

Lemierre's Syndrome: A Case Report

Hannah Doan¹, Sara Niyazi¹, April Burton¹, Ikramamul Nibir¹, Augustas Kavaliauskas²

1. Medicine, Lake Erie College of Osteopathic Medicine, Bradenton, USA 2. Internal Medicine, AdventHealth Florida, Sebring, USA

Corresponding author: Sara Niyazi, sniyazi49582@med.lecom.edu

Abstract

Lemierre's syndrome is a condition in which an oropharyngeal infection progresses to sepsis and thrombophlebitis of the internal jugular vein. Although the incidence of this syndrome has fallen dramatically since the widespread use of antibiotic therapy to treat streptococcal pharyngitis, it should still be suspected in otherwise healthy young patients presenting with the triad of prolonged pharyngitis, lateral neck pain, and septic symptoms. In this report, we explore a unique case of Lemierre's syndrome complicated by hypercoagulability and ineffective initial antibiotic therapy.

Categories: Internal Medicine, Obstetrics/Gynecology, Infectious Disease

Keywords: lemierre's syndrome, septic thrombophlebitis, necrobacillosis, jugular vein suppurative thrombophlebitis, post-anginal sepsis, postanginal sepsis

Introduction

Lemierre's syndrome, also known as jugular vein suppurative thrombophlebitis, post-anginal sepsis, and necrobacillosis, was first described by Andre Lemierre in 1936 and is characterized by thrombophlebitis of the internal jugular vein with concurrent sepsis [1,2]. The highest rates of Lemierre's syndrome were seen during the pre-antibiotic era, but since the use of antibiotics to treat oropharyngeal infections, it has become a fairly rare occurrence, with recent studies estimating a rate of approximately 3.6 cases per 1 million individuals per year [3]. It is most commonly preceded by pharyngitis, but other reported events leading to Lemierre's syndrome include dental infections, tonsillitis, infectious mononucleosis, and intravenous catheter insertion [4]. Infection usually descends from its primary site in the oropharynx to the carotid sheath vessels in the lateral pharyngeal space, causing septic thrombophlebitis of the internal jugular vein and sometimes the internal carotid artery [3]. As the infection progresses, it commonly spreads from the internal jugular vein to the lung, causing cavitary lung abscesses or empyema [5]. There have also been reports of spread to the joints, liver, muscle, pericardium, brain, and skin in some cases [6].

Lemierre's syndrome is usually a polymicrobial infection, with the most commonly implicated organisms being *Fusobacterium necrophorum*, *Eikenella corrodens*, *Porphyromonas asaccharolytica*, *Streptococcus pyogenes*, and *Bacteroides* [7-9]. Treatment requires appropriate antibiotic therapy combined with surgical intervention and anticoagulation when indicated [10]. It is recommended that empiric therapy include a beta-lactamase resistant beta-lactam antibiotic, such as ampicillin-sulbactam, piperacillin-tazobactam, or meropenem [11]. In rare instances, surgical ligation or excision of the internal jugular vein may be warranted [12].

Case Presentation

A previously healthy 21-year-old Caucasian female presented to the emergency department in a hypotensive and febrile state with associated weakness and vomiting. She reported having a sore throat and fever three days prior, for which she went to urgent care and was prescribed azithromycin for bilateral tonsillitis. Over the course of three days, the azithromycin did not relieve her symptoms, and instead, they progressed to also include nausea and vomiting. On the third day, she described a sensation of feeling "something burst" in her neck, which improved her sore throat but led to the presence of blood in her sputum, prompting her visit to the emergency department.

On initial assessment, she had a fever of 104°F, a blood pressure of 79/39 mmHg, a heart rate of 134 bpm, and a swollen and tender right neck. Her laboratory evaluation showed leukopenia and thrombocytopenia, and a contrast CT scan of the neck revealed enlarged bilateral palatine tonsils and a filling defect in the right internal jugular vein (Figures 1-2). The combination of her physical exam, laboratory, and image findings pointed to the diagnosis of Lemierre's syndrome with progression to sepsis. She was promptly admitted to the critical care unit and started on vancomycin and piperacillin/tazobactam.

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FIGURE 1: Initial CT with contrast demonstrating bilateral palatine tonsil enlargement.

