Primary Peritonitis Associated with Streptococcal Toxic Shock-Like Syndrome: Report of a Case

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Abstract
Several reports over the past 15 years describe severe group A streptococcal infections causing septic shock, soft-tissue necrosis, and multiple organ failure; a phenomenon known as streptococcal toxic shock-like syndrome (TSLS). However, primary peritonitis associated with TSLS is rare. We report the case of a 40-year-old man admitted with pain in both thighs, hypotension, and severe abdominal pain. His daughter had been diagnosed with streptococcal pharyngitis 3 days earlier. We performed an emergency laparotomy for peritonitis, and culture of the ascites was positive for group A β-hemolytic streptococcus (GAS). Further serotyping of the isolated GAS strain revealed the T-type 22 and the pyrogenic exotoxin gene, spe-C. The criteria for TSLS were clearly met, including the isolation of GAS from ascites, hypotension, liver failure, renal failure, coagulopathy, myositis, and a generalized erythematous macular rash with desquamation.

Key words Primary peritonitis · Toxic shock-like syndrome · Streptococcus pyogenes

Introduction
Many recent reports describe a disease caused by group A streptococcus (GAS) infection, characterized by sudden septic shock and necrosis of the soft tissues, which has been termed toxic shock-like syndrome (TSLS).1 Primary peritonitis rarely occurs in association with TSLS and, to our knowledge, only five cases have been described. We successfully treated a patient with TSLS associated with primary peritonitis, in whom culture of ascitic fluid obtained at laparotomy grew GAS.

Case Report
The patient was a 40-year-old Japanese man, whose 9-year-old daughter had been diagnosed by a local physician as having hemolytic streptococcal pharyngitis 3 days earlier. The patient began to experience femoral pain at about 18:00, a fever of 38°C at about 19:30, and vomiting and watery diarrhea at midnight. The next morning his temperature rose to 38.7°C and he visited the same physician, who gave him fluid replacement and an intestinal drug. Within a few hours of returning home, the femoral pain began to extend to both sides and became more severe, developing into numbness of both legs. He also began to suffer right lower abdominal pain which gradually increased in severity. He visited the same physician again at 21:00, who noted signs of peritoneal irritation and shock, with a systolic blood pressure of 66 mmHg on palpation. The patient was immediately transferred to our hospital. On admission, he was fully conscious and in obvious distress, complaining of severe lower abdominal pain which gradually increased in severity. He visited the same physician again at 21:00, who noted signs of peritoneal irritation and shock, with a systolic blood pressure of 66 mmHg on palpation. The patient was immediately transferred to our hospital. On admission, he was fully conscious and in obvious distress, complaining of severe lower right abdominal pain. His blood pressure was 87/40 mmHg, for which 10 µg/kg per minute of dopamine was continuously infused. His heart rate was 115 beats/min and regular, his respiratory rate was 20/min, and his heart and breath sounds were normal. He was anuric. There was marked tenderness, rebound tenderness, and muscular defense over the entire abdomen, mainly in the lower right abdomen, with no audible bowel sounds. He complained of pain at rest and tenderness in both femoral regions, especially in the right anterior area. An erythematous macular rash was noted, mainly over his chest. Blood counts revealed no
anemia, a white blood cell count of 3400/µl, and a C-reactive protein level (CRP) of 22.4 mg/dl. Laboratory data showed hepatic and renal dysfunction, with a very high creatine kinase (CK) level of 2984 IU/l (Table 1). Blood gas analysis showed metabolic acidosis. An abdominal X-ray film showed marked gaseous distention of the small intestine and paralytic ileus. There was no free air in the abdomen. An abdominal computed tomography (CT) scan showed distention of the small intestine, and an ultrasonogram of the abdomen showed no movement of the contents of the small intestine. In view of the physical and laboratory findings, we made a preoperative diagnosis of peritonitis caused by appendicitis, perforation of the gastrointestinal tract, or gastrointestinal tract necrosis, and an emergency operation was arranged. We performed laparotomy through a median incision in the lower abdomen, which revealed a small amount of yellow cloudy ascites. Intra-abdominal exploration revealed no appendicitis, no gastrointestinal tract perforation or necrosis, and no abnormalities in the other abdominal organs. After the ascites was sampled for culture, the abdomen was irrigated and an abdominal drain was placed. Although the operative findings indicated peritonitis of unknown origin, the fulminating course of the patient urged us to start treatment for septic shock. Dopamine was required to maintain blood pressure immediately after the operation, but was discontinued when his blood pressure stabilized 3 days postoperatively. Multiple organ failure (MOF) and disseminated intravascular coagulation (DIC) developed early during his postoperative course. However, these complications were successfully treated with intensive treatment, which consisted of gabexate mesilate, heparin sodium, freeze-dried concentrated human antithrombin III, the transfusion of platelets and fresh frozen plasma (FFP), antibiotics and freeze-dried sulfonated human normal immunoglobulin, endotoxin absorption therapy, and continuous hemodiafiltration (CHDF) (Fig. 1). The erythematous macular rash observed on the chest preoperatively extended from the side of the trunk to the inguinal region and disappeared by about 2 weeks. An erythematous macular rash with erosion and desquamation appeared on the scrotum and around the anus, but resolved within about 4 weeks. Thick desquamation was seen on both palms and soles. Culture of the ascites collected perioperatively grew GAS. The femoral pain our patient had suffered preoperatively was conceivably due to myositis because of increased CK. The diagnosis of TSLS was confirmed by characteristic findings, including isolation of GAS from ascites, hypotension, liver failure, renal failure, coagulopathy, myositis, and a generalized erythematous macular rash with desquamation, which met the draft diagnostic criteria of the Centers for Disease Control (CDC).2 Because these symptoms and signs developed within about 24h, the draft diagnostic criteria created by the study group of the Japanese Ministry of Health, Labor, and Welfare3 were also met. Moreover, GAS was not detected in repeated cultures of pharyngeal exudate, sputum, urine, or skin after the surgery. The isolated GAS had T antigens of type 22 and streptococcal pyrogenic exotoxin (SPE)-C producing type. The patient responded well to the intensive treatment and his postoperative course was favorable. He returned to this daily life without any sequelaes.

**Discussion**

Toxic shock-like syndrome was first described by Cone et al. in 1987.1 With successive reports in North America, the CDC proposed a draft of diagnostic criteria in 1993 (Table 2).2 The first case of TSLS in Japan was documented by Shimizu et al. in 1993,4 since when more than 200 cases have been reported in this country to date.5 The “Law Concerning the Prevention of Infectious Diseases and Patients with Infectious Diseases” introduced in April 1999 designates TSLS as a Category IV infectious disease in Japan. The ratio of men to women with TSLS in Japan is about 2:1, and there is a higher incidence of this disease in winter.5 Our patient was a man and the disease developed in March. It is postulated that SPE serves as the superantigen and leads to the exuberant inflammatory response associ-