Case Report

Lemierre Syndrome Presenting as Unilateral Neck Swelling with Coexisting Thyroiditis: A Rare Case Report

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Abstract

Lemierre syndrome (LS) is a rare but potentially life-threatening complication of oropharyngeal infection characterised by septic thrombophlebitis of the internal jugular vein, with or without metastatic septic embolisation. We report the case of a 54-year-old woman who presented with fever, painful left-sided neck swelling, and neck pain following a recent peritonsillar infection. Ultrasonography demonstrated a partially occlusive thrombus within the left internal jugular vein and diffuse thyroid enlargement with heterogeneous echotexture and increased vascularity, suggestive of coexisting thyroiditis. Contrast-enhanced computed tomography confirmed internal jugular vein thrombosis without deep neck abscess, mediastinal extension, or septic pulmonary emboli. A clinicoradiological diagnosis of LS was established, and the patient responded favourably to intravenous ceftriaxone without anticoagulation. This case highlights an uncommon presentation of LS with coexisting thyroiditis and emphasises the importance of early multimodality imaging and prompt antimicrobial therapy to prevent potentially life-threatening complications.

Keywords: Lemierre syndrome, Internal jugular vein thrombosis, Peritonsillar infection, Thyroiditis, Neck ultrasonography, Contrast-enhanced computed tomography

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Introduction

Lemierre syndrome (LS) is a rare but potentially life-threatening form of septic thrombophlebitis of the internal jugular vein (IJV) that typically develops as a complication of an oropharyngeal infection.[1] First comprehensively described by André Lemierre in 1936 as "post-anginal septicemia," the syndrome classically consists of a recent pharyngeal infection, IJV thrombosis, anaerobic bacteremia, and metastatic septic emboli, most frequently involving the lungs.[2] Fusobacterium necrophorum, an anaerobic Gram-negative bacillus that forms part of the normal oropharyngeal flora, remains the predominant causative organism, although other aerobic and anaerobic pathogens have also been implicated.[3,4]

Although the widespread availability of antibiotics markedly reduced its incidence during the latter half of the twentieth century, LS has re-emerged over recent decades, emphasising the need for continued clinical awareness.[5]

The pathogenesis involves extension of infection from the tonsillar or peritonsillar tissues into the parapharyngeal space, resulting in septic thrombophlebitis of the IJV with the potential for hematogenous dissemination.[1] Septic pulmonary emboli represent the most common metastatic complication and are reported in the majority of patients, whereas dissemination to joints, liver, bones, and the central nervous system occurs less frequently.[5]

Because the initial symptoms often resemble uncomplicated pharyngitis or tonsillitis, diagnosis may be delayed until thrombosis or septic embolisation becomes clinically evident. Contrast-enhanced computed tomography of the neck is considered the imaging modality of choice for confirming IJV thrombosis and delineating disease extent, while ultrasonography serves as an important first-line investigation because of its rapid availability, non-invasive nature, and ability to evaluate cervical venous flow.[6]

Thyroid involvement in LS is distinctly uncommon, and the coexistence of thyroiditis with IJV thrombosis has rarely been described. Furthermore, not all patients develop septic pulmonary emboli, making recognition of localised disease particularly challenging. Early diagnosis based on careful clinical evaluation and appropriate imaging allows timely initiation of antimicrobial therapy and may prevent life-threatening complications.[7]

We report a rare case of LS presenting after a peritonsillar infection with isolated left IJV thrombosis and coexisting thyroiditis in the absence of septic pulmonary emboli, highlighting the importance of maintaining a high index of suspicion even in atypical presentations.

Case Presentation

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A 54-year-old woman presented to the emergency department with a 4- to 5-day history of fever, progressive left-sided neck swelling, and neck pain following a recent episode of peritonsillar infection. On physical examination, she was febrile and had diffuse swelling with tenderness over the left side of the neck, extending along the expected course of the left IJV. Oropharyngeal examination was consistent with a resolving peritonsillar infection. No neurological deficits or signs of airway compromise were identified.

