



Ovarian necrosis due to *S. pyogenes* septic thrombophlebitis: A case report

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ABSTRACT

The incidence of systemic infection attributed to group A streptococci (GAS) is increasing, mainly in postpartum women. Such infections require multidisciplinary management and prompt treatment, but an atypical presentation can delay diagnosis.

We report the case of a 24-year-old woman admitted to the emergency department for evaluation. She had acute abdominal pain and fever 18 h after insertion of a levonorgestrel intrauterine device (IUD).

She had a normal vaginal delivery 45 days earlier, and no other significant medical background. In a few hours the symptoms worsened, with rapid progression towards multiorgan failure. Differential diagnoses of late ovarian thrombophlebitis and ovarian torsion were considered. Laparoscopic surgery revealed the absence of ovarian torsion. The microbiologic culture of the IUD showed colonization by GAS.

The sudden onset of shock-like symptoms in a postpartum woman with rapid progression towards multiorgan failure should prompt consideration of a diagnosis of GAS infection, so that appropriate treatment can be initiated to avoid the possible fatal consequences of this aggressive infection.

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1. Introduction

There has been a slow but steady increase in the incidence of severe infections with group A streptococci (GAS) in recent decades, particularly among postpartum women [1].

We report a case of streptococcal toxic shock after insertion of an intrauterine device (IUD) in an otherwise healthy young woman 45 days after vaginal delivery.

2. Clinical Case

A 24-year-old woman was admitted to the emergency department complaining of acute abdominal pain with sudden fever. She had a levonorgestrel IUD inserted without any complications 18 h earlier.

The only relevant medical history was a normal vaginal delivery 45 days previously; during her pregnancy, vagino-rectal cultures had been negative for group B streptococci.

The main symptoms at the time of admission were lower abdominal pain of mild intensity that began after insertion of the IUD, fever (39 °C), nausea and vomiting.

Abdominal tenderness was present in the lower quadrants but there was no evidence of peritonitis; complete blood count (CBC) indicated

moderate neutrophilia but no leukocytosis. The IUD was normally inserted in the uterine cavity, with no evidence of pelvic or adnexal masses or collections on gynecological ultrasound scan.

The initial diagnosis was pelvic inflammatory disease (PID). A 14-day course of oral antibiotic treatment with doxycycline was therefore prescribed and a dose of ceftriaxone was given intramuscularly.

Twelve hours later the patient was readmitted to the emergency department with general malaise, persistence of fever and intense abdominal pain localized to the lower right quadrant, as well as hypotension.

The IUD was removed and sent for microbiological culture. A gynecological ultrasound scan showed no pelvic abscess or free fluid in the lower abdomen. Hypotension and tachycardia were present as well as leukopenia, neutrophilia, elevated levels of C-reactive protein (CRP) and elevated levels of liver enzymes. The serum lactate level was 3.5 mmol/L, with prolonged coagulation times, in the context of initial disseminated intravascular coagulation (DIC).

Antibiotic treatment with meropenem was initiated.

An abdominal ultrasound scan revealed enlargement of the right ovary (45 × 45 mm in diameter) with an increase in echogenicity compared with the left ovary, suggestive of ovarian torsion.

A computerized tomography (CT) scan with intravenous contrast (Fig. 1) confirmed the ultrasound findings, with absence of blood flow in the right ovarian veins, reinforcing the diagnosis of ovarian torsion. Consequently, laparoscopic surgery was performed.

Upon laparoscopic exploration of the abdominal cavity, the macroscopic findings included an enlarged right ovary that appeared to be

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Fig. 1. Abdominal CT scan with IV contrast in coronal plane. Enlarged right adnexa with haemorrhagic changes and no contrast present, suggestive of necrosis. Absence of venous blood flow and engorgement of the venous territory on the right ovarian veins can be seen; suggestive of thrombophlebitis.

necrotic, with no sign of torsion but with evident thrombosis of the infundibulopelvic and ovarian suspensory ligaments (Fig. 2). There were no other pathological findings. A right adnexectomy was performed and the surgical piece was sent for anatomopathological diagnosis.

No thromboprophylaxis was initiated in the immediate postoperative period due to the presence of DIC with international normalized ratio (INR) 2.72. Low-molecular-weight heparin was initiated when coagulation parameters normalized.

The patient was admitted to an intensive-care unit for 24 h after surgery and treatment with meropenem was continued. She continued to



Fig. 2. Necrotic right ovary. Thrombotic mesovary with no evident torsion.

improve and was discharged on the 5th postoperative day with no further complications.

On the 6th postoperative day the results from the microbiological culture of the IUD were positive for *Streptococcus pyogenes* sensitive to penicillin.

3. Discussion

The patient's initial diagnosis was of pelvic inflammatory disease (PID), because of the presence of abdominal pain, fever, general malaise, tenderness to uterine and adnexal palpation, and neutrophilia in the context of the insertion of an IUD hours before. PID is usually related to the presence of genital bacteria in the otherwise sterile upper genital tract and the development of an infection in the fallopian tubes. The pathogens most frequently associated with this type of infection are *Neisseria gonorrhoeae* and *Chlamydia trachomatis* [2].

The rapid worsening of the patient's clinical condition, and the appearance of clinical signs of shock and multiorgan failure, led to the suspicion that a more serious infection might be involved and treatment was initiated with meropenem (a wide-spectrum antibiotic).

