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Case report

Treatment of infected calcific myonecrosis with chronically discharging sinus caused by iatrogenic aspiration: A case report

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ABSTRACT

Introduction and importance: Calcific myonecrosis (CM) is a rare, benign post-traumatic sequela which is often challenging to differentiate from soft tissue tumors. Infected CM is recalcitrant and sometimes requires invasive treatment despite its benign nature. We present a case of infected CM in which MRI and ^{201}Tl scintigraphy proved useful for diagnosis and intralesional debridement with prolonged placement of a suction tube allowed for successful treatment.

Case presentation: A 71-year-old man had undergone repeated aspiration for swelling of the lower leg and presented with a sustained pyogenic discharging wound. He underwent intralesional debridement of purulent necrotic tissue followed by prolonged suction tube placement. *Enterobacter cloacae* was detected in the discharge, and specific antibiotics were administered. Once the wound closed, a new sinus recurred four months after surgery, warranting reoperation with debridement of the remnant fascia and necrotic tissue with suction tube replacement. The wound healed eight months after the first surgery with no signs of recurrence.

Clinical discussion: CM can be diagnosed based on its unique imaging features and a history of compartment syndrome. To avoid infection, CM must be treated conservatively without surgical invasions, such as biopsy or aspiration. Extensive debridement with a myocutaneous flap is nevertheless recommended for infected CM treatment, despite significant invasion including intraoperative bleeding being problematic.

Conclusion: MRI and ^{201}Tl scintigraphy can help diagnose CM and avoid biopsy to exclude malignancy. Intralesional debridement of necrotic tissue with prolonged suction tube placement could be a valid treatment alternative to reduce the invasiveness of infected CM.

1. Introduction

Calcific myonecrosis (CM) is a rare post-traumatic sequela with only approximately 80 cases reported in the literature [1]. CM patients have a consistent history of high-energy trauma to the limb, followed by compartment syndrome, and eventual ischemia and fibrosis of the muscles [2]. The resulting mass expands over time due to recurrent intralesional hemorrhage within the chronically calcified mass [2]. On average, CM symptoms manifest 37 years after the initial injury [3]. History of trauma and radiologic features aid physicians to differentiate CM from neoplasia [1,2,4,5]. Failure to recognize this lesion prompts a biopsy, occasionally with devastating results. Interventions such as biopsy or aspiration can contaminate the sterile necrotic lesion of CM and convert it into an abscess, leading to a sinus tract and chronic discharge [2,6,7]. Although challenging, extensive removal of necrotic tissue is essential for infected CM [4]. Surgery for CM is associated with a high risk of secondary infections, which may require multiple surgeries and sometimes amputation [2,8]. Myocutaneous flap grafts are occasionally employed, but the flap can fail to adhere because of the infection and the hypovascularity of the recipient tissues [4]. We report a case of infected CM caused by repeated aspirations which was successfully treated after two debridement and irrigation surgeries, avoiding extensive flap grafts or amputation. Written informed consent was obtained from the patient to publish this case report, including any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request. This report fulfills the current SCARE 2020 criteria [9].

Abbreviations: CM, calcific myonecrosis.

