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Case Report

Acute toxic shock syndrome associated with intra-operative debridement and instrumentation removal for chronic osteomyelitis: A report of two cases

Michael Ulmer [a,*], Christopher R. Rehak [b], John C.P. Floyd [c], Robert M. Harris [c]

[a] Jack Hughston Memorial Hospital Residency, 4401 Riverchase Drive, Phenix City, AL 36867, United States of America
[b] Mercer University School of Medicine, 1633 1st Avenue, Columbus, GA 31901, United States of America
[c] Hughston Foundation Inc., Hughston Clinic, 6262 Veterans Parkway, PO Box 9517, Columbus, GA 31909-9517, United States of America

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ABSTRACT

Gram-positive organisms are known causative agents in toxic shock syndrome (TSS), an acute disease caused by bacterial exotoxins. During routine instrumentation removal for chronic osteomyelitis, intraoperative debridement, reaming, and irrigation can lead to cell lysis and subsequent dissemination of the bacterium exotoxin, which can result in acute cardiovascular compromise. We present two cases of chronic osteomyelitis in healed long-bone fractures that were treated with deep instrumentation removal and surgical debridement using a reamer-irrigator-aspirator (RIA) system. Both patients had positive *Streptococcus agalactiae* wound cultures and both developed acute intraoperative hypotension during the reaming/irrigation portion of the procedure. Case 1 experienced cardiac arrest and was resuscitated for several days in the ICU. Case 2 underwent intra-operative hypotension and was resuscitated appropriately. The RIA or standard reaming systems must be used with caution during debridement of osteomyelitis in the presence of known toxin producing bacteria. The risk of iatrogenic spread of infection or extravasation of intramedullary contents is present; a high index of suspicion with any change in vital signs and prompt response can help mitigate the effect of adverse outcomes associated with acute and severe intraoperative hypotension.

Introduction

Toxic shock syndrome (TSS) is a rare, acute, multi-system, toxin-mediated illness. First described in 1978, TSS was differentiated from the clinical syndrome of septicemia; it has been established as a spectrum of diseases caused by toxin-producing strains of *Staphylococcus aureus* and *Streptococcus pyogenes* (group A streptococcus). There is no single test for TSS; its diagnosis is based on clinical presentation. TSS is caused by the sudden and massive release of T-cell cytokines into the bloodstream. These bacterial toxins act as superantigens that stimulate immune-cell expansion and, in turn, can lead to overwhelming cytokine expression.

Long-bone osteomyelitis is a common diagnosis in a state of chronic infection [1]. The burden of infection parallels the increasing prevalence and severity of injuries, particularly due to motor-vehicle accidents [1,2]. In addition to a prolonged course of antibiotics,

* Corresponding author at: Jack Hughston Memorial Hospital Residency, The Hughston Foundation, Inc., 4401 Riverchase Drive, Phenix City, AL 36867, United States of America.
E-mail addresses: mulmer@auburn.vcom.edu (M. Ulmer), Rharris@hughston.com (R.M. Harris).

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the treatment of open injuries or established infections in long-bone fractures surgical intervention commonly involves wound debridement and fracture stabilization [3]. Reamer-irrigator-aspirator (RIA) systems are used in place of conventional reaming for long-bone intramedullary debridement [4–7]. Published case series and case reports document the efficacy and safety of the RIA system in managing osteomyelitis [4–7]. However, its widespread use in debridement of bony tissues to adequately remove intramedullary debris and prevent iatrogenic spread of infection is controversial [4–7]. With the expanded use of RIA, unanticipated complications have been seen. Intramedullary lavage combined with reaming may be related to the intraoperative release of bacterial toxins stimulating the cytokine release that causes tissue damage, disseminated intravascular coagulation, and organ dysfunction seen in TSS. No Tranexamic acid (TXA) was given pre and post-operatively. Patients were not on anti-platelet or anti-coagulation prior to surgery.