Initial laboratory investigations demonstrated leukocytosis, with a total leukocyte count of 14,000/mm3 (reference range: 4,000-11,000/mm3), consistent with an acute inflammatory process. Neck ultrasonography was performed as the initial imaging modality. Gray-scale imaging demonstrated an echogenic intraluminal thrombus within the left IJV causing partial luminal occlusion [Figure 1].

Color Doppler examination demonstrated residual blood flow surrounding the thrombus, consistent with a partially occlusive IJV thrombosis [Figure 2].

Ultrasonography of the thyroid gland demonstrated diffuse enlargement with heterogeneous echotexture and increased vascularity, suggestive of coexisting thyroiditis [Figure 3].

Multiple mildly enlarged cervical lymph nodes were identified at levels IB, II, and III with preserved fatty hila, consistent with reactive lymphadenopathy.

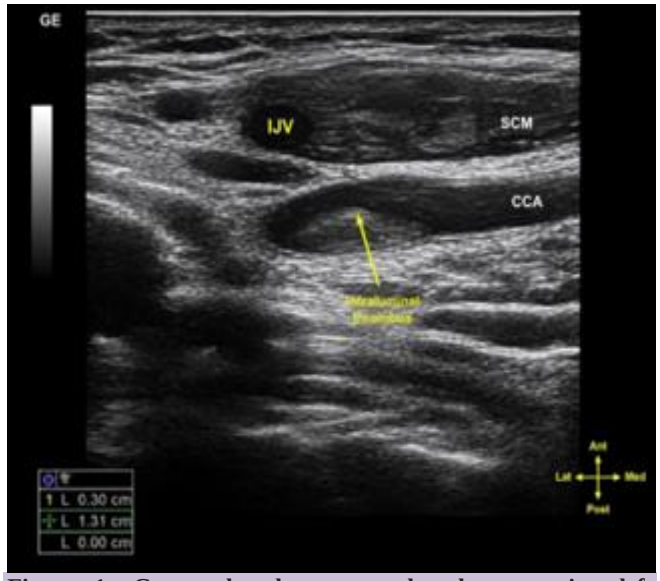

Figure 1: Gray-scale ultrasonography demonstrating left internal jugular vein thrombus. Gray-scale ultrasonography of the left neck demonstrating an echogenic intraluminal thrombus (arrow) causing partial luminal occlusion of the left internal jugular vein.

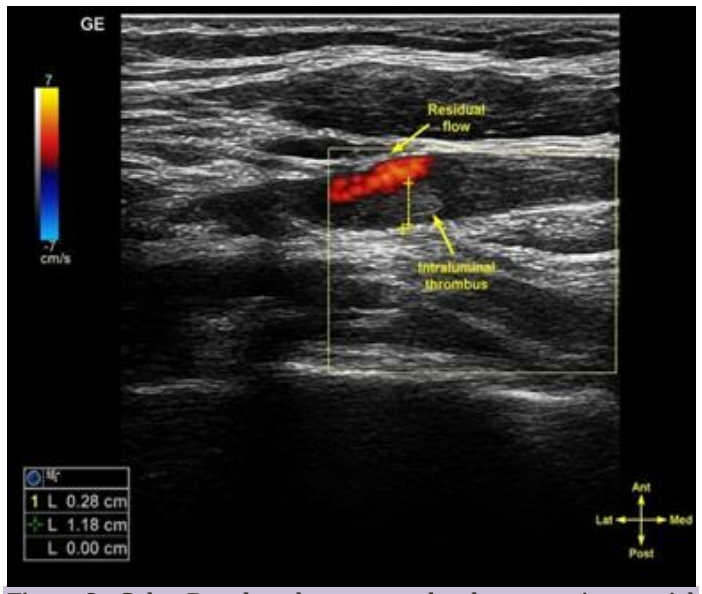

Figure 2: Color Doppler ultrasonography demonstrating partial recanalization of the left internal jugular vein. Color Doppler ultrasonography demonstrating residual blood flow surrounding the intraluminal thrombus (arrow), consistent with a partially occlusive left internal jugular vein thrombosis.

