# Unsupervised Exercise–Induced Myositis Ossificans in the Brachialis Muscle of a Young Healthy Male

## A Case Report

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Keywords: heterotopic ossification; eccentric exercise; ossifying fasciitis; sports injury; overuse injury

Extraskeletal bone formations within the soft tissues can be the presentation of benign self-limited ossifying processes such as heterotopic ossification  $(HO)^{24}$  or malignant neoplasms such as extraskeletal osteosarcomas.<sup>9</sup> Depending on the involved tissue, a number of synonymous terms have been established to describe these lesions. Myositis ossificans  $(MO)^2$  is the most common and refers to benign bone formation within skeletal muscles, whereas fasciitis<sup>14</sup> and panniculitis ossificans<sup>13</sup> refer to lesions in the fascia or the adipose tissue, respectively. The terms *heterotopic ossification* and *myositis ossificans* are often used interchangeably in the literature.

The etiology of MO is variable and classified as hereditary or acquired, with the latter being the most common type.<sup>26</sup> Hereditary MO includes fibrodysplasia ossificans progressiva and progressive osseous heteroplasia; both very rare, autosomal dominant inherited conditions associated with debilitating progressive heterotopic ossification.<sup>18</sup> The

The Orthopaedic Journal of Sports Medicine, 5(7), 2325967117718780 DOI: 10.1177/2325967117718780 © The Author(s) 2017 acquired MO is further subclassified into traumatic and nontraumatic depending on the presence of history of trauma. Although its underlying pathophysiology has not yet been clearly defined,<sup>19</sup> traumatic MO has been largely associated with various types of musculoskeletal injury, including fractures and soft tissue trauma, as well as orthopaedic procedures (hip, knee, and shoulder arthroplasties).<sup>1,16,24</sup> The most commonly affected site of traumatic HO is the quadriceps femoris.<sup>25</sup> Other causes of traumatic MO include central nervous system injury<sup>8</sup> and severe burns.<sup>6,20</sup> However, there are rare cases of MO that occur in the absence of a discrete traumatic etiology.<sup>10,17,29</sup> Despite being a benign condition, nontraumatic MO poses a significant diagnostic challenge, as its clinical presentation mimics that of sinister conditions such as osteosarcoma.<sup>28</sup>

Here, we present a unique case of HO in the right anterior brachialis muscle of a young healthy man with no previous history of trauma or injury, who had recently started working out at the gym. To the best of our knowledge, this is the first report of HO secondary to recreational exercise. Informed consent was obtained from the patient.

#### CASE REPORT

An 18-year-old right-handed male patient presented with a 6-month history of intensifying muscle soreness and a progressively growing palpable mass in the anterior distal third of the right humeral shaft. The mass had been growing within the anterior brachialis muscle, significantly limiting the range of motion of the right elbow joint. There was no history of antecedent trauma, previous surgery, or other predisposing conditions. The patient had no past medical history of note, and there was no family history of abnormal

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The authors declared that they have no conflicts of interest in the authorship and publication of this contribution.

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Summary of the Patient's Workout Regimen Involving His Biceps Muscles			
No. of Exercises for Biceps	Push-ups	Pull-ups	Barbell Curls
Repetitions	Until exhaustion	Until exhaustion	Until exhaustion
Sets	3	3	3
Intervals, s	60	60	60
Speed of repetitions	Quick	Quick	Quick
Range of motion	Limited in flexion and extension	Limited in flexion and extension	Limited in flexion and extension
Frequency per week	6 sessions	6 sessions	6 sessions

TABLE 1 337

bone formation. He had recently started working out at the gym on a regular basis, without professional supervision. His workout regimen involving the biceps brachialis muscles is summarized in Table 1. Physical examination revealed a  $3 \times 5$ -cm well-circumscribed, firm, and nontender mobile subcutaneous nodular mass. There were no associated ervthema, warmth, or skin changes. The right elbow joint range of motion was significantly decreased (80°-130°), but pronation and supination were not affected. His right arm was well perfused with an intact radial pulse, and neurological examination was unremarkable. Complete full blood count, biochemical analysis (including alkaline phosphatase), coagulation screen, and urinalysis did not reveal any abnormality.

