

## CASE REPORT

# A case of Lemierre-like Syndrome: internal jugular vein thrombosis secondary to *Staphylococcus aureus* sternoclavicular joint septic arthritis

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## Abstract

Lemierre syndrome is the internal jugular vein (IJV) suppurative thrombophlebitis, usually secondary to oropharyngeal infection. *Staphylococcus aureus* is an emerging responsible pathogen. We report a unique case of IJV thrombosis secondary to methicillin-susceptible *S. aureus* sternoclavicular joint septic arthritis. We review the existing literature on Lemierre syndrome: its various manifestations, causative pathogens, treatment and management.

## CASE PRESENTATION

A 52-year-old Chinese gentleman with a significant medical history of gout, hypertension, dyslipidaemia, asthma, ischaemic heart disease (IHD) and chronic kidney disease (CKD) with a baseline creatinine of 130 µmol/L presented to us with a swollen and painful left ankle of 2 days' duration. The diagnosis of flare of gout of the left ankle was made. He had already been admitted twice within the past 2 months for the same diagnosis. The current flare of gout happened while he was still taking prednisolone 20 mg once daily and colchicine 500 microgram twice daily. Due to his underlying conditions of IHD and CKD, non-steroidal anti-inflammatory drugs were not used. His prednisolone dose was increased to 30 mg daily. The rheumatologist injected 20 mg of intra-articular triamcinolone into his ankle on day 2 of admission. Left ankle joint fluid aspiration under ultrasound guidance was simultaneously performed, but this was unsuccessful as only minimal fluid was present. A magnetic

resonance imaging (MRI) scan of the left foot was performed as his gouty arthritis was frequent and recalcitrant. The MRI showed changes consistent with gout (Table 1).

The ankle pain did not improve much; however, the C-reactive protein (CRP) showed significant improvement (Table 2). Paracetamol and 400 mg of tramadol daily in divided doses provided minimal pain relief. The pain service team reviewed him and added oxycodone for pain control. Unfortunately, on day 20 of admission, he developed right neck, upper pectoral and sternoclavicular joint (SCJ) tenderness and swelling. A fever followed 2 days later. The CRP was remarkably raised (Table 2).

An ultrasonography (US) of the neck showed right internal jugular vein (IJV) thrombosis and right pectoralis muscle abscess. A computed tomography (CT) scan of the thorax showed right pectoralis muscle and right supraclavicular abscesses likely related to right SCJ septic arthritis, as well as thrombosis of the distal third of the right IJV (Figures 1–3). The first three sets of blood cultures grew methicillin-sensitive *Staphylococcus*

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**Table 1:** Relevant investigations

Parameter	Result
WBC (NR 3.37-8.38 x 10 <sup>9</sup> /L)	8.12
Absolute neutrophils (NR 1.49-4.67 x 10 <sup>9</sup> /L)	5.74 (high)
Hb (NR 13.3-16.6 g/dL)	14.9
Platelets (NR 172-378 x 10 <sup>9</sup> /L)	244
Sodium (NR 135-145 mmol/L)	140
Potassium (NR 3.5-5.1 mmol/L)	4.1
Ceatinine (NR 59-104 µmol/L)	135 (high)
Urea (NR 2.8-7.6 mmol/L)	11.6 (high)
Uric acid (NR 250-523 umol/L)	345
C-reactive protein (NR 1-5 mg/L)	106.7 (high)
Blood culture	MSSA
Transthoracic echocardiogram	No valvular vegetations. Normal left and right ventricular systolic function. Diastolic dysfunction grade 1 (prolonged relaxation). Normal chamber sizes. No regional wall motion abnormality. Normal valvular function. Visual ejection fraction 70%.
MRI ankle	Erosions involving the articulating surface of the distal tibia and fibula as well as of the talus along with soft tissue swelling and likely joint effusion, plantar calcaneal spur-appearances consistent with gouty arthritis.
US abdomen	-Fatty liver. No suspicious focal hepatic lesion. -Uncomplicated gallstone. -Simple cyst in the right kidney.
US doppler neck veins	-Thrombosis of internal jugular vein in lower third neck. It appears to be narrowed or obstructed by an inhomogeneous mixed echogenic lesion, suspicious for necrotic mass or abscess. -Elongated hypoechoic collection in the right pectoralis muscle suspicious for abscess or necrotic collection.
CT chest	-Abscess in the right supraclavicular region extending to the sternal head of right sternocleidomastoid muscle. -Hypodense collections in the right pectoralis major muscle. These abscesses are probably related to the right sternoclavicular joint septic arthritis. -Thrombosis of lower third of right IJV.

