Case Report

Two uncommon cases of Pneumococcal pyomyositis

Estelle Honnorat,1 Piseth Seng,1,2 Julie Finance,1 Benjamin Blondel3 and Andreas Stein1,2

1Centres de Référence Interrégionaux Sud-Méditerranée pour la Prise en Charge des Infections Ostéo-Articulaires Complexes, Service des Maladies Infectieuses, CHU de la Conception, 147, boulevard Baille, Marseille, France
2URMITE, CNRS UMR 6236, Faculté de Médecine, 27 Bd Jean Moulin, 13385 Marseille Cedex 05, France
3Unité de Chirurgie Orthopédique et Rachidienne, Hôpital Timone, 264, rue Saint-Pierre, 13385 Marseille, France

Introduction: Pneumococcal pyomyositis is a rare disease. To our knowledge, only 28 cases of this disease have been reported in adults.

Case presentation: We report two new cases of pneumococcal pyomyositis managed at an inter-regional referral centre for bone and joint infections in the south of France. One of our patients had heterozygous sickle-cell disease, and the second had no apparent immunodeficiency. The pneumococcal pyomyositis was localized primarily to the psoas muscle and was complicated by hip arthroplasty infection in one of our cases. In the other case, it was localized to the abductor muscle, which has not been reported previously.

Conclusion: We report two new cases of this disease with favourable outcomes following long-term antimicrobial treatment and surgery debridement.

Keywords: bacteria; human; pneumococcal pyomyositis; pyomyositis; Streptococcus pneumoniae.

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Introduction
Pyomyositis is a rare disease that is typically caused by Staphylococcus aureus (Bickels et al., 2002; Burdette et al., 2012). Pneumococcal pyomyositis occurs in 1% of cases of pyomyositis (4 of 452 cases; Bickels et al., 2002) and is usually reported in immunodeficient individuals, such as those with human immunodeficiency virus (HIV) infection, diabetes mellitus, cancer, connective tissue diseases, cirrhosis or asplenia (Burdette et al., 2012).

Here, we report two cases of pneumococcal pyomyositis managed in an inter-regional referral centre for bone and joint infections in the south of France. We also reviewed 28 cases of pneumococcal pyomyositis in adult patients reported in the literature between 1940 and 2014.

Case reports

Case 1
In August 2010, a 68-year-old man was admitted for fever, chills, dyspnoea, weight loss, and lumbar and right hip pain that occurred 1 month before admission. His medical history indicated that he had received a right hip prosthesis for tuberculosis coxarthrosis in 1976; the anti-tuberculosis treatment information was not available. He was treated for primary arterial hypertension by administration of candesartan cilexetil and hydrochlorothiazide. The patient did not have any malnutrition or alcoholism. This patient was born in Guinea and had lived in France for more than 30 years. He travelled for 3 months in Guinea and spent 1 month in Senegal for a family visit.

Upon admission, he presented with fever (38.5°C) and muscle pain that had migrated secondarily to his right hip prosthesis. Laboratory investigations revealed a high C-reactive protein level (299 mg l⁻¹; normal value ≤ 5 mg l⁻¹), an elevated leukocyte count (14 000 μl⁻¹, predominantly neutrophil granulocytes), an elevated platelet count (700 000 μl⁻¹).
and severe hypochromic microcytic aplastic anaemia (6 g haemoglobin dl\(^{-1}\) and 43 000 reticulocytes \(\mu^{-1}\)). Computed tomography scans of the chest, abdomen and pelvis revealed a bilateral psoas abscess associated with hip prosthesis infection (Fig. 1) and left basal pneumonia with pleural effusion. Trans-thoracic echocardiography revealed no evidence of infectious endocarditis or valve abnormalities.

Blood cultures were performed before antibiotics were administered and were sterile. A bacterial culture of purulent fluid aspirated from the psoas abscess with a percutaneous drainage needle was positive for \textit{Streptococcus pneumoniae}, as identified by matrix-assisted laser desorption-ionization time-of-flight mass spectrometry (MALDI-TOF MS) completed by an optochin disc and latex agglutination test for rapid detection of \textit{S. pneumoniae}. Laboratory tests found no immunosuppression. Screening by a sickle-cell test and capillary electrophoresis revealed heterozygous sickle-cell disease, which was the primary cause of the microcytic anaemia.

He was treated with administration of oral rifampicin (1.2 g day\(^{-1}\)) and amoxicillin (12 g day\(^{-1}\)) for 14 days. Antimicrobial treatment was then continued by oral monotherapy with amoxicillin (12 g day\(^{-1}\)) for 5 months without any intolerance being observed. Surgical drainage of the psoas abscess and two-stage prosthesis exchange were performed on day 7 of antimicrobial treatment. A new prosthesis was inserted during the fourth month of treatment. No relapses were observed during the 4-year post-antibiotic follow-up.

