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Calcific Myonecrosis Mimicking an Invasive Soft-Tissue Neoplasm

A CASE REPORT AND REVIEW OF THE LITERATURE*

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Investigation performed at University of California, Davis, Medical Center, Sacramento

Calcific myonecrosis has been reported as a late sequela of compartment syndrome, injury to the common peroneal nerve, and injury to the lower extremity without documented compartment syndrome or neurological injury^{1-8,13-19}. This rare condition has been reported to occur ten to sixty-four years after the initial injury and typically presents as an enlarging mass in the anterior compartment of the leg. The characteristic radiographic appearance is that of a large fusiform soft-tissue mass in the anterior compartment, with peripheral plaque-like calcifications and usually with a well defined border. The calcifications may extend along fascial planes¹³. Erosion of bone has been reported in only four patients^{8,13}. The benign radiographic appearance usually allows the lesion to be differentiated from an enlarging malignant mass in the soft tissues⁷. A sterile abscess usually is found at the time of operative treatment, but there is a high prevalence of chronic draining sinuses and secondary infection^{3,7,8,18}.

We report the case of a patient who was seen because of a painless, enlarging mass in the anterior and lateral compartments of the leg thirty years after he had been hit by an automobile. At the time of the initial injury, he had sustained damage to the knee, a partial sciatic-nerve palsy, and a probable compartment syndrome of the leg. The case of our patient differs substantially from previously reported cases of calcific myonecrosis in that there was extensive erosion of bone, giving the lesion the appearance of an invasive neoplasm.

Case Report

A forty-nine-year-old man was referred for evaluation of a slowly enlarging mass in the right leg. Thirty years previously, he had been struck by an automobile while walking. He stated that he had sustained an injury to the ligaments of the right knee as well as a partial loss of motor and sensory function in both divisions of the sciatic nerve, with associated weakness of dorsiflexion of the ankle. Soon after the injury, he had repair of the ligaments of the right knee. The

medical records from the time of the injury and the operation were not available, and the patient could not recall which ligaments had been repaired. Fifteen years after the injury, he noticed a mass in the anterolateral aspect of the leg; the mass slowly increased in size. The medical history was otherwise unremarkable, and the patient did not have diabetes.

Physical examination revealed massive enlargement of the right leg: the circumference of the calf was thirty-seven centimeters compared with twenty-four centimeters on the contralateral side. The mass predominantly involved the anterior and lateral compartments of the leg and had the consistency of a tense, fluid-filled cyst. The range of motion of the knee was 0 to 100 degrees of flexion. The ankle had a 5-degree equinus contracture and 5 degrees of plantar flexion from this position. There was no motion of the subtalar joint. There was clawing of all of the toes. Motor strength was grade 0 (of 5) for dorsiflexion of the ankle, grade 3 for plantar flexion of the ankle, grade 0 for dorsiflexion of the toes, grade 0 for plantar flexion of the toes, grade 0 for eversion of the hindfoot, and grade 0 for inversion of the hindfoot. Sensation was not present in the dorsum of the first web space and was subjectively diminished on the dorsal, plantar, medial, and lateral aspects of the foot. The contractures in the limb were consistent with the residua of a deep posterior compartment syndrome^{9,10}.

Imaging Studies

Radiographs of the right leg that had been made when the patient was forty-two years old (seven years before he was seen by us) showed a large mass. The middle third of the fibula was sclerotic, and the tibia appeared uninvolved (Fig. 1-A). Plain radiographs of the right leg that were made when the patient was seen by us showed a large fusiform soft-tissue mass with extensive plaque-like and amorphous calcifications. The middle portion of the fibula was eroded, and there was a small area of cortical erosion involving the lateral aspect of the tibia (Fig. 1-B). The pathological process appeared to have progressed markedly since the time of the previous radiographs. Magnetic resonance imaging demonstrated a non-homogeneous mass that had eroded and, in some areas, completely destroyed portions of the fibula (Fig. 2).

Operative Findings

The skin, subcutaneous tissue, and deep fascia appeared normal at the time of the operation. All of the tissues in the anterior and lateral compartments of the leg were completely necrotic, and bone spicules resembling rice bodies permeated the tissue, giving it a gritty texture. There was erosion of the anterior and lateral borders of the entire diaphysis of the fibula as well as scalloping of the posterior and medial borders, which were widely expanded into a thin shell over a distance of twelve centimeters. There was minimum erosion of the tibial cortex. No neurovascular bundles were present in either compartment. The blood loss was approximately 1600 milliliters, and two units of packed red blood cells were transfused during the operation.