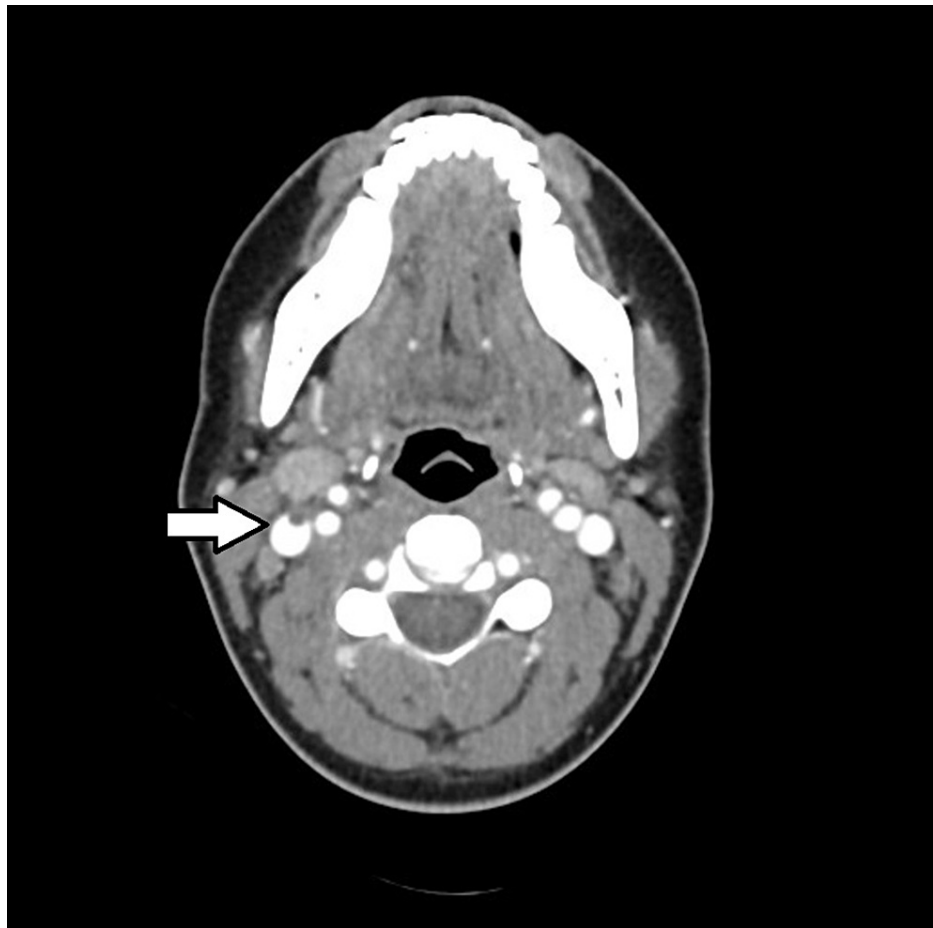


FIGURE 2: Initial CT with contrast demonstrating a filling defect in the right internal jugular vein.

On the second day of admission, her fever persisted and she developed acute hypoxic respiratory failure. A chest CT with contrast was performed and showed bilateral pleural effusion, patchy consolidations, and nodular opacities suggestive of lung abscess (Figure 3). She was then started on furosemide and IV meropenem to treat her pleural effusions and abscesses, respectively. Her fever, sore throat, and inflammation resolved in response to the antibiotics, however, her thrombus in the right internal jugular vein persisted. She was in a continual state of hypercoagulability, so the decision was made to have her Nexplanon® etonogestrel implant removed. Over the next two days, her condition started to improve with a decrease in her C-reactive protein (CRP) and an increase in her platelet count and blood pressure. At this point her throat pain, fever, and vomiting had resolved, but her right neck and ear were tender to palpation and she complained of mild chest tightness.

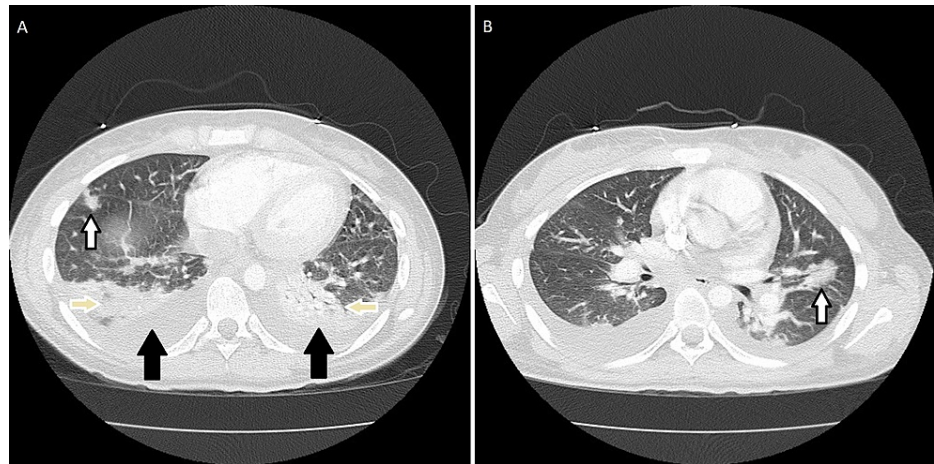


FIGURE 3: Chest CT with contrast demonstrating pleural effusion (black arrows), consolidation (yellow arrows) and nodular opacities (white arrows).

