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Myositis Ossificans Tramatica: A Series of 15 Patients Presented at End Stage Disease and Review of the Literature

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ABSTRACT

Background: Myositis Ossificans Traumatica (MOT) is self-limiting disorder characterized by bone and cartilage development in soft tissue after an injury. Its diagnosis is easier shortly after injury among professional sportsmen. But it becomes difficult in public particularly when present late to physicians. Although it is a self-limited disorder to shorten the period of healing, different non-surgical methods are applied. Sometimes the condition become so disturbing that surgical treatment becomes compulsory.

Cases presentation: Herein we repot a15 MOT patients who presented at disease end stage and treated surgically because they were beyond non-surgical treatment modality. It is a retrospective study in which all the patients' characteristics, age, gender, lesions locations and their pathological result were discussed.

To our knowledge, few series of MOT reported in the literature. All of the patients in those studies were professional sport practitioner whose diagnosis and treatment could be started at the very early stages of the disease and so surgical treatment was not mandatory. In contrast, our series contain normal public whose diagnosis was late and treatment was surgical because of their late presentation.

Conclusion: Myositis traumatica is a self-limited disorder, it's healing can be accelerated by non-surgical treatments models but when diagnosis made late, it can progress to the extent that needs surgical intervention

Keywords: Trauma; Hematoma; Injury; Sarcoma

INTRODUCTION

Myositis Ossificans (MO) has been historically used to describe a wide spectrum of pathological processes ranging from benign solitary lesions to congenital progressive disease states. The MO process is known to begin with inflammation and end with bone and cartilage formation in extra-skeletal soft tissue. Recently, MO has been

classified as hereditary or congenital (MO progressiva) and non-hereditary MO, the latter of which is examined in two types: non-traumatic (MO circumscripta) and traumatic (MO traumatica).[1-3]

Unlike MO progressiva (fibrodysplasia ossificans progressiva or Münchmeyer disease), which is an inherited autosomal dominant condition, MO traumatica is a benign, solitary, self-limiting condition leading to heterotopic ossification of connective or other soft-tissue .It was firstly described by Kransdorf et al..[4] It occurs as a complication in 9–20% of large hematomas caused by muscle strains or contusions, and is the most common type of MO. It is a well circumscribed lesion which shows a characteristic appearance consistent with bone formation in radiological imaging studies .It can result in prolonged pain, local tenderness, diminished flexibility and stiffness of nearby joints. Although it can develop in all kinds of soft tissue, the great majority of cases are seen in skeletal muscle tissue.[1-4]

Despite the fact that MO traumatica is the most common type of MO, it is still a rare condition that requires further investigation. The aim of the current study was to describe the characteristics of MO traumatica cases diagnosed at our center, and to put forth our experience with this disease.

MATERIALS AND METHODS

Files of 15 patients who were diagnosed as MO traumatica between 1997 and 2016 at Gören Pathology Centre, TOBB ETÜ University Medical Faculty were identified from departments' archives. Diagnosis was made via evaluation of clinical findings with respect to patient complaints, and was confirmed via imaging (X-ray, magnetic resonance imaging) or histopathology. Any patients with other types of MO, those concurrently diagnosed with other diseases affecting bone or soft tissue structures, and patients with missing critical data were excluded from the study. The complaints, symptoms at presenting, clinical characteristics (examination findings), and localization of lesions were recorded.

In patients who required histopathology for diagnosis confirmation, biopsies were performed and histopathological assessment and staging were reported by the pathology department with respect to the three well-defined stages: Excisional samples obtained after surgical treatment were also subjected to the same analytic procedure when deemed necessary. Patients presenting with mature lamellar bone organization were identified as being in the 'end stage' of the disease.

RESULTS

Out of the 15 patients, 7 (46.6%) were male 8 (53.4%) were female. The youngest patient was an 8-year-old boy and the oldest was a 38-year-old male; average age was 24.26 ± 8.91 years. The MO traumatica lesions were localized in the muscle group of the hips in 9 patients (3 right-sided and 6 left-sided), the leg muscles(gastrocnemius) in 2 patients, and the forearm (flexor carpi radials)muscles, trapezius muscle , right shoulder(deltoid) muscles and

metatarsal muscles–each in one patient. The gender distribution, patient ages, lesion localization presented in Table 1, histopathology results and treatment methods are presented in Table 2.

Table1: Summary of the patient ditails, gender, ages and lesions' location.

