

Management of Infected Calcific Myonecrosis: A Report of 2 Cases

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Summary: Calcific myonecrosis (CM) is a rare condition in which a large calcified mass develops after trauma. Generally, CM occurs in a lower extremity, and there have been no reports of its occurrence in the upper arm. We report 2 cases of infected CM, including a rare case of CM occurrence in the arm and a typical case in the leg. Case 1: An 84-year-old woman presented with a draining sinus and a large calcified mass in the arm and axillary region. The mass involved the neurovascular bundle; thus, complete resection was impossible. We performed surgical debridement and postoperative negative-pressure wound therapy with instillation and dwell. Case 2: A 43-year-old man presented with a large calcified mass in the right leg and 2 draining sinuses. After surgical debridement, negative-pressure wound therapy was initiated. However, the wound became infected, and we performed additional debridement, followed by a split thickness skin grafting. The infection was controlled in both patients, although complete resection was not feasible. Complete resection is generally considered the optimum treatment for infected CM, but it is difficult to achieve in some patients. Negative-pressure wound therapy with instillation and dwell appears as a good option for postoperative management if complete resection of infected CM cannot be achieved. (*Plast Reconstr Surg Glob Open* 2020;8:e2817; doi: [10.1097/GOX.0000000000002817](https://doi.org/10.1097/GOX.0000000000002817); Published online 21 May 2020.)

INTRODUCTION

Calcific myonecrosis (CM) is a rare condition wherein a large calcified mass generally develops after injury.^{1,2} Infected CM with a draining sinus is a difficult condition often requiring a complete resection. Reportedly, negative-pressure wound therapy (NPWT) is useful for the postoperative management of CM.^{3,4} However, NPWT with instillation and dwell (NPWT-id) appears to be more appropriate for treating infected CM. Here, we report a case of 2 patients with infected CM requiring surgical treatment. A good outcome was achieved in one patient using NPWT-id, but the infection recurred in the other patient despite postoperative NPWT.

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CASE REPORT

Case 1

An 84-year-old woman was referred to our hospital for a chronic ulcer on her right arm. She had fractured the right humerus and developed combined median and ulnar nerve paralysis at the age of 9 years. She received treatment at that time, but the details were unknown. Physical examination showed a skin ulcer (2 cm × 2 cm) with erythema on the medial aspect of the right arm, with an exposed calcified mass (Fig. 1). Radiographs revealed a large calcified fusiform mass in the subcutaneous tissue, with its long axis being parallel to the limb. Laboratory tests did not reveal any abnormalities in the thyroid-stimulating hormone, free T4, free T3, parathyroid hormone-related protein, calcium, and phosphorus levels. Her ulcer was managed at the outpatient clinic by irrigation with normal saline. Two months later, she developed a high fever and the ulcer began to show a calcified abscess; thus, she was admitted to our hospital and administered sulbactam/ampicillin (6g/d) intravenously. A bacterial culture test detected methicillin-susceptible *Staphylococcus aureus*. Therefore, cefazolin (3g/d) was continued for 2 weeks. Surgical debridement was performed under general anesthesia. A longitudinal skin incision was made parallel to the humerus (Fig. 2), resulting in drainage of a large volume of milky-white abscess. However, a hard-calcified mass involving the

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Fig. 1. Case 1: Imaging findings: anteroposterior radiograph shows a fusiform, densely calcified mass in the arm.

brachial artery remained. Moreover, the brachial artery could not be clearly identified. Additional debridement was performed but was difficult without a tourniquet in the axilla. Histopathological findings showed only hyalinized tissue and no malignant lesion. After daily wound lavage, NPWT-id (V.A.C. Ultra, KCI, USA) was initiated on postoperative day 10. Normal saline (0.9%, 20mL) was instilled with a dwell time of 10 minutes, and NPWT was applied at -75 mm Hg for 2 hours. Four weeks later, her wound was relatively clean and had decreased in size. She was scheduled for wound closure by skin grafting, but she unfortunately died of pneumonia 3 months later. During these 3 months, no recurrence of infected CM occurred.

Case 2

A 43-year-old man was referred to our hospital with chronic discharging sinuses on the right leg and ankle. His medical history was noteworthy owing to chronic renal failure and cerebral hemorrhage with residual drop foot. He had paraplegia and usually moved in a wheelchair. Physical examination revealed wound scars on the medial aspect of the right mid-leg and lateral aspect of the ankle with induration and a discharging sinus at each site. He was administered vancomycin (0.5 g/d twice a week) intravenously for 2 weeks. A bacterial culture test detected methicillin-resistant *S. aureus*.