CT = computed tomography; Hb = hemoglobin; MRI = magnetic resonance imaging; NR = normal range; US = ultrasound; WBC = white blood cells

**Table 2:** WBC and C-reactive protein trend

Date	WBC (NR 3.37-8.38 x 10 <sup>9</sup> /L)	Absolute neutrophils (NR 1.49-4.67 x 10 <sup>9</sup> /L)	C-reactive protein (NR 1-5mg/L)
Admission Day 1 (19 Mar 2018)	8.12	5.74	106.7
Day 4	12.02	10.68	32.6
Day 15			30.1
Day 20: Neck swelling occurred			
Day 22	11.49	9.73	207.9
Day 24: Cloxacillin started			
Day 27	10.13	9.13	73.8
Day 34	9.23	7.39	21.6
Day 40	8.21	6.17	19.7
Day 60	8.19	4.26	7.7
Day 86	4.78	2.54	5.3

NR = normal range; WBC = white blood cells

*aureus* (MSSA). The transthoracic echocardiogram showed no evidence of infective endocarditis. The patient did not have any skin lesions or notable entry points for the MSSA. Blood cultures were repeated every 48 hours until they were negative. He was initially planned for a trans-oesophageal echocardiogram (TOE). However, in view of subsequent negative blood cultures and the fact that he had no further fever spikes after commencing treatment, infectious disease specialists opined that a TOE was not necessary.

In view of the septic arthritis with concomitant bacteremia, he was treated with 6 weeks of intravenous cloxacillin. Cloxacillin was chosen based on culture and susceptibility results. In view of the proximity of the IJV thrombosis to the superior vena cava (SVC), and thus the concern that he might develop pulmonary embolism, he was anticoagulated, initially with enoxaparin (low-molecular weight heparin), followed by 3 months of rivaroxaban. The duration and choice of anticoagulation is based on the American College of Chest

