CASE REPORT
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An Unusual Presentation of a Patient with Covid-19 Infection—Lemierre's Syndrome

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ABSTRACT

Lemierre's syndrome refers to septic thrombophlebitis of the internal jugular vein that is rarely seen after an oropharyngeal infection. We present the case of a 64-year-old female patient with Covid-19 infection and Lemierre's syndrome. Contrast-enhanced computed tomography of the neck revealed thrombosis in the left internal jugular vein and non-contrast-enhanced computed tomography of the thorax was consistent with Covid-19 infection. The patient was immediately hospitalized. Systemic antibiotics, anticoagulation therapy, and favipiravir treatment for Covid-19 infection were started. Unfortunately, the patient died about two weeks after hospitalization. To the best of our knowledge, this is the first case of Lemierre's syndrome and Covid-19 overlap. Clinical suspicion of Lemierre's syndrome is important for rapid diagnosis. During this Covid-19 pandemic, we should keep in mind that any patient may have Covid-19 infection. In addition to the patient's primary disease, with clinical or laboratory suspicion of Covid-19 infection, diagnostic tests for it should also be conducted.

Keywords: Lemierre's syndrome, internal jugular vein thrombosis, Covid-19 infection, deep neck infection

INTRODUCTION

Lemierre's syndrome (LS) refers to a septic thrombophlebitis of the internal jugular vein (IJV) because of an oropharyngeal infection. The development of suppurative thrombophlebitis and neck pain are hallmarks of the disease. It is a systemic disease that originates from an oropharyngeal infection and may lead to septic clot fragments in the rest of the body (1). The causative agents are mostly normal oral flora organisms, and the most common organism among them is the fusobacterium species, especially necrophorum. We described the case of a patient presenting with severe neck pain, subsequently diagnosed with LS and COVID-19 infection simultaneously.

CASE PRESENTATION

A 64-year-old female patient was admitted to the hospital with severe pain on the left side of the neck and chin. The patient had had neck and jaw pain for four days. The patient had complained of a sore throat for 10 days. The left side of the neck was tender and painful with palpation, but no space-occupying lesion was palpated. Oral cavity and oropharynx examinations revealed no features other than oropharyngeal hyperemia.

The patient had no cough, chest pain, or shortness of breath. The patient had no known chronic lung disease and was a non-smoker, and saturation at room air was 95. In the laboratory tests of the patient, acute phase reactants were found to be high. Bilateral parenchymal infiltrations were detected in chest radiography. Non-contrast-enhanced thorax CT was consistent with COVID-19 Reporting and Data System (CO-RADS) category 5 (2). The contrast-enhanced CT of the neck showed edematous pharyngeal mucosa, microabscess formation in the retropharynx, inflammation of interplanar fatty tissue, and a total occlusion of the left IJV from the hyoid level to the origin

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of the brachiocephalic vein (Figure 1), consistent with LS. The patient was admitted to Covid services. The polymerase chain reaction (PCR) test for SARS-CoV-2 was positive, and the patient was assigned to CO-RADS category 6 (2). The test for blood culture was negative. The patient was given 2g ampicillin-sulbactam four times a day intravenously, 600mg clindamycin three times a day intravenously, 8mg dexamethasone orally, and a standard 5-day regimen of favipiravir for the COVID-19 infection. Enoxaparin and warfarin were started as anticoagulant therapy targeting an international normalized ratio (INR) between 2 and 3.

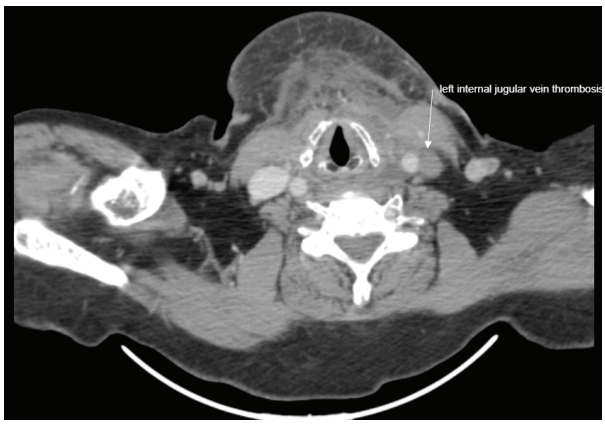

Figure 1: Contrast-enhanced neck CT. Left internal jugular vein thrombosis is seen at thyroid cartilage level

On the seventh day of hospitalization, retropharyngeal and parapharyngeal abscess formation was developed (Figure 2). The patient, whose condition worsened due to bacterial infection, was followed up in the intensive care unit (ICU) on the seventh day of hospitalization. Abscesses were drained surgically and recanalization of left IJV was observed during surgery. The abscess material was sent for microbiological analysis, but there was no growth in the abscess culture, possibly because the patient was under antibiotic therapy.

Figure 2: Non-contrast-enhanced neck CT.
The patient developed retropharyngeal abscess six days after admission to hospital.

