The Changing Face of an Old Disease: Case Report of Nonclassical Lemierre’s Syndrome Caused by a Panton-Valentine Leucocidin-Positive Methicillin-Susceptible Staphylococcus aureus Isolate

Chanin et al. published a review of methicillin-resistant Staphylococcus aureus (MRSA) causing Lemierre’s syndrome (4). We present another case of Lemierre’s syndrome caused by Panton-Valentine leucocidin (PVL)-positive methicillin-susceptible Staphylococcus aureus (MSSA) in a healthy 18-year-old Indian student. He had been a resident in the United Kingdom for a year. He presented with 3 days of right-sided facial swelling and fevers and a history of a sore throat in the 2 weeks prior to admission. Examination revealed a temperature of 38.9°C, a pulse of 135 beats per minute, blood pressure of 113/66 mmHg, and marked right facial and neck swelling with tenderness and induration of the right external jugular vein and pustules on his right cheek and lip. Examination of the oropharynx was unremarkable.

Investigations revealed negative HIV serology, a C-reactive protein level of 226 mg/liter (normal value, <5 mg/liter), and a white cell count of 17.1 × 10^9/liter (normal value, 4 × 10^9 to 11 × 10^9/liter), and a chest X ray showed airspace infiltrates and a cavitating lesion. Blood cultures taken on admission grew MSSA. Computerized tomography (CT) of the head and neck showed gross tissue edema in the face and neck and suggested a clot in the right jugular venous system. A Doppler ultrasound of his neck showed a free-floating nonocclusive thrombus extending from the right external jugular vein into the common jugular vein. A transthoracic echocardiogram revealed no cardiac vegetations.

A diagnosis of Lemierre’s syndrome caused by MSSA was made, and he was treated with 2 g of intravenous (IV) oxacillin four times a day (QD) for 14 days, followed by oral oxacillin for a further 2 weeks and warfarin therapy for 6 months. Toxin gene profiling of the initial blood culture isolate of MSSA by the reference laboratory confirmed that it was PVL positive.

The patient had a good response to treatment and made a full recovery.

As in most cases presented by Chanin et al., our case consisted of unilateral jugular vein thrombosis with metastases to the lungs and was caused by community-acquired S. aureus in an immunocompetent host.

We would like to reiterate the importance of early recognition of Lemierre’s disease, as emphasized by Chanin et al., to achieve earlier diagnosis and prevent complications. Despite the decline in cases of Lemierre’s syndrome in the later half of the 20th century (12), increasing prevalence of antibiotic-resistant pathogens, increasing virulence of organisms, and perhaps judicious use of antibiotics to curb the development of resistance mean that Lemierre’s syndrome is reported more frequently and less typically (2, 3, 7, 8). Lemierre’s syndrome is often described as a condition of the preantibiotic era; we suggest that Lemierre’s syndrome now belongs to the modern era of increasingly virulent and antibiotic-resistant organisms.

As noted by Chanin et al., S. aureus has been reported as a notable cause of Lemierre’s syndrome since 2002. Most isolates in the review were either PVL producers or MRSA, well recognized as significant pathogens. MSSA is emerging as a cause of Lemierre’s syndrome (Table 1), and pathogenic strains of MSSA are increasingly PVL producers (9, 14). PVL appears to have pathogenic synergism with alpha-hemolysin, which is almost ubiquitously expressed by staphylococci (5). The ability to lyse leukocytes and erythrocytes to assist immune evasion and bacterial replication seems to be a shared feature of PVL-producing S. aureus and Fusobacterium necrophorum, the pathogen classically associated with Lemierre’s syndrome (11), and may explain why both organisms appear to be capable of causing the constellation of clinical features described by Lemierre.

Our case reemphasizes the emergence of nonclassical Lemierre’s syndrome and raises awareness of S. aureus as a signifi-

### Table 1

<table>
<thead>
<tr>
<th>Yr</th>
<th>Case (reference)</th>
<th>Patient age (yr)</th>
<th>Bacterium</th>
<th>PVL</th>
<th>Internal jugular vein affected</th>
<th>Complications</th>
<th>Interventions other than antibiotics</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>2008</td>
<td>Puymirat et al. (10)</td>
<td>22</td>
<td>MSSA</td>
<td></td>
<td>Not reported</td>
<td>Multiple pulmonary nodules; cavitating; cavascular sinus thrombosis</td>
<td>SC heparin, excision of anterior and external jugular valves; IV heparin, activated protein C, warfarin</td>
<td>Recovered</td>
</tr>
<tr>
<td>2008</td>
<td>Shivashankar et al. (13)</td>
<td>32</td>
<td>MSSA</td>
<td>+</td>
<td>Left</td>
<td>Bilateral pulmonary nodules; pulmonary abscesses; cavascular sinus thrombosis; bilateral cerebellar, left frontal, and brain stem infarcts</td>
<td>None</td>
<td>Recovered</td>
</tr>
<tr>
<td>2009</td>
<td>Gokce Ceylan et al. (6)</td>
<td>80</td>
<td>MSSA</td>
<td></td>
<td>Not reported</td>
<td>Bilateral pulmonary nodular infiltrates; ribonucleoprotein accumulation</td>
<td>Anticoagulation therapy, abscess drainage</td>
<td>Died</td>
</tr>
<tr>
<td>2010</td>
<td>Aouad, Melkane, and Rassi (1)</td>
<td>4</td>
<td>MSSA</td>
<td></td>
<td>Not reported</td>
<td>Extension of thrombophlebitis to cavascular sinus; multisystem emboli to the brain, orbits, lungs, and heart valves</td>
<td>SC heparin, warfarin</td>
<td>Died</td>
</tr>
<tr>
<td>2012</td>
<td>Our case</td>
<td>18</td>
<td>MSSA</td>
<td>+</td>
<td>Right</td>
<td>Pulmonary infiltrates; cavitating</td>
<td>SC heparin, warfarin</td>
<td>Recovered</td>
</tr>
</tbody>
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a SC, subcutaneous.
cant cause. We also postulate that alpha-hemolysin plays a role in the pathogenesis of this aggressive condition.

REFERENCES