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Case Report

Myositis ossificans circumscripta of the thigh: A pediatric case report ☆,☆☆

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ABSTRACT

Circumscribed myositis ossificans is a benign process of focal heterotopic ossification of the soft tissues, occurring in young subjects, usually following trauma. We report a case of a 15-year-old patient who suffered a direct trauma to the thigh during a soccer match, and developed a hard mass in the anterior face of the thigh. The patient was diagnosed with myositis ossificans secondary to trauma. In this case report, we want to illustrate the different imaging aspects of this benign condition, both in conventional radiology, computed tomography and magnetic resonance imaging, as its clinical and radiological appearance can be misleading, suggesting a sarcomatous neoplastic process.

Introduction

Circumscribed myositis ossificans (MOC) is known as a nontumorous heterotopic proliferation of bone and cartilage tissue from the interstitial connective tissue within the soft tissues of the skeleton. It presents 0.7% of soft tissue pseudotumors. It affects the young subject, and sits mainly in the pelvic belt, scapula, thigh, and arm, frequently following a trauma. Because clinicobiological data are nonspecific, medical imaging is of interest as it is necessary for a positive diagnosis and subsequent care [1].

Case report

A 15-year-old patient, the second of a sibling of 3, right-handed, with no particular pathological history, presented to consultation for swelling of the anterior face of the right thigh,

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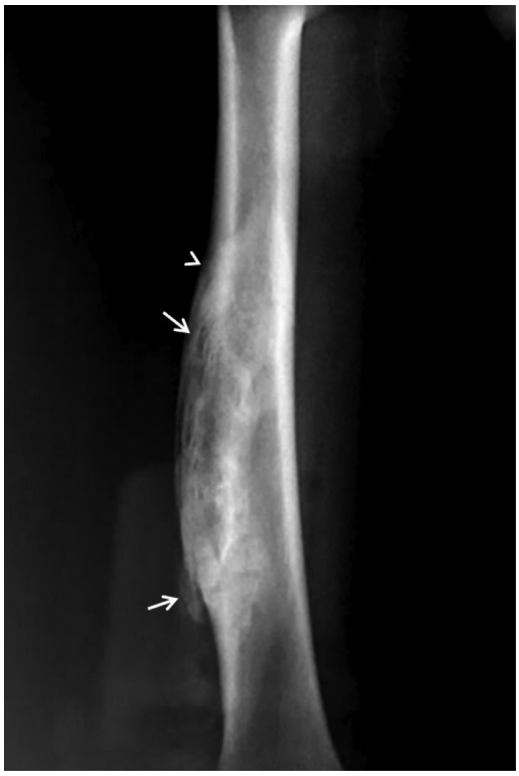

Fig. 1 – Femur X-rays showing a well-circumscribed calcified lesion (arrows), non separated from the underlying femur with multi-lamellar periostal reaction (head arrow).

not accompanied by functional impotence. The history of the disease dates back 3 months when the child suffered a direct trauma to the thigh during a soccer match.

The clinical examination finds a child of 55 kg, 160 cm. Examination of the musculoskeletal system found a painful and hard mass on the anterior surface of the right thigh, with no skin changes. The arc of motion of the hip and knee was complete. Vascular and nervous examination of the right lower limb, as well as the examination of the rest of the musculoskeletal system, was without abnormalities.

The X-rays showed a well-circumscribed peripherally calcified lesion, non separated from the underlying femur with multi-lamellar periostal reaction (Fig. 1). The surgical team then realized a CT scan which showed a peripheral ossification with a focal immature nonmineralized central area, with a zonal organization (Fig. 2).

The diagnosis of heterotopic ossification was evoked. Thus, an MRI was performed to analyze the extent of the mass within the muscle and soft tissues. It showed a well-defined oblong mass of the anterolateral side of the right thigh developing within the vastus intermedius muscle. It has irregular contours, a heterogeneous T2 and T1 signal surrounded by an asignal T1 and T2 border, without enhancement after Gadolinium. It is associated with a range of bone edema opposite with irregularity and thinning of the cortical bone and multi-lamellar periosteal reaction. Topographically, it invades the vastus intermedius muscle and pushes back the muscular fascia (Figs. 3 and 4).