Case report

Case 1: A 38-year-old African American male presented to the emergency department with a gunshot wound to his left femur. After initial resuscitation by the trauma team, the patient was diagnosed with a left intertrochanteric fracture that was treated with sharp excision and debridement of the gunshot wound followed by stabilization with a cephlomedullary nail. In the postoperative period, the patient continued to have drainage from the wound and was treated with IV antibiotics followed by a prolonged oral course of antibiotics by mouth. It was decided to suppress the infection until he could achieve a clinical union before dealing with the infected deep instrumentation and osteomyelitis. At 18 months postoperatively, the patient presented for draining sinus along the medial aspect of the knee and posterior lateral buttocks. His white blood cell (WBC) count was 9.6, C-reactive protein (CRP) of 2.7, and erythrocyte sedimentation rate (ESR) of 67. Radiographs demonstrated healing of the fracture (Fig. 1A and B).

The patient was taken to the operating room for irrigation, debridement, and cephlomedullary nail removal. After nail removal, an RIA system was used for debridement, and the canal was irrigated with saline and bacitracin (Fig. 2). During this portion of the procedure, the patient became acutely hypotensive and pulseless. Cardiopulmonary resuscitation began immediately, and the procedure was halted. The patient responded to chest compressions, epinephrine, and vasopressors. Once vital signs were stabilized, an antibiotic nail was placed without further reaming.

Postoperative labs showed elevated white blood cells (WBCs), decreased hemoglobin, and increased platelets (Table 1). Computed tomography with angiography ruled out pulmonary embolism. Intravenous fluids, ventilatory support, and vasopressors were required to maintain appropriate blood pressures for 48 h following the surgery. The patient was extubated after 24 h of ventilatory support. Intraoperative wound cultures grew *Streptococcus agalactiae* and *Peptostreptococcus teradius*; blood and urine cultures showed no growth. On postoperative day 5, the patient's white count had normalized, and he was discharged from the hospital. He was maintained on intravenous ceftriaxone for 6 weeks.

Case 2: A 52-year-old white male had sustained open tibia and fibula fractures following a motor vehicle accident. The patient had sustained an open mangled extremity with a highly comminuted tibia. He was treated with surgical debridement, an intramedullary

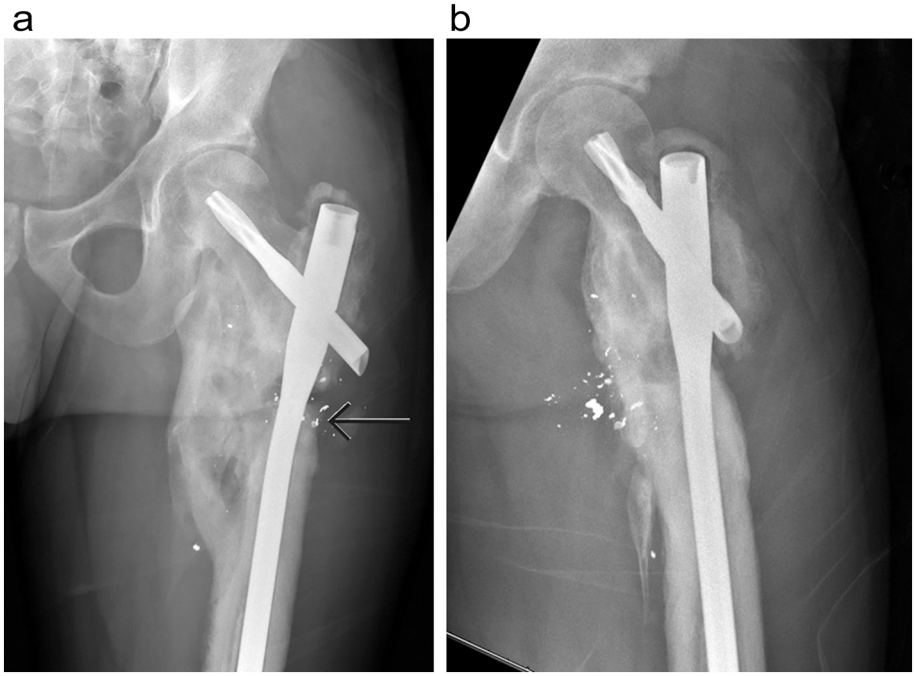

Fig. 1. A and B: Radiographs AP and lateral of the left femur demonstrates radiographic union.

