Case Report

Postpartum Toxic Shock Syndrome: A Report of a Case

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We describe a case of a 29-year-old Japanese woman with toxic shock syndrome which occurred 6 days after a normal vaginal delivery. This is the second case of postpartum toxic shock syndrome in Japan, so far as we know. Staphylococcus aureus was isolated from the uterine cavity. The isolate was coagulase type II and it produced both enterotoxin C and toxic shock syndrome toxin-1. The clinical course and laboratory data for this patient were in conformance with previously reported typical postpartum toxic shock syndrome, except for the onset time after delivery, because the previously reported postpartum TSS cases were clearly divided into two groups, early onset within 3 days of delivery and late onset 2 or more weeks after delivery.

Key Words: Toxic shock syndrome, Postpartum, Staphylococcus aureus, Toxic shock syndrome toxin-1.

Toxic shock syndrome (TSS), which was first reported in 1978, is characterized by high fever, erythematous rash with subsequent desquamation, shock, and multiple organ involvement. Staphylococcus aureus is thought to play a major role in the pathogenesis of TSS by producing exotoxin. Although TSS is known to occur predominantly in menstruating women who wear tampons, the proportion of cases not associated with menstruation has been gradually increasing. We describe here a case of a woman with postpartum TSS occurring 6 days after delivery.

CASE REPORT

A 29-year-old Japanese woman (gravida 4, para 2, abortus 1) was admitted to a maternity clinic for a term delivery. She had been a tampon user before pregnancy but she had never experienced any abnormal symptoms during menstruation. She had a normal vaginal delivery of a healthy child on May 20, 1986. She used a perineal pad and no intravaginal packings after delivery. On May 26, she had a fever of 38°C and developed watery diarrhea and general myalgia. Within one day her temperature had risen to 40°C and she developed generalized erythematous non-pruritic skin rash. Antibiotics were administered without resolution of the symptoms. Hypotension and restlessness developed, and she was transferred to the Hiratsuka City Hospital on May 28.

On admission, her heart rate was 120/min, respiration rate was 24/min, blood pressure was 60/40 mmHg, and body temperature was 37.5°C. There was diffuse edematous erythema over her trunk, face and extremities without petechiae. The palpebral conjunctiva and pharynx were hyperemic. No lymph nodes were palpable. The respiratory sound was normal. Heart sounds were normally audible. There was diffuse abdominal tenderness with slight guarding without rebound tenderness. Pelvic examination disclosed that the uterus was relatively large but not tender. The external genitalia were normal.

The following laboratory data were abnormal; (1) leukocytosis with absolute reduction of lymphocytes (white blood cells were 19,400/cmm with 49% segmented neutrophiles, 38% band forms, 9% metamyelocytes, 1% myelocytes, 1% lymphocytes and 2% monocytes), (2) hypoproteinemia of 4.2 g/dl and hypoalbuminemia of 2.0 g/dl, (3)
renal insufficiency (BUN and serum creatinine were 70 mg/dl and 2.6 mg/dl, respectively) associated with trivial proteinuria and 20 to 30 white blood cells and 1 granular cast per high power field, (4) hypocholesterolemia of 108 mg/dl, (5) serum electrolyte imbalances were: sodium, 125 mEq/L; potassium, 3.8 mEq/L; chloride, 98 mEq/L; calcium, 6.6 mg/dl; phosphate, 4.7 mg/dl; magnesium, 1.4 mg/dl. (6) mild anemia (hemoglobin, 11.4 g/dl) and mild thrombocytopenia (platelets, 12 x 10^4/cmm).

The bacterial flora of pharyngeal mucus and stool were normal, and the urine and blood cultures were negative, but a utero-cervical culture yielded a massive growth of Staphylococcus aureus.

The differential diagnosis at admission included septic shock, drug allergy and toxic shock syndrome.

She was treated with intravenous fosfomycin (2 g every 8 hours) and tobramicin (60 mg every 8 hours) and dexamethasone (16 mg every day). Despite the administration of antibiotics, fresh frozen plasma, massive crystalloid and dopamine hydrochloride, she remained febrile and hypertensive, and the erythema continued till the end of May. Severe thrombocytopenia developed (16,000/cmm on May 30), but no signs or symptoms of disseminated intravascular coagulation were observed. A test for anti-platelet antibody was negative in serum, and bone marrow aspiration showed normal megakaryocyte count. Although she did not complain of upper abdominal pain and an ultrasonogram showed no signs of acute pancreatitis, the serum amylase became gradually elevated (2096 u/L on June 3) and the pancreatic-type isoenzyme was dominant. Urinary amylase output was more than 4000 u/day.

