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Case Report

Four Limb Amputation Secondary to Symmetrical Peripheral Gangrene in a Polytrauma Patient: A Case Report

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Limb amputation · Gangrene · Disseminated intravascular coagulopathy · Inotropes · Trauma · Symmetrical peripheral gangrene

Abstract
Symmetrical peripheral gangrene (SPG) is a rare condition. At present, there is no clear etiology for this condition; however, it has been associated with multiple risk factors, including sepsis and coagulopathy. We present a patient who sustained a polytrauma with multiple organ injuries. During hospitalization, he developed septic and cardiogenic shock complications by disseminated intravascular coagulopathy, which led to ischemic changes in all four limbs. The patient underwent bilateral – trans-tibial and trans-radial – amputation. High level of suspicion and awareness are required to detect such a devastating condition in order to mitigate complications. We conclude that ischemic changes secondary to SPG are irreversible; subsequently, halting the disease progression is the mainstay of treatment.

Introduction

Symmetrical peripheral gangrene (SPG) is a rare and severe complication of disseminated intravascular coagulation (DIC) that frequently accompanies sepsis [1–4]. It is characterized by a sudden onset of peripheral gangrene in the absence of major vascular disease or occlusions, leading to ischemic changes in two or more extremities [5, 6]. Although clinically the peripheral pulses can be present and palpable, ischemic changes can still occur [5].

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Multiple limb amputation is uncommon, and numerous metabolic conditions have been reported in the literature to cause such problem [7–14]. Inotropes and vasopressin groups of drugs have been used in the treatment of hypotension, as well as cardiogenic and septic shock [6, 15–17]. Prolonged administration of these agents at high doses have been associated with the development of peripheral gangrene, which was first reported in 1973 by Holzer et al. [18]; it was also reported to cause 4 limbs gangrene [15, 19–22].

Management of such SPG is challenging and requires multidisciplinary team input. Early diagnosis and treatment is essential to halt the progression of this condition. However, about half the patients surviving this complication will require amputation of the affected limbs [4]. Ischemic changes secondary to SPG are irreversible, thus halting the disease progression is the form of treatment. We are presenting a case of a polytrauma patient who developed septic shock with DIC, leading to the development of 4 limbs ischemia and amputation.

Case Presentation

A 50-year-old male, with no medical history, presented to our trauma unit after a heavy object fell on him at his place of work. Upon initial assessment, the patient was conscious and oriented with a Glasgow Coma Scale (GCS) of 14/15, with a respiratory rate of 25 breaths/min. The patient's remaining vitals were within normal limits. An examination of the chest revealed that there was thoracic tenderness to the right side with crepitation and right anterior shoulder swelling, lacerations with a limited range of motion. Radiological examination of the chest confirmed multiple right-sided rib fractures and a small hemopneumothorax which was treated conservatively. There was also a right proximal humerus fracture with an ipsilateral scapula fracture; this, however, did not require any surgical intervention.

The abdominal examination was significant for right upper quadrant pain. Pan computed tomography (CT) scan revealed grade IV liver laceration, right lung contusion, and confirmed the presence of the right-sided hemopneumothorax. Initial laboratory results highlighted normal hemoglobin levels and normal renal function tests. Liver function tests revealed elevated levels of alanine transaminase (486 IU/L), aspartate aminotransferase (550 IU/L) and decreased albumin levels of 33 g/L.

The patient was admitted to the trauma intensive care unit (TICU) for further management. After 48 h of admission, his abdominal pain increased and stopped responding to analgesia; the patient's oxygen (O_2) saturation dropped to 92%. A new CT scan of the chest and abdomen with contrast showed an increase in perihepatic and intraperitoneal free fluids when compared to the initial scan. The new CT scan also showed bilateral pleural effusion and total collapse of the right lung, secondary to increasing hemopneumothorax. This was managed by the insertion of a right chest tube followed by intubation.

He suddenly had a cardiac arrest, immediately following the intubation. The acute trauma life support protocol was initiated and resuscitation was started. He was given atropine 0.5 mg, 3 mg of adrenaline, along with 1,000 mg of calcium chloride and 150 mmol of sodium bicarbonate. Normal sinus rhythm was regained, and a CT pulmonary angiogram did not show any pulmonary embolism. He was then put on vasopressin 0.03 unit/min for 24 h and noradrenaline 1.4 μg/kg/min for a total of 18 days.

Upon further investigation of his abdominal pain, there was an increase in the intra-abdominal pressure to 23 mm Hg, for which he underwent decompression laparotomy and application of negative pressure dressing. On the first postoperative day, he developed a high-grade fever of 38°C, hypotension of 103/31 mm Hg, tachypnea with a respiratory rate of 40 breaths/min, tachycardia with a heart rate of 140 beats/min. The patient's white blood count was 1.1; this was consistent with systemic inflammatory response syndrome. An empirical

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antibiotic was started, and a septic workup confirmed the diagnosis of *Escherichia coli* (*E. coli*) septicemia. The patient was shifted to intravenous meropenem 1 g every 8 h. With this change, the patient's parameters started to normalize. Unfortunately, he developed anemia with abnormal renal function values, indicating acute renal failure that required hemodialysis. In addition, the patient had abnormal coagulation profile with activated partial thromboplastin time of 45 s and prothrombin time of 19 s. The D-dimer was more than 35 and platelet count was 21,000ˆ3/μL. This was consistent with DIC. Red blood cells along with platelets and fresh frozen plasma units were transfused to the patient; he was then started on heparin infusion and kept on vasopressors to maintain his blood pressure. Supportive management was started, along with heparin infusion including IV fluids, analgesia for pain control, and antibiotics were resumed to treat the sepsis. These all contributed to the improvement of the patient's clinical condition.