	Sex	Age	Lesion location
1	Male	15years	Right hip
2	Female	25years	Right hip
3	Female	25Years	Right hip
4	Male	31years	Left hip
5	Female	25years	Forearm
6	Female	23Years	Left hip
7	Female	29Years	Left foot
8	Male	30Years	Left leg
9	Female	14Years	Left hip
10	Female	12Years	Left hip
11	Female	21Years	Left hip
12	Male	31Years	Right scapula
13	Male	8Years	Left hip
14	Male	37Years	Right shoulder
15	Male	38 Years	Left leg

Table 2: Demonstrates, biopsy obtainment pre-surgery (presurgical diagnosisi), type of treatment and finally histopathyological result.

Patient no	Biopsy before surgery	Treatment	Histopathology
1	No	Total excision	MOT
2	No	Total excision	MOT
3	No	Total excision	MOT
4	No	Total excision	MOT
5	Yes(FNB)	Total excision	MOT
6	Yes(tru-cut)	Total excision	MOT
7	No	Total excision	MOT
8	No	Total excision	MOT
9	Yes(FNB)	Total excision	MOT
10	Yes(FNB)	Total excision	MOT
11	Yes(tru-cut)	Total excision	MOT
12	Yes (FNB)	Total excision	MOT
13	Yes(FNB)	Total excision	MOT
14	No	Total excision	MOT
15	No	Total excision	MOT

FNB: Fine Needle Biopsy.

MOT: Myositis Ossificans Traumatica.

All patients complained of solid and painful lesions which restricted movement of the affected muscles and nearby joint at the time of presentation. Again, all patients had a history of major or repeated minor trauma at the site of the lesion. Twelve patients reported that they had sought medical help after major trauma, but had not attended follow-up studies until the development of symptoms.

All patients had undergone plain X-ray imaging (Figure1), and magnetic resonance imaging (Figure 2,3). Seven biopsies were ordered prior to treatment due to suspicion of malignancy (osteosarcoma). Ultimately, the diagnoses were confirmed via clinical and imaging findings in 6 patients, and via histological findings in 9 patients (7 biopsies, 2 excisional samples). All Histological examination revealed stage III MO traumatica in all cases. Grossly; the surface showed firm periphery, while the center showed soft and gelatinous, or hemorrhagic characteristics. Peripheral regions exhibited fresh bone trabecula rimmed by osteoblasts and mineralization (Figure 4). Patients attended follow-up for 2 years, and no recurrence was observed.

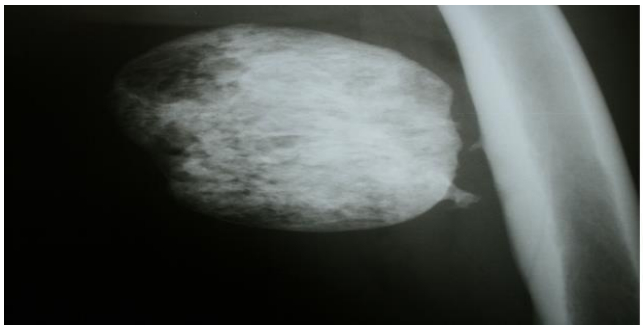

Figure 1: Direct Radiograph of the thigh demonstrating MOT

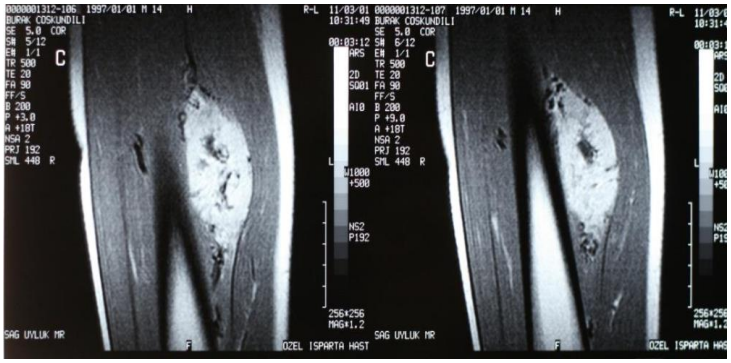

Figure 2: Coronal section MRI of the right thigh demonstrating MOT.

