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Bilateral Lower Extremity Calcific Myonecrosis: A Case Report

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Learning Point of the Article:

To consider calcific myonecrosis as a potential etiology for masses following trauma or large muscle group injury and understand that tenotomy or partial mass excision may be viable treatment options for alleviating associated symptoms.

Abstract

Introduction: Calcific myonecrosis is a rare soft-tissue disease where a single muscle or muscle compartment is replaced with central liquefaction and peripheral calcification. This disease usually occurs in a single limb after trauma. Until now, bilateral muscle involvement has not been previously reported.

Case Report: A 73-year-old woman presented with symptomatic masses in the soft tissues of bilateral anterior thighs. She had no known history of trauma but recalled a specific tearing sensation episode in both her legs while squatting when weightlifting in the distant past. The patient had calcified masses that had replaced the rectus femoris muscle bilaterally with associated effects on hip and knee range of motion. The patient underwent excision of a portion of calcific myonecrosis and two locations of rectus femoris tenotomies in one leg and only a simple rectus femoris tenotomy on the contralateral leg. The patient subsequently experienced a significant improvement in hip and knee motion and a reduction in associated pain on both sides.

Conclusion: Historical and radiographic information are key in making the diagnosis of calcific myonecrosis. Treatment decisions should be based on the patient's symptoms balanced with the morbidity of the expected procedures. Symptomatic patients should be considered for excision of the symptomatic involved areas. A tenotomy may be a viable option if a limitation in motion is the primary complaint. Asymptomatic patients with a stable lesion and imaging should undergo conservative management due to the high risk of infection and wound healing problems.

Keywords: Oncology, infection, calcific myonecrosis, tumor.

Introduction

Calcific myonecrosis is a rare soft-tissue disease that was first described by Gallie and Thompson in 1960 [1]. The pathogenesis is likely thought to be due to a traumatic event, oftentimes fracture that subsequently leads to compartment syndrome and years later evolves into calcific myonecrosis. The most common inciting injuries include fibular fracture, tibial shaft fracture, femoral shaft fracture, rupture of the femoral artery with ischemic muscle necrosis, and peripheral nerve injury [2,3]. The most common documented locations are in the lower leg muscles including the tibialis anterior, peroneus longus, and extensor hallucis longus. One difficult aspect of diagnosing this problem is the delayed presentation, as it can present decades after the inciting trauma. Radiographic and historical information are essential in making this diagnosis and

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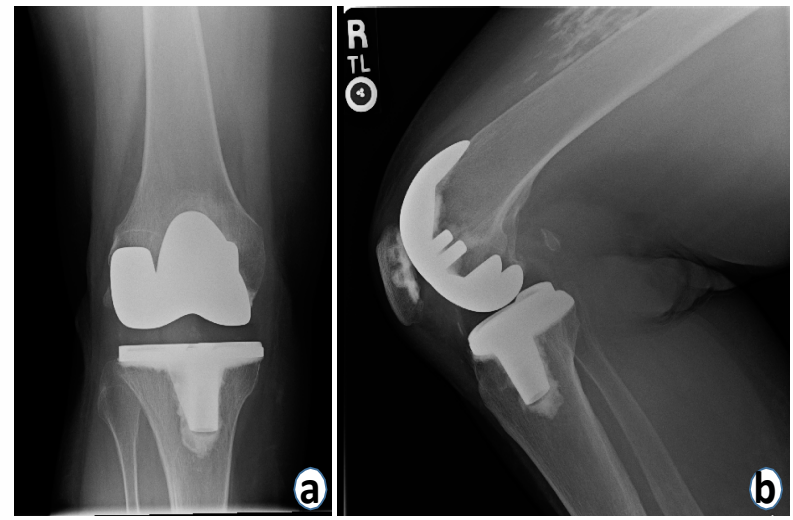

Figure 1: (a) Anteroposterior and (b) lateral radiographs of the right leg of a 73-year-old woman with calcified myonecrosis infiltrating the anterior compartment of the thigh.

delivering the appropriate management to your patient.

To enhance the literature, this article presents a 73-year-old woman with calcific myonecrosis of bilateral legs that developed without a prior diagnosis of trauma or related compartment syndrome. To the best of our knowledge, this is the first report of calcific myonecrosis in bilateral lower extremities without a previously diagnosed inciting factor.

