

Lemierre Syndrome—Who First Described It?

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Citation

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Abstract

Lemierre Syndrome is characterized by an anaerobic septic thrombus occluding the internal jugular vein with metastatic foci of infections. *Fusobacterium necrophorum* is the commonest infecting organism. People with Lemierre Syndromes often presented with chest infections. The oropharyngeal infections are easily missed if unaware. The following three cases illustrate the diagnostic difficulties of Lemierre Syndrome. The history of the Lemierre Syndrome will be discussed and explored. Reynolds SC, Chow AW, *Life-threatening Infections of the Peripharyngeal and Deep Facial Space of the Head and Neck, Infect Dis Clin N Am.* 2007;21:557-76

CASE 1

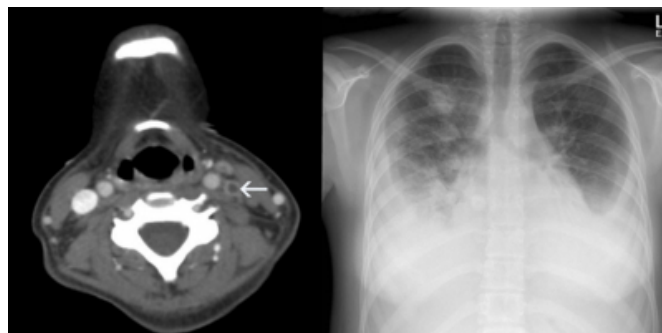
A 16-year-old female presented to the emergency department with right lower lobe pneumonia. She had recurrent pharyngitis and tonsillitis for 4 weeks that required multiple courses of oral antibiotics. She was admitted to the general medical ward for intravenous antibiotics.

She did not respond to antibiotic treatment and continued to have rigor and became febrile with tachycardia. She did not have leukocytosis or neutrophilia, but her C-reactive protein was 260 U/L with a platelet count of 11×10^9 /L.

Her situation deteriorated and she became supplementary oxygen dependent. Her blood culture performed on admission showed gram-negative bacilli. On day six of the admission, it was found that the consolidation was minimal after reviewing the initial chest X-ray. The primary infectious focus was directed to her tonsils. Computed Tomography (CT) of the neck and chest was performed, and it showed an abscess surrounding a thrombosed left internal jugular vein and multiple pulmonary abscesses with empyema (Fig. 1). Blood culture examination confirmed *Fusobacterium necrophorum* (1 of the 2 bottles in 24.2 hours).

Figure 1

Fig. 1. a) Left internal jugular vein thrombosis surrounded with abscess (arrow) and b) multiple pulmonary abscesses with bibasal empyema



Her intravenous antibiotic was changed to penicillin-G, which was guided by the culture sensitivity and verified by the minimal inhibitory concentration (MIC) test.

Anticoagulation was not commenced due to thrombocytopenia. Her neck abscess was explored and drained via a submandibular incision (Fig. 2). The internal jugular vein was not ligated.

Figure 2

Fig. 2. An intraoperative photo showing the drainage of purulent materials.



She improved clinically postoperatively. She was transferred to the ward and subsequently discharged with intravenous antibiotic and enoxaparin for 3 weeks.

CASE 2

A 46-year-old female presented to the emergency department with a 1-week history of right neck pain, severe right-sided headache, and general fatigue. She had leukocytosis, neutrophilia, and thrombocytopenia. Chest X-ray showed right lower lobe pneumonia. CT of the head was normal. Ultrasound of her neck showed right internal jugular vein thrombosis. Anticoagulation was not started due to the thrombocytopenia.

