CASE REPORT

Streptococcus salivarius meningitis and sphenoid sinus mucocele. Case report and literature review

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Introduction

Streptococcus salivarius is a member of the viridans group of streptococci, commensals of the human upper respiratory, gastrointestinal and female genitourinary tract, but most prevalent in the oral cavity. 1 Viridans streptococci frequently invade the bloodstream, but rarely cause meningitis because of their low virulence and a likely low affinity to the leptomeninges. Indeed, in most cases of viridans streptococcal meningitis, the source of infection is the patient’s endogenous flora and meningitis develops after an invasive procedure and/or because of local pathologic conditions. 1–5 S. salivarius meningitis has been related to risk factors or underlying conditions including neoplasia, 6–8 brain abscess, 4 cranial trauma, 4,9 and cerebrospinal fluid (CSF) fistula. 2,8,11-14 Moreover, S. salivarius appears to be an increasing cause of iatrogenic meningitis following different invasive diagnostic or therapeutic procedures. 2–5,7,8,10,11,13-29

We report a case of S. salivarius meningitis in a patient with CSF fistula due to sphenoid sinus mucocele and review the literature concerning meningitis caused by this uncommon aetiological agent.

Summary
We report a case of meningitis caused by Streptococcus salivarius in a 49-year-old woman with a previously undiagnosed cerebrospinal fluid fistula due to a sphenoid mucocele. We reviewed the literature concerning meningitis caused by this uncommon organism and to the best of our knowledge this is the first case of S. salivarius meningitis associated with sphenoid mucocele.

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Case report

In November 2002, a 49-year-old woman was admitted to the emergency department of a community hospital with a 1-day history of severe frontal headache and high fever followed by drowsiness and confusion.

On admission she was febrile (temperature, 39.8°C), lethargic, and confused. Her blood pressure was 130/90 mmHg, heart rate 100 beats/min, and respiratory rate 20/min. Neurological examination revealed signs of meningeal irritation (stiff neck, Kernig's and Brudzinski's signs). Brain computed tomography (CT) scan showed no abnormalities. The lumbar puncture (LP) yielded purulent CSF, but no laboratory examinations were performed on CSF specimens. After a clinical diagnosis of bacterial meningitis, the patient received ceftriaxone 2 g intravenously (IV) and was transferred to our "L. Spallanzani" National Institute for Infectious Diseases.

On admission, LP was done and three blood specimens were collected for culture. The CSF contained 3200 white cells/mm³ (90% neutrophils) and a protein concentration of 150 mg/dl; glycorrhachia was normal. CSF samples were submitted to the laboratory for Gram staining, bacterial antigens detection (latex tests for group B Streptococcus, Haemophilus influenzae type b, Streptococcus pneumoniae, Neisseria meningitidis group A, B, C, Y, W 135), and bacterial culture; all the tests were performed according to the standard procedures. Moreover, 4 ml of CSF were inoculated into both a Bactec Plus Aerobic/F and a Bactec Plus Anaerobic/F (Becton, Dickinson and Company, Sparks, U.S.A.) bottle at the patient's bed. During this procedure, personnel adopted aseptic measures including handwashing, surgical masks and gowns. The empirical antibiotic treatment included ceftriaxone 2 g IV q12h, plus ampicillin 3 g IV q6h.

CSF Gram staining and bacterial antigen detection assays resulted negative. The patient's clinical conditions dramatically improved 24 h later and the previously reported neurologic impairment disappeared. Forty-eight hours after her admission into our unit, all the standard CSF bacterial cultures remained negative, whereas both CSF Bactec cultures yielded an alpha-haemolytic Streptococcus, identified as S. salivarius by means of the standard methods. Blood cultures performed on admission resulted negative.

