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Non-traumatic myositis ossificans of the thigh. A case report

INTRODUCTION: *Myositis ossificans (MO) is an ectopic ossification characterized by an appearance of bone formation predominantly in muscle tissue. Trauma is the most common etiological factor, observed in almost 60-75% of cases, whereas a non-traumatic MO is rarely described in the literature. A diagnosis is based on clinical and radiological findings.*
PRESENTATION OF CASE: *A 75-year old female patient has been admitted to our clinic with a localized swelling of the posterior femoral compartment, presented on magnetic resonance as a calcification in the biceps femoris muscle. Laboratory test results were within the normal range. Surgical procedure consisted of excision of the tumor mass with primary wound reconstruction and drainage. The post-operative period was uneventful, and the patient was discharged from hospital on the seventh postoperative day. The pathohistology findings have shown the MO.*
DISCUSSION: *A non-traumatic MO is scarcely described in the literature. A chronic microtrauma, tissue ischaemia and inflammation are listed as causal mechanisms of a non-traumatic MO. MO non-traumatica occurs more often in patients with a parallel, subdural or epidural haemorrhage and a hip surgery. Our case did not present any family history, trauma or associated anomalies of hands or fingers.*
CONCLUSION: *Myositis ossificans should be considered as the differential diagnosis of all soft tissue tumor masses, even if known risk factors are not present in the anamnesis. Surgery is a reasonable therapeutic strategy in the presence of a tumor mass in soft tissues, and definite diagnosis can be set only based on pathohistological findings.*

KEY WORDS: Ectopic ossification, Non traumatic myositis, Surgery.

Introduction

Myositis ossificans (MO) is a benign condition, and a distinct formation of bones. In other words, it is a heterotopic ossification of the muscle with an increased tissue activity that leads to intramuscular bone formation [1].Myositis ossificans progressiva (MOP) is a dominant autosomal disorder, with the incidence of less than 1 in 10,000,000 people [2]. In most cases, trauma is the prevailing aetiology, and a non-traumatic MO is a very rare condition that is usually diagnosed in adults of 20-40 years of age, but is seldom observed in children and the elderly [3]. MO most frequently appears in a region with a high risk of injuries, such as the flexor muscle of the upper arm, the quadriceps femoris muscle and the abductor muscle of the thigh [4]. The clinical presentation includes the painful mass of soft tissue, usually without inflammatory symptoms. The diagnosis of this condition may be difficult and it requires radiologic and/or histologic results [5].

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Case Report

This paper presents a 75-year old female patient, sent to our clinic by her family doctor, in pain and with a swelling in the posterior compartment of the thigh, which she has been suffering from for several months. Three years prior to that, the patient had noticed a progressive increase in volume of the posterior compartment of the thigh, along with gradual sensorimotor difficulties. In the beginning, the swelling was insidious and associated with intermittent pain, but it gradually progressed with the pain which extended as far as the fourth and fifth fingertips. There had been neither a background of a blunt injury of the right thigh, nor had there been any bone fractures or family connexion.

The clinical examination revealed a large oval swelling measuring 25 cm x 15 cm, which was observed in the posterior and lateral compartment of the right thigh (Fig. 1).

The swelling had a hard consistency and was fixed to the skin, but minimally sensitive to the palpation. The examination of lower extremities showed a complete spectrum of movements without pain. The neurological findings were regular, with the motion range of the right knee showing a full extension to 130° flexion.

The laboratory blood test results, including alkaline phosphatase, serum calcium and phosphate levels, erythrocyte sedimentation rate (ESR) and C-reactive protein, were within the normal lab values. The magnetic resonance (MR) imaging revealed a significant increase in muscle volume of the right thigh with high intensity of signals related to the area of calcification, as well as a well-localised calcified mass which is clearly separated from the cortex of the adjacent thigh bone (Figs. 2 A,B).

A requirement for an operative treatment was established on the basis of the anamnesis, physical examination and MR findings. The patient was operated on under spinal anaesthesia. Intraoperatively, a hard tumorous formation was detected in the fatty tissue and between muscle fibers (Fig. 3).

