, vomiting and diarrhoea the third, fourth and fifth most common symptom after fever and dyspnoea. These features correlate nicely with the case history of our patient. However, this was not in keeping with our own pre-existing knowledge at the time and we suggest is not in keeping with clinical teams' understanding of the presentation of this syndrome. Therefore, this case serves to highlight gastrointestinal symptoms, as described in the Swedish study, as a feature of Lemierre's syndrome.

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