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2. Presentation of case

A 71-year-old man was referred to our hospital with swelling and sustained discharge from a wound in the anterolateral portion of the right lower leg (Fig. 1). He had undergone multiple surgeries for artery injury, cruciate ligament injury, and gastrocnemius necrosis of the right leg due to a skiing accident 46 years ago. Although his right ankle remained stiff, he could walk without assistance. Four years ago, his lower leg began to swell. Several aspirations were performed, and an ulcer wound with discharge formed on his lower leg. Three chronic wounds discharging chalky fluids were found in the lower part of the lower leg (Fig. 1). The patient had no fever. The initial bacterial culture results were negative. Plain radiography revealed an extensive speckled sheet-like calcification in the lateral lower leg (Fig. 2-A, -B). CT revealed an eggshell-like calcification surrounding the front and lateral compartments of the lower leg (Fig. 2-C, -D). The high-signal area inside the calcification and subcutaneous area on T2-weighted MRI revealed fluid collection (Fig. 2-E). No significant enhancement and ^{201}Thallium (Tl) uptake were found on enhanced MRI (Fig. 2-F) and ^{201}Tl scintigraphy. The ESR was 39 mm/h and the CRP level was 0.3 mg/dL. The complete blood count was within normal limits. The surgery was performed under general anesthesia by two well-trained orthopaedic surgeons. During surgery, after cutting the thickened deep fascia (Fig. 3-A), the anterior and posterior muscles were necrotic and had changed into rotten-wood-like materials and chalky white deposits (Fig. 3-B, -C). The entire necrotic tissue was removed to avoid sacrificing the peroneal nerve (Fig. 3-D, -E). Two drain tubes were inserted into the anterior and lateral spaces, and primary closure of the wound was performed to repair the three ulcer lesions (Fig. 3-F). Histological evaluation confirmed a wide range of necrosis of the muscle, fat tissue, and vasculature, with coagulation and scattered calcification deposits (Fig. 4). No atypical cells were found. After surgery, the fluid continued to drain through the drainage tube, and even though the wound after the drainage was removed. The wound healed two months after surgery (Fig. 5-A). However, a new sinus formed with discharge contaminated with *Enterobacter cloacae*, which was sensitive to cefdinir administered postoperatively (Fig. 5-B). Thus, reoperation with debridement and irrigation was performed four months after the initial surgery. The infected granulation tissue and the remaining fascia were completely removed and irrigated with large amounts of normal saline. Two drains were placed, and the wound was closed (Fig. 5-C, -D). *Enterobacter cloacae* was detected in the fluid collected intraoperatively, and levofloxacin was administered to the patient after surgery. The patient was discharged two weeks after surgery and continued wound treatment almost once a week for a small amount of discharge at the outpatient department. Levofloxacin was switched for sulfamethoxazole trimethoprim for long-term oral administration until the discharge stopped. The discharge did stop, the wound was completely healed, and no fluid collection was found on MRI eight months after the first surgery (Fig. 5-E, -F). There were no signs of recurrence or infection at the final follow-up one year after the initial surgery. The patient was satisfied to be able to walk without assistance.

3. Discussion

CM is a benign entity that develops from originally sterile necrotic tissue as a result of compartment syndrome, ischemia, and eventual fibrosis [10–12]. CM can be confidently diagnosed by a history of high-energy trauma with compartment syndrome and its unique imaging characteristics [2,10]. Radiography or CT images show peripheral linear plaque-like or dense eggshell-like calcification within an atrophied muscular compartment [2,3]. MRI shows a well-circumscribed