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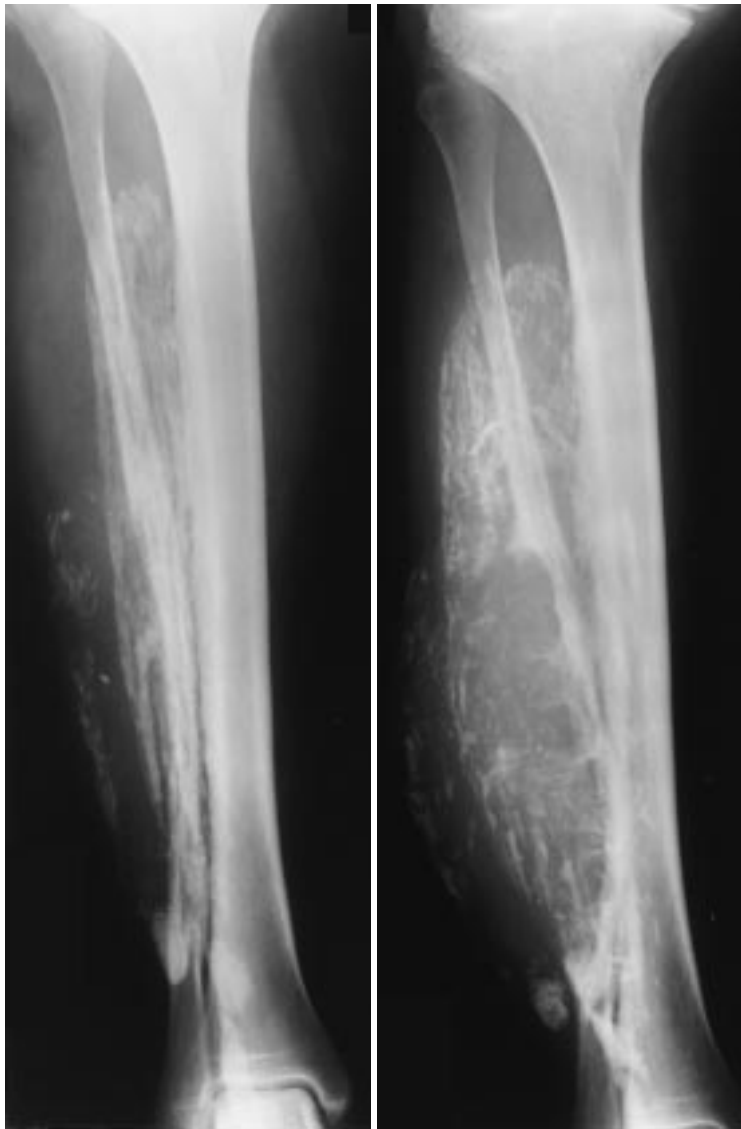


FIG. 1-A

FIG. 1-B

Fig. 1-A: Radiograph made seven years before the patient was seen by us, demonstrating a mass with plaque-like and amorphous calcifications. The middle third of the fibula was sclerotic.

Fig. 1-B: Plain anteroposterior radiograph of the right leg, made when the patient was seen by us, showing a large fusiform soft-tissue mass with extensive plaque-like and amorphous calcifications. The mass was much larger than it had been seven years previously. The middle third of the fibula was partially destroyed. The lateral cortex of the tibia was thickened and sclerotic, with peripheral erosion.

Frozen sections of multiple samples of tissue were evaluated in order to rule out malignant disease. Analysis of all of the specimens revealed necrotic debris, blood, fibrin, and multifocal areas of calcification. Specimens were sent for aerobic, anaerobic, fungal, and mycobacterial culture; all cultures were negative. The wound was closed over a suction drain, and a bulky compressive dressing was applied to the leg. Postoperatively, while still in the hospital, the patient was managed empirically with intravenous administration of antibiotics, including cefazolin (one gram every eight hours for three days) and gentamicin (400 milligrams every twenty-four hours for two days). The wound healed without drainage or infection. At the fourteen-month follow-up evaluation, the patient had no additional problems and no evidence of recurrence.

Pathological Findings

The pathological evaluation revealed that the major resection specimen consisted of necrotic tissue, skeletal muscle, and scant fragments of bone; the specimen was twelve by five by five centime-

ters. Histological evaluation demonstrated mostly necrotic soft tissue and bone admixed with blood and fibrin. Extensive calcification was noted. The wall of the cyst was densely fibrotic. The histological diagnosis was necrotic tissue.

