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*...
The transthoracic echocardiogram showed 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.

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

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

| Date                          | WBC (NR 3.37-8.38 x 10^9/L) | Absolute neutrophils (NR 1.49-4.67 x 10^9/L) | C-reactive protein (NR 1-5 mg/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 | 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

CT = computed tomography; Hb = hemoglobin; MRI = magnetic resonance imaging; NR = normal range; US = ultrasound; 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.
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**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...
| 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 |
| 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 |
| 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 | Drainage of SCM abscess | Heparin then 3 months of warfarin | Recovered |
| Pitsiou et al, Jan 2013 | 25 yo Male | Previously healthy | MSSA in blood & tracheal aspirates | 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 | 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 | Nil | Left internal jugular vein | 3 weeks of antibiotics, Drainage of abscess | Anticoagulated | Septic shock requiring inotropes |
| Marulasiddappa et al, Nov 2013 | 24 yo male | Previously healthy | S. aureus in pus & tissue cultures | Left parapharyngeal abscess | Nil | Right internal jugular, and external jugular vein | Right jugular vein | Nil | Recovered |
| Kidambi et al, 2015 | 24 yo Female | Intravenous drug abuse | MRSA in blood cultures | Right retropharyngeal abscess | Pulmonary septic emboli | Septic emboli to lungs and brain | Deceased |
| Jariwala et al, 2017 | Paediatric patient | 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 |
| Raggio et al, Mar 2018 | 5 week old female | Previously healthy | MRSA in blood cultures | Left parapharyngeal 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.

FUNDING

The author(s) received no financial support for the research, authorship, and/or publication of this article.

CONSENT

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

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