

Sepsis-Induced Disseminated Intravascular Coagulation:

A Rare Complication of Sacrococcygeal Pilonidal Sinus Disease

THE TERM "SACROCCYGEAL PILONIDAL SINUS" describes one or more midline openings in the natal cleft. It was first reported by Mayo in 1833, and although in the 1950s pilonidal sinus disease (PSD) was thought to be of congenital origin, it is now widely accepted to be an acquired disorder. PSD is a common hair-containing sinus or abscess, and initially 50% of patients present with a pilonidal abscess. During the last century physicians focused their efforts on etiology and medical management of PSD (1–3).

A 22-year-old white male, a beach cleaner by profession, was admitted to the emergency unit of the Clinical Hospital of Split because of fever and coma. In the previous few weeks he had occasionally complained of headache, but his medical history, elicited from the patient's mother, did not reveal a history of illness or drug abuse, or a family predisposition to any disease. The day before admission his body temperature was 37.3°C and he complained of headache and dizziness. On admission, he was perspiring profusely, his pulse rate was 144/min, blood pressure was 100/70 mm Hg, respiratory rate was 27/min, and body temperature was 41.5°C. He was in a coma, with a Glasgow Coma Scale (GCS) score of 3. His body mass index was 32.

Upon admission, he vomited coffee-ground-colored matter (hematemesis), so nasogastric tube lavage was performed to clear the stomach of clots and fresh blood. Endoscopy revealed erosions of the middle and lower parts of the esophagus and duodenal postbulbar segment. Nasogastric suction content sent for chemical-toxicology analysis was negative. The patient's laboratory parameters indicated sepsis with disseminated intravascular coagulation: prothrombin time 41% (normal range 75–100), INR 1.65 (0.9–1.08), activated partial thromboplastin time 45 s (21–32), platelet count $33 \times 10^9/L$ (140–440), bands 26% (0–5), fibrinogen 4.07 g/L (2–4.1), D-dimer 1,535 $\mu g/mL$ (<250), factor V 25% (60–140), factor VII 30% (60–140), factor VIII 48% (50–200), factor X 58% (60–140), factor XII 44%

(60–140), antithrombin III 70% (80–130), and C-reactive protein 188 (0–3).

Urine sent for qualitative drug abuse screening (benzodiazepines, opiates, methadone, amphetamines, cocaine, barbiturates, phencyclidine and cannabinoids) and a test for alcohol in the blood were negative. Blood samples for microbiological analysis were taken. Urine and chest X-ray didn't reveal any pathological findings. Spinal fluid sent for microbiological analysis was negative. Physical examination revealed inflamed sacrococcygeal pilonidal sinus (erythema and swelling over the sacrococcygeal region), but physicians didn't consider the PSD to be a cause of sepsis.

The patient was intubated and a Foley-urine catheter was placed. Computed tomography of the brain revealed massive edema, especially in the mesencephalon, with a smaller amount of subarachnoidal and marginal petechial hemorrhage. The patient was sent to the intensive care unit, where mechanical ventilation (IPPV), TV=750 mL, f=12, FiO₂=0.40), crystalloid and colloid solutions, dexamethasone 4 × 1 amp i.v., analgesic sedation medications (fentanyl, midazolam), and 6 U of packed platelets were administered. Antibiotics (cefepim 3 × 2 g i.v., gentamicin 240 mg i.v., and metronidazole 3 × 500 mg i.v.) were given.

On the second day, an inflamed pilonidal sinus (6 × 4 cm) of the sacrococcygeal area became more apparent. Before the surgical procedure, purulent sinus samples were sent for microbiological analysis. Radical excision of the pilonidal sinus down to the presacral fascia was done, without primary closure of the wound. Laboratory examination showed pathological values: AST = 1920 IU/L (1–38), ALT = 2133 IU/L (1–41), urea 10 mmol/L (2.5–8.3), creatinine 231 mmol/L (54–116), lactate dehydrogenase = 2,833 IU/L, and still low platelets = $31 \times 10^9/L$, so 6 U of packed platelets and low-molecular-weight heparin (enoxaparine, Clexane, Aventis Pharma, 0.8 mL/80 mg bid s.c.)

were administered. HIV and hepatitis B and C viral infection were excluded.