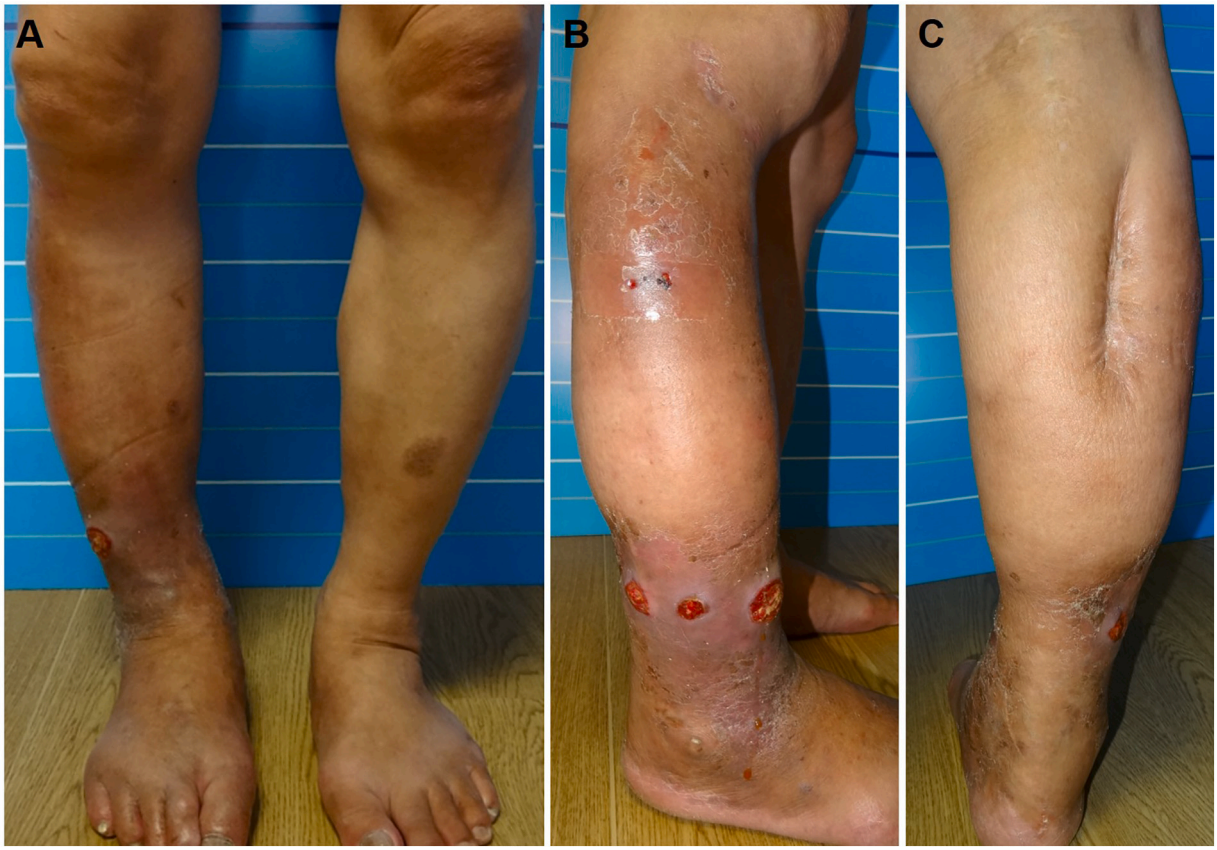

Fig. 1. Clinical photographs of the lower legs. Frontal view (A). The lateral view of the right lower leg shows three discharging sinuses (B). The rear view of the right lower leg shows an old surgical scar (C).