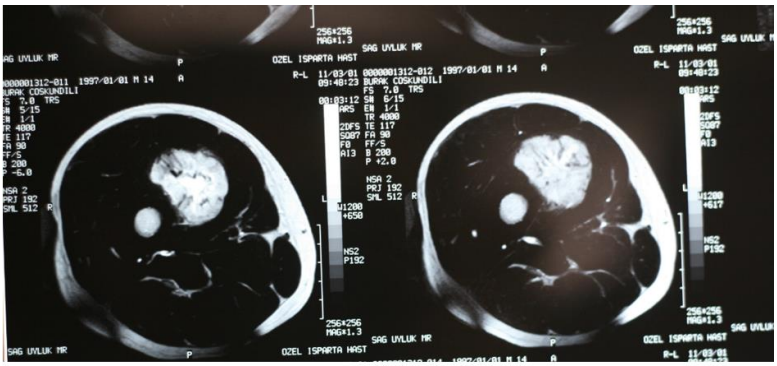

Figure 3: Axial section MRI of the right thigh demonstrating MOT.

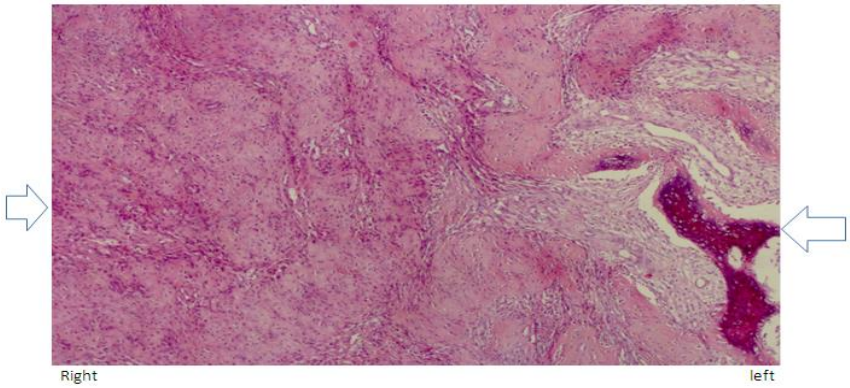

Figure 4: Showing the histological features MOT: Histologically there is characteristic zonal pattern with cellular fibroblastic cells in the central part (left) and mineralized bone trabeculae (right) HEX100.Left border demonstrating the inner zone while the right border demonstrating the inner zone (mature bone formation)

DISCUSSION

MO traumatica, as the name clearly explains, is the posttraumatic formation of bone and cartilage in soft tissues – often preceded by intramuscular hematoma. It is relatively more common in athletes as a complication after muscle injury, but it can occur in any individual after major or recurrent minor trauma. Although its pathogenesis is not completely understood, ıt has been hypothesized that MO traumatica develops due to the liquefaction of the center of the hematoma, leading to the formation of a sheet of nonspecific cells. In anoxic conditions, these cells rapidly proliferate and may differentiate into osteoblasts, resulting in bone formation.[5-7] The clinical presentation of MO traumatica is variable, most patients are able to recall specific injuries or repetitive minor trauma at the site. MOT should be clinically suspected when the injury-associated symptoms are sustained up to 10-14 days after rest. Differential diagnosis from osteosarcoma is a very important issue since the first is a self-limited disorder, while osteosarcomas are very aggressive cancers by definition.[2,4] However, especially in patients with MOT who present at later disease stages (such as our patients), surgical intervention may be required.

Most of the case series on this topic show a male predominance in MO traumatica, but no significant sex predominance was observed in our group of patients. This difference can be explained by the fact that most of the other studies accrued data from athletes and professional sports players, which are patient groups with male sex predominance. To our knowledge, our series is the only one completely comprised of non-athlete patients. Prior data on such patients are largely from various case reports including single patients.

The patients included in our study were non-athletes and were not involved in amateur or professional contact sports. Therefore, the variation in localization (60% of lesions were in the hips) can be attributed to falls on their hips (with respect to their history), rather than sports-related trauma which is common in prior studies.[8,9] Taken from another perspective, being professionally involved in sports causes a high likelihood of early presentation to healthcare facilities after injury and commitment to follow-up evaluations, which greatly increases the chance for

early diagnosis and treatment; whereas the same attention may not be present in our group of patients.[10] Therefore, although the literature reports very few surgical interventions for MO traumatica, all of our patients required surgery due to being in the end stage of disease.

Some serum tests as C-reactive protein and erythrocyte sedimentation rate were found to be elevated at the initial stages of MO traumatica, but, similarly, they are not specific to the condition.[10-11]

Radiological imaging plays a vital role in MOT diagnosis. Ultrasonography is often used as an initial diagnostic method, especially in early disease stages where bone formation is not observed via X-ray. But at a late stage of the disease calcified lesion can be very obvious on plain X-ray, Figure 1. Thomas et al. evaluated the role of ultrasonography in MO diagnosis, and described an inner, a central and an outer concentric zone in the lesions of patients.[11] Ultrasonography was not performed in any one of our patients. Because all of our patients were admitted at a very late stage of disease due to delayed presentation, the masses were calcified in the great majority – necessitating differential diagnosis from malignant lesions. Figure 2 and Figure 3 demonstrate calcified lesions on MR.

