CASE REPORT

A Case Report on Toxic Shock Syndrome Complicated with Symmetrical Peripheral Gangrene after Insertion of Intrauterine Contraceptive Device

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Abstract

Background: Toxic shock syndrome (TSS) is a lethal complication, which is rare. It is mediated by a toxin and can rapidly produce shock and multiorgan failure. It can lead to symmetrical peripheral gangrene (SPG), which is defined as ischemia or two or more limbs without obstruction of a large vessel or any vasculitis. Toxic shock syndrome can occur in some other clinical situations as well, like burns, nasal packing, and menstrual issues. The incidence of this syndrome is extremely low in gynecology, especially after intrauterine contraceptive device (IUCD) insertion.

Case description: A 34-year-old female patient (Para 4 plus1), postpartum with lactational amenorrhea, was admitted as uterine perforation from the ER after IUCD insertion. The main complaints were abdominal pain, nausea, and vomiting. The patient's vital signs at admission were normal, but she developed shock within the next few hours. The patient was started on IV fluids, antibiotics, and analgesia and kept under observation. The patient became hemodynamically unstable. Official ultrasound revealed IUCD inside the uterus, marked fluid reaching up to Morrison's pouch, left ovarian cyst 2 × 2 cm, and pouch of Douglas contained free fluid. The patient underwent exploratory laparotomy. Mild to moderate hemoperitoneum was seen. Intrauterine contraceptive device was removed. She developed TSS, disseminated intravascular coagulation (DIC), multiple organ dysfunction syndrome, and SPG in the next few days. Multidisciplinary teams were involved in the management. The patient experienced discoloration of the hands, which worsened to dry gangrene.

Conclusion: Symmetrical peripheral gangrene due to this syndrome is very rare in gynecology. The mortality rate due to TSS is high, and SPG may also follow it. Early diagnosis and aggressive treatment are important to save the patient.

Clinical significance: Symmetrical peripheral gangrene caused by TSS following IUCD insertion is a rare and unsuspected occurrence. This fact should be kept in mind while managing gynecology patients.

Keywords: Case report, Multiple organ dysfunction, symmetrical peripheral gangrene, Toxic shock syndrome.

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Introduction

Toxic shock syndrome (TSS) was first described in 1978.[1] It can quickly develop into multiorgan failure. Toxic shock syndrome is related to infection of postoperative wounds.[2] If elective procedures were clean, the odds of postoperative wound infection were very low. The majority of TSS occurs because of Staphylococcus aureus, where presenting symptoms include high-grade fever, nausea, a diffuse rash, and desquamation of the skin. Symmetrical peripheral gangrene (SPG) is defined as ischemia of two or more limbs, without obstruction of a large vessel or any vasculitis. It is a rare complication of disseminated intravascular coagulation (DIC).[3] Symmetrical peripheral gangrene can end in amputation of limbs.[4] The incidence of TSS with SPG after intrauterine contraceptive device (IUCD) insertion is extremely low. Here, the authors present a case of a lady with TSS that resulted in multiple organ dysfunction syndrome, DIC, and SPG after IUCD insertion.

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Case Description

A 34-year-old patient (Para 4, plus 1), postpartum with lactational amenorrhea, was admitted as uterine perforation from the ER at 02:00 am. She had a history of IUCD insertion one day before, followed by vomiting and acute abdominal pain. Her past medical history was unremarkable. There was no history of fever, flu-like illness, dysuria, or altered bowel habits, and a history of use of intravaginal tampons, sick contacts, travel, or uncommon foods. There was no history of inflammatory bowel disease. Her husband said she was prescribed parenteral antibiotics in a private hospital before admission. Past surgical history was unremarkable. On examination, her temperature was 37° C, heart rate was 89/minute, BP was 100/70 mm Hg, and O_2 saturation was 99% on room air.