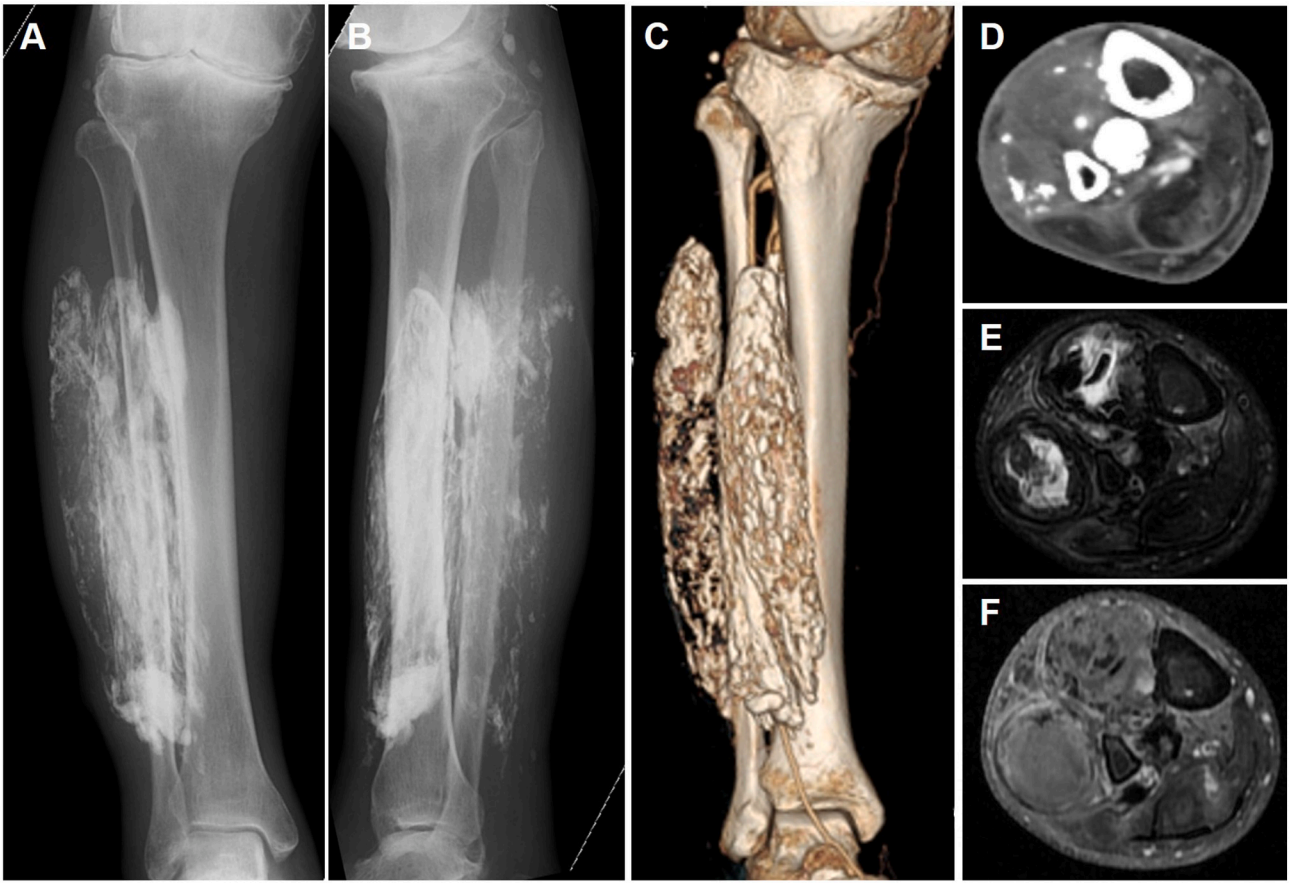

Fig. 2. Plain radiography of the right lower leg. Frontal view (A) and lateral view (B). 3D-CT image (C). Axial image of enhanced CT shows eggshell-like calcification of anterolateral side and bulk calcification in tibiofibular area (D). Axial T2-weighted image with fat suppression shows high-signal area reflecting fluid collection (E). Coronal gadolinium contrast image shows no significant enhancement (F).