Unfortunately, her condition declined once again the following day, with recurrent fever and worsening neck pain. Repeat CT showed progression of the right internal jugular vein thrombosis into a fully occluded vessel, with wall thickening and hyperemia indicating thrombophlebitis (Figure 4). The thrombophlebitis extended inferiorly to the infrahyoid muscle and supraclavicular fat, and her temperature continued to increase from 100.7°F to 101.1°F. While the thrombophlebitis was extensive, there were no signs of neck abscess or acute odontogenic infection. At this point, heparin was started as her thrombocytopenia had improved since intake.

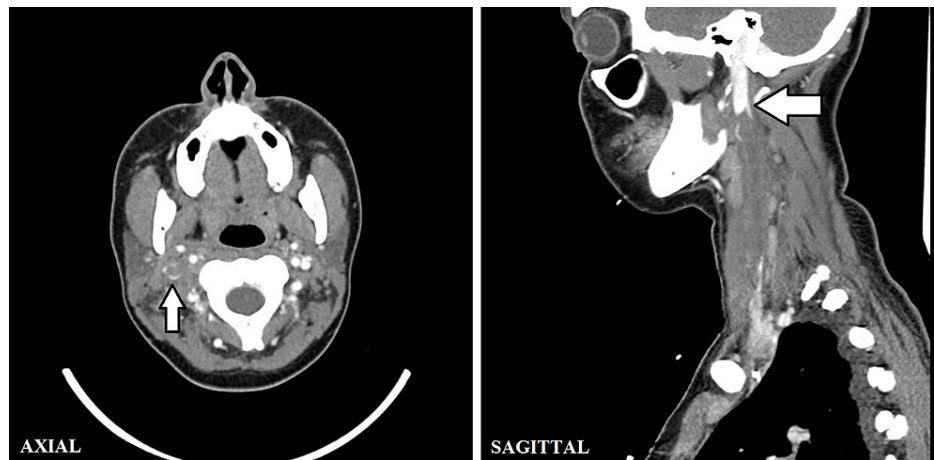


FIGURE 4: Axial and sagittal view of CT with contrast demonstrating a fully occluded right internal jugular vein (white arrows).

In an effort to prevent septic embolism, vascular surgery was consulted and a surgical ligation was planned. However, during the ligation procedure, the patient was found to have severe inflammation and purulent material within the right internal jugular vein, so drainage and a suppurative thrombectomy were performed instead of the initially planned ligation. The thrombus and purulence from the internal jugular vein were retrieved and sent for culture and pathology, which showed signs of extensive acute inflammation but no organism growth. The thrombectomy was successful, and over the next five days, the patient's condition greatly improved. She was discharged home with clopidogrel, apixaban, IV meropenem, and vancomycin, which were found to be the optimal antibiotic choices based on sensitivity testing.

Discussion

Due to the low incidence of Lemierre's syndrome, it is not commonly seen and therefore may not be considered as a complication when treating patients for oropharyngeal infections. This can potentially lead to inadequate treatment regimens, missed diagnosis, and increased disease complications, as seen in our

patient. When she arrived at urgent care she was treated with azithromycin, most likely because they did not suspect complications that could potentially result in Lemierre's syndrome. However, when a patient presents with symptoms of tonsillitis or other oropharyngeal infections, an alternative treatment to consider is penicillin, as it has been shown to provide stronger coverage against certain oropharyngeal microbes, such as *Fusobacterium necrophorum* [13].

Furthermore, a well-known risk of various birth control methods is the possibility of venous thromboembolic events and coagulopathies [14]. Isolated cases of thrombotic thrombocytopenia have also been reported in patients with progestin contraceptive implants [15]. Our patient had an indwelling progestin implant when she developed Lemierre's syndrome, which may have contributed to her rapid decline and disruption of hemostasis. On the day of implant removal, her platelet count was 49×10^3 mL, and following removal, her platelet count began to steadily increase and normalize to 257×10^3 mL four days following removal. These findings suggest a possible connection between her progestin implant and her hematologic changes.

Conclusions

Hormonal contraceptives may cause changes in hemostasis and put patients at an increased risk for thromboembolic complications. It is possible that the use of a hormonal birth control implant increased our patient's risk of thrombus formation and played a role in her disease severity. This is evidenced by the fact that, in conjunction with the use of antibiotics, removal of the birth control implant appeared to aid in hemostasis. Furthermore, this case brings attention to the urgency of initial antibiotic therapy for oropharyngeal infections and highlights the importance of choosing a regimen that provides adequate coverage for the multitude of oropharyngeal bacteria, including those with increased resistance to more commonly used antibiotics.

