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CASE REPORT

Initially unrecognised group A streptococcal pelvic inflammatory disease in a postmenopausal woman

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ABSTRACT

Invasive group A *streptococcal* infection is a severe disease with high mortality. Invasive group A *streptococcal* infection may arise after pelvic inflammatory disease. Pelvic inflammatory disease in postmenopausal women is rare. Here, we report a unique case of a postmenopausal woman with fatal invasive group A *streptococcal* infection due to pelvic inflammatory disease and an extraordinary course of diagnosis.

KEYWORDS

Group A *streptococcus*, pelvic inflammatory disease, severe sepsis

What was known on this topic?
Invasive group A *streptococcal* infection is a severe and contagious disease. In postmenopausal women, invasive group A *streptococcal* infection due to pelvic inflammatory disease is extremely rare.

What does this add?
We describe a unique case of a postmenopausal woman with fatal group A *streptococcal* toxic shock syndrome due to pelvic inflammatory disease. This is also a cluster of fatal invasive group A *streptococcal* infection in one family.

INTRODUCTION

Streptococcal toxic shock syndrome (STSS) is a severe clinical illness characterised by shock and organ failure associated with a high mortality of up to 65%.[1] Apart from intensive medical support, STSS requires prompt recognition to enable source control and reduce toxin synthesis by the bacteria with intravenous penicillin and clindamycin.[2] In humans, the known reservoir for group A *streptococcus* (GAS) is the skin and mucous membranes. GAS causes various manifestations of acute infections including pharyngitis, tonsillitis, scarlet fever, cellulitis, erysipelas, necrotising fasciitis, and STSS. Pelvic inflammatory disease can be caused by GAS. Vaginal infection with GAS in adult women has been reported with menopausal vaginal atrophy as a prominent risk factor.[3] Currently there are no literature reports about pelvic inflammatory disease caused by GAS in postmenopausal women. We describe a case of unrecognised pelvic inflammatory disease caused by invasive GAS infection with toxic shock syndrome. The fatal occurrence of invasive GAS infection with necrotising fasciitis in a household member two weeks later became the lead to the diagnosis of the index case.

CASE REPORT

Patient A

A 72-year-old woman was admitted to hospital because of fever, abdominal pain, and diarrhoea for four days. She was on thyrax treatment for hypothyroidism. Upon presentation the patient was in shock with a heart rate of 120 beats/min and blood pressure of 70/40 mmHg. Her temperature was 37.3 °C, respiratory rate was 25

breaths/min, and oxygen saturation was immeasurable due to poor circulation. On physical examination there was diffuse abdominal pain without abdominal guarding. Her hands were blue-coloured; no other skin abnormalities were observed. Pelvic examination was not performed. Blood cultures could not be obtained because of her poor circulation. Initial vascular access was impossible and initial fluid resuscitation was achieved by the intra-osseous route. Urine analysis and culture were not taken because of anuria. Laboratory findings after initial resuscitation were: haemoglobin 8.3 mmol/l, platelets 149 x 10^9/l, white cell count 1.3 x 10^9/l, creatinine 246 µmol/l, bilirubin 38 µmol/l, aspartate aminotransferase 183 U/l, alanine aminotransferase 70 U/l, lactate dehydrogenase 461 U/l, alkaline phosphatase 54 U/l, gamma glutamyltransferase 99 U/l, creatinine kinase 5499 U/l, lactate 6.1 mmol/l, and procalcitonin > 100 ng/ml. Arterial pH was 7.19 and bicarbonate was 10 mmol/l. No abnormalities were seen on a chest X-ray. A CT scan of the abdomen showed a nonspecific swelling of the duodenum, descending and sigmoid colon and also free fluid in the pouch of Douglas. There were no signs of perforation, ischaemia or pelvic inflammatory disease. The patient was admitted to the intensive care unit (ICU) with a severe sepsis of unknown origin; antibiotic treatment included cefuroxime, metronidazole, and tobramycin. Despite treatment her condition deteriorated and she died 12 hours after admission.

Patient B

Two weeks after the death of patient A, her 82-year-old husband visited the emergency department because of excruciating pain in his right leg and shock. His medical history included diabetes mellitus and erythroderma due to cutaneous T-cell lymphoma. His medication included azathioprine, ciclosporin, and prednisone. He was diagnosed with necrotising fasciitis and after surgery he was admitted to our ICU. Blood cultures showed GAS. This patient developed multi-organ failure and died six days after admission.

Additional information

Autopsy was performed in patient A, showing signs of severe sepsis without a definite focus in the initial macroscopic report. Shortly after the death of patient B, a vaginal culture of patient A taken by the general practitioner because of vulvodynia a few days before her admission showed GAS. The final microscopic results of the post-mortem examination of patient A showed bilateral ovarian abscesses (*figure 1*) with Gram-positive cocci (*figure 2*). The isolate from the vaginal culture of patient A had not been stored and was therefore not available for genotypical comparison with the isolate from patient B.