The CT and ultrasound findings were consistent with ovarian torsion [3,4]. Ovarian torsion is uncommon; it typically presents with abdominal pain in the lower quadrants or lateralized to the iliac region of the abdomen on the side of the affected ovary. Usually an adnexal mass, either solid or cystic, can be identified on ultrasound; the mass produces torsion of the enlarged ovary around its vascular supply, and abdominal pain is a typical symptom, often of sudden onset. The torsion can spontaneously reverse, in which case the patient may present with intermittent symptoms and often spontaneous resolution of the torsion [5].

If a gynecological ultrasound scan leads to the suspicion of ovarian torsion, a CT scan with intravenous contrast can confirm the diagnosis or suggest another ovarian pathology that should be considered in the differential diagnosis. The other pathology considered in this case was septic ovarian thrombophlebitis (SOT), given that the patient had a vaginal delivery 45 days earlier and that she had no risk factors for ovarian torsion and that no adnexal masses were identified on the initial gynecologic ultrasound scan. The incidence of SOT is around 1/600 to 1/2000 deliveries. Symptoms typically begin within 15 days of delivery, with a higher incidence in the first postpartum week. They include abdominal pain of moderate to severe intensity, low-grade fever and general malaise. In 90% of cases it develops on the right side, possibly due to reverse blood flow in the right ovarian veins after changes in the circulation during pregnancy [6].

In the typical clinical scenario, SOT produces no clinical signs of shock, nor of multiorgan failure, and the course is milder than in the case presented here.

The diagnosis of SOT is often made after radiologic imaging, such as CT with intravenous contrast, which has a sensitivity of 100% for this ovarian pathology.

In the case presented, there was a high radiologic suspicion of ovarian torsion and the rapid onset of septic shock with multiorgan failure led to exploratory gynecologic laparoscopy. There was no evidence of torsion of the vascular supply of the right ovary, even though the macroscopic appearance was necrotic with thrombosis of the ovarian and uteroovarian arteries (Fig. 2).

The possibility of spontaneous de-torsion was dismissed due to the rapid progression of the clinical symptoms, with septic shock developing within 18 h of insertion of the IUD. De-torsion of the ovary would have led to spontaneous dramatic improvement in the clinical condition and disappearance of abdominal pain.

The presence of thrombi in the ovarian veins, evident during surgery, made the diagnosis of SOT very likely but it was initially dismissed in light of the rapid progression towards multiorgan failure.

The results of the IUD microbiologic culture, in which a group A streptococcus was isolated, led to a definitive diagnosis of toxic streptococcal shock syndrome (TSSS).

Historically, beta-hemolytic group A streptococci (GAS) were usually responsible for postpartum sepsis. GAS were initially identified by Pasteur in the 19th century and were considered responsible for two-thirds of postpartum deaths in the 18th and 19th centuries [7].

The incidence of postpartum sepsis attributable to GAS in modern times has decreased to around 0.06 cases per 1000 live births. Nonetheless, in recent decades there has been a slight increase in the incidence of systemic infection by GAS, and there have been epidemic outbreaks in health-care centers, all involving the same strain.

It appears that the increase in the virulence of some strains of GAS is related to the production of exotoxin A and, more specifically, the re-emergence of strains M1 and M3 [8,9].

In TSSS, progression towards multiorgan failure occurs within hours of the initial onset of symptoms; consequently, a mortality rate of nearly 60% is reported in some studies. TSSS is directly related to the production of endotoxin A. The condition progresses through three clear stages. In the first, there is a proliferation of GAS, with an increasing number of bacteria at the inoculation site, but with non-specific, mild symptoms such as local erythema and pain. In up to half of cases the inoculation site is not evident. Many patients at this first stage take non-steroidal anti-inflammatory drugs (NSAIDs) for treatment of local pain, up to 92% of patients in one study [10]. This, though, can delay the diagnosis and so allow the GAS to continue proliferating and the patient progresses to the next stage of the disease.

The second stage is characterized by the production of exotoxin A by the GAS. This triggers the release of cytokines locally at the inoculation site and increases the capillary permeability, which allows the passage of exotoxin A directly into the bloodstream. At this stage the predominant clinical features are vomiting, nausea, diarrhea, general malaise, shivering and fever. Without treatment, the TSSS progresses to the third stage, characterized by tachycardia, hypotension, high-grade fever, leukopenia and alteration of clotting times; there is a high risk of cardiomyopathy. Symptoms and signs of hepatic or cardiac failure may be present.

According to case reports, being in the postpartum period is considered a risk factor for the development of GAS infection, as is gynecologic instrumentation, such as the insertion of an IUD [11], dilation and curettage [12] or hysteroscopies [13]. There are even case reports of TSSS secondary to vulvovaginitis [14].

If a GAS infection leads towards septic shock, treatment usually consists of beta-lactam antibiotics and basic vital support until the elimination of the exotoxin A allows for clinical improvement.

The use of clindamycin favors quicker resolution due to the interruption of exotoxin production [8].

Life support measures usually include fluid therapy, replacement of blood and blood components such as platelets or coagulation factors, and in some serious cases even dialysis for faster toxin elimination.

Where there is no clinical improvement after antibiotic treatment is begun, surgery should be considered, because in most of the case reports and studies reviewed a hysterectomy or adnexectomy was needed to achieve clinical resolution of the TSSS.

Contributors

Alicia Guntiñas contributed to analysis and discussion of the case and drafted the paper.

Janette L. Kirk contributed to analysis and discussion of the case and drafted the paper.

M. Teresa Blanco contributed to analysis and discussion of the case and drafted the paper.

Javier Pérez contributed to analysis and discussion of the case.

Miguel A. Rodríguez contributed to analysis and discussion of the case.

Conflict of Interest

The authors declare that there is no conflict of interest regarding the publication of this case report.

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