Additional Information

Disclosures

Human subjects: Consent was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

References

- Riordan T, Wilson M: Lemierre's syndrome: more than a historical curiosa. *Postgrad Med J*. 2004, 80:328-34. [10.1136/pgmj.2003.014274](https://doi.org/10.1136/pgmj.2003.014274)
- Eilbert W, Singla N: Lemierre's syndrome. *Int J Emerg Med*. 2013, 6:40. [10.1186/1865-1380-6-40](https://doi.org/10.1186/1865-1380-6-40)
- Hagelskjaer Kristensen L, Prag J: Lemierre's syndrome and other disseminated *Fusobacterium necrophorum* infections in Denmark: a prospective epidemiological and clinical survey. *Eur J Clin Microbiol Infect Dis*. 2008, 27:779-89. [10.1007/s10096-008-0496-4](https://doi.org/10.1007/s10096-008-0496-4)
- Sinave CP, Hardy GJ, Fardy PW: The Lemierre syndrome: suppurative thrombophlebitis of the internal jugular vein secondary to oropharyngeal infection. *Medicine (Baltimore)*. 1989, 68:85-94.
- Karkos PD, Asrani S, Karkos CD, Leong SC, Theochari EG, Alexopoulou TD, Assimakopoulos AD: Lemierre's syndrome: a systematic review. *Laryngoscope*. 2009, 119:1552-1559. [10.1002/lary.20542](https://doi.org/10.1002/lary.20542)
- Chirinos JA, Lichtstein DM, Garcia J, Tamariz LJ: The evolution of Lemierre syndrome: report of 2 cases and review of the literature. *Medicine (Baltimore)*. 2002, 81:458-465. [10.1097/00005792-200211000-00006](https://doi.org/10.1097/00005792-200211000-00006)
- Jensen A, Hagelskjaer Kristensen L, Prag J: Detection of *Fusobacterium necrophorum* subsp. *funduliforme* in tonsillitis in young adults by real-time PCR. *Clin Microbiol Infect*. 2007, 13:695-701. [10.1111/j.1469-0691.2007.01719.x](https://doi.org/10.1111/j.1469-0691.2007.01719.x)
- Celikel TH, Muthuswamy PP: Septic pulmonary emboli secondary to internal jugular vein phlebitis (postanginal sepsis) caused by *Eikenella corrodens*. *Am Rev Respir Dis*. 1984, 130:510-3. [10.1164/arrd.1984.130.3.510](https://doi.org/10.1164/arrd.1984.130.3.510)
- Morizono S, Enjoji M, Sonoda N, et al.: Lemierre's syndrome: *Porphyromonas asaccharolytica* as a putative pathogen. *Intern Med*. 2005, 44:350-3. [10.2169/internalmedicine.44.350](https://doi.org/10.2169/internalmedicine.44.350)
- Escher R, Haltmeier S, von Steiger N, Dutly AE, Arnold A, Kickuth R, Schiemann U: Advanced Lemierre syndrome requiring surgery. *Infection*. 2008, 36:495-6. [10.1007/s15010-008-8216-7](https://doi.org/10.1007/s15010-008-8216-7)
- Seidenfeld SM, Sutker WL, Luby JP: *Fusobacterium necrophorum* septicemia following oropharyngeal infection. *JAMA*. 1982, 248:1348-1350. [10.1001/jama.1982.03330110044024](https://doi.org/10.1001/jama.1982.03330110044024)
- Baker CC, Petersen SR, Sheldon GF: Septic phlebitis: a neglected disease. *Am J Surg*. 1979, 138:97-105. [10.1016/0002-9610\(79\)90248-4](https://doi.org/10.1016/0002-9610(79)90248-4)
- Christopher D, Alyson P, Katherine H: Lemierre's syndrome: the result of an incorrect penicillin allergy. *Int Med Care*. 2017, 1:1-3. [10.15761/IMC.1000106](https://doi.org/10.15761/IMC.1000106)
- Lidegaard Ø, Nielsen LH, Skovlund CW, Løkkegaard E: Venous thrombosis in users of non-oral hormonal contraception: follow-up study, Denmark 2001-10. *BMJ*. 2012, 344:e2990. [10.1136/bmj.e2990](https://doi.org/10.1136/bmj.e2990)
- Fraser JL, Millenson M, Malynn ER, Uhl L, Kruskall MS: Possible association between the Norplant

contraceptive system and thrombotic thrombocytopenic purpura. *Obstet Gynecol.* 1996, 87:860-865.