A biopsy was performed by direct approach under general anesthesia by an orthopedic surgeon-oncologist. The delay between the first consultation and the biopsy was 2 weeks.

The anatomopathological study of the surgical specimen showed striated muscle tissue, with calcifications, mature regular bony lamellae, and locally hyalinized fibrovascular tissue returning in favor of a circumscribed myositis ossificans.

On the basis of CT, MRI and pathological findings, the diagnosis of circumscribed myositis ossificans was retained. The patient was put on indomethacin in 15-day courses spaced 1 month apart (3 courses) with monitoring of tc-99m scintigraphic fixation. Surgery was proposed after no scintigraphic fixation. The patient was followed for 15 months with good clinical and biological evolution. Radiological and scintigraphic follow-up shows no structural changes.

Discussion

Myositis Ossificans (MO) is a benign condition characterized by the abnormal growth of bone and cartilage tissue in soft tissues of the skeleton, occurring in abnormal locations. This rare lesion accounts for 0.7% of soft tissue pseudotumors [2]. MO may occur at any age, but it is diagnosed most frequently in individuals in their second and third decades of life [3]. The condition predominantly affects males [4].

The most frequent sites for myositis ossificans are the major muscle groups of the thigh and upper arm and it typically manifests more frequently in the anterior compartments than in the posterior ones. The uncommon locations where MO can occur include the neck, scapula, axillary region, hand, foot, hip, chest, and abdominal wall [5,6]. The quadriceps femoris and gluteus muscles in the lower extremity are the most frequently affected sites, while in the upper extremity, it is the brachialis muscle that is commonly impacted. Myositis Ossificans typically occurs following trauma in 60% to 75% of cases and is occasionally referred to as Myositis Ossificans Traumatica (MOT) [7].

Histological findings vary according to the age of the lesion:

- **In the early stage:** there is an active mesenchymal proliferation of fibroblasts with significant mitotic activity, along with hemorrhaging and necrosis [8].
- **In the intermediate stage:** heterotopic osteoblasts appear, producing an osteoid matrix and forming a fibrous capsule known as the "zone phenomenon". This stage consists of 3 distinct zones: the *central zone* (mesenchymal tissue, fibroblasts, mitoses, hemorrhages, necrosis), the *intermediate zone* (osteoblasts, immature bone islands), and the *outer zone* (traces of mature bone). Ossification occurs centripetally [9].
- **In the late stage**: peripheral bone formation typically begins around 6 to 8 weeks but can sometimes happen earlier. By the late phase, around 5 to 6 months, the lesion may completely ossify, developing a cortex and marrow spaces. As the lesion matures, it commonly shrinks in size and in approximately 30% of cases, may eventually resolve spontaneously [9].

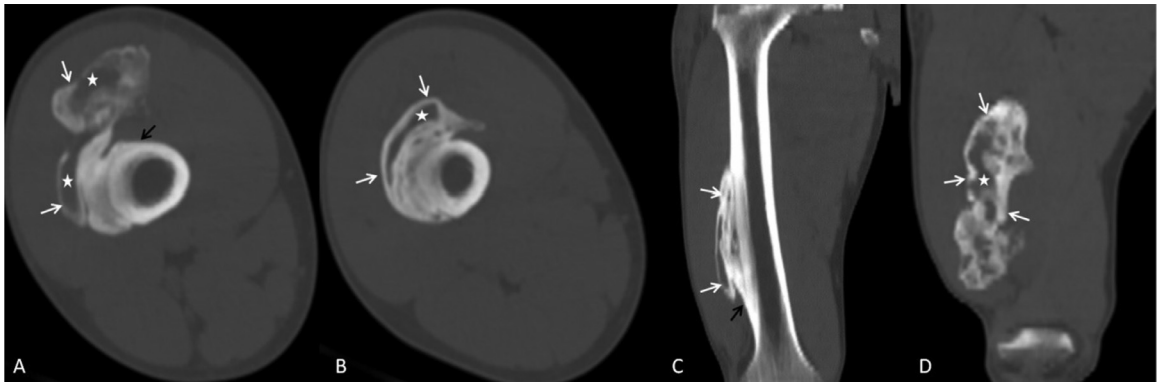

Fig. 2 – Axial (A, B) and coronal (C,D) CT scan of the thigh showing a well-defined mass with peripheral ossification (white arrows) with a focal immature nonmineralized central area (), associated with periostal reaction (black arrows).

