The Changing Face of an Old Disease: Case Report of Non-Classical Lemierre's Syndrome caused by a PVL-Positive Meticillin-Susceptible *Staphylococcus aureus*

Keywords

Staph aureus, MSSA, PVL, Lemierre's syndrome, septic emboli, jugular thrombus

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Chanin et al published a review of meticillin-resistant *Staphylococcus aureus* (MRSA) causing Lemierre’s syndrome [4]. We present another case of Lemierre’s syndrome caused by Panton-Valentine Leucocidin (PVL)-positive meticillin-susceptible *Staphylococcus aureus* (MSSA) in a healthy 18-year-old Indian student. He had been resident in the UK 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, pulse of 135 beats per minute, blood pressure of 113/66mmHg 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, C-reactive protein 226mg/liter (<5 mg/liter), white cell count 17.1x10⁹/L (4-11x10⁹/L) and chest X-ray showed airspace infiltrates and a cavitating lesion. Blood cultures taken on admission grew MSSA. Computerised tomography (CT) of the head and neck showed gross tissue oedema in the face and neck and suggested a clot in the right jugular venous system. Doppler ultrasound of his neck showed a free-floating non-occlusive thrombus extending from the right external into the common jugular vein. Transthoracic echocardiogram revealed no cardiac vegetations.

A diagnosis of Lemierre’s syndrome caused by MSSA was made and he was treated with IV flucloxacillin 2g QDS for 14 days followed by oral flucloxacillin 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 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 development of resistance means 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 pre-antibiotic 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-recognised as significant pathogens. MSSA is emerging as a cause of Lemierre's syndrome (see table), pathogenic strains of MSSA are increasingly PVL producers.[9, 14] PVL appears to have pathogenic synergism with alpha-haemolysin, which is almost ubiquitously expressed by Staphylococci.[5] The ability to lyse leucocytes 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 re-emphasises the emergence of non-classical Lemierre’s syndrome and raises awareness of *S. aureus* as a significant cause. We also postulate that alpha-haemolysin plays a role in the pathogenesis of this aggressive condition.
References

<table>
<thead>
<tr>
<th>Year</th>
<th>Case</th>
<th>Age</th>
<th>Bacteria</th>
<th>PVL</th>
<th>Internal Jugular Vein</th>
<th>Complications</th>
<th>Interventions in addition to antibiotics</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>2008</td>
<td>Puymirat[10]</td>
<td>22yrs</td>
<td>MSSA</td>
<td>Not reported</td>
<td>Right</td>
<td>Multiple pulmonary nodules, cavitation, cavernous sinus thrombosis</td>
<td>SC heparin, excision of Internal and External Jugular</td>
<td>Recovered</td>
</tr>
<tr>
<td>2009</td>
<td>Ceylan[6]</td>
<td>80yr</td>
<td>MSSA</td>
<td>Not reported</td>
<td>Right</td>
<td>Bilateral pulmonary nodular infiltrates, bibasal pleural effusions</td>
<td>None</td>
<td>Died</td>
</tr>
<tr>
<td>2010</td>
<td>Aouad[1]</td>
<td>4yr</td>
<td>MSSA</td>
<td>Not reported</td>
<td>Right</td>
<td>Extension of thrombophlebitis to cavernous sinus, multivisceral emboli to the brain, orbits, lungs and heart valves</td>
<td>Anticoagulation therapy, Abscess drainage</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 and cavitation</td>
<td>SC heparin, Warfarin</td>
<td>Recovered</td>
</tr>
</tbody>
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