Discussion

To our knowledge, eighteen cases of calcific myonecrosis of the leg were reported in the English-language literature between 1960 and 1996^{2-8,13,15-17,19}. The patient who was seen earliest was reported on by Gallie and Thomson and was followed from 1914 until 1952. The condition is rare, and no individual surgeon has had extensive experience with it; the largest reported series consists of only three patients¹³. The differential diagnosis of calcific myonecrosis includes malignant tumors that undergo calcification, such as epithelioid sarcoma,

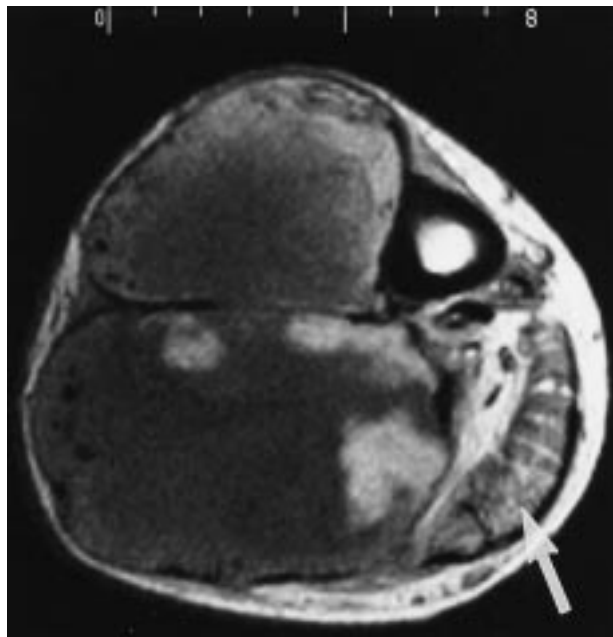


FIG. 2

Axial spin-echo T1-weighted magnetic resonance image made after the administration of gadolinium, showing a lack of enhancement of a bilobular, sharply defined non-homogeneous mass that had completely destroyed the posterolateral aspect of the fibula. The nerves and vascular structures also were affected. At this level, almost all of the muscles were replaced by the necrotic mass. The only spared muscle was the medial head of the gastrocnemius (arrow).

synovial sarcoma, and soft-tissue osteosarcoma¹³. Seven of the eighteen patients who were reported on in the literature had complications of treatment, such as persistent drainage, infection, and poor wound-healing necessitating multiple débridements^{7,8,13,17,18}.

A history of trauma to the extremity with ischemia or neurological injury seems to be a prerequisite for the development of calcific myonecrosis. A fracture of the tibia preceded the development of calcific myonecrosis in eight of the patients who were described in the literature. Five of these eight patients had a definite history of compartment syndrome or frank vascular injury, or both^{4,6,13,15}. The remaining three patients had associated physical findings that were suggestive of a possible ischemic complication of the tibial fracture or a neurological injury; specifically, one patient had a footdrop³, another had weakness of dorsiflexion of the ankle¹³, and the third had associated weakness of the muscles of the anterior compartment and decreased sensation in the first web space¹⁹.

A femoral fracture preceded the development of calcific myonecrosis in five patients. Two of these patients had a specific history of ischemia^{2,5}. The remaining three patients had associated physical findings that were suggestive of ischemia. Specifically, one patient had atrophy of the calf, a fixed equinus deformity of the ankle, and clawing of the toes¹⁹; another had a severe equinovarus deformity and absent muscle power in the anterior compartment of the leg⁵; and the third had a

footdrop that had developed during the treatment of the initial injury¹⁷. These clinical findings are consistent with a history of unrecognized ischemia of the anterior or deep posterior compartment, or both^{9,10}.

The initial injuries in the remaining five patients included a dislocation of the knee necessitating vascular reconstruction³, a gunshot wound to the thigh with an aneurysm of the femoral artery⁸, blunt trauma with associated compartment syndrome¹⁷, fractures of the femur and the ipsilateral tibia necessitating vascular reconstruction⁶, and a fracture of the fibular neck with transection of the common peroneal nerve⁷. On the basis of the latter case, Janzen et al. reported that calcific myonecrosis appeared to be associated with peripheral nerve injury with or without associated compartment syndrome or ischemia. However, most of the reported cases suggest that the common denominator of calcific myonecrosis is ischemia of the muscles. Myonecrosis of the muscles of the leg may occur without a history of trauma in a patient who has diabetes¹².

Calcific myonecrosis typically presents as an enlarging mass in the anterolateral aspect of the leg. Eight of the eighteen patients who were described in the literature had had the mass for one year or less before they sought medical attention^{1,3,8,13,17}; two others had had the mass for more than ten years⁵. The time between the initial injury and the clinical presentation of calcific myonecrosis has ranged from ten to sixty-four years^{2-8,13,15-17,19}. A common finding at the time of the physical examination is a tense fusiform mass that may replace the entire anterior compartment of the leg. In fourteen of the eighteen known cases, the mass was confined to the anterior compartment^{2-5,7,8,16,17}; in two, it was found in all four compartments^{13,19}; in one, it was located primarily in the anterior compartment and partially in the deep posterior compartment⁶; and in one, it was confined to the posterior compartment¹⁵. Seven of the patients had pain in association with the mass, and ten noticed progressive enlargement of the lesion.