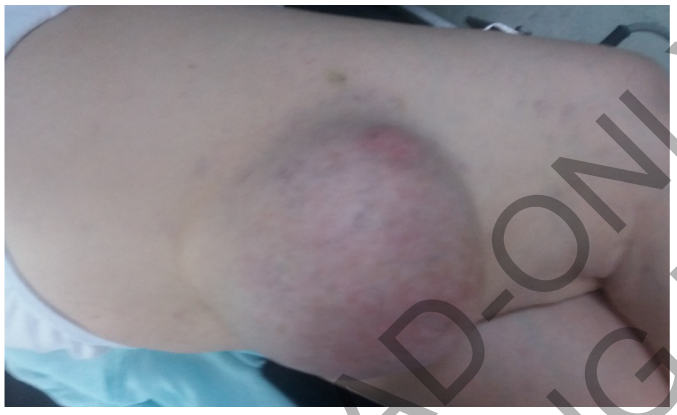
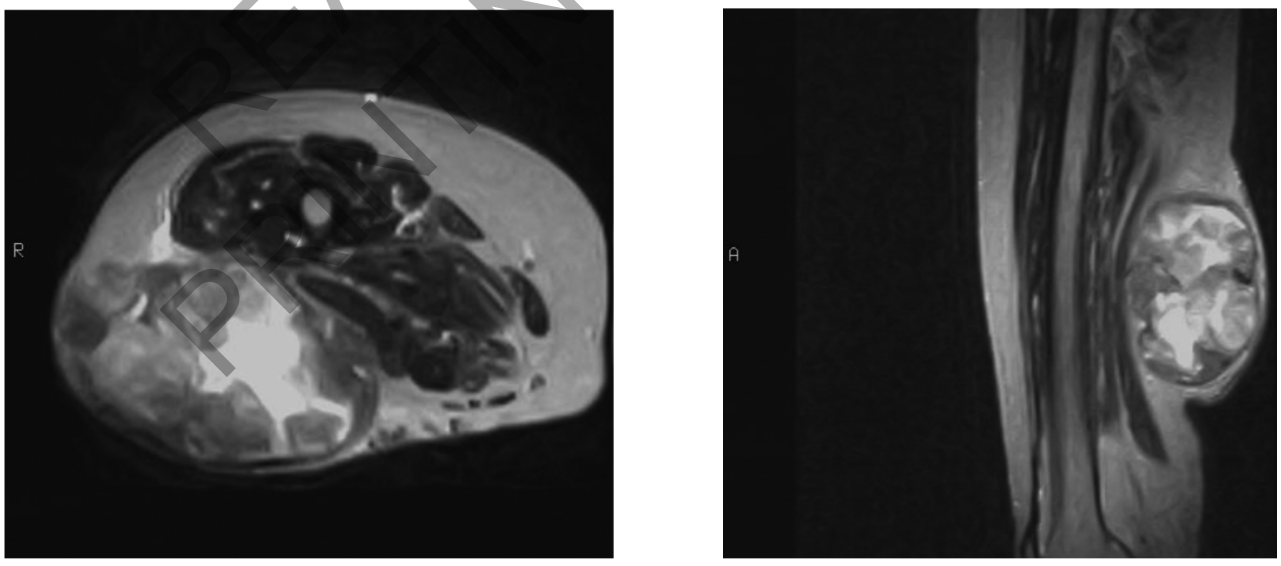


Fig. 1: Clinical aspect of the right thigh; tumorous formation measuring 25 x 25 cm in the posterior compartment of the thigh.



A B

Fig. 2: MR scan showing the volume of myositis ossificans which measures about 20 × 10 × 10 cm in the right thigh and lateral head of the biceps femoris muscle (A). An obvious zone (B) is present between the femur and the mass.

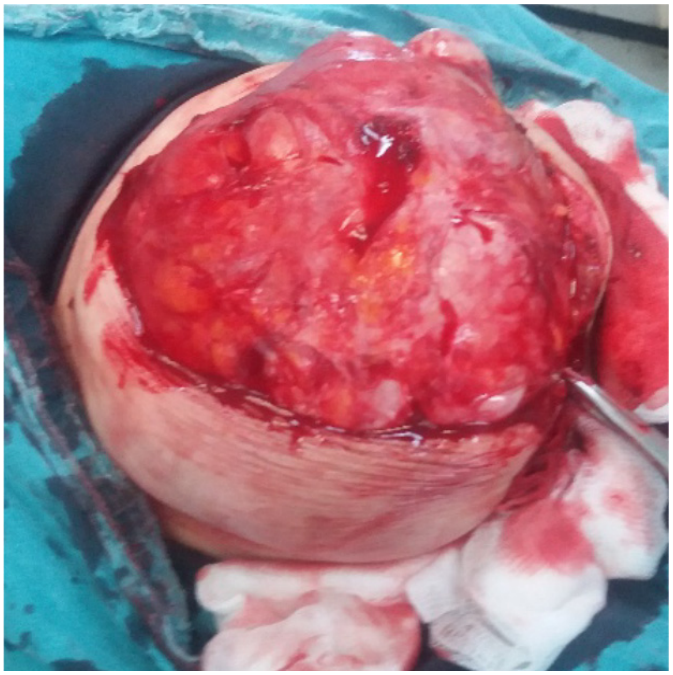


Fig. 3: Peroperative findings include a clearly defined mass present in the fatty tissue and between the fibers of the biceps femoris muscle.