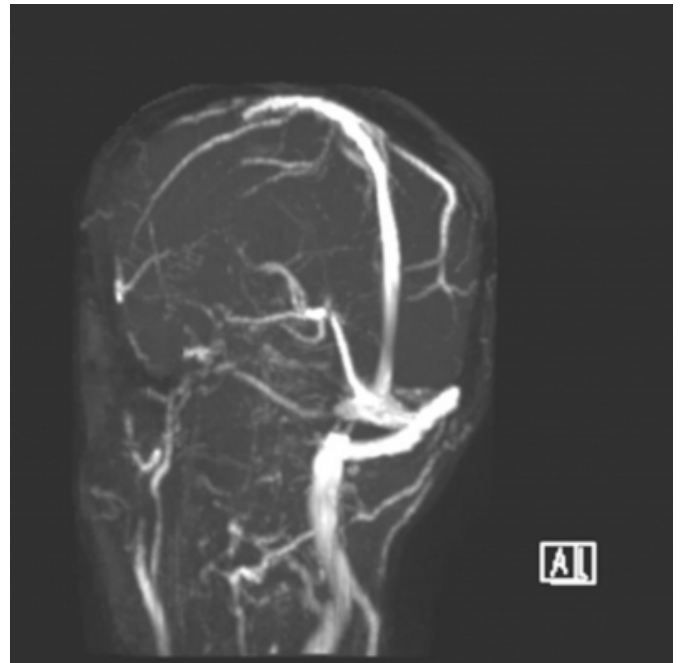
Her condition did not improve with antibiotic treatment. A concern of pulmonary emboli was raised. CT pulmonary angiography was negative for emboli, but showed complete right internal jugular vein thrombosis with surrounding thrombophlebitis. The blood culture performed on admission showed *Fusobacterium necrophorum* in 1 of the 2 bottles at 20.4 hours. Lemierre syndrome and an oropharyngeal source of infection were suspected. An orthopantomogram was performed, which revealed dental abscess. The tooth was removed and the antibiotic was changed to penicillin-G, which was guided by the culture sensitivity and verified by the MIC test.

Her condition, however, deteriorated with increasing confusion. A magnetic resonance imaging scan showed that the thrombus extended to the right sigmoid sinus up to the level of the transverse sinus (Fig. 3). Warfarin was initiated.

She responded to antibiotics and anticoagulation treatment slowly, and she was discharged with oral antibiotics and warfarin on day 10.

Figure 3

Fig. 3. Magnetic resonance reconstruction showing complete obliteration of the right sigmoid sinus.



HISTORY

Lemierre syndrome was named after André Alfred Lemierre (1875–1956), a professor of bacteriology in Paris, ¹ who originally described the disease as peritonsillar abscess causing pulmonary infarcts and septic arthritis in 1936. He also commented that the disease was so characteristic and should constitute a syndrome.

He acknowledged that a number of people had described the concept of distance suppurative lesions due to septic emboli. Hugo Schottmüller (1867–1936) was given credit as the first person to describe such a condition. However, Lemierre did not cite Schottmüller's article. Since then, a number of journal articles claimed Schottmüller as the first person who described Lemierre syndrome in 1918.²

The timing of this German publication was unusual since it was the year that WWI finished. We verified with the publishers, and they confirmed that there was only an entry of the announcement in the journal. Hugo Schottmüller, however, as a founder of the concept of sepsis, described the concept of distance metastasis of infection from an oral source to organs at a distance in 1922.³ Lemierre accepted the hypothesis and postulated a possible mechanism of the

syndrome that he described as “a thrombophlebitis of the tonsillar and peritonsillar veins which can spread to the internal jugular vein.” He acknowledged that the concept was from the work of E. Frankel in 1919, but did not cite that article as well. Therefore, it is a fair statement to say that there were a number of people who contributed in the establishment of Lemierre syndrome, while Lemierre was the one who first pulled all the pieces of puzzle together and suggested suppurative thrombophlebitis of an internal jugular vein from an oropharyngeal infection causing distance septic emboli constitute a syndrome.

CONCLUSION

Two cases of Lemierre syndromes were presented as a series. The difficulties in early diagnosis lead to morbidity and sometimes mortality. Patients who are otherwise healthy

and presented with severe bilateral pneumonia and pleural effusions raise concerns of systemic septic emboli such as Lemierre syndrome. This case series also clarifies the origin of Lemierre syndrome.

ACKNOWLEDGEMENTS

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References

1. Lemierre A., On certain septicemia due to anaerobic organisms. *Lancet*. 1936; 1:701-703
2. The article that did not exist: Schottmüller H, “Ueber die Pathogenität anaërober Bazillen.” *Dtsch Med Wochenschr*. 1918; 44:1440
3. Schottmüller H, Ueber den angeblichen Zusammenhang zwischen Infektionen der Zähne und Allgemeinerkrankungen. *Dtsch med Wochenschr*. 1922; 48: 181-182

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