Case Report

Iatrogenic-induced Peripheral Gangrene: From Multiple Limb Amputations to Prosthetic Restoration

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ABSTRACT

Objectives: Multiple limb amputations are an uncommon complication from inotrope-induced peripheral gangrene.

Case Presentation: A 20-year-old lady with valvular heart disease had septic shock secondary to infective endocarditis and required prolonged cardiopulmonary resuscitation. Despite aggressive fluid resuscitation, the patient had triple strength intravenous noradrenaline to maintain hemodynamic stability. On day 5 of post-shock, dry gangrene occurred in distal parts of all limbs, and inotrope was stopped. Although the gangrenous changes were non-progressive, she required a significant degree of assistance with mobility and daily function performance. The patient was counseled for multiple limb amputations to promote walking and hand function through prosthetic restoration. Five months after the event, she had a right transtibial amputation, left Chopart amputation, left wrist disarticulation, and right second, third, fourth, and fifth fingers amputation. Three specific goals for inpatient rehabilitation were independent short-distance ambulation with prostheses, performing basic activities of daily living with adaptive devices, and independent wheelchair propulsion for long-distance mobility using a right-sided transtibial prosthesis, left-sided Syme's prosthesis with Kingsley's foot, right-sided silicone-based cosmetic glove and left-sided body-powered transradial prosthesis.

Discussion: Although an uncommon complication, inotrope may lead to multiple limb amputations secondary to peripheral gangrene. Following amputation, the ultimate rehabilitative goal is to restore the mobility and capacity to perform daily functions through prosthetic restoration, whether walking for lower amputees or functioning hand for upper limb amputees. Prescribing prosthesis in a single limb loss is relatively straightforward, but restoring multiple limb amputations bears many challenges toward successful recovery of walking and functions.

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Highlights

• Inotropic use leads to multiple limb amputations secondary to peripheral gangrene.

• We present the chronology of events restoring mobility and capacity to perform daily functions through prosthetic restoration.

• There are challenges in restoring multiple limb amputation in developing countries.

Plain Language Summary

This case report describes the chronology of an event in a young lady with underlying heart disease complicated with sepsis. She survived a near-death complication but was left with gangrene of all four limbs leading to amputations. To restore her function to perform daily activity, prosthetic restoration, and rehabilitation was done. Many challenges were faced along the process in terms of physical, mental, resource, and financial aspects, especially in a third-world country.

1. Introduction

Large vessel diseases such as atherosclerosis, diabetes mellitus, and long-term smoking are common disorders that lead to peripheral gangrene [1]. Infrequently, microvascular angiopathy secondary to autoimmune vasculitis or connective tissue diseases causes similar complications [2]. Uncommonly, inotrope, burns, purpura fulminans, cryoglobulinemia, and cocaine abuse may lead to multiple limb amputations secondary to peripheral gangrene [3].

From a rehabilitation standpoint, prosthetic restoration is a major part of restoring function in amputated persons with a holistic approach as the lynchpin of managing a person with limb(s) amputation [4]. This article highlights the chronology of events and challenges to restoring mobility functions and abilities to perform activities of daily living (ADLs) in a young person who underwent multiple limb amputations.

2. Case Presentation

A 20-year-old lady with underlying valvular heart disease had septic shock secondary to infective endocarditis after presenting with two weeks of fever, palpitation, and cardiac failure symptoms, including shortness of breath, orthopnoea, and reduced effort tolerance in early 2017. Her situation got complicated with prolonged cardiopulmonary resuscitation, and despite aggressive fluid resuscitation, triple strength intravenous noradrenaline was prescribed to maintain hemodynamic stability (lowest recorded blood pressure was 33.17 mmHg). On day 5 of post-shock, she had dry gangrene complication in distal parts of all four limbs, and inotrope was immediately stopped. Investigations included blood tests to rule out diabetes mellitus, autoimmune vasculitis, cryoglobulinemia, and clotting investigation to exclude disseminated intravascular coagulation (DIC) as the cause of peripheral gangrene. The pulmonary function test did not suggest scleroderma or any restrictive lung patterns connective tissue disease. She is a non-smoker with no prior history of substance abuse to suggest other causes of gangrene.

Although the gangrenous changes were non-progressive, she required significant assistance for mobility and ADLs performance. She could not stand due to the neuropathic pain complication and had to be fed by the caregiver. The patient and caregivers were counseled on the option of multiple limbs amputation to promote walking and hand function through prosthetic restoration. As the patient comes from a low-income family living in the rural area of Sabah, the option of prosthetic restoration is very costly for her, let alone the logistic expenses and treatment fees. She was qualified for total financial aid from the welfare section under the government and eligible for fully subsidized prostheses. The patient had to be counseled several times before agreeing to multiple limb amputations. Five months after the event, she underwent right-sided transtibial amputation, left-sided Chopart amputation, left-sided wrist disarticulation, and right-sided second, third, fourth, and fifth fingers amputation (Figure 1). There was a period before the amputation was done when she was clinically depressed and underwent counseling sessions to improve her psychological state. The post-operative was uneventful.