The unusual aetiological agent that is commonly resident in the oral cavity led to the likelihood of an abnormal communication between upper airways and subarachnoid space. The patient denied any history of head trauma, sinusitis, and otitis, but reported that during the previous 2 months she presented a persistent nasal secretion, that was explained as an allergic rhinitis by her general practitioner. Moreover, she referred that rhinorrhea mainly occurred when she bent her head forward. This information reinforced our suspicion of a CSF fistula.

Cranial CT (Fig. 1) and nuclear magnetic resonance scans were immediately performed: both revealed an enlarged sphenoid sinus communicating...
with the posterior cranial fossa. The patient recovered in 2 weeks and was transferred to the Neurosurgery Unit for surgical correction of the fistula. During the neurosurgical intervention, a sphenoid mucocele was found, drained off, and treated with an obliterative procedure after repair of the CSF fistula.

The patient was discharged in good general conditions, with no residual neurological signs or symptoms. No recurrence of CSF leak has been observed after a 2-year follow-up.

Discussion

To the best of our knowledge, this is the first report on a *S. salivarius* meningitis secondary to a sphenoid sinus mucocele with a CSF fistula. In our patient, the development of a sphenoid mucocele caused a bone defect resulting in transdural communication and CSF leakage. As a consequence, the spread of endogenous flora (*S. salivarius*) from the patient’s upper respiratory tract to the CSF was enabled.

The patient was discharged in good general conditions, with no residual neurological signs or symptoms. No recurrence of CSF leak has been observed after a 2-year follow-up.

Table 1  Risk factors for *Streptococcus salivarius* meningitis as reported in the literature

<table>
<thead>
<tr>
<th>Underlying condition/procedure</th>
<th>Number of cases (total = 43)</th>
<th>Reference no.</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gastrointestinal tumor</td>
<td>2</td>
<td>[6,7]</td>
</tr>
<tr>
<td>Leiomyosarcoma of the uterus</td>
<td>1</td>
<td>[8]</td>
</tr>
<tr>
<td>Brain abscess</td>
<td>1</td>
<td>[4]</td>
</tr>
<tr>
<td>Cranial trauma</td>
<td>3</td>
<td>[4,9]</td>
</tr>
<tr>
<td>Fistula</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Spontaneous</td>
<td>2</td>
<td>[11,12]</td>
</tr>
<tr>
<td>Posttraumatic</td>
<td>1</td>
<td>[2]</td>
</tr>
<tr>
<td>Mucocele</td>
<td>1</td>
<td>PR</td>
</tr>
<tr>
<td>Iatrogenic</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Postneurosurgical fistula</td>
<td>3</td>
<td>[8,13,14]</td>
</tr>
<tr>
<td>Thermocoagulation of gasserian ganglion</td>
<td>1</td>
<td>[11]</td>
</tr>
<tr>
<td>Spinal anesthesia/analgesia</td>
<td>14</td>
<td>[3,5,7,10,16,17,22-24,27-29]</td>
</tr>
<tr>
<td>Diagnostic lumbar puncture/myelography</td>
<td>11</td>
<td>[4,18-21,25,26]</td>
</tr>
<tr>
<td>Gastrointestinal endoscopy</td>
<td>3</td>
<td>[2,11,15]</td>
</tr>
</tbody>
</table>

PR, present report.

A mucocele is an uncommon benign, epithelial-lined mucous-filled cystic structure that can develop in the paranasal sinuses. It can progressively grow and cause bone destruction resulting in an abnormal communication between sinusal and subarachnoidal space, thus increasing the risk of meningitis. The causes of sphenoid sinus mucocele include congenital anomaly, trauma, infection, allergy, and surgery of the sphenoid sinus. In our patient, a previously undiagnosed congenital anomaly is the most likely cause of mucocele.

In conclusion, this case is of particular interest due to the unusual organism responsible for meningitis, the predisposing anatomical condition, and the microbiological diagnosis. The isolation of *S. salivarius* from CSF should induce clinicians to research the above reported conditions predisposing to this uncommon infection; the direct inoculation of CSF into culture medium should always be performed in order to increase the probability of aetiological diagnosis of meningitis.

References