Bacteroides fragilis was isolated from hemoculture and sinus fluid samples, so amoxicillin and clavulanic acid (3 × 1.2 mg i.v.) and clindamycin 3 × 600 mg i.v. were introduced. Brain CT showed reduced edema, without sign of intra- or extracerebral hemorrhage.

After 7 days in the intensive care unit, the patient was hemodynamically stable, in good condition, with laboratory values within the normal range. Microbiological analysis of blood samples and purulent material from the sinus identified the same pathogenic bacteria, *B. fragilis*, so we concluded that acute inflamed pilonidal sinus was the cause of sepsis. Bioptic material showed encapsulated nests of hair with purulent infection. The wound healed, without any sign of inflammation spreading to the surrounding area. Histological analysis revealed foreign body granuloma. After 10 days the patient was released and there were no long-term sequelae.

Although pilonidal disease has been known since 1833, only a few published reports about life-threatening complications of PSD have appeared. Verdu et al. described the rare case of a male diabetic patient with a recurring pilonidal cyst, who developed a lumbar osteomyelitis and epidural abscess three weeks after pilonidal cyst excision with epidural anesthesia, with a fatal outcome despite emergency treatment (4). Borer et al. described septic arthritis due to *B. fragilis* after pilonidal sinus resection, in a patient with rheumatoid arthritis (5). Velitchkov et al. published the rare case of a previously healthy adult male who developed necrotizing fasciitis and toxic shock syndrome, both of which complicated neglected sacrococcygeal PSD associated with *Streptococcus pyogenes* and *B. fragilis* (6). Shlasko et al. described a case of toxic shock syndrome due to staphylococcal infection occurring in a previously healthy young man after elective surgery for a pilonidal cyst (7). In 1982 Friis and Madsen described recurrent toxic shock syndrome in a man, after surgery for a pilonidal cyst, and Cobb et al. published a case report of toxic-shock syndrome in a young man with a pilonidal abscess (8, 9).

Risk factors for PSD are white race, male sex, family predisposition, increased sweating, activity associated with sitting and buttock friction, sedentary lifestyle, poor personal hygiene, obesity, and local trauma. The incidence rate of pilonidal disease is approximately 0.7%, the disease occurs 2.2–4 times more often in men than women, and the average age of patients at presentation is 21 years (1, 2). PSD involves a combination of skin

and perineal flora. *Staphylococcus aureus* is the most common organism and *B. fragilis* is the most common anaerobe. Although numerous randomized clinical studies have evaluated different treatments, no clear consensus has been reached as to the optimal medical or surgical treatment (10).

Until now there have been only four published case reports about toxic shock syndrome in male patients with PSD, usually after surgical treatment, and associated with *Staphylococcus*, *Streptococcus* and *B. fragilis* infections. We have presented a case of disseminated intravascular coagulopathy (multiple organ failure: liver and renal failure, and massive brain edema with subarachnoidal and marginal petechial hemorrhage, and erosions of esophagus and duodenum) in a patient with severe sepsis due to neglected pilonidal abscess, which successfully responded to antibiotics, and symptomatic and surgical treatment. *B. fragilis* was isolated from his hemoculture and sinus fluid samples.

Previously published case reports of unusual complications of this illness have also concerned males (4–9), perhaps because the incidence of PSD is higher among males. The case is remarkable for its favorable and quick outcome and its rarity.

The case serves as a reminder that neglected pilonidal abscess can lead to unusual and life-threatening consequences, even in a previously healthy patient. When a patient presents with sepsis of unknown source, physicians should consider PSD as a possible cause.

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