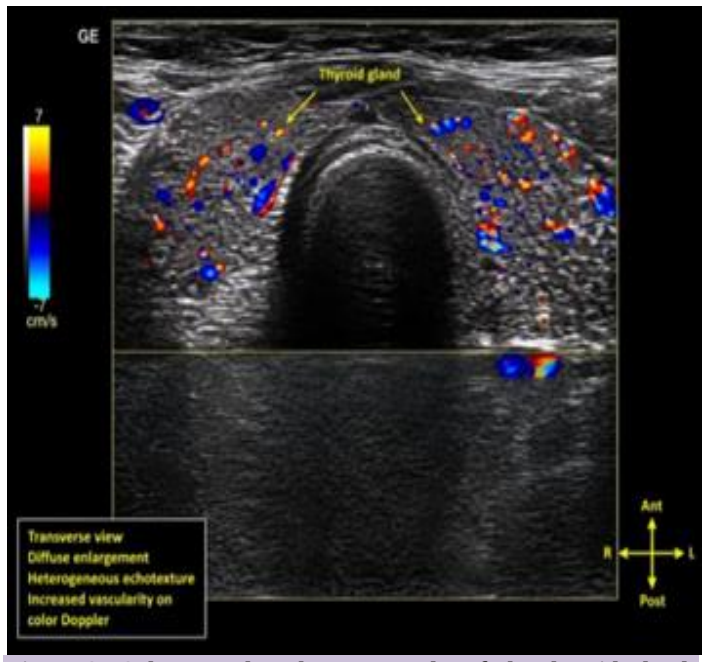

Figure 3: Color Doppler ultrasonography of the thyroid gland. Color Doppler ultrasonography demonstrating diffuse enlargement of the thyroid gland with heterogeneous echotexture and increased vascularity, suggestive of coexisting thyroiditis.

Contrast-enhanced computed tomography (CT) of the neck was subsequently performed to further delineate the extent of venous involvement. CT demonstrated a non-enhancing intraluminal filling defect within the left IJV, confirming IJV thrombosis [Figure 4]. No drainable deep neck abscess or mediastinal extension was identified. Contrast-enhanced CT of the chest demonstrated no evidence of septic pulmonary emboli or other thoracic septic complications.

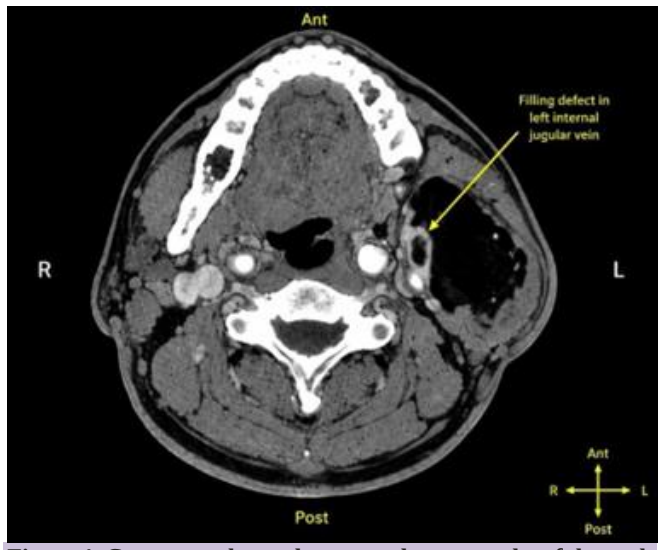

Figure 4: Contrast-enhanced computed tomography of the neck showing left internal jugular vein thrombosis. Axial contrast-enhanced computed tomography image demonstrating a non-enhancing intraluminal filling defect within the left internal jugular vein (arrow), consistent with internal jugular vein thrombosis.

Based on the recent history of peritonsillar infection together with the characteristic clinical and radiological findings, a diagnosis of LS with isolated left IJV thrombosis and imaging findings suggestive of coexisting thyroiditis was established.