**Case 2**

In April 2014, a 72-year-old man was admitted to the Emergency Department of the University Hospital in Marseille for fever. He had a medical history of multiple degenerative age-related arthrosis, which happened 10 years before the current episode. He did not have trauma or apparent immunodeficiency state. He had practiced aquatic gymnastics during the week before his admission. A clinical examination did not reveal any abnormalities. Laboratory investigations showed a high C-reactive protein level (299 mg l\(^{-1}\)), an elevated leukocyte count (21 000 \(\mu^{-1}\), predominantly neutrophil granulocytes), a normal platelet count (187 000 \(\mu^{-1}\)) and normal haemoglobin (13 g dl\(^{-1}\)). Blood cultures were positive for \textit{S. pneumoniae}, as identified by MALDI-TOF MS completed by optochin disc and latex agglutination tests for rapid detection of \textit{S. pneumoniae}. The pneumococcal urinary antigen test was positive.

He was treated with the oral antibiotic amoxicillin-clavulanate acid (3 g day\(^{-1}\)). He developed first crackles in his right lung base. Chest radiology did not reveal any abnormalities. On day 3 of treatment, he developed painful erythema of the left thigh and left groin pain, and a fever of 39 °C. Laboratory investigations showed the following changes: an elevated C-reactive protein level (206 mg l\(^{-1}\)), an elevated leucocyte count (13 000 \(\mu^{-1}\), predominantly neutrophil granulocytes) and a normal platelet count (150 000 \(\mu^{-1}\)). Magnetic resonance imaging of the left hip revealed arthritis and an abscess of the left adductor brevis muscle (Fig. 2).

Surgical debridement of the adductor was performed using a direct internal approach. He was treated with a combination of intravenous amoxicillin and gentamicin for 3 days, and the treatment was then switched to intramuscular ceftriaxone (2 g day\(^{-1}\)) and oral rifampicin (900 mg day\(^{-1}\)) for 1 month. The patient was discharged at 4 weeks after surgical drainage and was prescribed the oral antibiotics amoxicillin (9 g day\(^{-1}\)) and rifampicin (900 mg day\(^{-1}\)) for 5 weeks without any intolerance being observed. We did not observe any relapse at the 6-month post-antibiotic follow-up. Echography at the 4-month post-antibiotic follow-up revealed a normal adductor brevis muscle.

**Discussion and Conclusion**

Here, we report two cases of pneumococcal pyomyositis managed in our centre. In PubMed/Medline, Web of Science and Google Scholar, we used the following keywords (`pneumococcal pyomyositis`, `\textit{Streptococcus pneumoniae}` and `pyomyositis`) with limits of the English and French language.

With the exception of paediatric cases, two hypocomplementaemia cases (Ekdahl \textit{et al}., 1995) and one case report of a 75-year-old woman that has been cited in previous studies (Collazos \textit{et al}., 1996), only 28 cases of pneumococcal pyomyositis have been reported in the literature (Table 1).

Of these 28 adult cases of pneumococcal pyomyositis that have been reported, four of the patients (14 %) were over 65 years of age and 16 (57 %) were immunodeficient, comprising three (11 %) patients with splenectomy, three (11 %) with HIV infection, seven (25 %) with chronic alcoholism, one with diabetes mellitus, one with Hodgkin’s lymphoma recurrence (this case had also underwent a splenectomy), one with metastatic thyroid neoplasia and one
with inflammatory rheumatism. Nine cases reported in the literature did not identify any risk factors. With regard to our two cases, the first had heterozygous sickle-cell disease and the second had no apparent immunodeficiency. Both of our patients had arthritis and one of our patients had a hip prosthesis, which has not been reported in the literature as a risk factor of pneumococcal pyomyositis. It should be considered as a risk factor for future observation.

Our two patients have not been vaccinated with pneumococcal vaccine. Zadroga et al. (2012) reported one case of pneumococcal pyomyositis in an immunocompromised patient who had previously received a *S. pneumoniae* vaccine. It would be interesting to study, in the perspective of infectious diseases, the effectiveness of pneumococcal vaccination in preventing this unusual disease.

Pneumococcal pyomyositis is frequently localized to the psoas muscle, as reported in 10 cases in the literature (36%) (Baddour et al., 2001; Bruggeling & Houwing, 1992; Collazos et al., 1996; Jimenez-Lucho & Quinn, 1985; Levine et al., 1982; Nakazato et al., 1999; Oliver et al., 2000; Orrison et al., 1977; Scott & Schmidt, 1989; Simpson et al., 2009) (Table 1). One of our cases had primary pyomyositis localized to the psoas muscle complicated with hip arthroplasty infection. In the other case, it was localized to the abductor muscle, which has not been reported previously.

