



Calcific myonecrosis

Kalsifik miyonekroz: Olgu sunumu

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Kalsifik miyonekroz, ekstremitte travmasının yıllar sonra görülebilen nadir bir komplikasyonudur. Genellikle alt ekstremitede kompartman sendromu ardından geç dönemde gelişen distrofik kalsifikasyon ile kendini gösteren bir patolojidir. Bu yazıda, ateşli silah yaralanmasını takiben sol uylukta kompartman sendromuna yönelik cerrahi girişimlerden 35 yıl sonra kalsifik miyonekroz gelişen 66 yaşında bir erkek hasta sunuldu. Hasta sol bacağına ağrı ve şişlik yakınmasıyla başvurdu. Muayenede, sol kruris orta hat anterolateralinde, eklemle ilişkisi olmayan, 20 x 8 x 6 cm boyutlarında, sınırları belirgin, hareketsiz yumuşak kıvamlı bir kitle görüldü. Radyografik incelemede sol kruriste çok sayıda lineer kalsifikasyon odağı saptandı; kemik patolojisi yoktu. Ameliyat sırasında tibia anterior kas kompartmanındaki tüm liflerin kalsifiye olduğu gözlemlendi. Kemik dokuya yapışıklık göstermeyen kitleye eksizyonel biyopsi uygulandı. Histopatolojik değerlendirme sonucu kalsifik miyonekroz ile uyumlu bulundu. Birinci yıl sonunda yapılan kontrolde semptomsuz olan hastada kitleye ait herhangi bir nüks görülmedi.

Anahtar sözcükler: Kalsinoz/etyoloji; kompartman sendromu/komplikasyon; bacak yaralanması; kas hastalığı; nekroz.

Calcific myonecrosis is a rare complication of limb trauma, that may occur after many years. It is characterized by dystrophic calcification that develop in the late period following compartment syndrome usually in the lower limb. We present a 66-year-old man who developed calcific myonecrosis 35 years after surgical intervention for compartment syndrome secondary to a gunshot injury to the left thigh. He presented with pain and swelling in the left leg. On physical examination, there was a well-defined and immobile mass lesion in the anterolateral part of the left crus, soft in consistency and 20x8x6 cm in size, showing no relation with the joint. Radiographic evaluation showed linear calcifications in the left crus without osseous pathology. At surgery, all the fibers in the anterior tibial compartment were calcified and there was no attachment to the bone. The patient underwent an excisional biopsy. Histopathological evaluation of the specimens revealed calcific myonecrosis. At the end of one-year follow-up, the patient was symptomless, without any recurrence.

Key words: Calcinosi/etiology; compartment syndromes/complications; leg injuries; muscular diseases; necrosis.

Calcific myonecrosis is a painful pathology presenting as a soft tissue mass with slow growth rate which usually occurs after a trauma in lower extremities, and classified within the dystrophic calcification group. Initially, it has been described by Gallie and Thompson^[1] in 1960 and to date, more than 40 cases have been reported in foreign literature.^[2-15] Most commonly af-

ected site is known to be anterior compartment of leg. However, cases showing foot^[11] and upper extremity^[14] involvement have been reported, as well. Compartment syndrome occurs as a result of various traumas such as fracture, crush, blunt trauma, firearms injury, knee ligament injuries; and establishes the appropriate ground for myonecrosis development. While the exact reason

has not been described yet, post-traumatic muscular ischemia and cystic degeneration are believed to cause the underlying physiopathology.^[16] In the present report, a case with calcific myonecrosis localized in the cruris which occurred 35 years after a compartment syndrome developed due to firearms injury in thigh, is presented.