On plain roentgenograms, a large calcified mass was found surrounding the tibia and fibula (Fig. 3). Surgery was performed under general anesthesia. The anterior compartment was found to be filled by a calcified



Fig. 2. Case 1: Operative findings: initial debridement and calcified necrotic tissue.

mass and necrotic tissue, which were resected (Fig. 4A). Histopathological findings showed only hyalinized tissue and no malignant lesion. Daily wound lavage was performed postoperatively, and the wound became relatively clean. Thereafter, NPWT was initiated on postoperative day 30 (NPWT-id was not available in Japan then) and continuously applied at -125 mm Hg. Because his wound became infected again 2 weeks later, debridement was performed. Three months later, a split thickness skin graft was applied to cover the defect. After 5 years, functional results did not change and there was good wound healing with no signs of recurrent infection (Fig. 4B).



Fig. 3. Case 2: Imaging findings: anteroposterior radiograph shows a densely calcified mass extensively involving the leg.

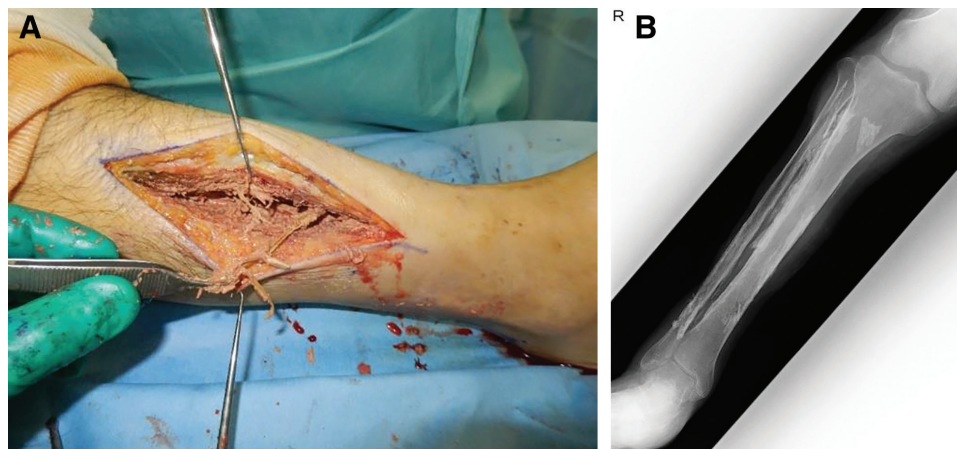


Fig. 4. Case 2: Operative findings and postoperative radiograph. A, excision at the lateral and medial aspect of the leg. B, anteroposterior radiograph shows that almost all of the calcified mass was removed.

DISCUSSION

CM was first described by Gallie in 1960.¹ It characteristically presents itself as a slowly enlarging painful mass in the lower extremity.² Almost all patients with CM can recall previous trauma, particularly lower extremity fractures, but the interval from injury to lesion development often spans several decades (range: 10–64 years).² The etiology of CM remains unknown. Once calcium regulation is impaired, calcium begins to be deposited. The deposited mass is enlarged by repeated intralesional hemorrhage. It has been hypothesized that compartment syndrome after trauma causes ischemia with recurrent bleeding and necrosis, resulting in fibrosis and subsequent calcification.⁵ Only 2 cases involving the forearm^{6,7} have been reported, and our case 1 is the first case of CM affecting the arm. CM is reportedly associated with compartment syndrome,² which is rare in the arm, and this may explain why CM is also rare in the arm. CM can be diagnosed on radiographs because of its characteristic peripheral calcification and central liquefaction.^{8,9} Diagnostic biopsy is generally avoided owing to a high risk of infection (~30%).⁷ The general consensus is that asymptomatic CM should be conservatively managed, and it can be considered a “do not touch” lesion.² In the case of infected CM, antibiotic treatment via the bloodstream is insufficient because of its avascularity, and extensive debridement is required.¹⁰ Simple debridement and direct closure or staged closure using skin grafts with NPWT appears to be the most frequently used surgical technique.³ Regarding postoperative management, NPWT appears to be useful for treating CM after complete resection. However, for infected wounds, NPWT-id is more suitable than NPWT. Although the best treatment for infected CM is additional debridement, it is impossible in some cases. Therefore, we believe that NPWT-id is a good alternative for its postoperative management.

CONCLUSIONS

We reported the cases of 2 patients with infected CM. Although complete resection of infected CM is preferable, some difficult cases exist. In such cases, we recommend postoperative NPWT-id to control infection after surgical debridement.

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