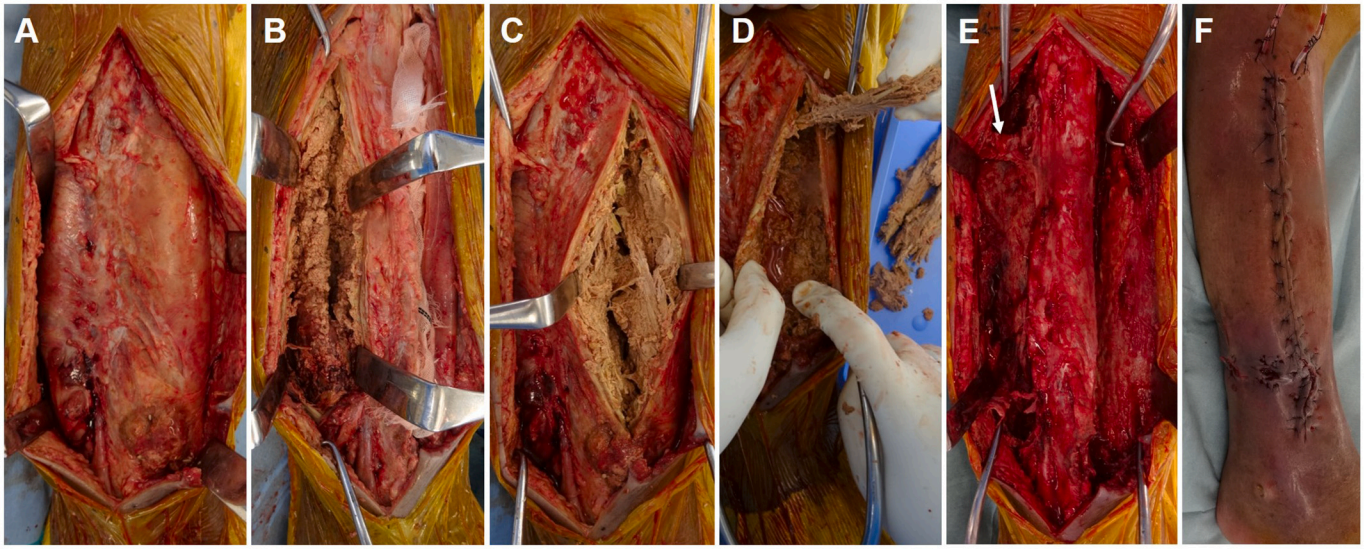

Fig. 3. Intraoperative photographs. Thickened deep fascia (A). Cutting of the anterior compartment fascia (B) and lateral compartment fascia (C). Debridement and curettage of the necrotic muscle fibers (D). All necrotic tissue was removed, and the peroneal nerve (arrow) was preserved (E). Wound closure with two drains (F).

isointense mass, with the muscles on T1- and T2-weighted images appearing hyperintense, reflecting central liquefaction [12–14]. Soft tissue tumors presenting calcification, such as epithelioid sarcoma, synovial sarcoma, and soft tissue osteosarcoma, should be excluded [1,13]. In the present study, contrast MRI and ^{201}Tl scintigraphy were performed to exclude malignant soft-tissue tumors, and no contrast enhancement or Tl-uptake was observed, supporting our diagnosis.

CM must be treated conservatively, without any surgical invasion, including aspiration or biopsy, after diagnosis [2,14,15]. Since CM is composed of abundant avascular necrotic tissue, once contaminated, the

