

2022

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### Recommended Citation

Qureshi, Anum; Bailey, Theodore; Atkinson, Michael; and Tayabali, Khadija (2022) "Atypical Lemierre's syndrome. A case report and review of Literature," *Journal of Community Hospital Internal Medicine Perspectives*: Vol. 12: Iss. 1, Article 7.

DOI: 10.55729/2000-9666.1006

Available at: <https://scholarlycommons.gbmc.org/jchimp/vol12/iss1/7>

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# Atypical Lemierre's Syndrome. A Case Report and Review of Literature

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## Abstract

Lemierre's syndrome (LS) is a rare disease entity, which can be catastrophic if organism-directed treatment is not initiated early. Lemierre's syndrome is frequently caused by *Fusobacterium* infection which is frequently susceptible to clindamycin. Evidence suggests there is an increase in the incidence of cases of drug resistant *Fusobacterium* species. Through this case we present a unique case of a 45-year-old Caucasian female with Lemierre's Syndrome due to polymicrobial organisms that were resistant to clindamycin thus developing recurrent infections despite being on antibiotics.

**Keywords:** Lemierre's Syndrome, *Fusobacterium necrophorum*, *Prevotella Bucca*, Meropenem, Ertapenem, Internal jugular vein thrombosis

## 1. Introduction

Lemierre's syndrome (LS) is a rare disease entity caused by bacterial oropharyngeal infection with the constellation of acute tonsillopharyngitis often leading to thrombophlebitis of the internal jugular vein and pulmonary septic emboli as the commonest presentation of disseminated infection. The first case of Lemierre's Syndrome was described in 1900 and is known to be a life-threatening disease in the absence of early treatment. *Fusobacterium necrophorum*, an anaerobic gram-negative bacillus is the most common causative organism that causes necrobacillosis in 90% of cases, however other less reported causative organisms in the literature include *Bacteroides* and *Eikenella Genera*.

We are reporting a case of LS with an initial classic presentation that later went on to develop severe septic complications manifesting in form of lung abscess and native valve endocarditis postulated to be due to resistance to Clindamycin, typically the agent of choice for oral anaerobes.

## 2. Case presentation

45-year-old Caucasian female patient with past medical history of thyroid cancer status post

thyroidectomy who presented to emergency department with fever (Tmax 103.7 F), shortness of breath and midsternal chest pain. Her symptoms was positive for fever, chills, sore throat, light-headedness, loss of appetite, malaise, shortness of breath and chest pain. Of note, 2 weeks prior to admission she was treated for a sore throat and tender lymphadenopathy with Azithromycin, and steroids which was later switched to oral clindamycin due to no improvement in her symptoms. This was then stopped due to GI side effects in 3 days.

On admission, she was septic, febrile (39.4 C) tachycardic with a heart rate of 137 and tachypneic with RR 37 and hypotensive with BP 77/44 and required pressors briefly. She was hypoxic to 86% on room air. Physical Exam was notable for the patient being in respiratory distress, having dry mucous membranes, tender right cervical lymphadenopathy extending 10cm posteriorly, and decreased bilateral breath sounds. Heart sounds were normal with no murmurs auscultated. Pertinent lab findings included no leukocytosis but significant elevation in procalcitonin of 22.7, elevated inflammatory markers (CRP 33.4) and a lactate of 5.4. Initial COVID swap was negative. Imaging revealed bilateral pleural effusion with atelectasis, and bilateral patchy

Received 17 May 2021; revised 17 October 2021; accepted 25 October 2021.  
Available online 31 January 2022

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<https://doi.org/10.55729/2000-9666.1006>

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densities on CXR. Our differential ranged from community acquired pneumonia, COVID 19 pneumonia, pulmonary embolism, pericarditis, pulmonary edema, upper airway obstruction or fungal pneumonia. Blood cultures were drawn, and the patient was initially started on IV ceftriaxone, metronidazole, and azithromycin for severe sepsis.

CT neck without contrast (contrast avoided due to elevated creatinine) obtained was not suggestive of any abscess or thrombosis. However, due to the persistence of tender lymphadenopathy, upon improvement of creatinine CT, neck with contrast was obtained which showed marked right neck soft tissue infection with extensive phlegmon, adenopathy, abscess and septic thrombosis of the right internal jugular vein, subclavian and left cephalic vein thrombosis befitting criteria for Lemierre's syndrome prompting initiation of anticoagulation. Her initial blood culture data from the day of admission grew *Streptococcus Viridans*, *Fusobacterium*, and *Peptostreptococcus*, and antibiotics were switched to clindamycin to offer anaerobic and anti-toxin coverage. However when she underwent US guided aspiration of the right neck fluid cultures grew *Prevotella Buccae* and *Streptococcus Constellatus* both of which were resistant to Clindamycin. Prompting change from Clindamycin to meropenem. Despite this change, patient was still spiking fevers which prompted a transesophageal echocardiogram revealing an aortic valve endocarditis which is a rare secondary complication of Lemierre's syndrome. Her hospital course was then complicated by development of bilateral loculated pleural effusions with considerable compressive atelectasis requiring tPA dornase and was eventually discharged home on IV meropenem via PICC line.