Case report

A 66-year-old male patient presented to our polyclinic due to swelling and pain in the left leg. The complaints had started 7-year previously, swelling had become larger and the pain had reached higher levels during the last year. The patient who had an entry hole in his abdomen and lateral portion of his left hip and who also had both entry and exit holes in his left inguinal region due to firearms injury, had been subjected to vascular repair for circulatory problems arising as a result of femoral artery injury and left thigh fasciotomy due to compartment syndrome. Physical examination revealed a palpable, immobile, remarkable mass of soft consistency with 20x8x6cm size localized anterolaterally to median line of left cruris (Figure 1a). Plain radiography showed no osseous pathology; multiple linear calcifications surrounding tibia were determined on the area consistent with tibialis anterior location (Figure 1b). Magnetic resonance imaging (MRI) revealed a fluid deposit which was demarcated from the soft tissue by a clear capsula and including calcified components inside (Figure 1c). Peripheral arteriography and MR angiographic evaluation showed no

vascular pathology (Figure 1d). Laboratory workout exhibited normal erythrocyte sedimentation rate, serum calcium, phosphorus, alkaline phosphatase, and parathyroid hormone levels.

Under spinal anesthesia, longitudinal incision was applied through the mass localized anterolateral to the median line of left cruris. Following the accession to the cutaneous and subcutaneous tissues, the mass which has been demarcated from the adjacent osseous and soft tissues by a remarkable capsula, was reached (Figure 2a). Capsula was opened and all the fibers in tibialis anterior muscle compartment were observed to be calcified (Figure 2b). Excisional biopsy was carried out on the mass except the portion which was exhibiting adhesion to the vessel-nerve package.

Postoperative follow-ups did not show skin necrosis or infection. No growth was observed in the culture of the tissue obtained during the operation. The histopathological results of the tissue samples found to be consistent with calcific myonecrosis (Figure 3). At the end of the first postoperative year the patient had no swelling or pain complaint; obtained radiograph showed no new focus other than the region adjacent to the vessel-nerve package (Figure 1e-g).

Discussion

Calcific myonecrosis is a rare event seen following a trauma and most commonly occurs as a dystrophic calcified soft tissue mass. An extremity trauma suffe-



Figure 1. (a) Clinic and (b) radiographic view of soft tissue mass localized in the left leg of the patient. (c) a well-demarcated heterogeneous intensity rise including calcified foci in the magnetic resonance section (d) No vascular relation of the mass is seen in the peripheral angiographic examination. In 12th month of the postoperative period (e) clinical appearance and (f) antero-posterior and (g) lateral radiographs.



Figure 2. During the operation (a) a well-demarcated mass; (b) calcified appearance of tibialis anterior muscle fibers after the opening of the capsula.

red long before (10-64 years) the presentation plays a role in the etiology and all the cases have a compartment syndrome or neurovascular injury (particularly peroneal nerve damage) history suffered subsequent to a trauma.^[16] Ischemic contractures and nerve damage

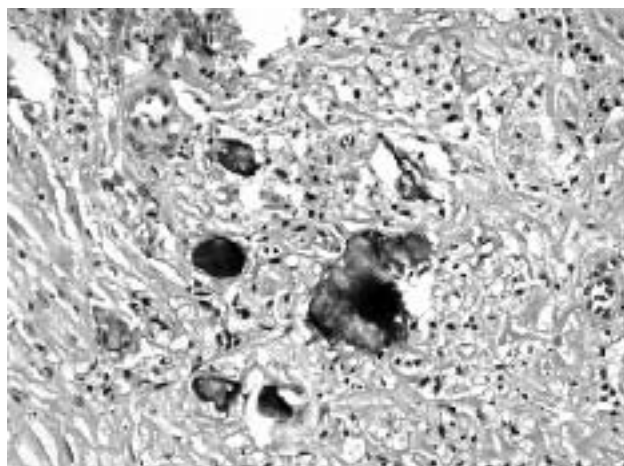


Figure 3. Calcification foci surrounded by fibrous tissue, macrophages, and chronic inflammatory cells in the histopathological section (H-E x 200)

are well known complications after compartment syndrome. However calcific myonecrosis is an uncommon sequela of compartment syndrome. Current conjecture is that compartment syndrome leads to necrosis and fibrosis by diminishing the blood circulation of a certain area, and recurrent hemorrhages cause growth of the mass along with calcification, in time.^[4]

Physical examination revealed a painless or painful growth of a mass.^[5,12,16] There are also reports in the literature which indicate no finding of a mass arising from the replacement of the necrotic muscle by the pathology.^[17] In the present case, pain complaint had started seven years before presentation and a growth of mass had taken place in the cruris anterior during the course of the time.