Figure 1. *Detailed view of abscess formation with bacteria. Black arrow: abscess. Open arrow: bacteria. Haematoxylin and eosin (HE) 10 x*

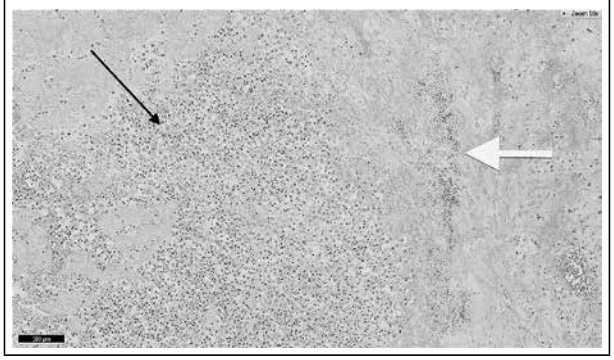
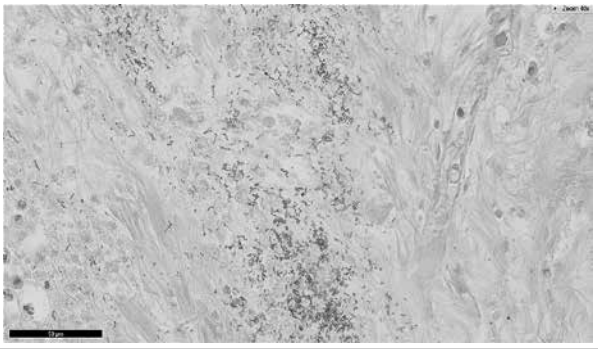


Figure 2. *Detailed view of coccoid bacteria. Haematoxylin and eosin (HE) 40 x*



DISCUSSION

This report describes two household members with fatal GAS infection. The cause of the fulminant septic shock of patient A was determined after the death of her husband, patient B. Patient A took care of the daily nursing of patient B because of his comorbidity, which may have facilitated the bacterial transfer. Although the presence of septic shock in patient A was clear, the diagnosis of toxic shock syndrome was not considered. Her history included an episode of blunt abdominal trauma due to a fall just before her fever started. Therefore, the first hypothesis was traumatic abdominal sepsis. In retrospect, the diagnosis might have been missed initially, and adequate source control may have been hampered due to several factors. Firstly, carefully reviewing the patient's history would have revealed the vulvodynia. Secondly, pelvic examination should have been performed as part of a meticulous physical examination, as this is warranted in all patients with septic shock of unknown origin. Thirdly, no blood cultures were taken, not immediately upon presentation and not after admission to the ICU.

This case report illustrates the importance of a complete medical history. If the recent history of vulvodynia had been known and the vagina culture had been examined in an affiliated laboratory, and thus noticed, we could have considered the diagnosis of STSS earlier. An extensive electronic health record would be an important solution resulting in improved communication between different organisations in our medical system.

Pelvic inflammatory disease caused by GAS in non-pregnant women is rare. A few cases about pelvic inflammatory disease and GAS infection in non-pregnant women are reported. The ages of these women ranged from 23-41 years.[4-7] There are no literature reports about postmenopausal women and pelvic inflammatory disease caused by GAS. The exact incidence of pelvic inflammatory disease in postmenopausal women is unknown; in one study less than 2% of women with tuba-ovarian abscess formation were postmenopausal.[8] Physiologically, the cervical mucus of postmenopausal women is more tenacious and serves as a mechanical barrier to ascending infections.[9] In postmenopausal women, microorganisms most frequently encountered in pelvic inflammatory disease or tuba-ovarian abscess were *E. coli* and *Klebsiella*.[8] In postmenopausal women with pelvic inflammatory disease, mortality rates of 25% have been reported despite adequate antimicrobial and surgical treatment.[10]

As we were not aware of the origin of the sepsis and its causative pathogen in patient A, her husband did not receive antibiotic prophylaxis. In the Netherlands, antibiotic prophylaxis is recommended for household members of a person with invasive GAS infection in case of necrotising fasciitis or STSS.[11] The efficacy of antibiotic prophylaxis for prevention of severe invasive GAS infection is not very well established. Although antibiotic prophylaxis is efficient in the eradication of GAS from the respiratory tract, firm evidence that antibiotic prophylaxis prevents invasive GAS infection is lacking.[12,13] However, no invasive GAS infection has been described in patients receiving antibiotic prophylaxis.[14] Risk factors for increased risk of invasive GAS infection are age exceeding 65 years, chronic cardiac and pulmonary disease, diabetes mellitus, varicella infection, human immunodeficiency virus (HIV) infection, cancer, usage of corticosteroids, injection drug use, and alcohol abuse.[15] Patient B had at least three of these risk factors.

In conclusion, we present a unique case of a postmenopausal woman with STSS due to pelvic inflammatory disease. We want to emphasise the severity of invasive GAS infection and possible difficulty in diagnosing this clinical illness. A complete medical history and meticulous physical examination is essential in postmenopausal women presenting with severe sepsis.

DISCLOSURES

This case report was written without external financial report. No conflict of interests declared.

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