Recanalization of the left IJV was also seen in the neck CT after abscess drainage (Figure 3). There was no septic embolism because the IJV thrombophlebitis was detected. The patient's general condition deteriorated each day, and unfortunately, she died from sudden cardiac arrest on the tenth day of ICU admission.

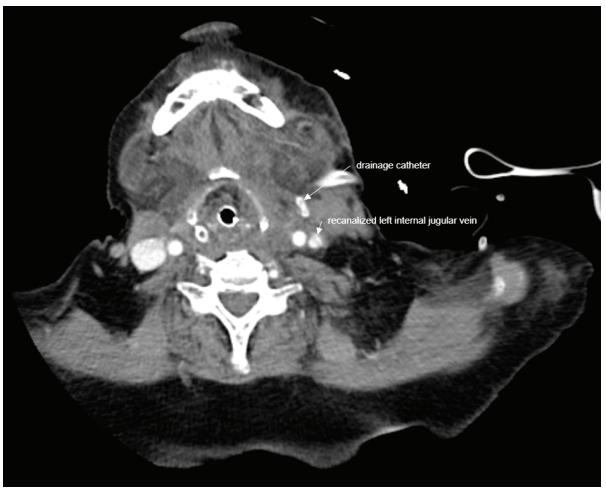

Figure 3: Contrast-enhanced neck CT
Near-total recanalization of left internal jugular vein is seen.

Her husband provided written consent after being informed about the aim of the study.

DISCUSSION

LS is a very rare disease, with a reported incidence of 0.6–2.3 per million (3). Since it was described in 1936 by Andre Lemierre, the incidence of LS has fallen dramatically because of antibiotic usage, to the extent that it has been called the forgotten disease. Primary infection in most cases of LS is associated with oropharyngeal infection as in our case. In patients with LS, primary infection is followed by a local invasion of the pharyngeal space and IJV, causing septic thrombophlebitis, with a 1–3 week time interval (4). The time interval between sore throat and LS in our patient was about 7–10 days. Common signs and symptoms of septic thrombophlebitis of IJV are pain, induration, or swelling at the ipsilateral angle of the mandible of the neck extending along the sternocleidomastoid muscle together with high fever and trismus (5). A thrombosed IJV is rarely palpable, and there may be no significant neck findings upon presentation. Therefore, in case of doubt, especially if the patient has symptoms such as severe neck and jaw pain and signs of tender and painful neck with palpation, neck imaging should be performed. A study has shown that IJV thrombophlebitis was detected in 59% of patients with increased use of imaging (6).

Covid-19 infection is a disease that predisposes to coagulopathy, and an increase in plasma D-dimer levels is the most frequently described report related to Covid-19 coagulopathy (7, 8). In hospitalized patients for Covid-19 infection, a coagulation profile should be performed, including D-dimer, PT, PTT, platelet count and fibrinogen (9). In patients with a Covid-19

infection, coagulopathy can be varying degrees from high levels of D-dimer to severe disseminated intravascular coagulation characterized by thrombocytopenia, prolonged PT, and elevated D-dimer (10). The worsening of laboratory parameters related to coagulation indicates progression in the severity of the Covid-19 infection. In this case, there was only an increase in the D-dimer level among the coagulation parameters. Therefore, clinical deterioration of our patient was mainly due to a bacterial infection and subsequent retropharyngeal abscess formation.

Management of IJV thrombosis in LS has changed over time. Before the use of antibiotics, ligation, or resection of the IJV was common, but today, systemic antibiotherapy is the main method of treatment. Ligation or resection of the IJV is indicated in cases of uncontrolled sepsis or ongoing septic emboli despite antibiotics (11). In a recent study, 8% of patients with LS required IJV ligation or resection (12). Anticoagulation therapy is not routinely used in LS, but under some circumstances, such as extensive thromboses, acute setting, and when thrombosis has the potential for retrograde progression to the cavernous sinus, anticoagulation therapy is recommended (13, 14). In this case, because of acute setting and extensive thrombosis, the patient was administered anticoagulation therapy. In most patients, recanalization occurs in the thrombosed vein with aggressive systemic antibiotherapy and surgery in necessary cases. Anticoagulation should be considered for high-risk patients if there is no contraindication to anticoagulation. Recanalization of the thrombosed vein may take several weeks or months. In our case, recanalization occurred seven days after diagnosis, and this recanalization was seen both during the surgical operation for abscess drainage and the contrast-enhanced neck CT scanning after the operation.

CONCLUSION

To our knowledge, this is the first case of Lemierre's syndrome presenting simultaneously with SARS-CoV-2. These two diseases on their own can have severe consequences for patients. Thus, the coexistence of the two may aggravate the devastating effects. Prompt diagnosis, early hospitalization, initiation of intravenous antibiotherapy, and a multidisciplinary team approach are crucial in the management of such patients. The COVID-19 pandemic is an ongoing global health problem. Therefore, nowadays, it should be kept in mind under any circumstances, as in this case with Lemierre's syndrome.

Informed Consent: Written informed consent was obtained from the patient's family for this case report.

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