Case Report

A 73-year-old female presented to our clinic with a chief complaint of progressive right leg pain which localizes to the lateral hip and radiates down the side of her right leg. She also presented with low back pain and abnormal gait. The patient describes bilateral palpable masses/lumps in the anterior aspect of both legs which are bothersome and limit range of motion. Medical history is significant for hypertension, anxiety, chronic pain syndrome, right total hip, and total knee arthroplasties. Her right total hip arthroplasty procedure required revision due to a leg length discrepancy. She participated in body building with heavy weights while she was in her forties, and she recalls one specific event where she felt a tearing sensation in both thighs while squatting heavy weight.

On initial examination, the patient appeared to have a slightly longer right leg compared to left, and her lumbar spine was hyper lordotic. That patient was exquisitely tender over her greater trochanter and IT band. Examination showed symmetric palpable masses along the anterior aspect of bilateral thighs, consistent with the rectus femoris muscle. Her right knee had 2° of hyperextension and flexion to 120° when her hip was allowed to also flex. However, her right knee would only flex to 30° when her hip was kept at neutral extension. Her knee was stable to ligamentous examination. Her right hip was stable with minimum discomfort on motion testing. She did have an equivocal straight right leg raise test. She also had numbness along the right lateral thigh consistent with the lateral femoral cutaneous nerve distribution. The left leg was neurovascularly intact throughout and non-tender to palpation. The range of

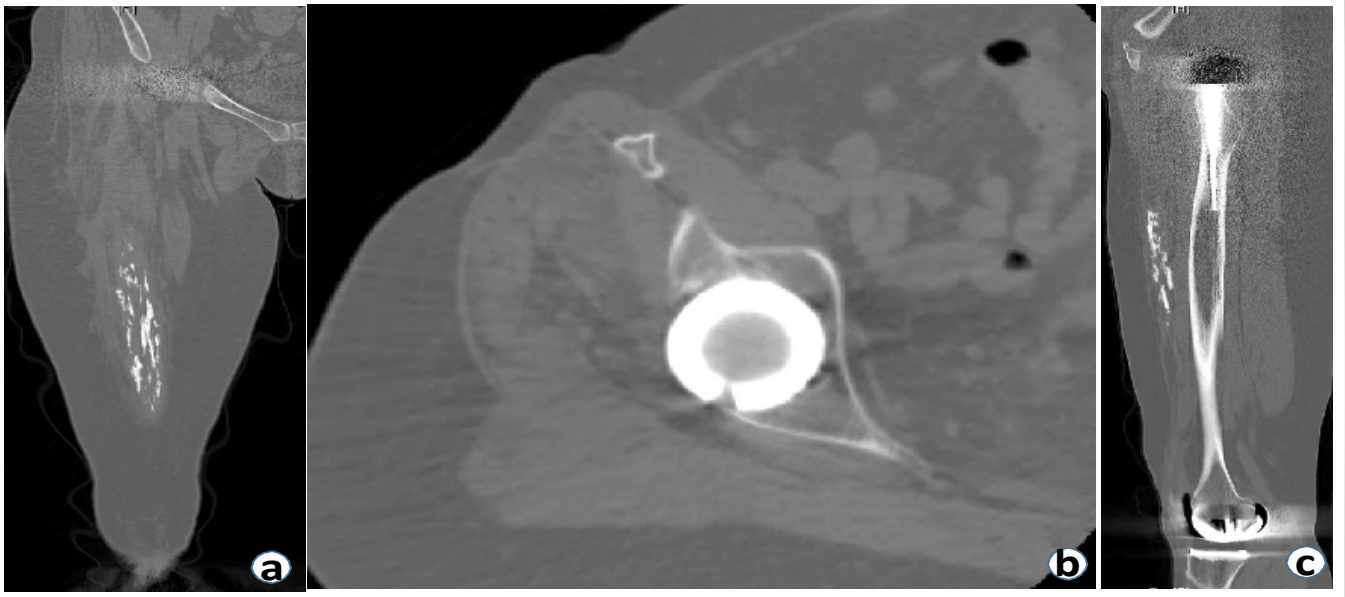

Figure 2: (a) Coronal, (b) axial, and (c) sagittal computerized tomography scan of the right leg show a calcified fusiform mass with peripheral plaque-like calcifications linearly oriented in the anterior compartment of the thigh. Figure (b) specifically shows the most proximal portion of the calcification and the anterior inferior iliac spine avulsion.