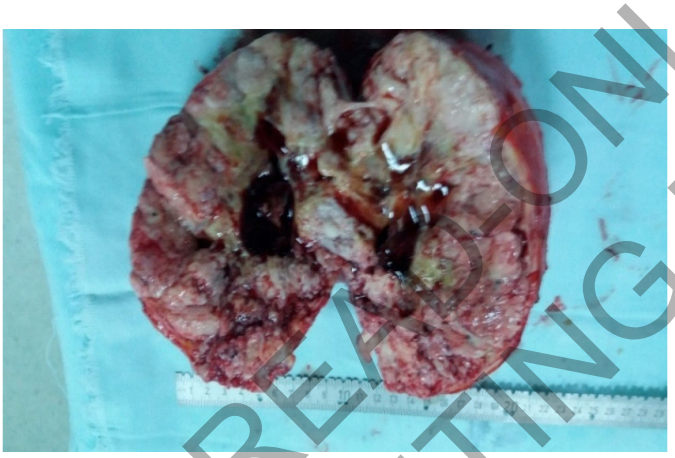


Fig. 4: Vertical section of surgically excised mass (20 x 10 cm)

A surgical excision of the change was performed, as well as drainage and multiple-layer suturing. The post-operative period was uneventful and the patient was discharged home on the seventh day after surgery (Figs. 4, 5).

Discussion

MO is a rare non-neoplastic condition characterised by a heterotopic ossification of soft tissue. Even though trauma is present in about 60-75 % [6], there are also non-traumatic formations, just as is the case of our patient. MO non-traumatica may usually be observed in younger patients, with a well-localised lesion [7,8]. Although not proven, some other aetiological mechanisms are also possible. One of the theories claims that osteoblasts, released from the periosteum and trapped in soft tissue, act as a trigger agent of MO [9]. Another mechanism is a theory of "ectopic calcifying islands" which argues that the periosteal tissue shifts into soft tissue due to the effects of the MO-causing trauma [10].

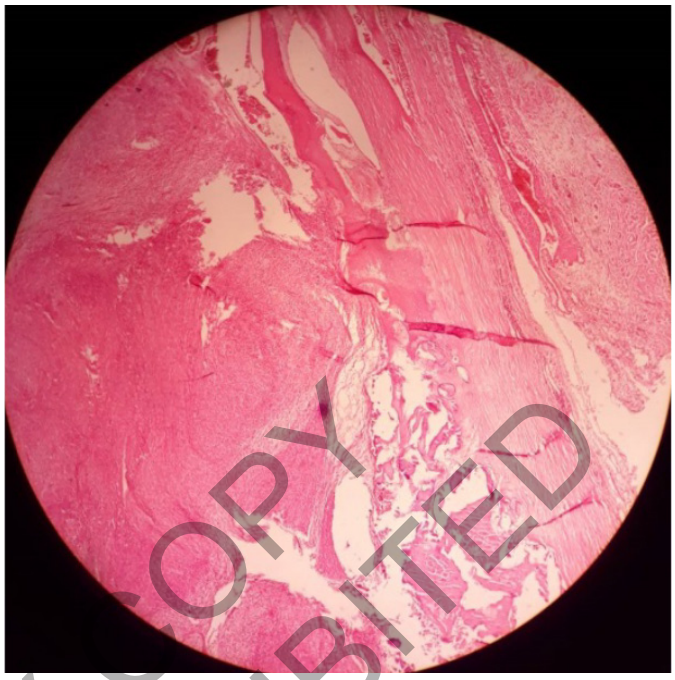


Fig. 5: PH results revealing a mature bone tissue within the fibroadipose and muscle tissue, and thus confirming the MO diagnosis.

A non-traumatic MO is scarcely described in the literature. A recurring microtrauma, tissue ischaemia and inflammation are listed as causal mechanisms of a non-traumatic MO [11]. MO non-traumatica occurs more often in patients with a parallel, subdural or epidural haemorrhage and a hip surgery. Fibrodysplasia ossificans progressiva is a disorder associated with an anomaly of the thumb or fingers, and it frequently occurs along with a non-traumatic MO [10,12]. Our case did not present any family history, trauma or associated anomalies of hands or fingers.

