



## Ludwig Angina and Lemierre's Syndrome: A Rare Co-presentation

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**Running Head:** Ludwig Angina and Lemierre's Syndrome: A Rare Co-presentation

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### Abstract

This case report highlights a unique co-presentation of two rare infections, Lemierre's syndrome and Ludwig angina in a patient, caused by a polymicrobial infection involving unusual pathogens, *Streptococcus anginosus* and *Candida dubliniensis*. Ludwig angina is a severe submandibular infection known for its rapid spread, posing a risk of airway obstruction due to edema. On the other hand, Lemierre's syndrome typically arises as a complication of a peritonsillar abscess, wherein the infection extends into the parapharyngeal and carotid spaces and even reaches the internal jugular vein. This case report describes a 50-year-old white male patient who presented with left submandibular swelling, extending to the anterior neck, lower left facial droop, and severe trismus. A neck CT scan revealed substantial dental caries, thrombophlebitis, and multiple submental abscesses. The patient underwent three incision and drainage procedures for the abscesses, excision, and ligation of the left internal jugular vein and common facial vein, as well as dental tooth extractions. Microbial cultures from the abscesses identified *Streptococcus anginosus* and *Candida dubliniensis*, while blood cultures revealed *Prevotella buccae*. Notably, *Fusobacterium necrophorum*, the typical causative agent of Ludwig angina, was not isolated. Treatment involved a six-week course of ceftriaxone, metronidazole, and micafungin, followed by six months of fluconazole. The patient showed improvement in trismus, neck swelling, and pain; however, he continued to exhibit a lower left facial droop. This case highlights important clinical insights, emphasizing the importance of promptly recognizing Ludwig's angina and its potential progression to Lemierre's syndrome.

### Introduction

Ludwig angina and Lemierre's syndrome are two serious infections that can affect the head and neck, but they generally develop separately from each other. Ludwig angina is a rapidly progressing infection that primarily affects the sublingual and submandibular spaces. It typically shows up as a severe infection causing tissue death in the floor of the mouth, accompanied by swelling under the tongue, which can potentially block the airway and pose a serious threat. On the other hand, Lemierre's syndrome typically presents as a condition where a blood clot forms in the internal jugular vein due to an infection, most caused by *Fusobacterium necrophorum* originating from an oral cavity infection or a peritonsillar abscess.

### Case Report

A 50-year-old Caucasian man presented to the emergency department with inability to open his mouth, left facial weakness,

a swollen neck, and associated facial pain. His past medical history was only significant for hypertension. He hadn't visited the dentist for over a decade and had poor dental hygiene. Two weeks prior, he had been diagnosed with a dental abscess at an outpatient clinic and was initially prescribed ciprofloxacin. However, after five days, his condition worsened, and he returned to the clinic with increasing neck swelling and left-sided facial weakness. Then, His primary care physician switched his antibiotic to clindamycin. Three days later, the patient presented at our emergency department with severe swelling on the left side of his neck, extending to the front without experiencing stridor. CT neck with contrast showed large multiloculated abscesses in the left sublingual, submental, submandibular, parapharyngeal, and carotid spaces with significant mass effect, as well as a thrombus in the left internal jugular vein and multiple punctate foci of air consistent with septic thrombophlebitis (Figure A).



**Figure A: CT neck with contrast showing left internal jugular vein thrombus with multiple punctate foci of air compatible with septic thrombophlebitis.**

He was taken urgently to the operating room for incision and drainage of his various abscesses by the otolaryngology team. Intraoperative findings included purulent fluid encountered both superficial and deep to the superficial investing fascia of the neck and in the carotid sheath. Vascular surgery was consulted for the management of septic thrombophlebitis of the left internal jugular vein. The internal jugular vein was ligated proximally and distally and excised, including the common facial vein. Surgical drains were placed, and the final wound measured 10 x 8 x 6 cm. Following surgery, the patient was admitted to the intensive care unit (ICU) for airway monitoring. The panorex revealed large carious lesions and retained root tips with the periapical disease of the lower left side of the patient's mouth. He was later evaluated by a dentist who performed tooth extractions during his hospitalization. Abscess cultures grew *Streptococcus anginosus* and *Candida dubliniensis*. One of the two blood cultures was positive for *Prevotella buccae*. He was treated with six weeks of ceftriaxone, metronidazole, and micafungin followed by six months of fluconazole. Four weeks after his initial presentation, the wound was healing well but the patient had persistent left facial droop and loss of sensation.

## Discussion

A thorough literature search on PubMed in October 2023 for cases of "Lemierre's syndrome and Ludwig angina" revealed only one case report.<sup>1</sup> Ludwig angina and Lemierre's syndrome

are both life-threatening head and neck infections, often triggered by dental infections. Approximately, 70% of Ludwig angina cases arise from the spread of infection from molars. Other potential causes include oral injuries or fractures in the jawbone.<sup>2,3</sup> *Streptococcus*, *Staphylococcus*, and *Bacteroides* are commonly associated with Ludwig angina, although less common species like *Klebsiella* and *Pseudomonas* may also be implicated. Diagnosis of Ludwig angina is typically based on clinical assessment, but it can be complemented by imaging methods such as CT, MRI, or neck ultrasound. The main priority in Ludwig angina treatment is to secure the patient's airway.<sup>4</sup> Subsequently, broad-spectrum intravenous antibiotics must be administered to cover both gram-positive and gram-negative bacteria, as well as anaerobes. Antibiotic coverage should also include *Pseudomonas* and methicillin-resistant *Staphylococcus aureus* (MRSA), especially for immunocompromised patients.<sup>5,6</sup> If the patient develops fluid collections, surgical procedures like aspiration, or incision and drainage need to be considered in a timely manner. For cases of suspected Lemierre's syndrome, further diagnostic imaging, such as a CT scan or MRI of the neck is crucial to determine the extent of internal jugular vein involvement. Decisions regarding conservative vs surgical interventions depend on several factors such as airway stability, abscess formation, and clinical improvement after broad-spectrum

antibiotics.<sup>7,8</sup> While anticoagulation is typically used, the evidence for its effectiveness is mixed in the literature.

The causative agents for these two serious conditions often differ. *Fusobacterium necrophorum*, an anaerobic gram-negative bacterium commonly found in the mouth, is the most reported cause of Lemierre's syndrome. For Ludwig angina, strep, and staph species are more frequently involved.<sup>1-4</sup> Notably, in our patient's case, *Streptococcus anginosus*, *Candida dubliniensis*, and *Prevotella buccae* were isolated from blood cultures, which is unusual for both Lemierre's syndrome and Ludwig angina. This led to tailoring his treatment regimen including ceftriaxone, metronidazole, and micafungin. The simultaneous presentation of Ludwig angina and Lemierre's syndrome is quite unique. It is worth remembering that both infections can progress rapidly due to the abundance of lymphatic, vascular, and neural structures deep within the superficial investing fascia of the neck. The infection initially in the submandibular space (Ludwig angina) may have spread through the parapharyngeal space, connecting with the lateral pharyngeal, retropharyngeal spaces, and the areas around the carotid sheath, eventually leading to thrombophlebitis of the internal jugular vein (Lemierre's syndrome).

## Conclusion

This exceptional case highlights a critical point: when Ludwig angina is not promptly treated, it has the potential to progress into Lemierre's syndrome. Clinicians must remain vigilant about rare yet life-threatening infections, as any delay in diagnosis and appropriate management can lead to severe complications, including airway obstruction or, in extreme cases, death.

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