The characteristic features of calcific myonecrosis are known to be well-defined, large fusiform soft tissue appearance including peripheric plaque-like or thin linear calcification foci localized generally in the cruris anterior compartment.^[4,5,12,16] Along with mild periosteal reaction, destruction may be observed in the adjacent bone.^[3,5,7,9] In magnetic resonance imaging, the replacement of the normal muscle tissue is observed as well-defined heterogeneous signal intensity in T₂-weighted sequences and more homogeneous signal intensity in T1-weighted sequences.^[16] Three phase bone scintigraphy shows increased uptake in soft tissue and bone phases.^[13] Laboratory workout often reveals normal levels of serum calcium, phosphate, alkaline phosphatase, and parathyroid hormone.^[16] Histopathological analyse shows mixed blood and fibrine, necrotic soft tissue, and bone calcification material.^[5,8,12]

Differential diagnosis of calcific myonecrosis should include all the neoplastic and inflammatory diseases that can cause soft tissue calcification. Moreover, along with malignities such as sinovial sarcoma, epithelioid sarcoma, soft tissue osteosarcoma, and parosteal osteosarcoma; pathologies like myositis ossificans, trauma-related pseudoaneurysm, dermatomyositis-polymyositis, tumoral calcinosis, and diabetic myonecrosis should be considered, as well.^[8,9,11,12] The fact that these malignities are seen in younger ages and follow a more aggressive and destructive course, is important for differential diagnosis of soft tissue sarcomas.^[4] Furthermore, while these tumors reveal a gadolinium enhancement in MRI, calcific myonecrosis does not show such an enhancement.^[5] Dermatomyositis and polymyositis present with large calcification foci and oftenly syste-

mic findings accompany those diseases.^[16] Myositis ossificans is a pathology that can develop after a trauma, as well. However, progressive growth of the mass and destruction of the adjacent bone are not observed.^[5] Calcification do not incline to include the whole of the muscle in pathologies such as dystrophic calcification associated with infection, calcification associated with connective tissue diseases, diabetic necrosis and trauma-related pseudoaneurysm.^[16]

Majority of the cases associated with calcific necrosis have been reported to be localized in cruris. Several cases including development of calcific myonecrosis in foot^[11] and forearm^[14], have also been reported in the literature. All the cases have an history of a trauma related to the localization of the pathology. Ryu et al.^[7], reported a case with calcific tenosynovitis in extensor hallucis longus muscle accompanied by a calcific myonecrosis in the tibialis anterior muscle developed after a fibula or tibia condyle fracture. Holobinko et al.^[13], reported a case with calcific myonecrosis spread to the muscles of the foot compartment following a tibial fracture. In the present case, fasciotomy had been applied against the compartment syndrome developed after a firearms injury which caused development of calcific myonecrosis after 35 years in cruris distal to the trauma area.

Cases in which infection has developed after the application of biopsy for histopathological diagnosis, have also been reported.^[3,4,9,11,12] Due to the risk of causing infection in the sterile necrotic tissue by biopsy, biopsy has been recommended only if surgical excision is to be applied subsequently.^[16] While in asymptomatic cases, conservative therapy and monitorization should be carried out, in symptomatic cases, complete excision of the mass along with reconstructive flap repair for closing the open space should be planned. In the present case, after reaching the prediagnosis of calcific myonecrosis following history, clinical examinations and radiographic evaluations; complete excision has been applied except the portion adjacent to the vessel-nerve package and no recurrence has been seen during the 12-months follow-up of the patient.

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