The patient was treated with intravenous ceftriaxone (2 g once daily). Supportive management, including intravenous fluids and analgesics, was provided as required. Blood culture results and additional microbiological investigations were unavailable. Anticoagulation was not initiated because there was no radiological evidence of thrombus progression or metastatic septic complications.

The patient demonstrated gradual clinical improvement, with resolution of fever and progressive reduction in neck pain and cervical swelling during hospitalisation. She remained hemodynamically stable throughout her hospital stay and developed no evidence of septic pulmonary emboli or other metastatic complications. Intravenous ceftriaxone was continued for a total duration of seven days. She was discharged after five days of hospitalisation in stable clinical condition to complete the remaining antibiotic course. Follow-up imaging and subsequent outpatient clinical data were unavailable.

Discussion

LS is a rare but potentially life-threatening complication of an oropharyngeal infection characterised by septic thrombophlebitis of the IJV, with or without metastatic septic embolisation. First described by Lemierre in 1936 as "anaerobic post-anginal septicemia," the syndrome became known as the "forgotten disease" following the widespread introduction of antibiotics for pharyngitis.[8,9] However, Harper et al. and Lee et al. reported that the number of documented cases has increased over the past two decades, possibly owing to greater clinical awareness, improved diagnostic imaging, conservative antibiotic prescribing practices for uncomplicated pharyngitis, and emerging antimicrobial resistance.[8,10] Although the estimated incidence remains approximately one case per million population annually, Manuja et al. emphasised that clinicians should maintain a high index of suspicion because delayed diagnosis continues to contribute to significant morbidity and mortality.[11]

In classical presentation, LS starts with an infection of the oropharynx, usually tonsillitis or peritonsillar infection, with contiguous spread to the parapharyngeal space and septic thrombophlebitis of the IJV. Harper et al., Vincent et al., and Qureshi et al. described how disruption of the pharyngeal mucosal barrier permits bacterial invasion of the lateral pharyngeal tissues and jugular venous system, resulting in thrombus formation and subsequent hematogenous dissemination.[10,12,13] Fusobacterium necrophorum remains the predominant pathogen, accounting for the majority of reported cases; however, Harper et al., Agonafir et al., and Im et al. noted that Streptococcus, Staphylococcus, Bacteroides, Eikenella, and polymicrobial infections have also been implicated. Furthermore, blood cultures may remain negative or unavailable because anaerobic organisms are difficult to isolate or because antimicrobial therapy is initiated before microbiological sampling.[10,14,15] In our patient, microbiological confirmation was unavailable, and therefore the diagnosis was established based on the characteristic clinical presentation together with radiological evidence of IJV thrombosis following a recent peritonsillar infection.

LS tends to occur in adolescents and young adults, particularly ages 15-30. Harper et al, Im et al, and Garcia Romero et al noted that older patients are not typical for the syndrome of LS, and our patient was a 54-year-old woman.[6,10,15] However, she presented with persistent fever, unilateral neck pain, and cervical swelling shortly after her peritonsillar infection, which was a typical presentation of the disease as described in the published literature. Therefore, LS should always be one of the major differentials if compatible clinical features are present, regardless of patient's age, this case being one of them.

The most frequent complication of LS is considered to be metastatic septic embolisation, and in 80-90% of patients the lungs are involved. Qureshi et al. reviewed the literature and concluded that septic embolisation in the lungs occurs in 85-97% of cases,[13] Vincent et al. and Harper et al. also found the lungs to be one of the most common sites of metastatic infection. Interestingly, in our patient, left IJV thrombosis was isolated without evidence of septic pulmonary emboli on contrast computed tomography (CT) of the chest.[10,12] It is suggested that this is due to having been noticed early and starting antimicrobial treatment before the process went on to metastasis.