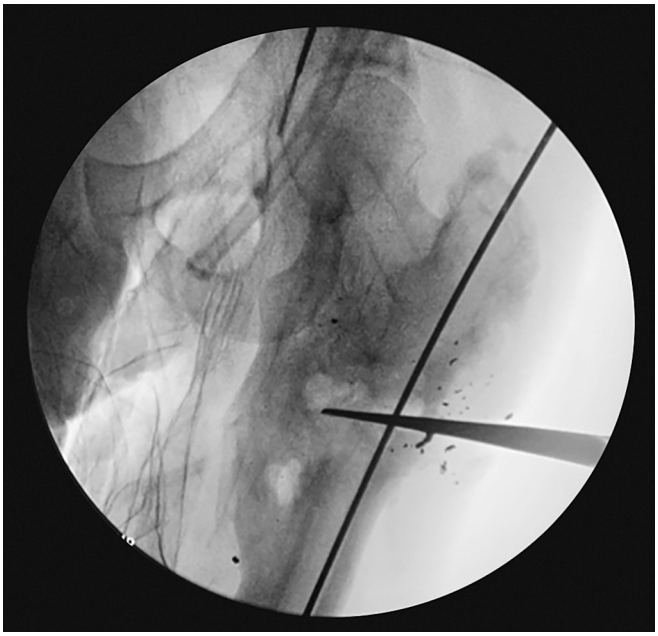

Fig. 2. Intraoperative fluoroscopy shows debridement of the femoral intramedullary canal.

Table 1
Demonstrates Case 1 labs post-operatively.

Case 1	POD 0	POD 1	POD 2	POD 3	POD 4	POD 5
WBC	21.14	19.61	15.04	13.49	12.38	11.69
Hgb	6.9	8.1	7.3	6.9	7.5	7.5
Hct	21.9	24.4	23.2	21.9	24.0	23.4
Plt	506	335	260	287	450	485

nail and plate technique and long-term intravenous antibiotics. Two years following this initial surgery, he was diagnosed with chronic osteomyelitis and underwent rod removal and a second course of intravenous antibiotics. Four years following the initial injury and surgery, the patient represented with erythema tenderness and drainage from the proximal anterior tibia fracture region. Radiographs showed a lucency in the proximal tibia (Fig. 3A and B). A magnetic resonance imaging (MRI) scan demonstrated a nidus formation with involucrum (Fig. 4A and B), consistent with osteomyelitis.

The patient was returned to the operating room for irrigation and debridement. A RIA reamer was placed to debride the sequestrum in the proximal portion of the tibia and the entire tibial shaft. Coincident with the reaming and pulsed lavage, the patient's blood pressure dropped from 110/80 to 60/40 mmHg, but improved with IV fluids and epinephrine. The surgery was then continued. The tibia was irrigated and pulse lavaged; again, the patient's blood pressure dropped to 70/30 mmHg, which again responded to epinephrine. Once stabilized the surgery was concluded.

He was extubated 4 h after the surgery and did not require vasopressors for blood pressure control (Table 2). The patient was started on IV Vancomycin and Meropenem. Intraoperative wound cultures grew *Streptococcus agalactiae*; blood cultures showed no growth. The patient was discharged with long-term antibiotics.

Discussion

TSS is an acute, multi-system, toxin-mediated disorder that can result in multi organ failure and death most commonly caused by *S. aureus*. Toxic Shock-Like Syndrome (TSLS) closely resembles TSS in that it causes fever, rash, desquamation, hypotension, and multi-system organ dysfunction, though TSLS is most commonly caused by *S. pyogenes* [8]. TSS and TSLS following orthopaedic surgery are rare; however, it is important to promptly recognize and diagnose these syndromes intraoperatively or in the acute post-operative setting in order to initiate appropriate treatment without delay. TSS management must provide the appropriate hemodynamic support.