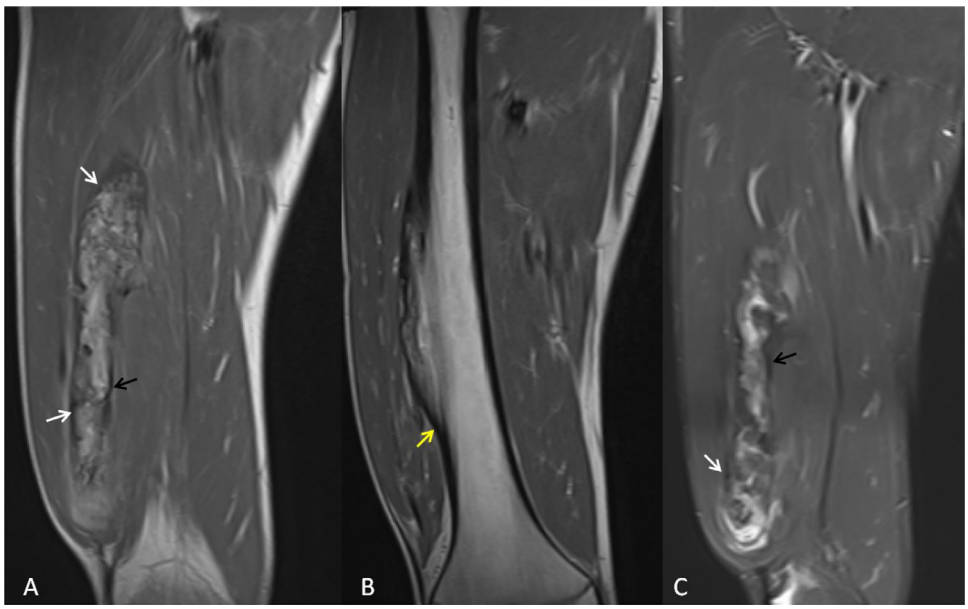

Fig. 3 – Coronal T1W (A, B) and STIR (C) MRI shows an oblong mass in the vastus intermedius muscle (white arrows), heterogenously hyperintense in the center on T1-WI with a halo in hypo signal T1 and STIR (black arrows) . Note the periosteal reaction (yellow arrow).

Upon suspicion of heterotopic ossification on imaging, it has been suggested to perform a biopsy to confirm the diagnosis. However, current recommendations advocate for imaging follow up after 4 weeks, which together with the history of trauma can confirm the diagnosis [10]. When the characteristic calcification pattern or clinical findings are not typical and the biopsy is done because the differential diagnosis is uncertain, it is crucial to be mindful of the potentially misleading histological appearance that can resemble a malignant lesion in early stages of MO. Additionally, if the biopsy sample is solely taken from the center of the lesion and does not display the characteristic zonal pattern, caution should be exercised [9]. Biopsy of the central and intermediate layers of the MO lesion can mistakenly suggest sarcoma, with potentially disastrous consequences [11]. Hence, the biopsy should be performed by using ultrasonography and should include the full length of the target lesion (complete sampling of the lesion) [12].

At an early stage, the lesion is soft and painful, progressing to a firm and typically painful mass within a few weeks within the impacted muscles. This mass undergoes maturation over a period of 6 to 12 months, eventually calcifying and becoming asymptomatic [7]. Depending on its location and size, the lesion may lead to restricted movement capabilities.