Abdominal examination showed lower abdominal tenderness but no palpable mass. On per speculum examination, no thread was seen and few blood clots were removed. Initial hemoglobin was 145 g/L (normal: 115–150 gm/L), WBC 10.2×10^9 cells/L (normal: $3.5–9.5 \times 10^9$/L), and platelets 46×10^9 cells/L (normal: $125–350 \times 10^9$/L). The patient was started on IV fluids, antibiotics, and analgesia and kept under observation. Official ultrasound done at 08:00 am revealed IUCD inside the uterus, marked fluid reaching up to Morrison's pouch, left ovarian cyst 2×2 cm, and the pouch of Douglas showed free fluid. The patient's condition deteriorated. BP fell to 80/60 mm Hg and heart rate increased to 115/min. Her body temperature was 38.2° C. A rapid resuscitation team was activated, and arrangements were made for urgent exploratory laparotomy. Operative findings revealed a hemoperitoneum amounting to 350 cc of blood with clots. There was generalized tissue edema and congestion with blue discoloration of the bladder wall and pelvic organs. The picture was suggestive of DIC secondary to sepsis. There was no pus collection or pus exudate. Uterine walls were found to be intact. There was a small abrasive area in the left mesosalpinx with no active bleeding. There was a small ruptured left ovarian cyst. There was no bowel injury as explored by the general surgery team. Thorough peritoneal irrigation was done with warm saline. A drain was inserted. Intrauterine contraceptive device thread was not seen vaginally, so IUCD was removed with forceps vaginally (as the possible source of sepsis). Throughout the procedure, IV fluid resuscitation was done with crystalloids after the insertion of the CVP line. Urine was found to be bloody during the procedure. The patient was shifted to the intensive care unit (ICU). She was treated with imipenem, tigecycline, clindamycin, blood transfusion, and lots of fluids. The patient's condition worsened the next day. There were infiltrates in both lungs on the chest X-ray. She was intubated due to severe tachypnea and hemodynamic instability. Her condition remained critical. She was commenced on continuous pumps of norepinephrine and vasopressin. Her condition further deteriorated; blood analysis suggested severe metabolic acidosis. Her WBC count was very high, hemoglobin was low, platelet count was low, prothrombin time, and activated partial thromboplastin time were prolonged, the international normalized ratio (INR) was higher than the upper normal limit, serum potassium was toward the lower side of the normal limit, serum calcium was low, and serum creatinine was high. Aspartate aminotransferase and alanine aminotransferase were three times higher, and the total bilirubin level was two times higher than their upper normal limits. Serum albumin was low, and the serum lactate and creatine kinase (CK) were much higher than their upper normal limits. Both blood and urine cultures before antibiotic therapy were negative. On subsequent investigations, the INR increased, platelets dropped further, and WBC, CK-MB, myoglobin, and C-reaction protein kept rising. Her C-reactive protein, CK, myocardial-bound creatine kinase (CK-MB), muscle-type creatine kinase (CK-MM), creatinine, and serum lactate were also high. On abdominal ultrasound, the liver was normal in size, the gallbladder was contracted, and both kidneys and renal collecting system were normal in appearance and echogenicity. During the first 10 days of admission, 2200 mL of red blood cells, 1200 mL of fresh frozen plasma, 28 units (U) of platelet concentrate, and 8U of cryoprecipitate were transfused to the patient. Wound site care was done, and the drain was removed as produced minimally. Her condition improved slowly. However, on day four of admission, the patient experienced discoloration of the hands and developed diffuse erythematous lesions on her thighs,

Fig. 1: Federation symmetrical peripheral gangrene of both hands

back of hands, and big toes, and a few petechial skin lesions were seen on her buttocks and thighs. Besides, the patient developed conjunctival hemorrhages, diarrhea, disorientation, and alteration in consciousness, followed by bilateral digital cyanosis of both hands. Vascular and plastic surgery were consulted. Vasopressin was stopped, and noradrenaline was reduced to a minimal dose followed by stoppage. The patient was started on heparin, and INR was kept at 1.5–2 by the ICU team. Her culture reports revealed no growth, possibly due to the commencement of parenteral antibiotics in a private hospital before the admission. The urine culture showed no growth, and the blood culture was negative. Blood cultures were negative for any organisms throughout. Nose swab results revealed no pathogen identified. Even the IUCD culture did not show any growth of bacteria. The virology screen was negative. On the 9th day of admission, sputum culture and growth revealed growth of Klebsiella pneumoniae, sensitive to tigecycline. No change of antibiotics was done in liaison with the infection control team. The patient was shifted to step-down HDU on 12.03.2023 for continued supportive care by ICU as the patient's condition improved significantly. Although the petechial skin lesions on the region of buttocks and thighs subsided, peripheral ischemia worsened on her fingers, resulting in dry gangrene. The patient was shifted to the ward on 18.03.2023. Physiotherapy was started. The patient was followed by the GS/plastic surgery team. She was discharged from the gynecology unit side in stable condition on 26.03.2024 and transferred to plastic surgery care. She is for follow-up in the outpatient clinic. Her hemoglobin level, WBC count, hematocrit, platelet, C-reactive protein, CK level, CK-MB, CK-MM, INR, aspartate transferase, alanine aminotransferase, and total bilirubin levels improved on discharge. The plastic surgery consultant findings on discharge day were: Left hand had dry gangrene of all fingers at the level of the distal palmar crease and of thumb at the interphalangeal joint level. The right hand had dry gangrene of all fingers at the level of proximal phalanges and of thumb at the distal phalangeal level. Gangrenous parts were dry, with no pus or any discharge. There was some area of desquamation of the skin of the dorsum of the left hand, which was treated with dressing and was in the healing phase. Advice was made for hand hygiene. The plan was to wait for the demarcation of viable and nonviable tissue and then perform amputation of dead tissues. Amputation was advised, which was planned after discharge. She

was seen 1 month after admission in good health. Counseling was done to the patient and family, and they opted to do the procedure in a private hospital (Fig. 1).