Over the following 48 h, the patient developed cyanosis with cold peripheral pulses which were palpable on clinical examination and audible on Doppler. The cyanosis progressed into frank dry gangrene over 6 weeks as shown in Fig. 1.

The patient was kept in the TICU for close observation and supportive treatment that included IV fluids, antibiotics, and hemodialysis. During his TICU stay, staged amputation of his four limbs was planned. He first underwent bilateral trans-tibial amputation. One week later, he underwent bilateral trans-radial amputation (Fig. 2).

After the 97-day stay in the TICU, the patient was transferred to the trauma ward for rehabilitation. Initial rehabilitation involved chest physiotherapy, passive, as well as active range of motion exercises for the knees, hips, elbows, and shoulder joints to avoid stiffness and contractures. After the patient had completely healed from the amputation, stump fitting of bilateral endoskeletal modular trans-tibial prosthesis with silicon liner socket and shuttle lock took place to start ambulation of the patient. First, the patient was educated regarding bed to chair transfer and sitting to standing positions with minimal support. This enabled the patient to sit in an electric wheelchair for mobilization. Ambulation training was started and was strictly in the presence of a physiotherapist. Gradually, the patient achieved minimal dependence ambulation on flat surfaces for at least 100 m. The patient was, however, completely dependent on Canadian walking frame. The patient continues to receive care including psychological support and physiotherapy.

Discussion

SPG is commonly associated with septic and cardiogenic shock [5]; its occurrence has been reported in the literature with the association of multiple conditions [6, 23–26]. All reported cases are usually associated with a pro-inflammatory state in which the inflammatory cytokines trigger DIC and potential microthrombosis formation. The pathological examination of amputated limbs in SPG has often revealed underlying thrombi concentrated in small vessels but not in large vessels [3, 27]. Clinical features that give rise to the suspicion of SPG include cold erythematous extremities followed by dusky skin discoloration and finally well-demarcated gangrenous changes over 2-week duration. These changes usually affect the lower limbs first followed by the upper limbs, despite the presence of peripheral pulses [2, 5, 6, 27, 28].

The patient in our report had no risk factors for peripheral vascular disease. However, the significant trauma he sustained including the grade VI liver laceration put him in a high pro-inflammatory condition. As a result, this has put the patient at risk of developing microthrombosis by reducing hepatic production of natural anticoagulants, protein C, and anti-thrombin [1, 16, 17, 29]. Moreover, he had developed septic shock secondary to an

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Fig. 1. Peripheral gangrenous changes of the patient showing both upper limbs and lower limbs.

Fig. 2. Four limb amputation, bilateral trans-tibial and bilateral trans-radial amputations.

abdominal visceral infection that necessitated the use of inotropes to manage the associated hypotension. Noradrenalin is dose dependent, and with high doses, it can lead to vasoconstriction of the vessels. All these devastating events that this patient had suffered led to the development of DIC and microthrombus formation within small vessels of all four limbs and ischemic changes which resulted in the amputation of all four limbs.

Management of SPG should involve a multidisciplinary approach, with the involvement of multiple caregivers such as critical care specialist, hematologist, general or orthopedic surgeons, physiotherapist. It is vital that the patient be admitted to the critical care unit. To date, there are no established guidelines for the management of this condition; once ischemic changes occur, they cannot be reversed, and this is associated with high morbidity and mortality rates [30]. There is a consensus that the goal of treatment should be directed to prevent disease progression by removing causative factors, preventing secondary infection, and removing nonviable tissues. Close monitoring for peripheral ischemic changes should be carried out whenever dopamine is initiated in patients with septic or cardiogenic shock, who have developed DIC. Once ischemic changes have occurred, the use of dopamine should be minimized when possible, with early surgical removal of the necrotic tissues when well demarcation has developed.

Conclusion

SPG is a rare and challenging condition with high morbidity and mortality rates. Physicians should be fully aware of this condition, as early identification and prompt management may help in preventing devastating consequences. Ischemic changes secondary to SPG are irreversible; halting the disease progression is the mainstay of treatment. Management of this condition should be undertaken in an intensive care unit with the involvement of multidisciplinary teams. The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see www.karger.com/doi/10.1159/000528255) [31].

Statement of Ethics

This study protocol was reviewed and approved by the Hamad Medical Corporation Institutional Review Board with approval number: MRC-04-22-433. A written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict of Interest Statement

The authors have no competing interests to declare.

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Author Contributions

Yahya Alborno: investigation and writing original draft – review and editing. Isam Sami Moghamis: investigation and writing – review and editing. Hammam Kayali and Elhadi

Babikir: investigation, writing, and writing – review and editing. Shamsi Hameed: investigation, project administration, and writing – review and editing. Mohammed Al-Ateeq Al-Dosari: investigation, supervision, project administration, original draft, and review and editing.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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