The patient had intensive inpatient rehabilitation at eight months post-amputation aiming for three specific goals: independent short-distance ambulation with a walking aid, performing basic ADLs with adaptive devices, and independent wheelchair propulsion for long-distance mobility. Unfortunately, the baseline modified Barthel index (MBI) score at admission was only 18. The road to recovery was initially an uphill battle for the patient, as she had to come to terms with losing her four limbs. Initially diagnosed with mild depression, her mood significantly improved with psychological intervention incorporating counseling and strong family support throughout the inpatient stay lasted for 1 month.

She was prescribed a right-sided transtibial prosthesis (pin and lock suspension type and single-axis foot) and a left-sided Syme's prosthesis with Kingsley's foot. One of the main challenges for restoring prosthesis in Chopart amputation is limb length discrepancy. However, this issue may not be significant because the patient has bilateral amputations. While waiting for the latter, the patient was given a temporary ankle-foot orthosis (AFO) to accommodate the left Chopart amputation (Figure 2). Mobility training included quadriceps and hamstring strengthening, balance training, and sit-to-stand exercises. As her balance and standing endurance improved, she donned a right transtibial prosthesis and temporary AFO using a walking frame with forearm supports (Figure 3 left).

To improve her right-hand function, the right second, third, fourth, and fifth finger amputations would render her non-functional without prostheses. Hence, the patient was counseled for a mini Ilizarov procedure of the right forefinger with modification of the ipsilateral thumb creating web spaces. This procedure would achieve opposition movement and significantly promote hand function without needing a prosthesis. However, she was not keen on another round of surgical interventions and still required psychological support. Therefore, she used a modified adaptive device to assist her with several basic ADLs. She received a right-sided silicone-based cosmetic glove and a left-sided body-powered transradial prosthesis (for the left wrist disarticulation) for long-term use (Figure 3 right). Using this prosthesis would mean that she would have a functional hand. Before discharge, the MBI (The modified Barthel Index) score increased to 62, and no adverse events occurred during this inpatient stay.

Although young and expected to recover quickly, she needed an extended period of training in the community setting before achieving a community ambulator status to allow social integration. For this purpose, she used a lightweight wheelchair, and propulsion is made possible with an adaptive device so that the patient would achieve earlier social integration, especially for community-based rehabilitation training since she resides remotely. Approximately 18 months after the initial septic event,

Figure 1. Multiple limb loss 8 months after amputations

Figure 2. Right transtibial prosthesis and temporary ankle-foot orthosis to accommodate the left-sided chopart amputation

she used all her prostheses, and rehabilitation was continued as home-based training with interval follow-up in an outpatient setting. At the time of writing, prostheses were mainly confined to the home setting and mobilized with a wheelchair for outdoor activities to reduce energy consumption.

3. Discussion

Our case had dry gangrene complications in the distal parts of all four limbs from the use of inotropes intended for managing septic shock. As a result, she had to endure multiple limb amputations. Despite being an uncommon complication, inotrope may lead to multiple limb amputations secondary to peripheral gangrene. Dopamine and noradrenaline are frequently used to treat hypotension secondary to shock. In this case, she received triple strength intravenous noradrenaline, and at such a high dose with prolonged administration (more than 24 hours), the risk for peripheral gangrene would be increased. This complication was first reported in 1973 by Holzer et al. followed by four similar cases in the 1990s; all led to all four limb amputations [5, 6].

However, a recent case report by Ki Jin Jung et al. in 2018 stated that symmetrical peripheral gangrene might not be as rare as many have assumed [7]. A population-

Figure 3. Left: Donned right-sided transtibial prosthesis and temporary left-sided ankle-foot orthosis for gait training in rehabilitation ward; Right: Donned a right-sided silicone-based cosmetic glove and a left-sided body-powered transradial prosthesis.