Radiological imaging is essential to confirm LS. Harper et al., Manuja et al., and Im et al. noted that ultrasonography is a quick, inexpensive, non-invasive initial investigation that can identify IJV thrombosis, although this might be limited by the need for operator dependence and its inability to visualise beneath the mandible and clavicle. In contrast, contrast-enhanced CT is widely regarded as the imaging modality of choice because it accurately demonstrates intraluminal filling defects, evaluates disease extent, and identifies associated complications such as

deep neck abscesses and septic pulmonary emboli.[10,11,15] In our patient, ultrasonography readily identified an echogenic thrombus with partial recanalisation within the left IJV while simultaneously demonstrating diffuse thyroid enlargement with heterogeneous echotexture and increased vascularity. Subsequent contrast-enhanced CT venography confirmed the thrombus and excluded mediastinal extension, deep neck abscess, and pulmonary septic embolisation. These findings illustrate the complementary roles of ultrasonography for early diagnosis and CT for comprehensive disease assessment.

An unusual feature of the present case was the coexistence of thyroiditis with LS. Thyroid involvement has been only rarely reported in association with LS. Manuja et al. described thyroid enlargement presenting as goitre in a patient with LS,[11] whereas Im et al. reported abnormal thyroid imaging that initially raised concern for an alternative diagnosis.[15] In our patient, sonography revealed that the thyroid gland was diffusely enlarged with heterogeneous echotexture and high vascularity, which is characteristic of inflammatory thyroiditis but not of inflammatory thyroid abscesses. While it is unclear if there is a direct connection between thyroiditis and LS from an individual case, the association does expand the list of inflammatory patterns of the neck in association with LS and underlines the need for comprehensive workup of nearby structures during imaging.

Intravenous antibiotics are still the mainstay of treatment. The patient received IV ceftriaxone, with a resolution of fever and gradual improvement of neck pain and swelling without clinical evidence of worsening infections, within 21 to 28 days of starting treatment, which aligns with the recommendations in previous publications for use of a combination of β-lactam/β-lactamase inhibitor antibiotics or carbapenems or ceftriaxone plus metronidazole for adequate anaerobic cover, with a recommended duration of treatment between 3 and 6 weeks depending on the severity of illness and metastatic complications.[6,10,14,15] Combination therapy is often suggested where Fusobacterium species are known to dominate, but in our patient this did not happen and suggests that early diagnosis and prompt antibiotic treatment may be an effective treatment for localised disease without the onset of metastatic complications. However, since microbiologic confirmation was not possible, there was no ability to form a determination of pathogen-specific susceptibility.

The use of anticoagulation is still debatable. Our patient had isolated IJV thrombosis, and she seemed to have improved clinically within a few days with antibiotic therapy alone, so anticoagulation was not used, as some studies have indicated, because we should not routinely give anticoagulant drugs when there is no evidence of a randomised clinical trial supporting this approach.[6,10,13-15]

This is in line with the accumulating evidence that some patients with uncomplicated LS can be treated successfully without anticoagulant therapy.

The most significant feature of the case was the timely diagnosis that was made by using complementary ultrasonography and contrast-enhanced CT scan before the septic pulmonary emboli or other metastatic disease developed. Moreover, isolated IJV thrombosis, as another uncommon radiological finding, is also included in the growing clinical spectrum of LS. Successful conservative treatment without anti-coagulation drugs further demonstrates that treating patients on a case-by-case basis may yield excellent results based on clinical severity and imaging findings.

There are also some caveats. However, blood cultures and microbiological investigations were unavailable, and the diagnosis was not microbiologically confirmed, but remained clinicoradiological. Follow-up imaging was not performed in order to document complete resolution of the thrombus, and long-term follow-up was not available. With the above limitations, the temporal association with a recent peritonsillar infection along with those imaging features makes the diagnosis of LS clinically probable.

Conclusion

This case illustrates the importance of the use of LS in older and younger patients with perioral infection present for more than 21 days despite persisting fever and neck swelling, regardless of age. The early use of ultrasonography, then followed by contrast-enhanced CT, allows the prompt diagnosis of IJV thrombosis and may result in the identification of unusual associated findings, such as concurrent thyroiditis before the onset of metastatic events. Management is still essentially the same, but other treatment modalities such as the use of anticoagulants should be decided individually based on the development of the thrombus and whether septic or intracranial complications are present. Early diagnosis and timely treatment are essential to avoid any potential life-threatening sequelae.

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Conflicts of interest

There are no conflicts of interest.

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