By June 2, she became afebrile and normotensive, and the erythematous skin rash disappeared. Within a few days, desquamation of the skin developed over the trunk and then thick membranous desquamation was observed on the palms and soles. The Staphylococcus aureus isolated from the uterine cavity was found to be coagulase type II and it produced both enterotoxin C and toxic shock syndrome toxin-1 (TSST-1). This history and the findings were compatible with the diagnosis of toxic shock syndrome.

Because the utero-cervical cultures remained positive for Staphylococcus aureus despite the clinical improvement, endometrial curettage and uterine cavity lavage with povidone iodine were performed several times and minocycline was administered every day. On June 10, the utero-cervical cultures became negative. All abnormal laboratory data became ameliorated except for amylase, which was at a level slightly higher than normal. She was discharged on June 25, and was followed up as an outpatient. No recurrence was observed to the end of 1986. Her baby was doing well and no infectious diseases were observed.

**DISCUSSION**

We describe a case of a 29-year-old woman with postpartum TSS occurring 6 days after a normal vaginal delivery.

Todd et al, first reported 7 cases of TSS in 1978, involving children aged 8 to 17 years old1). In 1980, there was a large increase in the number of cases in young menstruating women who were using tampons in the United States. A case definition of TSS for epidemiologic study was published by the Center for Disease Control in 19804) and was revised in 19825). There are six criteria for the diagnosis of TSS; (1) fever over 38.9°C, (2) diffuse macular erythematous skin rash, (3) desquamation, (4) hypotension, (5) involvement of three or more of the organ systems including gastrointestinal, muscular, mucous membrane, renal, hepatic, hematological, neurological systems, (6) absence of evidence of other causes of illness. Our patient met all these criteria, making the diagnosis of TSS definite.

Although the pathogenesis of TSS is not well understood, staphylococcal exotoxin has been thought to play a major role1). Over 90% of the staphylococcal strains isolated from TSS cases were found to produce a common toxin6), which has been given the name TSST-17). Though the toxin is pyrogenic in rabbits, it has not been proved to develop erythematous rash and hypotension. The role of TSST-1 in human TSS is still debated, and the existence of another exotoxin has also been postulated8). However, TSST-1 is a useful marker in the diagnosis of TSS. Staphylo-
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coccus aureus isolated from this patient also was proved to produce TSST-1 in high concentration by the reversed passive latex agglutination method.

It is true that TSS has occurred predominantly in menstruating women, but the proportion of cases not associated with menstruation has been increasing steadily since 1980. About 30% of the cases were not associated with menstruation in 1983. TSS is now recognized in association with staphylococcal infection at any sites in many settings.

So far as we have found in the literature, 30 cases of postpartum TSS have been reported, including only one Japanese case. TSS occurred after vaginal delivery (18 cases), cesarean section (5 cases), therapeutic abortion (2 cases) and no information on the type of delivery (5 cases). All cases except one are divided into two groups according to onset time. One group includes the patients with onset within 3 days of delivery and the other group includes the patients with onset 2 or more weeks after delivery. The one exception is a case occurring 10 days after delivery associated with postpartum staphylococcal endometritis. In patients with early onset nearly all occurred within 24 hours after delivery. In these cases intrauterine infections such as chorioamnionitis may have been preexisting and the staphylococcal exotoxin may have been absorbed immediately after delivery from endometrium damaged on delivery. In cases of late onset almost all patients used tampons after delivery for lochia, therefore the mechanism of developing TSS may be the same as that in menstruating cases.

In the present patients, there was no evidence of preexisting intrauterine infection and no history of postpartum tampon use. Postpartum intrauterine manipulation was thought to be the most probable cause of intrauterine staphylococcal infection, and this may have been the cause of the atypical onset time. Another unexplained point is that she lacked clinical sympotms of intrauterine infection such as pain and tenderness of uterus or abnormal lochia, and the infection was relatively resistant to systemic antimicrobial therapy combined with endometrial curettage and lavage. It is still unknown whether this atypicality in the infection had some relation to the exceptional onset time after delivery.

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REFERENCES