The serum alkaline phosphatase (SAP) level starts off as normal but rises after 3 weeks, correlating with bone formation, particularly in patients with clinically significant Myositis Ossificans (MO). It peaks around 10 weeks before returning to normal levels by 18 weeks. During the acute phase, levels of C-reactive protein, erythrocyte sedimentation rate, and serum prostaglandin-E2 are elevated in the early stages of MO. Initially, the serum calcium level typically drops briefly, return-

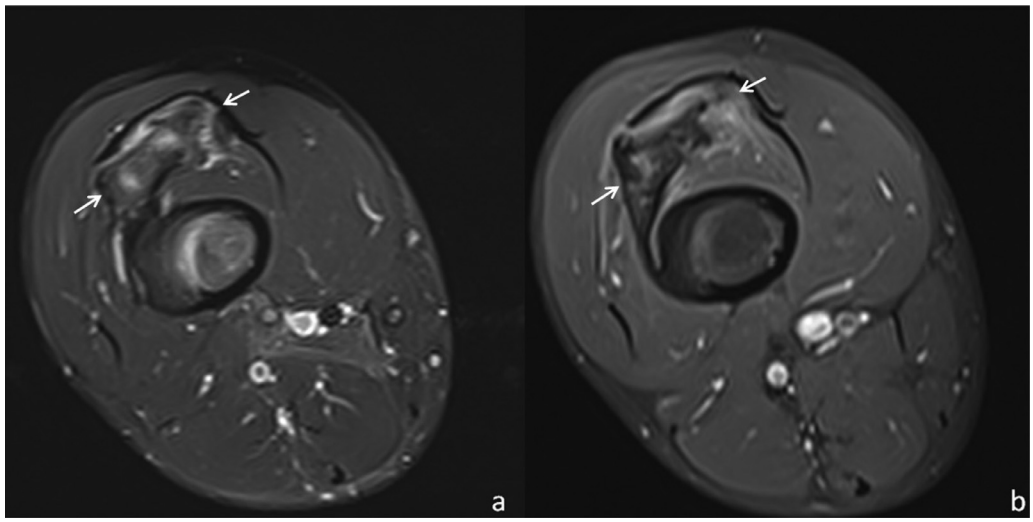

Fig. 4 – Axial STIR (a) and T1 FAT SAT WI after Gadolinium (b) which shows the absence of enhancement within the mass (arrows).

ing to normal before the increase in SAP levels. Elevated creatine phosphokinase levels usually indicate muscle damage and may serve as a predictive factor for the development and severity of the damage.

The serum alkaline phosphatase (SAP) level starts off as normal but increases after 3 weeks along with bone formation, particularly in patients with clinically significant (MO) [9]. Peaking at around 10 weeks and returning to normal at 18 weeks [13]. In the acute phase, the C-reactive protein level, the erythrocyte sedimentation rate, and the serum prostaglandin-E2 level are elevated during the initial stages of MO. The serum calcium level usually decreases for a short time and then returns to normal before SAP increase [13]. The level of creatine phosphokinase is usually elevated if there is muscle damage and may be predictive of the development and severity of the damage [14].

Radiology plays a pivotal role in the diagnosis of MO, with imaging findings evolving in conjunction with the 3 stages of the condition.

In the acute phase: X-rays may appear normal or show soft tissue swelling along with periosteal reaction near the bone, but without any calcification. Ultrasound may reveal muscle echogenicity abnormalities and altered vascularization without specific features [15]. CT scans typically display soft tissue swelling without calcification, with enhanced images after contrast product injection concerning the adjacent bone. On MRI, the mass usually appears isointense in T1-weighted images, heterogeneously hyperintense in T2-weighted images, and shows slight enhancement after gadolinium injection, often with muscular edema extending beyond the swelling limits [16].

The primary differential diagnosis to consider is *soft tissue sarcoma*, although the marked muscle edema seen in the early stages of post-traumatic myositis ossificans is not typically present in soft tissue sarcoma. It also has a more aggressive appearance on MRI (irregular margins, rapid growth) and less likely to have the zonal pattern seen in MO [17]. The edematous muscle aspect can also indicate conditions such as *abscess, hematoma, focal myositis, or rhabdomyolysis*.

In the case of an *intramuscular abscess*, ultrasonography reveals an irregular hypoechoic or anechoic lesion. CT scans show a heterogeneous hypodense lesion before liquefaction and a hypodense lesion postliquefaction. MRI demonstrates heterogeneously hyperintense signals in T2-weighted images for abscesses and homogenously hyperintense signals in T2-weighted images for liquefied abscesses, with wall enhancement post gadolinium injection. *Intramuscular hematoma* will appear hyperechoic in ultrasonography and hyperdense in CT scans. In MRI, they are hyperintense on T1-weighted images, show varying signals on T2- weighted images, and lack post-contrast enhancement [18].