Respiratory infection has been reported to be the main infection associated with pneumococcal pyomyositis, as reported in six cases in the literature (Collazos et al., 1996; Ejlertsen & Døssing, 1997; Granowitz et al., 1992; Nakazato et al., 1999; Oliver et al., 2000; Peertmans et al., 1993), followed by meningitis, which has been reported in five cases of pneumococcal pyomyositis (Levine et al., 1982; Orrison et al., 1977; Robertson-Mackay & al-Hillawi, 1993; Scott & Schmidt, 1989; Simpson et al., 2009).

Of the 28 cases of pneumococcal pyomyositis, 56% were documented by identification of *S. pneumoniae* in deep biopsy cultures. *S. pneumoniae* has rarely been isolated from blood cultures (only 4 of 18 cases) (Collazos et al., 1996; Ejlertsen & Døssing, 1997; Levine et al., 1982; Robertson-Mackay & al-Hillawi, 1993). One of our cases had a positive blood culture, and the other had a positive needle aspiration culture.

The two isolates of *S. pneumoniae* in our cases were identified by MALDI-TOF MS as described previously (Seng et al., 2013), as well as an optochin test and a latex agglutination test. To the best of our knowledge, MALDI-TOF MS [i.e. Microflex with Biotyper (Bruker Daltonik) and the VITEK MS system (bioMérieux)] is an early and accurate tool to identify *S. pneumoniae* from other *Streptococcus mitis* groups (Branda et al., 2013; Dubois et al., 2013; Werno et al., 2012).

In general, pneumococcal pyomyositis has a good outcome with antibiotic treatment (Table 1). Of the 28 cases of pneumococcal pyomyositis that have been reported, one died (Orrison et al., 1977), 20 were cured following...
## Table 1. Review of 28 cases of pneumococcal pyomyositis in adult patients reported in the literature