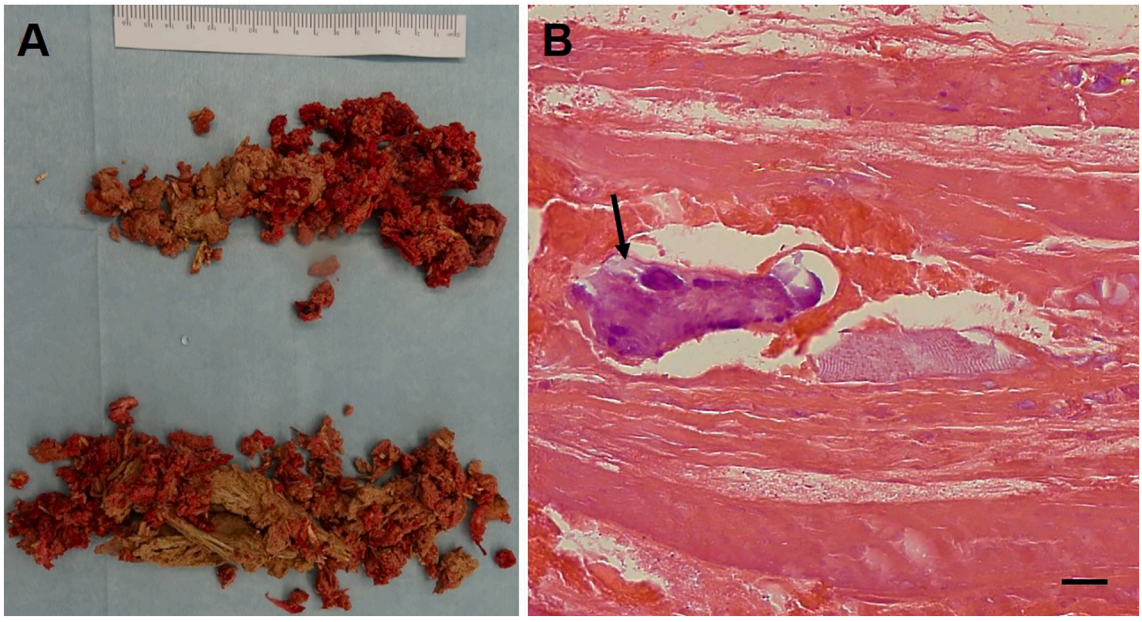

Fig. 4. Curetted necrotic tissue (A). Hematoxylin and eosin staining shows necrotic muscular fiber and calcification deposits (arrow) without atypical cells (B). Scale bar indicates 100 μm.