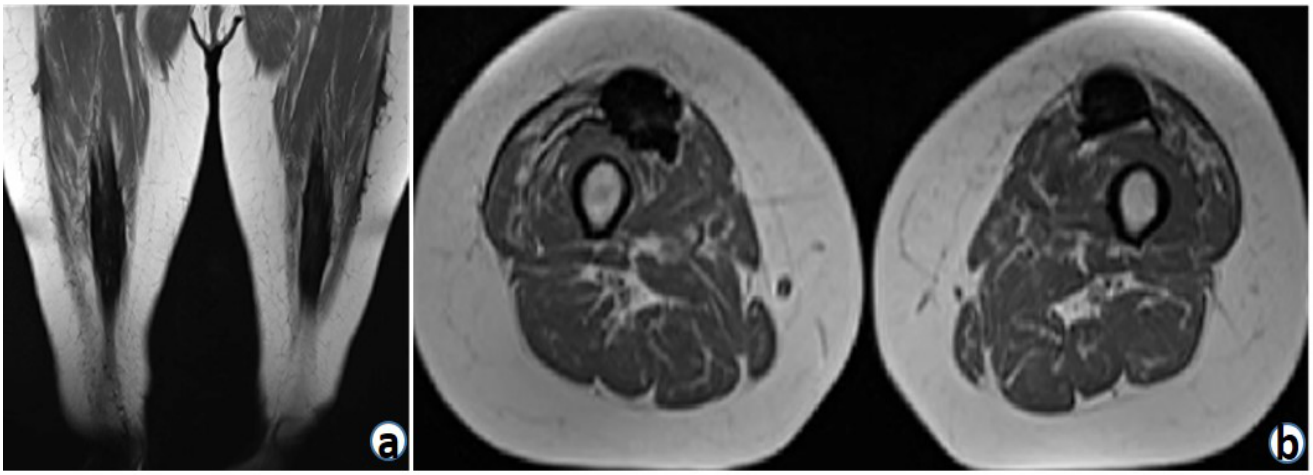

Figure 3: Coronal (a) and axial (b) magnetic resonance images showing the bilateral nature of this patient's calcific myonecrosis. The low T1 signal throughout the course of the rectus femoris muscles is consistent with calcification and necrosis.

motion examination of her left knee was very similar with flexion to 120º when her hip was allowed to flex and 30º if her hip was held in neutral extension.

Radiographs of the right knee showed a stable total knee implant with no evidence of loosening, and the lateral radiograph showed linear calcifications in the anterior muscle compartment of the thigh (Fig. 1).

Computerized tomography (CT) scan of the right leg showed a calcified fusiform mass with peripheral plaque-like calcifications linearly oriented throughout the entire course of the rectus femoris muscle. (Fig. 2). The CT scan also demonstrated an avulsion of her anterior inferior iliac spine.

Axial and coronal magnetic resonance (MR) images of bilateral legs showed a heterogenous lesion that was predominantly dark on all imaging sequences representing a calcified mass that has replaced the entire rectus femoris muscle (Fig. 3).

No mechanical issues were identified with the hip or knee arthroplasty implants. Initial treatment of trochanteric bursitis relieved her pain about her greater trochanter, but she continued to have significant limitations due to the masses and secondary effects on hip and knee motion. The option of removing a portion of the calcific myonecrosis and releasing the restrictive band was offered to the patient.

Treatment

The patient initially had the right leg addressed with a proximal anterior approach to the hip with excision of a 3 cm portion of the calcific myonecrosis and avulsed AIIS with associated release of the proximal rectus femoris tendon (Fig. 4 and 5). The

Figure 4: Clinical image prior before surgical intervention showing the limitations in knee range of motion when the hip is kept extended.