While on IV meropenem, she returned 6 weeks later with complains of shortness of breath, foul smelling productive cough, pleuritic chest pain and hemoptysis and found to have a new large left lower lobe abscess concerning for empyema that required drainage. Meropenem was switched to ertapenem for feasibility and easy dosing and two weeks into her hospital stay, after significant clinical and radiologic improvement and with adequate drainage achieved her chest tube was successfully removed and was eventually discharged home back on IV Ertapenem with infectious disease follow up.

### 3. Discussion

Lemierre's Syndrome (LS) is a triad of oropharyngeal infection, thrombosis of internal jugular vein and bacteremia due to *F. necrophorum*

(commonest anaerobe) but other aerobic pathogens like *E. coli*, *Staphylococcus* and *Streptococcus* have also been implicated as causative agents<sup>1</sup>

*F. necrophorum* is a gram-negative bacillus found as normal flora in oropharynx of healthy adults. It generates liposaccharide endotoxin, virulence factor which leads to abscess formation.<sup>2</sup> In pharyngitis this pathogen penetrates through the oropharyngeal tissue to the vascular structures and leads to thrombosis, thrombophlebitis, septic emboli, and metastasis.<sup>1</sup>

Symptoms initially present as sore throat and pharyngitis in 85% cases, and otitis media and orthodontal infection in <2%.<sup>3</sup> In a study of 38 cases of LS done by Sinave et al., 97% of cases had pulmonary involvement, whereas the incidence of septic arthritis and osteomyelitis was 13% and only 2.6% of patients presented with findings of meningitis.<sup>4</sup> Similarly, Leugers and Clover study of 39 patients found all patients with F bacterium as a causative agent, remote area of involvement most commonly pulmonary lesions with an incidence of 85%, 26% with septic arthritis and osteomyelitis and only 2.5% of cases with meningitis.<sup>5</sup> Roiden et al. in his study also mentioned remote areas of involvement in descending orders pulmonary 92%, septic arthritis 11%, osteomyelitis 5% and meningitis 1%.<sup>6</sup>

Infective endocarditis is an uncommon complication seen with anaerobic bacteremia with reported incidence ranging from 1 to 16%.<sup>1</sup> Incidence of infective endocarditis is frequently seen with *Bacteroides Fragilis* and *Streptococci* but extremely rare with *Fusobacterium* as a causative agent.<sup>7</sup> A review of 8 patients with endocarditis due to *F. necrophorum* done by Samant et al. demonstrated only 2/8 having underlying heart disease and an equal distribution in involvement of native mitral and aortic valve endocarditis.<sup>8</sup>

Diagnosis of Lemierre Syndrome is usually clinical supported by imaging, preferably CT/MRI neck with contrast to identify abscesses and potentially look for thrombosis of the neck vessels.<sup>9</sup> In the pre-antibiotic era, LS was associated with mortality rate of 32–90%, and the incidence of embolic events was 25%, 12.5% specifically for endocarditis. However, with the advent of antibiotics which is the definitive management, the mortality rate significantly reduced to 17% albeit still a notable number. Antibiotic of choice is normally penicillin, but due to high resistance beta lactams/beta lactamase inhibitors with addition of metronidazole or clindamycin for anaerobic coverage is preferred<sup>9</sup>

Our patient unfortunately had a polymicrobial bacteremic infection (*Streptococcus Viridans*, *Fusobacterium*, *Peptostreptococcus* susceptible to clindamycin) but then culture data from the neck

abscess growing different anaerobes (*Prevotella Buccae*, *Streptococcus Constellatus*) the latter of which was resistant to clindamycin supporting our hypothesis that this is what led to the development of endocarditis and recurrent pulmonary involvement. Badr et al. in a study done on Clindamycin susceptibility to different gram-negative anaerobes showed *Bacteroids* 65%, *Bacteroids fragilis* 81%, *Fusobacterium* 100%, *Prevotella* 84.3%, *Porphyromonas* 80%, *Veillonella* species 95% covered by clindamycin.<sup>10</sup> Overall evidence suggests that Clindamycin should suffice, but our patient went on to develop several septic complications despite appropriate coverage.

The role of anticoagulation in Lemierre's syndrome is controversial since no clinical trials have been done to support this. A study by Ls. Bondy et al. in 2008 showed that role of anticoagulation in LS is unclear,<sup>11</sup> but Ridgway et al. supported the use of anticoagulation<sup>12</sup> even though it increases the risk of bleeding. Ultimately the decision to initiate anticoagulation is therefore variable and based on a clinician's experience and preference rather than supported by evidence.

Lastly it is important to note that despite antibiotics being the cornerstone of management, surgical drainage of neck and pulmonary abscesses results in robust recovery and shortens duration of antibiotics thus reducing unexpected toxicities.

#### 4. Conclusion

Lemierre's Syndrome leads to septic thrombophlebitis of neck veins and multiorgan organ involvement involving lungs, liver, kidney, and brain. It is a life-threatening condition and delay in treatment is associated with worse prognosis. This case highlights how early inadequate treatment resulted in more complicated and deadly disease leading to multidrug resistant – did this lead to x drug resistant or were the organisms already resistant to begin with? Organism infections and requiring higher up therapy and treatment- what does this mean?? We also suggest that further research on modernizing treatment protocols of Lemierre disease. Role of anticoagulation is not

evidence based as well, and more randomized trial are needed for supporting the use of anticoagulation in thrombophlebitis.

#### Conflict of interest

No potential conflict of interest was reported.

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