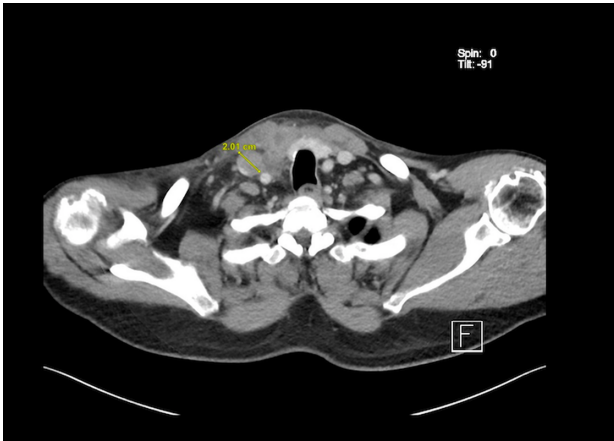


Figure 1: Thrombosis in distal right IJV

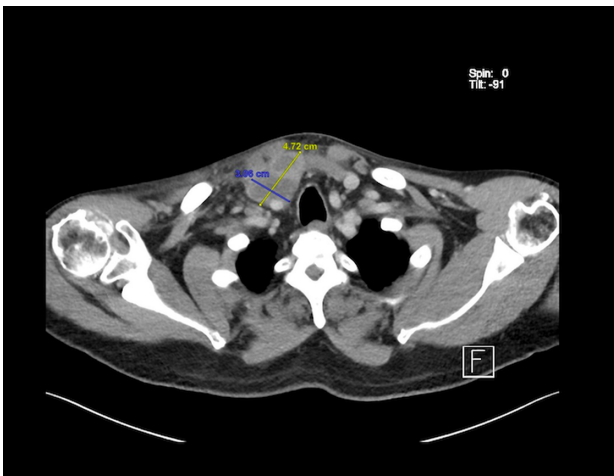


Figure 2: Abscess in right supraclavicular region

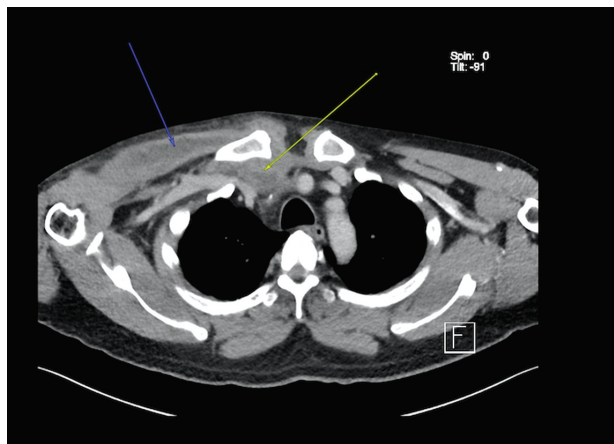


Figure 3: Abscess in right pectoralis muscle (blue arrow) and posterior to right clavicle (yellow arrow) related to right SCJ arthritis

Physicians (ACCP)'s guidelines on upper extremity DVT (refer to [Discussion](#)). The patient was transitioned to rivaroxaban as it afforded him more convenient outpatient therapy.

Cardiothoracic and orthopedic surgery opinions were sought. No surgical intervention was deemed necessary. A follow-up CT

of the thorax after 6 weeks of antibiotics showed resolution of the pectoral abscess with persistent IJV thrombosis. His CRP normalized.

## DISCUSSION

Lemierre's syndrome has the following characteristics [1]:

- (i) follows an oropharyngeal infection;
- (ii) there is a presence of suppurative thrombophlebitis of the IJV with metastatic septic emboli;
- (iii) is usually caused by fusobacterium; and
- (iv) affects mainly young and previously healthy patients.

However, many variants of the syndrome have been described.

Apart from oropharyngeal infection, trauma [1], primary infections in the sinuses [2] or face [3] and head and neck malignancies [1] can also cause IJV thrombosis.

Apart from *Fusobacterium spp.*, other causative bacteria include *Eikenella*, *Proteus*, *Bacteroides*, *Peptococcus*, *Peptostreptococcus*, *Klebsiella*, *Streptococcus* and *Staphylococcus* (both MSSA and MRSA) [1].

Common clinical presentations include sore throat, cervical lymphadenopathy, neck pain, swelling, fever, chills and rigors [4]. The most common site of metastatic infection is the lungs (80–97% of cases) [4].

Contrast-enhanced CT is the imaging study of choice [4] to diagnose IJV thrombosis. Ultrasound is a rapid, low-cost, non-invasive, radiation-free alternative, but it can miss thrombus above the mandible or below the clavicle [5].

*Staphylococcus aureus* has been reported to cause Lemierre syndrome. Chanin *et al.* [6] noted 11 cases from 2002 to 2011. All but one case involved young patients, and all cases featured metastatic spread of infection to the lungs.

Since then, there have been 11 further reported cases (2011 to present; Table 3). However, none of the cases was caused by SCJ septic arthritis or pectoral abscess.

Treatment involves removing the focus of infection—with surgical drainage if necessary—and several weeks of appropriate antibiotics [7].

To date, there are no controlled studies regarding the role of systemic anticoagulation [6]. Some authors suggest that anticoagulation could limit seeding of septic pulmonary thromboemboli and prevent retrograde clot extension from the IJV into the sigmoid or cavernous sinuses [5]. Most would consider anticoagulation if there is extensive internal jugular venous thrombosis or extension despite antimicrobial therapy [8].

Our patient was in his middle age and had multiple comorbidities. His IJV thrombosis was secondary to SCJ septic arthritis and pectoralis muscle abscess rather than an oropharyngeal infection. There was no evidence of septic emboli based on the CT chest and US abdomen. He responded well to antibiotic treatment.