Radiographs alone can be diagnostic for MOT, particularly in the mature phase of lesions when the patient's history and clinical presentation are correlated. During the first 2 weeks after injury plain radiographs may look normal even if MO is developing. But, occasionally periosteal reaction can be demonstrated. The calcification of soft tissue becomes apparent on the radiographs around the 3rd to 4th weeks after injury. It is also possible to utilize CT in diagnosis. However, since it is best to show the zonal pattern of calcification before the calcification develops, magnetic resonance imaging is generally accepted as the best radiologic modality of soft-tissue evaluation.[11-17] (Figure 2) In some patients, although radiologic diagnosis was indicating MO traumatica, biopsies were obtained because of the need for differential diagnosis between MO traumatica and sarcomas, particularly in younger patients. Scintigraphy will show increased uptake in injured muscles due to calcium salt presence, and thus, scintigraphy is the most sensitive imaging method for the determination of heterotopic bone formation at early stages. Although diagnosis is usually straightforward with sufficient medical history and imaging studies, differential diagnosis may become mandatory in some cases. Abscess formation, periosteal reaction, recurrent giant cell tumor and melorheostosis are among the benign lesions that should be considered when no clear history of trauma and no clear radiological imaging zones are seen or reported. On the other hand, extra-skeletal osteosarcoma, soft-tissue sarcoma and parosteal osteosarcoma are among the malignant lesions that should be considered.[13,14,19] Atypical locations can also make differential diagnosis crucial. In a study comprised of 23 patients with atypical MO presentation, Nuovo et al. reported that 15 lesions were in unusual locations, such as the fingers and chest wall.[20] We also had some cases with unusual localization, one in the metatarsal muscle and the other with a lesion located on the trapezius muscles. The same study also found that three of their patients were younger than 10 years. We had one patient 8 years of age, 3 patients 10-15 years old.

Despite being a self-limiting disease which can sometimes resolve on its own, MO traumatica has a number of effective treatments. Authors tend to emphasize the importance of early non-surgical treatment.[21,22] Nonsurgical treatment aims to minimize the symptoms , improve the functions of affected muscles or nearby joints and shortening the period of rest or been away from activities. For initial treatment, a short period (3-7 days) of immobilization, rest, ice application and elevation is advised. Minimizing of intramuscular blood flow via cryotherapy is also applied. Another form of nonsurgical treatment is extracorporeal shock wave therapy .It was declared that this treatment had shortened the healing period. Acetic acid phonophoresis is another recent addition to the nonsurgical treatment methods.[23]

Surgical treatment should be preserved for symptomatic lesions which have not responded to non-surgical treatment. The aim of treatment, whether surgical or nonsurgical, is to improve function and reduce lesion-associated symptoms. Surgical treatment should be indicated in cases with intractable pain resulting from mechanical irritation of nearby anatomic structures, and lesions that cause compression of neurovascular structures that lead to restriction of motion and daily activities. Unnecessary aggressive resection should be avoided since the disease is benign, but at the same time some recurrence probability should be kept in mind while performing resection.[11-25] All of our patients were treated surgically due to delayed presentation and diagnosis. It is likely that our patients were unaware of their lesion until mass formation and increased pain which led to discomfort. Although motion restriction was reported by all subjects, it appears that the extent of restriction was acceptable for them –possibly because most lesions were not in close proximity to joints. But all 15 patients were fearing malignancy.

CONCLUSION

MO traumatica is commonly seen in persons involved in professional or semi-professional sports, and usually after muscular trauma that causes hematoma development. Despite being uncommon in the general public, our group of patients demonstrates that it may still be encountered. We believe it is critical to note that physicians should be highly aware of such possibilities in patients, evidenced by the fact that none of the patients included in the current study could be diagnosed in the early stage of disease. The differential diagnosis of the condition, especially from sarcomas, is important for treatment. The first-line treatment of MOT is advised to be non-surgical, and there appear to be important developments on this topic. Surgical treatment should be preserved for lesions in which non-surgical treatment has failed, or for those who present with solid and very painful masses –as was the case in all 15 of our patients.

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