In their review of their RIA experience in 32 patients, Jakma et al. noted a 31 % rate of complications, including 6 % of pulmonary emboli [9]. In a randomized control trial on different reaming techniques, Hall et al. demonstrated that there was an embolic load with the use of the RIA system [10].

The RIA system offers benefits of removing large amounts of intramedullary content, employing concurrent irrigation and aspiration, and decreasing the risk of debris extravasation are important considerations [4–7]. Nonetheless, as illustrated above, it is

Fig. 3. A and B: Radiographs left tibia demonstrate this healed fracture with radiolucency along proximal tibia.

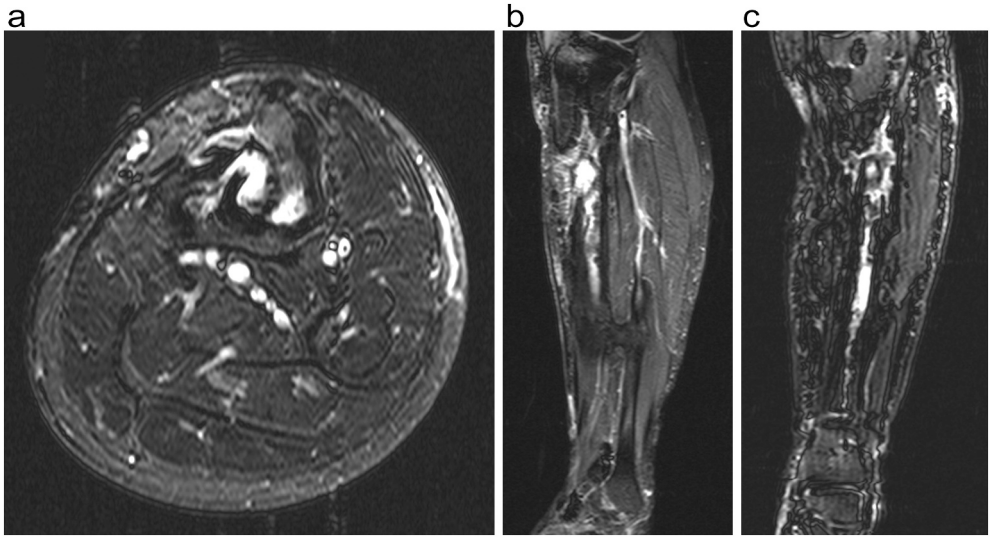

Fig. 4. A, B, and C: MRI demonstrates this nidus formation with involucrum with sinus tract formation to the proximal tibia.

important to note that the risk of extravasation is not completely eradicated. Therefore, our osteomyelitis cases during debridement, we now irrigate slowly to maintain lower intramedullary pressures, and we continually check the aspirator for blockage as prior authors have experienced intramedullary contents blocking the aspirator. We are also more vigilant during the reaming and irrigation portion of the surgery, and we inform our anesthesia colleagues of this increased risk to be better prepared in case of intraoperative hypotension.

This case report demonstrates the potential for intra-operative hypotension, cardiac arrest, and end organ damage from the use of a

Table 2
Demonstrates Case 2 labs post-operatively.

Case 2	POD 0	POD 2
WBC	12.79	5.89
Hgb	13.1	10.5
Hct	41.0	34.1
Plt	198	163
PT	13.5	n/a
INR	1	n/a
PTT	25	n/a

RIA reamer to debride known toxin producing bacteria from a chronic osteomyelitis in a long bone. This may be the result of a toxic shock or toxic shock like syndrome produced by embolization of the bacteria and/or bacterial toxins into the body's systems. With known toxin producing bacteria in similar future clinical situations, notifying the anesthesia provider and preparation by the surgical team for such a potential intra-operative event may prevent severe complications.

Declaration of competing interest

John C.P. Floyd, MD, has stock or stock options in Bongiovi Medical and Carbofix. Robert M. Harris, MD, is a paid consultant for NexScience. He also consults and has received research support from Citieffe. Additionally, Dr., Harris is a board or committee member for the Clinical Orthopaedic Society. For the remaining authors, none were declared.

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