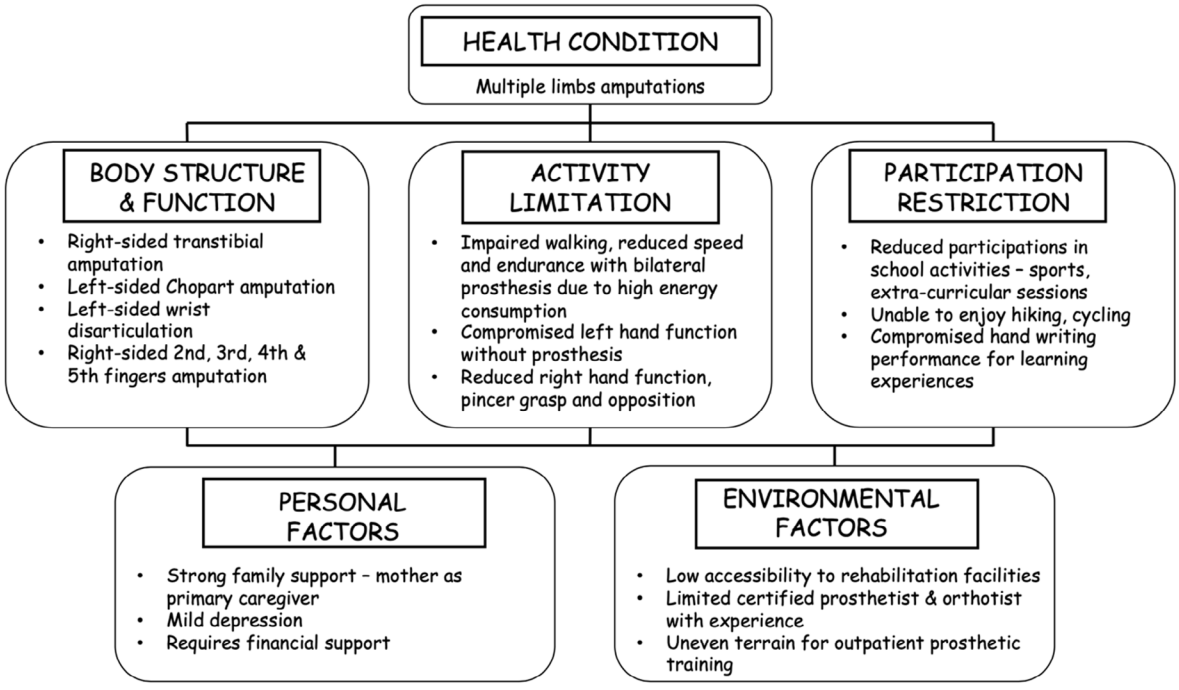

Figure 4. The international classification of functioning, disability, and health model as applied to multiple limb amputations reported in this case

based cohort study in South Korea concluded that 0.8% of sepsis survivors underwent limb amputation to treat peripheral gangrene. They also found that sepsis survivors with chronic pre-morbid conditions such as diabetes mellitus and peripheral arterial disease or those who received specific vasopressor treatments (epinephrine and dopamine) were more likely to undergo limb amputation [8].

Following an amputation, the ultimate rehabilitative goal is to restore mobility and abilities to perform ADLs through prosthetic restoration. In most amputation cases, prescribing a prosthesis for a single limb loss is relatively straightforward by considering the physical impairments, the expected level of functioning, and the anticipated social roles. However, restoring multiple limbs amputation bears a higher level of challenges toward successfully recovering the functional capacities. Figure 4 displays the issues identified based on the ICF (international classification of functioning, disability & health) model.

According to the World Health Organization (WHO), 0.5% of the population in a developing country has some degree of physical disabilities that require rehabilitation services and the need for prosthesis or orthosis to support their daily activities [9]. WHO survey analysis demonstrated that people with disabilities were more than twice as likely to seek healthcare expertise due to inadequate equipment to meet their basic daily needs and almost three times as likely to be denied healthcare access or services [9].

There are 6 practicing rehabilitation physicians in the state of Sabah, where our case resides, with only one dedicated inpatient ward for intensive rehabilitation. The number of certified prosthetists and orthotist is also low. Hence, accessibility to comprehensive rehabilitation services for people in rural areas is limited. The people receive financial subsidiary from the government to support the funding of prostheses but only cover the basic functional prosthesis. Therefore, practitioners and patients must use available options within the permitted budget, although the prescribed prostheses might not be the optimal type [10].

The social environment is an important factor that influences an amputee to participate in the recovery process, especially with the performance of social activities [11]. Inaccessibility issues and stigma towards people with disabilities present architectural barriers for amputees to achieve improved functional levels as they would have achieved in a rehabilitation ward. Furthermore, donning double lower limb prostheses would be challenging for successful community integration. Here lie the needs for universal design in sustainable community development.

4. Conclusion

Although an uncommon complication, inotrope may lead to multiple limb amputations secondary to peripheral gangrene. Following amputation, the ultimate rehabilitation goal is restoring mobility and the capacity to perform daily functions through prosthetic restoration. Prescribing prosthesis in a single limb loss is relatively straightforward, but restoring multiple limb amputations bears many challenges toward successfully recovering functions.

Ethical Considerations

Compliance with ethical guidelines

Ethical consideration was not required due to the nature of the write-up, with no identifiable personal details in the manuscript.

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Authors' contributions

Conceptualization: Fatimah Ahmedy, Jamie Joseph, Syahiskandar Sybil Shah, and Kang Bee Lee; Writing the original draft: Linda Joehari, Fatimah Ahmedy, and Vanessa Chua; Writing, review, and editing: Farhana Harzila Mohd Bahar and Jamie Joseph; Resources: Kang Bee Lee; Supervision: Fatimah Ahmedy and Syahiskandar Sybil Shah.

Conflict of interest

All authors declared no conflict of interest.

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