<table>
<thead>
<tr>
<th>Age (years)</th>
<th>Sex</th>
<th>Risk factor(s) and co-morbidities</th>
<th>Site</th>
<th>Clinical association</th>
<th>Outcome</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>47</td>
<td>M</td>
<td>Alcoholism, splenectomy, Gaucher’s disease</td>
<td>Iliopsoas abscess</td>
<td>Meningitis</td>
<td>Died</td>
<td>Antibiotics and surgical drainage</td>
</tr>
<tr>
<td>50</td>
<td>M</td>
<td>Alcoholism</td>
<td>Psoas abscess</td>
<td>Meningitis</td>
<td>Cured</td>
<td>Chloramphenicol, penicillin and surgical drainage</td>
</tr>
<tr>
<td>40</td>
<td>M</td>
<td>Alcoholism</td>
<td>Psoas abscess</td>
<td>Trauma</td>
<td>Cured</td>
<td>Penicillin and surgical drainage</td>
</tr>
<tr>
<td>66</td>
<td>F</td>
<td>None</td>
<td>Psoas abscess</td>
<td>Meningitis</td>
<td>Cured</td>
<td>Penicillin and surgical drainage</td>
</tr>
<tr>
<td>69</td>
<td>F</td>
<td>None</td>
<td>Psoas abscess</td>
<td>None</td>
<td>Cured</td>
<td>Clindamycin and surgical drainage</td>
</tr>
<tr>
<td>44</td>
<td>M</td>
<td>None</td>
<td>Lower leg and pectoralis abscess</td>
<td>Arthritis and pneumonia</td>
<td>Cured</td>
<td>Penicillin, ceftriaxone and surgical drainage</td>
</tr>
<tr>
<td>54</td>
<td>F</td>
<td>Inflammatory rheumatism</td>
<td>Gluteus abscess</td>
<td>Pneumonia</td>
<td>Cured</td>
<td>Ampicillin and surgical drainage</td>
</tr>
<tr>
<td>36</td>
<td>M</td>
<td>Splenectomy</td>
<td>Deltoid abscess</td>
<td>Meningitis</td>
<td>Cured</td>
<td>Penicillin and surgical drainage</td>
</tr>
<tr>
<td>Unknown</td>
<td>M</td>
<td>None</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
</tr>
<tr>
<td>72</td>
<td>F</td>
<td>None</td>
<td>Shoulder abscess</td>
<td>Respiratory tract infection</td>
<td>Cured</td>
<td>Penicillin</td>
</tr>
<tr>
<td>39</td>
<td>M</td>
<td>None</td>
<td>Unknown</td>
<td>Respiratory tract infection</td>
<td>Unknown</td>
<td>Unknown</td>
</tr>
<tr>
<td>53</td>
<td>M</td>
<td>None</td>
<td>Shoulder and biceps abscess</td>
<td>Arthritis, pneumonia</td>
<td>Cured</td>
<td>Penicillin and surgical drainage</td>
</tr>
<tr>
<td>Unknown</td>
<td>M</td>
<td>Thyroid neoplasia</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
</tr>
<tr>
<td>47</td>
<td>F</td>
<td>Alcoholism</td>
<td>Psoas abscess</td>
<td>Respiratory tract infection, septic shock</td>
<td>Cured</td>
<td>Imipenem-cilastatin, penicillin and surgical drainage</td>
</tr>
<tr>
<td>62</td>
<td>F</td>
<td>None</td>
<td>Psoas abscess</td>
<td>Cellulitis</td>
<td>Cured</td>
<td>Meropenem, cefuroxime and percutaneous drainage</td>
</tr>
<tr>
<td>64</td>
<td>M</td>
<td>None</td>
<td>Psoas abscess</td>
<td>Respiratory tract infection</td>
<td>Unknown</td>
<td>Metronidazole, tobramycin, cefotaxime and surgical drainage</td>
</tr>
<tr>
<td>Unknown</td>
<td>–</td>
<td>None</td>
<td>Iliopsoas and iliacus abscess</td>
<td>Sacroiliitis and osteomyelitis</td>
<td>Unknown</td>
<td>Unknown</td>
</tr>
<tr>
<td>Unknown</td>
<td>–</td>
<td>HIV</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
</tr>
<tr>
<td>32</td>
<td>F</td>
<td>HIV</td>
<td>Unknown</td>
<td>Cured</td>
<td>Antibiotics and surgical drainage</td>
<td></td>
</tr>
<tr>
<td>44</td>
<td>M</td>
<td>None</td>
<td>Unknown</td>
<td>Cured</td>
<td>Surgical drainage</td>
<td></td>
</tr>
<tr>
<td>68</td>
<td>M</td>
<td>Alcoholism</td>
<td>Unknown</td>
<td>Cured</td>
<td>Antibiotics and surgical drainage</td>
<td></td>
</tr>
<tr>
<td>48</td>
<td>M</td>
<td>Alcoholism</td>
<td>Unknown</td>
<td>Cured</td>
<td>Antibiotics and surgical drainage</td>
<td></td>
</tr>
<tr>
<td>59</td>
<td>M</td>
<td>Chronic obstructive pulmonary disease</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Cured</td>
<td>Antibiotics and surgical drainage</td>
</tr>
<tr>
<td>40</td>
<td>F</td>
<td>HIV</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Cured</td>
<td>Antibiotics and percutaneous drainage</td>
</tr>
<tr>
<td>37</td>
<td>M</td>
<td>None</td>
<td>Psoas abscess</td>
<td>Meningitis</td>
<td>Cured</td>
<td>Amoxicillin, ceftriaxone and metronidazole</td>
</tr>
<tr>
<td>55</td>
<td>M</td>
<td>Diabetes mellitus</td>
<td>Quadriceps and adductor abscess</td>
<td>Unknown</td>
<td>Cured</td>
<td>Amoxicillin, gentamycin and surgical drainage</td>
</tr>
<tr>
<td>47</td>
<td>M</td>
<td>Hodgkin’s lymphoma and splenectomy</td>
<td>Shoulder abscess</td>
<td>Unknown</td>
<td>Cured</td>
<td>Cephalaxin</td>
</tr>
<tr>
<td>52</td>
<td>M</td>
<td>Alcoholism, hepatitis C virus infection</td>
<td>Knee abscess</td>
<td>Trauma</td>
<td>Cured</td>
<td>Ceftriaxone and moxifloxacin</td>
</tr>
</tbody>
</table>

M, male; F, female.
treatment and the outcomes were unknown for seven. Surgical drainage was performed in 17 cases and needle aspiration in two (Table 1). Antibiotic treatment was given in 22/23 cases, and one case was treated with surgery only, without antibiotic treatment. β-Lactams were used in most cases. Multidisciplinary management is necessary for the successful treatment of pneumococcal pyomyositis. One patient in this case series required a second surgery with a large debridement because of septic shock and necrosis of the muscle (Guerrier et al., 2011). Our two patients benefitted from surgical drainage and long-term antibiotic treatment, after which they were cured and did not experience recurrence, as determined several months later. None of the 28 reported cases in the literature showed a reduced sensitivity to penicillin. Similarly, the pneumococcal isolates from our patients showed no resistance to penicillin.

In summary, pneumococcal pyomyositis is a rare disease. We have reported two new cases of this disease with favourable outcomes following long-term antimicrobial treatment and surgery debridement.

Acknowledgements

This study was approved by the institutional research ethics board, and a written informed consent form was signed by the patients. The authors declare they have no conflicts of interest.

References


