

Bilateral overuse myositis ossificans of the triceps: report of an unusual case

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SUMMARY: Bilateral overuse myositis ossificans of the triceps: report of an unusual case.

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Overuse myositis ossificans is considered an extremely rare diagnosis, with only a few cases reported so far. A case of a 54-year-old firefighter, involved regularly in training exercises, with bilateral myositis ossificans of the triceps, is presented. The patient had a 4 year history of painful masses at the posterior region of both arms. He never experienced any traumatic event, while physical examination revealed firm masses extending longitudinally within the lateral head of the triceps, bilaterally. The patient had painful restricted range of motion at both

sides. Plain X-ray views of the humerus showed linear lobulated heterogeneous masses posterolateral. Surgical resection of both masses, which extended intramuscularly through the whole lateral triceps' head, was performed. Histological examination confirmed the diagnosis of myositis ossificans. Myositis ossificans represents a benign, relative rare clinical entity, defined as heterotopic ossification of the soft tissues. Only 5 cases of myositis ossificans due to overuse have been described so far. The present case is unique, since it is the first one describing bilateral appearance of two masses which extended through the whole length of the lateral head of the triceps. A detailed medical history, as well as imaging examination seem to be necessary in order to establish the diagnosis. Treatment should be decided upon the stage of the lesion. For mature lesions surgical treatment is advised.

KEY WORDS: Heterotopic ossification - Overuse injury - Myositis ossificans.

Introduction

Myositis ossificans represents a form of heterotopic ossification, usually affecting active young adults. It is most commonly the result of a direct traumatic event or intramuscular hematoma. It usually presents near the long bone diaphysis, with the thigh being the most common region of appearance (1).

It is of utmost importance to exclude malignancies from the differential diagnosis, such as soft tissue sarcomas (2). The bilateral appearance of this clinical entity is considered rare.

A case of a 54-year old male presenting with atraumatic bilateral myositis ossificans of the triceps, treated operatively is presented.

Case presentation

A 54-year old-male firefighter, with a 4-year history of painful masses at the posterior region of both arms, presented at the out-patient clinic of the orthopaedic department. He could not remember any specific onset of pain, while he stated that this masses had become larger over the last 6 months. The patient was involved as a trainer in an intensive fire-fighting training program during the last 10 years. He experienced some fatigue, especially during exercises involving the upper extremities. However, he never experienced any traumatic event. His rest medical history was unremarkable. Physical examination revealed firm masses extending longitudinally within the lateral head of both triceps. The patient had painful restricted range of motion at both sides (right side: extension limited at 65° and flexion limited at 120°; left side: extension limited at 70° and flexion limited at 125°). Neurovascularly both upper

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extremities were intact. The quickDASH score was found to be 29.5.

Plain X-ray views of the humerus showed linear lobulated heterogeneous masses at the posterolateral region (Figure 1). The differential diagnosis of soft tissue sarcoma and myositis ossificans was established. Due to the fact that the lesion had occurred over a long period of time (4 years) and was bilateral the diagnosis of sarcoma was highly unlikely.

At that point surgical resection was advised. The patient, under general anesthesia and in supine position underwent resection of both masses. Intra-operatively it was revealed that both masses were located intramuscularly, within the lateral head of the triceps. En bloc resection was performed (Figure 2). Due to the fact that the masses involved the whole lateral head of the triceps, the whole head was necessary to be resected at both sides.

The patient made a satisfactory recovery. He was discharged on the 4th postoperative day. He was then subjected to physiotherapy, involving the first 2 weeks passive and then active elbow flexion and extension. The patient was also commenced on in-

domethacin 75 mg/day for 6 weeks.

The histological examination of the masses showed spindle cell lesions with moderate cellularity. The cells were relatively uniform, with elongated nuclei, small nucleoli, and indistinct borders. There was mild mitotic activity without atypical mitoses. No necrosis was present. In part of the periphery of the lesion, reactive bone production with intense osteoblastic rimming was observed. On immunohistochemical staining, the spindle cells were positive for vimentin and mildly positive for α -smooth muscle actin. There was no positivity for desmin, caldesmon, S-100 protein, β -catenin, CD34, CD117/c-kit or CD57. The pathologic features were consistent with myositis ossificans.

Due to absence of a traumatic event and the patient's participation into firefighting training programs for the last decade the diagnosis of overuse bilateral myositis ossificans was established.