All but one of the case reports on calcific myonecrosis have included a radiographic description of the lesion. The typical finding is a large fusiform soft-tissue mass in the anterior compartment of the leg that often completely replaces a muscle. The lesion frequently has peripheral plaque-like calcifications, which may extend along fascial planes¹³. Gravitational settling of the calcifications was noted in one patient⁷. Erosion of the bone adjacent to the mass was noted in four patients^{8,13}. One of these patients, who was reported on by Malisano and Hunter, had scalloping of the tibia. The other three patients, all of whom were reported on by O'Keefe et al., had varying degrees of erosion; specifically, one patient had smooth erosion of the tibia, one had minimum erosion of the tibia associated with a smooth periosteal reaction, and one had extensive erosion of the fibula with chronic reactive periostitis and cortical thickening of the tibia.

Our patient had destruction of the posterolateral aspect of the fibula, beginning several centimeters distal to the fibular head and extending all the way to the ankle joint. The invasive appearance of the lesion led us to suspect a calcifying soft-tissue sarcoma, such as synovial sarcoma, epithelioid sarcoma, or soft-tissue osteosarcoma¹³. The radiographic appearance of destruction of the fibula in our patient was similar to that observed by O'Keefe et al., but the appearance of the fibular destruction in our patient led to a presumptive diagnosis of a secondary aneurysmal bone cyst.

Operative treatment of calcific myonecrosis has been associated with serious complications. Three of the patients who were described in the literature sought medical attention after one to eight months of persistent drainage following an open biopsy^{3,17}. Both of the patients in the report by Snyder et al. needed serial débridements, and one needed wound closure with use of a rectus abdominis free flap, before healing occurred. One of the two patients in the report by Viau et al. had chronic intermittent drainage two and a half years after débridement. The other patient in that study had repeat débridement four months after the original procedure; an infection with *Staphylococcus aureus* developed despite this treatment, and a below-the-knee amputation was done seven months later. Malisano and Hunter reported on a patient who, despite incision and drainage, continued to have growth of several microbacterial organisms on culture of specimens that were obtained from a chronic draining sinus five years after excision of the mass. Janzen et al. reported on a patient who had a chronic draining infection after an open incisional biopsy. O'Keefe et al. reported the case of a seventy-two-year-old man who lost 800 milliliters of blood during the excision of a calcified necrotic mass in the leg. The patient became hypotensive and had a cardiac arrest at the conclusion of the procedure; two months later, after a prolonged hospital course, he died of gastrointestinal complications.

The pathophysiological mechanism that leads to late calcific myonecrosis is not known. It has been hypothesized that an initial compartment syndrome increases the pressure and decreases the circulation within a limited space, leading to necrosis and fibrosis. With time,

the mass may enlarge as a result of repeated intraleisional hemorrhage¹³. Mentzel et al. reported on four patients who had a similar pathological condition that had occurred in or over the tensor fasciae latae muscle; in each case, the mass had developed six weeks to twenty years after an episode of blunt trauma. Histological examination of all of the specimens demonstrated blood clots and necrotic debris in the subcutaneous, perifascial, and intramuscular cavities. Other findings included a chronic inflammatory infiltrate and dystrophic calcification consistent with chronic organizing hematoma associated with reactive inflammation¹¹. These histological findings are similar to those associated with calcific myonecrosis of the leg^{13,18}. Mentzel et al. hypothesized that the initial injuries in their patients had continued to bleed into the tight fascial space of the tensor fasciae latae. Those authors postulated that a cycle of repeated hemorrhage into this space, coupled with increased pressure caused by stretching of the iliotibial tract, was responsible for the ultimate cystic degeneration. They suggested that the similar clinicopathological entities of calcific myonecrosis, chronic expanding hematoma¹⁴, and posttraumatic cyst of soft tissues^{1,18} should all be considered together as a single entity, which they called ancient hematoma¹¹.

In conclusion, calcific myonecrosis of the leg may present as a slowly enlarging mass in the anterior compartment, months or years after an episode of blunt trauma. The history or physical appearance may suggest that the involved muscles in the compartment sustained ischemic damage as a result of the initial injury. Plain radiographs are usually diagnostic, with the lesion appearing as a fusiform, peripherally calcified soft-tissue mass. However, in isolated cases, such as the one described here, the presence of extensive fibular destruction may be highly suggestive of an invasive soft-tissue neoplasm. If an operation is performed, we believe that all necrotic tissue should be completely removed in order to avoid such problems as chronic drainage and delayed wound-healing. If only the anterior compartment is involved, partial elimination of the anterior dead space and restoration of some control to the foot by transfer of the posterior tibialis muscle through the interosseous membrane may be performed⁸.

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