Figure 5: Image demonstrating the improvement in knee range of motion after the proximal and distal tendon releases and partial calcific myonecrosis excision.

author then moved to the distal third of the anterior thigh and excised a 4.5 cm portion of the calcified distal rectus femoris with distal rectus femoris tenotomy (Fig. 6). In both instances, fluoroscopic guidance was used to identify the location of calcified fragments. The deep fascia in both operative sites was closed without filling the extra space left over from the mass excision, which is similar to a previous study by Chun and Shim [3]. Intraoperatively, after completion of the proximal and distal portions of the procedure the patient's range of motion improved from 30 º to 90° of knee flexion with her hip extended.

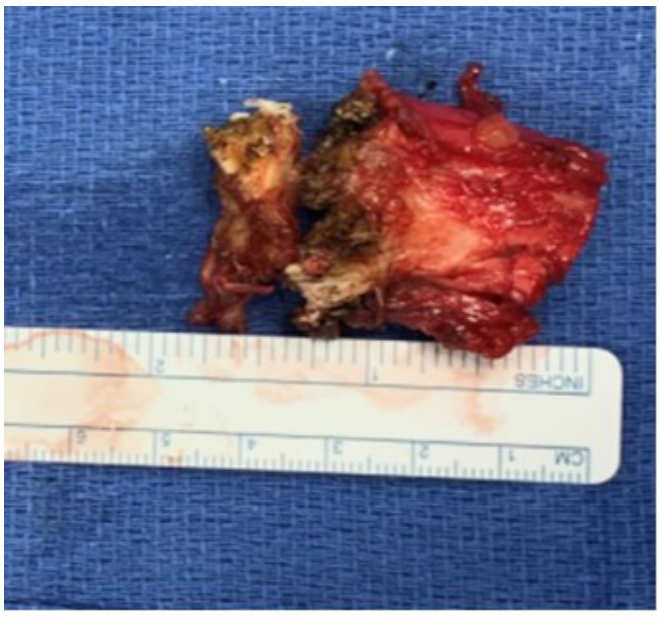

Figure 6: Image demonstrating the 4.5 cm excised portion of calcified tissue.

Postoperatively, there were no complications, and the patient reported significant discomfort relief and maintained the range of motion gains.

Due to a successful outcome on the right leg, the patient elected to proceed with a similar procedure on the contralateral side. 2 months later the patient returned to the operating room and an incision was made 6 cm proximal to the patella and dissected down to the rectus femoris tendon. A tenotomy of the rectus femoris and underlying vastus medialis was performed without mass removal. Once both tendons were released, the patient could flex her knee to 90° with hips fully extended, and this was equal to the outcome on the contralateral side. The hip was not approached on this side as the patient did not have the same mechanical symptoms associated with the AIIS avulsion identified on the contralateral side. Postoperatively, there was no evidence of infection or wound healing problems. The patient maintained her range of motion gains.

The patient was followed postoperatively until the 1-year mark and was noted to subjectively be happy with her lack of pain and greatly improved level of function. Her intraoperative range of motion was maintained. She reported an improvement in her back symptoms as it was suspected the effects on the range of motion of her hips was leading to excessive lordosis and stress in her lumbar spine.

Discussion

Calcific myonecrosis is a rare disease that was first reported by Gallie and Thompson in 1960 [1]. It is thought to arise slowly for many decades following compartment syndrome which occurred as a sequela of major trauma [4, 5]. The average mean time between injury and diagnosis of calcific myonecrosis in several documented cases has been 37 years [6]. The vast majority of documented cases occur in the lower extremity, specifically the anterior compartment of the lower leg [2, 3, 5-15]. However, there are a few cases in the literature where the upper extremity is the location of pathology [16, 17].

In this condition, an entire single muscle, or muscle compartment is replaced with central liquefaction and peripheral calcification resulting in a central cystic mass in the muscle. The cyst consists of necrotic muscle with cholesterol crystals, fibrin, and thrombus (hemosiderin-laden macrophages) [2]. The mass is commonly noted to expand, and this is possibly due to the recurrent intralesional hemorrhage which may occur [18]. The pathophysiology of this condition has not been fully delineated, but it is plausible that the necrosis, fibrosis, and liquefaction stems from the initial

insult of a compartment syndrome.

The differential diagnosis for this condition should include soft-tissue sarcomas that have a propensity for calcifications or ossifications such as synovial sarcoma, epithelioid sarcoma, and extraskeletal osteosarcoma. Benign lesions such as myositis ossificans, diabetic myonecrosis, inflammatory disease, posttraumatic pseudoaneurysms, dermatomyositis, and polymyositis should also be considered [10, 11, 19, 20].