Focal myositis presents as a hypoechoic mass with thickened muscle fibers on ultrasonography and may also exhibit hyperechoic characteristics [19,20]. MRI typically shows a defined mass within a single muscle or an enlarged appearance of the affected muscle without calcifications. T1-weighted images usually reveal an isointense or slightly hypointense mass, while T2-weighted images show iso- or hyperintense signals with homogeneous or heterogeneous patterns [18]. *Focal myositis* often mimics the early imaging features of myositis ossificans, with the key distinction being its progression towards healing without calcification.

In the intermediate phase: X-rays and CT scans reveal floccular calcifications at the periphery of the lesion (crown). The ossification is demarcated from the bone by a radiolucent band (safety edging). Ultrasound displays the 3-zone phenomenon: the *outer zone* appears hypoechoic, indicating hyperemia and perilesional edema. The *second zone* is hyperechoic due to calcification, while the *innermost zone* is hypoechoic, representing the mass content. MRI imaging in this phase depicts the mass as T1 hypointense and T1 and T2 hyperintense at the center, with a halo showing hypo signal in both T1 and T2 sequences (indicating calcifications) [18]. Oc-

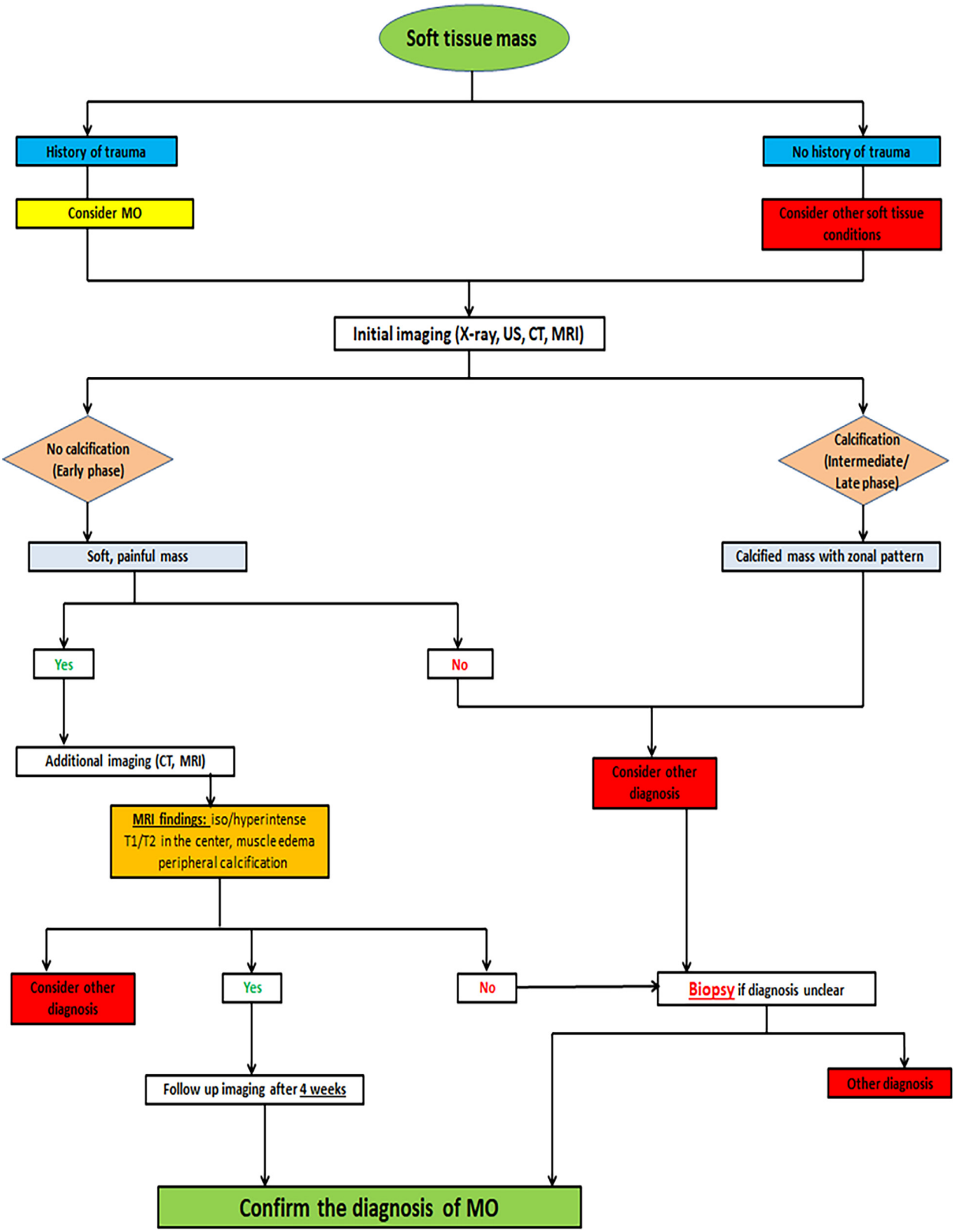

Fig. 5 – Diagnostic flowchart for myositis ossificans.

Table 1 – Differential diagnoses for Myositis Ossificans, with key features and imaging characteristics and how each condition can be differentiated from MO.

Differential diagnosis	Key features	Imaging characteristics	Differentiation from MO
Soft Tissue Sarcoma	Aggressive, rapid growth; irregular margins	MRI: Irregular, heterogeneous signal; may show necrosis/hemorrhage	Lacks zonal pattern; Biopsy needed, avoiding central-only sampling
Abscess	History of infection, fever	Ultrasound: Hypoechoic/anechoic; CT: Hypodense lesion postliquefaction	Central liquefaction with peripheral enhancement; fluid-filled cavity
Hematoma	Recent trauma; bleeding disorders	MRI: Hyperintense on T1, variable on T2, no enhancement	No calcifications; hyperdense on CT; hyperechoic on ultrasound