At the 3-year follow-up the patient continues his everyday activities without any sign of a disease, while no recurrence has occurred. The quickDASH score at the final follow-up was found to be 6.8 (Figure 3).

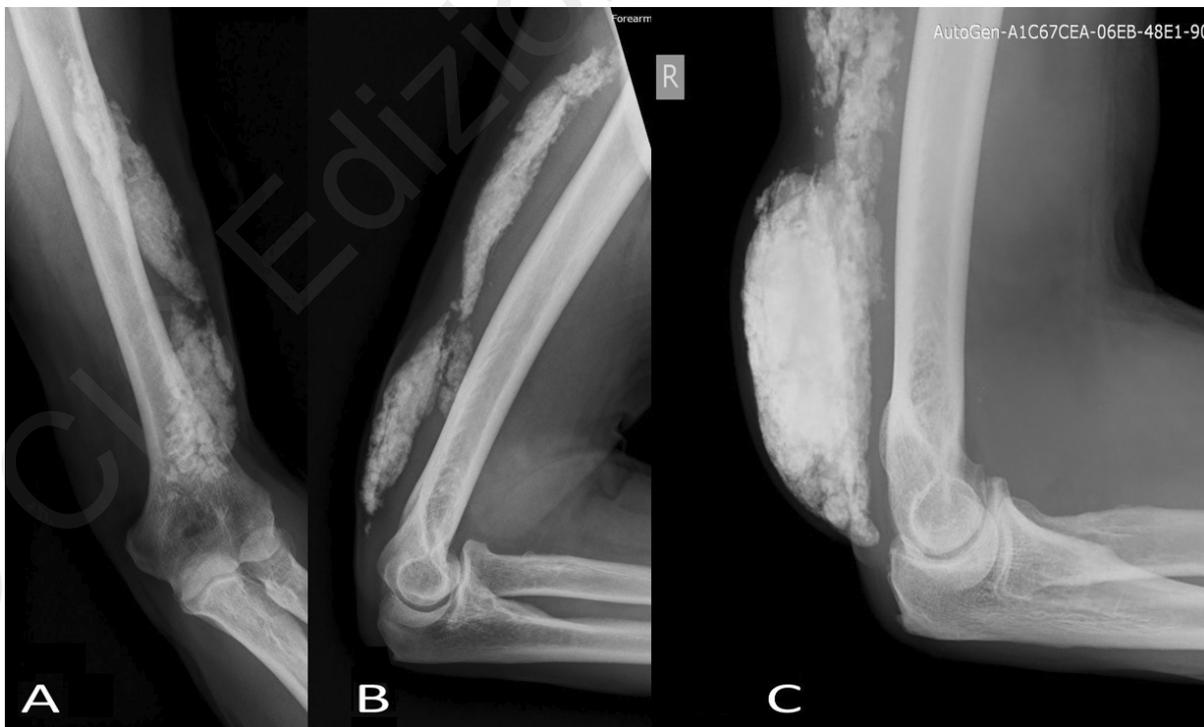


Figure 1 - X-ray views of both arms. A, B) Left side, C) right side. Linear lobulated heterogeneous masses at the triceps region are evident.



Figure 2 - Intraoperatively picture showing the masses' resection. The masses were located intramuscularly, extending through the whole length of the lateral triceps' head bilaterally.

Discussion

Myositis ossificans represents a benign, relative rare clinical entity, defined as heterotopic ossification of the soft tissues (1, 2). It is a pseudoinflammatory tumour originating from skeletal muscle and corresponding to a heterotopic, metaplastic, non malignant bone tumour. It most commonly appears in active young adults, especially during the second or third life decade. However, there have been cases of elderly as well as pediatric patients (1-3).

The majority of myositis ossificans cases have been associated with trauma. The most commonly affected locations are the anterolateral aspect of the thigh and upper arm (4). A muscle contusion is adequate enough to cause muscle damage and lead to

hemorrhage, as well as muscle necrosis. An inflammatory response is then produced, stimulating fibroblasts and undifferentiated stromal cells to migrate to the connective tissue. The release of multiple enzymes at the place of injury may trigger the differentiation of those fibroblasts into osteoblasts and therefore to calcification formation (4, 5). Increased vascularity, metabolism and excess calcium due to hyperemia represent the main elements that may lead to ectopic bone formation (4, 6).