Discussion

Toxic shock syndrome is very rare. Since patients undergoing gynecological procedures are usually healthy, any kind of fatal complication is hard to accept. The authors reviewed the literature, which disclosed a few cases of TSS related to inserted IUCD. The first case of septic shock caused by an IUCD was reported in 1966.[5] A case of fatal TSS due to an IUCD that was infected by *Staphylococcus aureus*,[6] and another fatal case of TSS due to an IUCD infected with a strain of streptococci.[7] The patient in the latter case report presented without a rash, which was similar to our patient. Another case of TSS was due to streptococci from an IUCD that got colonized due to oral sex.[8] The patient underwent two exploratory laparotomies, a bilateral salpingectomy, and a total abdominal hysterectomy. The first case of TSS caused by Group A streptococcus was reported after IUCD insertion, resulting in a total abdominal hysterectomy with bilateral salpingectomy.[9] Other complications after IUCD insertion include severe adnexitis, peritonitis, and salpingitis.[10] One case was associated with septicemia caused by *Escherichia coli* due to Dalkon Shield IUCD and resulted in septic abortion. The Dalkon device was withdrawn from the US market in 1974 because several septicemia cases and deaths were reported.[11] Staphylococcal septicemia was also reported in one case after Lippes Loop insertion.[12] Meningococcal septic shock has also been reported in association with an IUCD.[13] One case of SPG was associated with TSS in a lactating lady who had experienced an abortion a couple of months ago. Infections due to IUCD rarely present as TSS. This makes diagnosis very difficult. The mechanism by which IUCDs cause infections is not known. It is suggested that the vaginal string of the IUCD might be the cause of breaking the protective barrier of the cervical mucosa.[14,15] It is also postulated that the injury caused by IUCD results in disruption of the cervical mucosa and hence infection. Group A *Streptococcus pyogenes* (GAS)[16] and *Staphylococcus aureus* are frequently related to TSS. There is a rapid development of multiple system organ failure and dermatologic consequences following a short period of TSS. The condition requires large amounts of fluids and vasopressors to maintain blood pressure. The Centers for Disease Control and Prevention (CDC) suggest different criteria for diagnosis. Although the rash is a compulsory criterion for a confirmed case of TSS, only about 10% of the cases of TSS caused by GAS develop a rash, followed by desquamation.[17] This fact makes it challenging to diagnose. Our patient's clinical manifestation and subsequent course were in accordance with the TSS, 2011, Case definition by the CDC. Cultures of blood, urine, and throat are peculiarly negative. Positive blood culture results are scarce in staphylococcal TSS, where only less than 5% are positive.[18] Regardless of a strikingly increased WBC count and CRP, the blood culture reports are invariably negative. Symmetrical peripheral gangrene is mostly associated with the use of vasopressors in DIC.[19] Hypotension because of sepsis is probably another cause of SPG and is believed to be exaggerated due to hypotensive therapy.[20] Disseminated intravascular coagulation can result in intravascular thrombosis and hence infarction of the skin and extremities.[21] Symmetrical peripheral gangrene causes characteristic symmetric necrosis of the skin and extremities, which is followed by gangrene in two or more distal sites. Around 18–40% mortality rate and a high frequency of limb amputations have been reported.[22] An early amputation is contraindicated. This is because firstly, secondary infection of necrotic tissue is common, and secondly, the limits of ischemic lesions occur over time, and amputation at a lower level might result in another amputation surgery when gangrene extends proximally. There is no fully useful treatment. Identification and treatment of the underlying cause might help to halt and prevent further progress of SPG. Early acknowledgment is the most important factor in the management of SPG.

Conclusion

Toxic shock syndrome can occur due to unsuspected reasons like IUCD insertion. Early diagnosis and timely management are the ways to prevent amputation and mortality. Even without a sign of local infection, active fluid resuscitation and intensive care support should be provided whenever TSS is suspected.

Clinical Significance

Toxic shock syndrome following IUCD insertion is a rare and unsuspected occurrence. This fact should be kept in mind while managing gynecology patients.

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