The differential diagnosis is related to the universal idiopathic calcinosis, dermatomyositis, osteosarcoma and illnesses caused by a change in calcium metabolism. Myositis ossificans may be often perplexing, but it has to be distinguished from osteosarcoma. The pain and swelling associated with osteosarcoma are persistent and progressive, with the periosteal elevation and cortical destruction appearing in X-rays, along with anaplasia in microscopic findings [13].

The symptoms include a localised swelling, pain and a feeling of reduced motion range in the affected extremity. At an early stage of the illness, the lesion is soft and painful, and in several weeks it develops into a hard and painful mass in the affected muscles [14]. A common clinical presentation includes the presence of a painful mass

in muscles and a palpable bone formation, but it can be erroneously diagnosed as a tumour-osteogenic sarcoma [15]. Our patient presented at a later stage, with the MR characteristics of a mature MO already set in, which helped us to exclude an osteogenic sarcoma.
There are no specific radiological findings of a non-traumatic MO. MRI is a technique of choice for assessing soft tissue lesions [16]. MRI results show heterogeneity due to the histological structure of an MO lesion, with the appearance of the hyperintense ring round the hypointense core being a sign of the maturation of the mass [17].
A nonoperative treatment is recommended solely if the lesion is completely immature, because removal of an immature bone may cause an extensive local recurrence. Some studies argue that the application of prophylactic indomethacin and etidronate may be useful for reducing a postsurgical ectopic calcification [18]. The application of bisphosphonates is effective at the initial stage of treatment, but later on their effectiveness is gradually and completely decreased [19]. Our patient could not have benefitted from physio- or medicamentous therapy due to the deterioration and maturity of the mutation.
Pharmacological interventions, such as indomethacin and other nonsteroidal anti-inflammatory drugs (NSAID) may supplement the therapy [19]. Surgery is recommended for patients presenting a mature change with functional limitations, as well as a prominent mass or lasting chronic pain. An operation is performed in the case of a complete bone maturity, according to the assessed presence of cortex in the radiography or MRI [20]. As our patient presented a large mutation, chronic pain and resultant functional difficulties, we decided upon a surgical excision. After one year of observation, the patient did not show any signs of a relapse, and she has fully recovered with no more pain.

Conclusion

A non-traumatic MO, present in our patient, is an extremely rare condition, for it has no previous trauma and includes an isolated involvement of the biceps femoris, while sparing other muscles. This case has not been documented in the literature known and accessible to us.
Our case presents an elderly female person, whereas most case reports deal with affected young men.
A successful postoperative recuperation serves as proof that a mature change necessitates an inevitable surgical intervention, with no additional radio- or medicamentous therapy.

Riassunto

La miosite ossificante (MO) è un'ossificazione ectopica caratterizzata da un'apparizione di formazione ossea prevalentemente nel tessuto muscolare. Il trauma è il fattore eziologico più comune, osservato in quasi il 60-75% dei casi, mentre un MO non traumatica è raramente descritta in letteratura. La diagnosi si basa su risultati clinici e radiologici.
Qui si riferisce di una paziente di 75 anni, ricoverata nella nostra clinica con un gonfiore localizzato nel compartimento femorale posteriore, risultante alla RMN come calcificazione nel muscolo bicipite femorale. I dati di laboratorio erano nell'ambito della normalità. Si è proceduto alla rimozione chirurgica della massa tumorale con ricostruzione e drenaggio, con un decorso postoperatorio regolare, e dimissione della paziente in settima giornata post-operatoria. Il risultato anatomo-patologico e istologico è stato di MO.
In letteratura ci sono pochi esempi di MO senza una causa traumatica nell'anamnesi. Un microtrauma cronico, l'ischemia tissutale e l'infiammazione sono elencati come meccanismi causali di una MO non traumatica, che capita più spesso in pazienti con emorragia parallela, subdurale o epidurale e chirurgia dell'anca.
Il nostro caso non ha presentato alcuna storia familiare, traumi o anomalie associate di mani o dita.
Dunque la miosite ossificante deve essere considerata come la diagnosi differenziale di tutte le masse tumorali dei tessuti molli, anche se i fattori di rischio noti non sono presenti nell'anamnesi. La chirurgia è una strategia terapeutica ragionevole in presenza di una massa tumorale nei tessuti molli, e la diagnosi definitiva può essere stabilita solo sulla base di reperti anatomo-patologici.

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