Focal Myositis	Localized muscle pain; no calcifications	MRI: Iso/hyperintense mass on T2; homogeneous signal	No calcifications; thickened muscle fibers on ultrasound
Rhabdomyolysis	Muscle pain, weakness, dark urine	MRI: Diffuse muscle edema	No focal mass; elevated creatine kinase (CK) levels; Central calcifications
Synovialosarcoma	Near joints; potential calcifications	MRI: Heterogeneous mass; possible fluid-fluid levels	Irregular and central calcifications; joint involvement
Calcific Tendinitis	Localized pain near tendons	X-ray/CT: Discrete calcifications near tendons	Calcifications without a well-defined mass; near tendon insertions
Ewing's Sarcoma	Diaphyseal bone involvement; soft tissue mass	MRI/CT: Central mass with extensive bone destruction	Permeative pattern with lamellated periosteal reaction Central calcification
Osteosarcoma (Extraskeletal/Paraosteal)	Dense, central calcification; aggressive bone involvement	X-ray/CT: Central calcification; MRI: heterogeneous signal	Central calcification versus peripheral in MO; bone involvement

casionally, fluid/fluid levels may be present, indicating hemorrhage within the immature central portion of the lesion [16].

During this period, if the lesions are located around the joint, the primary differential diagnoses include *rhabdomyosarcomas* and *synovialosarcomas* where the calcifications are typically central and scattered, often involving the adjacent bone In contrast, circumscribed myositis ossificans exhibits peripheral and continuous calcifications in relation to the bone. *Calcific tendonitis* may also be considered, but it is distinguished by calcifications without a distinct mass [16]. Extensive muscle edema and continuous periosteal reaction in the femur may indicate malignancy like *Ewing's sarcoma*, which calcify outward and invade surrounding tissues unlike MO, showing bone marrow abnormalities and cortical bone destruction [21].

In the late stage, X-rays and CT scans reveal a completely calcified lesion with trabecular bone formation, which can occasionally blend with the cortex of the neighboring bone. On MRI, the lesion appears largely hypointense across all sequences due to calcification. The center of the lesion may show areas with intensity similar to that of normal bone marrow [22]. Perilesional oedema is absent.