The mechanism of overuse leading to new bone formation still remains unknown. It has been suggested that is continuous activity causes microscopic muscle injury and damage, initiating an inflammatory reaction (4, 7-9).

Myositis ossificans should be differentially diag-

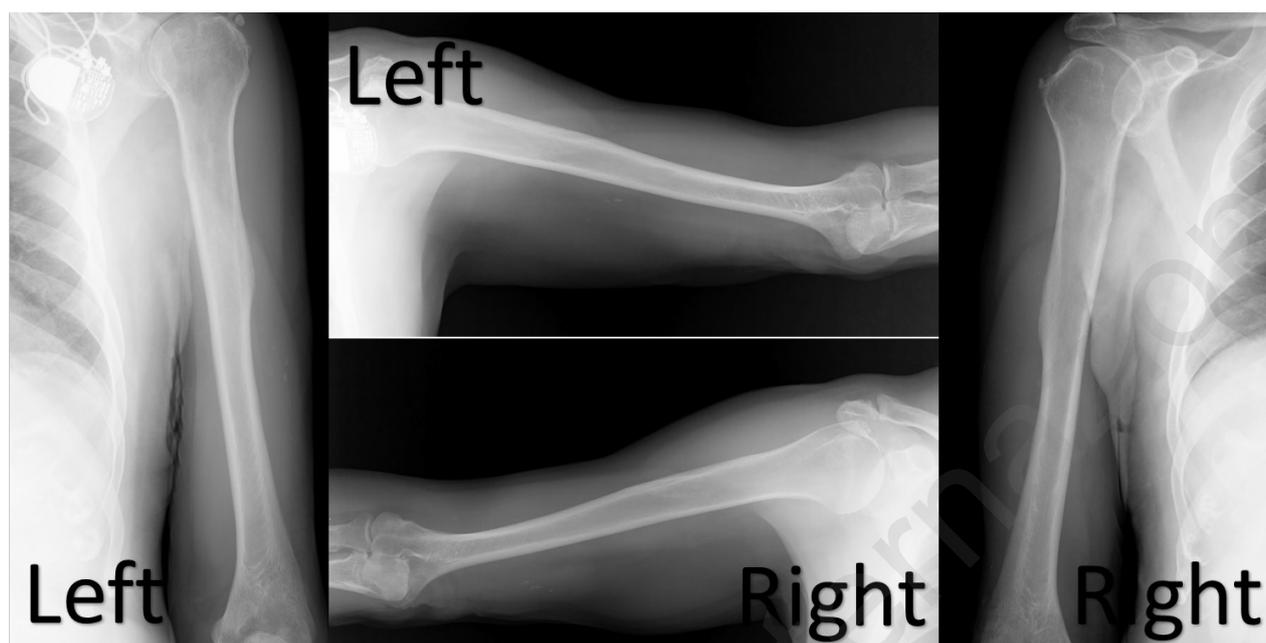


Figure 3 - Postoperative X-rays of both arms at the 3-year follow-up.

nosed from osteomyelitis and malignancy, such as osteosarcoma or soft tissue sarcoma. Computed tomography is the gold standard for diagnosis of myositis ossificans in the early stages of the lesion (1-4). Typical signs are muscle edema and perilesional edema. Calcification of the lesion is characteristic in the periphery. Firm diagnosis is performed through histological examination (4, 10, 11). Therefore, it is of paramount importance to resect lesion with high suspicion for malignancy, such as cases with minor fever and palpable adenopathy. The reported patient did not exhibit such symptomatology. However, due to the painful range of motion restriction, as well as the extension of the lesions, surgical resection was performed.

Atraumatic myositis ossificans is considered relative rare, while overuse as a causative factor has only been described in a few cases. After a meticulous electronic literature review, using the PubMed and Medline databases, myositis ossificans due to overuse seems to have been reported in 5 cases. Gast et al. in 1987 first described the appearance of this clinical entity in a 16-year old female, taking part in running races (12). Webner et al., 20 years later, reported on a 42-year-old recreational runner, without a history of a traumatic event (13). Defoort et al. in 2012 described a case of myositis ossificans circumscripta of both triceps due to overuse in 32-year old

female swimmer (4). In 2014, a case of myositis ossificans of the psoas muscle due to overuse in a 16-year old female gymnastic was also reported (14). Furthermore, in 2017 a similar case of overuse myositis ossificans in the brachialis muscle in an 18-year old male was described (15). These 5 cases described the overuse as a causative factor for myositis ossificans. However, only one case involved both triceps (4). In the present patient, the ectopic bone formation had affected the whole lateral triceps head bilaterally. These characteristics make this case unique.