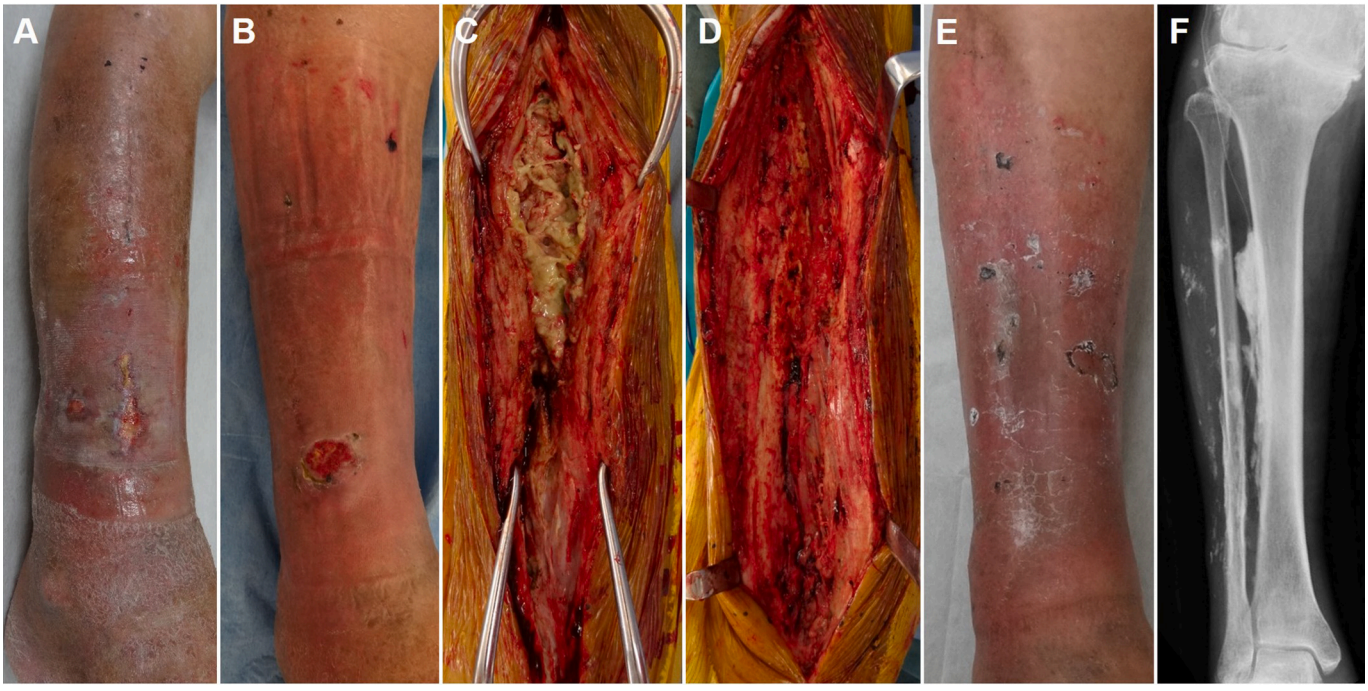

Fig. 5. Two months postoperative clinical photographs. There was only little discharging and the wound was almost healed (A). Large amounts of discharge recurred and a new sinus formed four months after surgery (B). Second surgery; the infected granulation tissue was covered by thickened fascia (C). Debridement of granulation and fascia (D). Completely healed wound after eight months from the first surgery (E). Plain radiography at the final follow-up one year after the first surgery (F).

infection spreads throughout the lesion, forming one or more abscesses within it, resulting in intractable fistulas [2,6,7]. It has been reported that most infected CM cases had previously undergone invasive procedures, including biopsy [2,8]. A biopsy of a CM lesion may be performed on suspicion of a soft-tissue tumor [10]. A previous report described CM as a "don't-touch" type of lesion, because of the challenges associated with its treatment [2].

High rates of postoperative complications have been reported. Approximately 30% of these relate to infection or fistulas formation [10,16]. To avoid secondary infection, suction drain tubes placed under the wound and compression dressing are recommended [6]. For patients with aggressively infected CM, extensive debridement and myocutaneous flaps are reported to be a valid treatment alternative [4,17]. However, extensive debridement of CM is associated with a risk of severe intraoperative bleeding [13]. Collateral arteries develop in the long term after interruption of blood flow to the muscle as a result of contusion injury to the muscle, and often to the main arteries as well [13]. Okada and colleagues reported an intraoperative bleeding of 2.4 L

in a CM patient who underwent extensive debridement [13]. In the present case, collateral arteries from the popliteal artery and an arterial aneurysm, which are often observed in CM patients, were detected in the posterior tibial artery near the deep calcified lesion.

The use of a myocutaneous flap after extensive debridement has been reported, although mycocutaneous flaps require invasion of a donor site, despite the fact that CM is a benign entity [4]. Several authors reported successful treatment of CM with the use of prolonged suction drain tube placement, followed by the application of a bulky compressive dressing [1,10,15]. The use of limited access dressing or vacuum-assisted closure has been reported to be effective in managing empty spaces left after debridement [3,18,19]. In the present case, two drain tubes placed in the dead space after debridement proved to be effective in both surgeries.

4. Conclusion

Asymptomatic CM should be managed conservatively. To avoid unnecessary surgical invasion, which can lead to infected CM, physicians should know and recognize this as a “don't touch” lesion when encountering patients with the characteristic manifestations and history of compartment syndrome. The absence of accumulation on enhanced MRI or ^{201}Tl scintigraphy aids diagnosis and avoids biopsy. In case of infected CM, intralesional debridement with prolonged suction drain tube placement and adequate antibiotic administration could be a valid and preferrable treatment option to reduce its invasiveness. However, extensive debridement with a myocutaneous flap might be a necessary treatment alternative in cases of massive infections, skin infection, and soft tissue spreading to multiple compartments, though only when treatment of intraoperative damage to collateral arteries and blood transfusions can be performed.

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Ethical approval

For this type of study, ethics approval was waived by the Ethics Committee of the Kanazawa University Hospital.

Consent

Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images.

Author contribution

Takashi Higuchi: Participated in the surgery, data collection, case analysis, and writing of the manuscript. Norio Yamamoto: Supervised and analyzed the case data. Hideji Nishida: Participated in the surgery and collected the data. Katsuhiro Hayashi: Validated the case data. Akihiko Takeuchi: Validated the case data. Hiroyuki Tsuchiya: Conceptualization and supervision.

Research registration

This is not a research study.

Guarantor

Norio Yamamoto, MD, PhD.

Provenance and peer review

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Declaration of competing interest

No benefits, in any form, have been received or will be received from a commercial party related directly or indirectly to the subject of this article.

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