In this stage, the main differential diagnoses include *extraskeletal and paraosteal osteosarcomas*. In MO, calcification begins at the periphery and advances towards the center, a centripetal pattern, whereas in osteosarcomas, calcification tends to be dense, initially central, and progresses outward centrifugally [23].

The scintigraphy shows increased uptake in soft tissues during the acute and subacute phases which diminishes as the condition enters the chronic phase [11].

It is noted that our patient was in the intermediate phase of progression.

To assist clinicians in the diagnostic process, we have developed a comprehensive flowchart that outlines a step-by-step approach for evaluating patients with soft tissue masses. This flowchart integrates key clinical and imaging findings (Fig. 5) and considers important differential diagnoses, including soft tissue sarcomas, intramuscular abscesses, hematomas, and other conditions that may mimic myositis ossificans (MO), as summarized in Table 1.

Myositis ossificans typically follows a course of stabilization, regression, or complete disappearance of the lesion [24]. Sarcomatous transformation is rare once an initial diagnosis of myositis ossificans is confirmed [2].

Although MO treatment is usually conservative, its incidence, location of the bone formed, and resulting complications can show marked variations. Reported complications include functional impact, pressure effect on nerves or blood vessels, peripheral nerve entrapment, pressure ulcers, and functional impairment due to ankyloses. Cases of vessel occlusion have also been reported [25]. Nonselective nonsteroidal anti-inflammatory drugs such as indomethacin can halt the progression of MO [23]. Surgical intervention is typically reserved for cases where the lesion has matured to reduce the risk of recurrence [26].

Previous therapeutic approaches for post-traumatic myositis ossificans have typically centered around conservative methods like rest, ice, compression, and elevation (RICE), along with passive stretching and strengthening routines. However, these traditional approaches often require a longer period of treatment and may not be as effective for cases

of myositis ossificans that are unresponsive to conservative care. In contrast, recently, extracorporeal shockwave therapy (ESWT) has shown effectiveness in managing MO [26].

ESWT has been utilized effectively for conditions like calcification of tendons and pseudo arthroses, showcasing biological actions through microdisruption of avascular tissues. ESWT can suppress local nociceptors, stimulate tissue repair, and trigger neovascular angiogenesis. The treatment is believed to provide analgesia through hyperstimulation effects, leading to lasting pain relief [27].

In a case-series by Buselli et al., ESWT demonstrated successful treatment for myositis ossificans (MO) athletes, showing improvement in symptoms and function. ESWT was applied every other week for a total of 3 treatments, leading to significant pain reduction and improved range of motion (ROM). The treatment was well-tolerated, with patients experiencing notable analgesic effects post-treatment and enhanced ROM [26].

While ESWT was effective in reducing pain and swelling in MO patients, especially in athletes, some side effects like pain, skin bruising, and swelling were reported. No serious adverse effects were noted, with most discomfort subsiding within hours to days post-treatment [26]. However, the efficacy of ESWT on MO is supported mainly by descriptive case reports, lacking validation in controlled trials. Further research is needed to establish treatment protocols, safety profiles, and effectiveness across different patient populations.

Myositis ossificans poses significant long-term challenges, impacting both functional abilities and quality of life. Functional limitations such as restricted range of motion, muscle weakness, and chronic pain can hinder daily activities, work performance, and social engagement. Psychological effects like anxiety and depression, alongside social and occupational challenges, further compound the condition's impact. To address these complexities, a holistic approach involving multidisciplinary care, tailored rehabilitation programs, and regular follow-ups is crucial for managing the condition effectively, improving outcomes, and enhancing the overall well-being of individuals affected by MO [28].

Conclusion

MOC is a benign affection of the soft tissues, Early in its development, the clinical and paraclinical appearance may mimic a malignant process. The lack of typical imaging features can lead sometimes to a surgical biopsy for diagnosis. The treatment remains essentially medical, it is rarely surgical.

Patient consent

Written informed consent for the publication of this case report was obtained from the parents of the patient.

Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

Guarantor of submission

The corresponding author is the guarantor of submission.

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