Sarcomas and osteosarcomas may radiographically appear similar to calcific myonecrosis, but close examination shows different patterns of calcifications. Calcific myonecrosis usually presents radiographically with peripheral calcifications and central liquefactive necrosis, while sarcomas have calcifications distributed throughout the mass [11]. In addition, a CT scan often shows a cystic lobulated component and more compartmental involvement [20]., while MR imaging (MRI) will show the lesions to be heterogenous on T1- and T2-weighted images with the outer rim showing low-signal intensity due to the calcification and fibrosis [21]. Alternatively, myositis ossificans normally shows calcifications with the absence of an intralesional trabecular pattern and marrow signal on MRI, and it usually will not have progressive enlargement [19]. Polymyositis, pseudoaneurysms, dermatomyositis, and diabetic myonecrosis will usually present with a host of other symptoms which make the diagnosis more probable than calcific myonecrosis. When considering biopsy, the surgeon must be cautious and aware that both open and closed biopsies can lead to complications such as infection and poor wound healing [19]. However, biopsy should be used for histological diagnosis in the scenario where the mass appears to resemble a sarcoma both radiographically and clinically.

This case of calcific myonecrosis is unusual, because our patient presented without a known cause of trauma or compartment syndrome. The only significant history our patient had was body building in her forties and reported to have a significant "tearing" sensation in bilateral quads while squatting. This very well could have been the inciting event which led to the onset of gradual fibrosis, necrosis, and calcification in her bilateral anterior thigh compartments.

Due to the rarity of this condition, there is limited guidance on treatment options and outcomes. Various authors recommend surgical excision for symptomatic patients [2, 18]. Other physicians suggest treating symptomatic patients with aggressive debridement followed by flap coverage [9, 13, 19]. Early et al. reported satisfactory results in two patients who underwent complete resection of the calcified area and had the remaining empty space filled with tibial muscle [8]. Chun and Shim did a complete removal of the mass with wound closure and no filling of the dead space with soft tissues, and they had no complications postoperatively [3]. Renwick et al. reported a cystic degeneration in the superficial posterior compartment which was managed with complete excision and closed primarily over a suction drain with great outcomes [12]. Finally, Viau et al. suggested that needle decompression of these cystic masses might be beneficial [22].

Our treatment was unique in that the focus was on symptomatic relief of the patient's limited range of motion. Preoperatively, her bilateral knees could only be flexed to 30° with both hips extended, but our surgical procedures were able to restore this to 90° of flexion. This highlights the importance of tailoring the treatment to the specific complaints of the patients. Complete removal of the entire calcified rectus femoris muscle would have been associated with significantly increased morbidity and was not performed on either side for the patient in our report. The range of motion gains and symptomatic improvements were similar for both sides and partial excision of the involved area of calcific necrosis was performed one side, and no excision was performed on the other. This raises the possibility that complete excision of the area of calcific necrosis may not be necessary and should be considered on an individual case basis.

Historically, infection is a very common complication associated with surgical treatment of calcific myonecrosis [9, 14, 15, 17, 19, 21, 23]. Incomplete excision and packing of the wound with dressings has been associated with increased risk of infection and chronic drainage [14]. If infection occurs, patients often require multiple surgical procedures and extended antibiotic courses. The sequelae of infection can be so severe and difficult to treat that it has been reported to result in an amputation. Due to the high risk of complications with surgery, especially infection, asymptomatic patients should undergo conservative management with follow-up surveillance [7].

Conclusion

Treatment should be tailored to the nature and severity of the patient's symptoms. Symptomatic patients can be considered for excision or tenotomy based on the clinical scenario. Asymptomatic patients with characteristic imaging and stable lesions should be treated conservatively and with observation due to the high risk of infection and wound healing problems associated with surgery.

Clinical Message

Orthopedic practitioners should tailor the treatment of patients with calcific myonecrosis based on the nature and severity of their symptoms. Asymptomatic or minimally symptomatic patients should be treated conservatively due to the high rate of complications associated with surgery. Complete excision, partial excision, or simple tenotomy can be successful treatment options for patients with calcific myonecrosis.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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