In the early stages of myositis ossificans the proper management is considered to be conservative, including rest and anti-inflammatory drugs such as indomethacin and ibuprofen (16). It is of note that such regimens have been applied not only for the treatment of myositis ossificans, but also for the prophylaxis of heterotopic ossification, following surgical resection. In addition to anti-inflammatory drugs, radiation has also demonstrated similar efficacy as postoperative prophylaxis for heterotopic ossification (17). The present case suffered from a mature lesion, extending intramuscularly through the whole lateral head of both triceps. Surgical resection was the treatment of choice, while postoperatively he received indomethacin for 6 weeks. Three years following the operation he remains free of disease.

In conclusion myositis ossificans represents a benign clinical entity that should be differentially diagnosed from malignancy. A detailed medical history, as well as radiological examination seem to be necessary in order to establish the diagnosis.

Congresses

None.

Contributors

None.

Conflict of interest

The Authors declare that there is no conflict of interest.

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Authors' contributions

SG, CK for the literature search and analysis, and manuscript writing. CK for the final manuscript revision. All Authors have read and approved the final manuscript.

References

1. Micheli A, Trapani S, Brizzi I, Campanacci D, Resti M, de Martino M. Myositis ossificans circumscripta: A paediatric case and review of the literature. *Eur J Pediatr.* 2009;168:523-9.
2. Ogilvie-harris DJ, Fornasier VL. Pseudomalignant myositis ossificans: Heterotopic new-bone formation without a history of trauma. *J Bone Joint Surg Am.* 1980;62:1274-83.
3. Paterson DC. Myositis ossificans circumscripta, report of four cases without history of injury. *J Bone Joint Surg Br.* 1970;52:296-301.
4. Defoort S1, Arnout NA, Debeer PD. Myositis ossificans circumscripta of the triceps due to overuse in a female swimmer. *Int J Shoulder Surg.* 2012 ;6:19-22.
5. Estwanik JJ, McAlister JA. Contusions and the formation of myositis ossificans. *Physician Sportsmed.* 1990;18:53-7.
6. Chadha M, Agarwal A. Myositis ossificans traumatic of the hand. *Can J Surg.* 2007;50:21-2.
7. Cheung K, Hume P, Maxwell L. Delayed onset muscle soreness: treatment strategies and performance factors. *J Sports Med.* 2003;33:145-64.
8. Toumi H, Best TM. The inflammatory response: Friend or enemy for muscle injury? *Br J Sports Med.* 2003;37:284-6.
9. Miller AE, Davis BA, Beckley OA. Bilateral and recurrent myositis ossificans in an athlete: A case report and review of treatment options. *Arch Phys Med Rehabil.* 2006;87:286-90.
10. Saussez S, Blaivie C, Lemort M, Chantrain G. Non-traumatic myositis ossificans in the paraspinal muscles. *Eur Arch Otorhinolaryngol.* 2006;263:331-5.
11. Ehara S, Nakasato T, Tamakawa Y, Yamataka H, Murakami H, Abe M. MRI of myositis ossificans circumscripta. *Clin Imaging.* 1991;15:130-4.
12. Gast W, Hagen F-damage caused by excessive stress in performance sports? *Sportverletz Sportschaden.* 1987;1:59-63.
13. Webner D, Huffman GR, Sennett BJ. Myositis ossificans traumatica in a recreational marathon runner. *Curr Sports Med Rep.* 2007;6:351-3.
14. Masquijo JJ, Sartori F. Myositis ossificans circumscripta of the psoas muscle due to overuse in an adolescent gymnast. *J Pediatr Orthop B.* 2014;23:529-32.
15. Galanis N, Stavrika C, Valavani E, Kirkos J. Unsupervised Exercise-Induced Myositis Ossificans in the Brachialis Muscle of a Young Healthy Male: A Case Report. *Orthop J Sports Med.* 2017;5:2325967117718780.
16. Shehab D, Elgazzar AH, Collier BD. Heterotopic ossification. *J Nucl Med.* 2002;43:346-53.
17. Baird EO, Kang QK. Prophylaxis of heterotopic ossification - an updated review. *J Orthop Surg Res.* 2009;4:12.