This unprecedented case of IJV thrombosis secondary to SCJ septic arthritis highlights the following:

- (i) IJV thrombosis does not occur only after an oropharyngeal infection. It may occur if there is an infective process near its draining site. In this case, the right SCJ septic arthritis and pectoralis muscle abscess led to the narrowing of the IJV, precipitating thrombosis.
- (ii) High clinical suspicion for Lemierre syndrome in the setting of head and neck infections is important as surgical

Table 3: Staphylococcus aureus as a cause of Lemierre Syndrome

Reference & date	Demographics	Past medical history	Organism isolated	Source of infection	Metastatic infection	Venous thrombosis	Treatment for sepsis	Anticoagulation	Outcome
Chanin et al May 2011	22 yo Caucasian Female	Previously healthy	MRSA in blood & cerebrospinal fluid (CSF) cultures	Oropharyngeal	Necrotising pneumonia, Cerebral infarcts	Bilateral IJV	6 weeks of antibiotics	Nil	Required intubation and ventilatory support. Recovered Recovered
Molloy et al, Sept 2012	18 yo Indian	Previously healthy	MSSA on blood culture	Oropharyngeal	Nil	Right IJV	4 weeks of antibiotics	6 months of warfarin	Recovered Recovered
Abhishek et al, Nov 2012	24 yo Male	Previously healthy	MRSA in blood, wound & sputum cultures	Dental infection which seeded to traumatic hematoma in right sternocleidomastoid (SCM) muscle	Necrotising pneumonia	Right IJV extending to subclavian confluence	6 weeks of antibiotics Drainage of SCM abscess	Heparin then 3 months of warfarin	Required intubation and ventilatory support. Recovered
Pitsiou et al, Jan 2013	25 yo male	Previously healthy	MSSA in blood & tracheal aspirates	Nil apparent	Pulmonary septic emboli	Inferior vena cava up to the level of external and internal iliac veins	Antibiotics	Anticoagulated	Required intubation, ventilatory support and inotropes. Deceased
Root et al, Jan 2013	10 month old infant	Previously healthy	MSSA on blood culture	Neck abscess	Septic emboli to lungs and brain, pericardial tamponade secondary to purulent pericarditis	Right internal jugular vein, extending into her left ventricular outflow tract	Drainage of neck abscess, pericardial window 6 weeks of nafcillin	Heparin then enoxaparin	Septic shock, requiring vasopressor support, intubation Recovered with minimal permanent sequelae

Continued

Table 3: (continued)

Reference & date	Demographics	Past medical history	Organism isolated	Source of infection	Metastatic infection	Venous thrombosis	Treatment for sepsis	Anticoagulation	Outcome
Stauffer et al, Feb 2013	18 yo male	Previously healthy	MRSA on blood, sputum, eye, retropharyngeal abscess	Retropharyngeal abscess	Nil	Right internal jugular extending into the right sigmoid and transverse sinuses Bilateral cavernous sinus with potential extension into the ophthalmic veins	Antibiotics Unsuccessful surgical drainage of abscess	Anticoagulated	Binocular vision loss secondary to bilateral ophthalmic vein occlusion and optic nerve ischemia
Kizhner et al, Jul 2013	16 yo male	Previously healthy	MRSA	Oropharyngeal					
Marulasiddappa et al, Nov 2013	24 yo male	Previously healthy	<i>S. aureus</i> in pus & tissue cultures	Left parapharyngeal abscess	Nil	Left IJV, left transverse and sigmoid sinuses	3 weeks of antibiotics Drainage of abscess	Anticoagulated	Septic shock requiring inotropes
Kidambi et al, 2015	24 yo Female	Intravenous drug abuse	MRSA in blood cultures	Right retropharyngeal abscess	Pulmonary septic emboli	Right internal and external jugular vein	4 weeks of antibiotics Drainage of abscess	Nil	Recovered
Jariwala et al, 2017	Paediatric patient	Previously healthy	MRSA						
Raggio et al, Mar 2018	5 week old female	Previously healthy	MRSA in blood cultures	Left peritonsillar/-parapharyngeal space abscess	Septic emboli to lungs and brain	Left internal jugular vein	Antibiotics, bedside abscess aspiration	Nil	Deceased

drainage of collection wherever possible and prolonged antibiotics are necessary.

- (iii) More prospective studies are required to clarify the role of anticoagulation in Lemierre syndrome. The IJV is close to the SVC. Thus, pulmonary embolism is a potential sequela. Some consider IJV thrombosis a form of upper extremity thrombosis. Should anti-coagulation be required, 3 months of anti-coagulation should be adequate based on ACCP's guidelines on upper extremity DVT [9]. The anticoagulant agent should be chosen to suit the patient's clinical circumstances [9].

### CONFLICTING OF INTERESTS STATEMENT

None declared.

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### CONSENT

Consent has been obtained from the patient for the purposes of this case report.

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