Plain radiographs of the right arm revealed a welldefined ovoid structure with a prominent mineralized periphery and a radiolucent center at the distal third of the humerus with intact cortex (Figure 1).

Magnetic resonance imaging (MRI) revealed no continuity between the mass and the underlying bone. The mass had an intermediate- to low-intensity signal on T1-weighted images and a heterogeneous high-intensity signal on T2-weighted images. The bony structures of the right elbow joint were not affected.

Total excision of the mass was performed with an anterior approach. Macroscopically, the mass measured 5  $\times$  5  $\times$ 4 cm and had a gray-white appearance (Figure 2). It had firm to fragile consistency with a thin bony rim in part of its periphery and contained jelly-like matrix. There was no continuity with the surrounding tissues.

Histological examination revealed a spindle cell lesion with moderate cellularity. The cells were relatively uniform, with elongated nuclei, small nucleoli, and indistinct borders. There was mild mitotic activity without atypical mitoses. The cells were arranged in fascicles and whorls (Figure 3). The stroma contained abundant collagen fibers, with focal myxoid and hyaline changes. Focal lymphocytic infiltration and mild hemorrhage were also observed. In small areas, the cellularity and mitotic activity were increased (there were up to 6 mitotic figures per high-



Figure 1. Plain radiograph showing a well-defined mass with mineralized periphery and radiolucent center at the distal third of the right humeral shaft.



Figure 2. Intraoperative image showing a well-circumscribed mass with a gray-white appearance.

power field). 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  $\alpha$ -smooth muscle actin. There was no positivity for desmin, caldesmon, S-100 protein, β-catenin, CD34, CD117/c-kit, ALK-1, or CD57. The pathologic features were consistent with ossifying fasciitis.

Five years after the operation, there was no sign of recurrence, and the patient had full recovery of function returning to his normal exercising routine and was advised to follow specialist guidance for his future workout routines.



Figure 3. Microscopic appearance of the lesion.

### DISCUSSION

MO is a benign process of ectopic bone formation within the soft tissues<sup>26</sup> with a yet-unclear pathophysiology.<sup>19</sup> Although its etiology has been classified into traumatic, neurological, and genetic causes, its exact pathophysiology yet remains unclear. It has been suggested that the condition occurs through the aberrant differentiation of mescenchymal stem cells into osteoblastic precursor. This process is mediated by the dysregulated release of osteoinductive factors such as bone morphogenetic proteins and transforming growth factor– $\beta$  in response to tissue injury and consequent inflammation.<sup>11</sup>

MO is a fairly common sequela of musculoskeletal injury and is rather prevalent in young people involved in strenuous physical exercise, particularly athletes.<sup>16</sup> Very few cases of nontraumatic HO have been published to date involving the paravertebral<sup>10,22</sup> and iliopsoas<sup>7</sup> muscles and thigh.<sup>3</sup> Apart from direct trauma, repetitive microtraumas during physical activities can also lead to the development of HO. Such cases have been described in the deltoid muscle of young military men due to execution of shoulder-arm exercises and rifle firing<sup>12</sup> and in the adductor muscle of a young football player presenting as chronic groin pain.<sup>5</sup> In this report, we present a rare case of HO in a young healthy man with no previous history of trauma, who had been intensely working out in the gym. To the best of our knowledge, this is the first report to describe HO within the brachialis muscle as a result of unsupervised recreational workout. We believe that a hypertrophy-oriented program without supervision of a specialist entails the risk of misinterpretations that could lead to injury. In our case, we speculate that the major mistakes were overloading the muscle group responsible for the flexion of the elbow joint and, additionally, the limited range of motion combined with quick repetitions during the eccentric phase.<sup>23</sup>

Such presentations of MO pose a diagnostic challenge to the clinicians, as they mimic sinister conditions such as malignant soft tissue sarcoma,<sup>27</sup> which is associated with poor prognosis and which requires radical surgical treatment. Other conditions that have similar clinical presentation and should be excluded include aneurysmal bone cyst, calcified fibromatosis, and local infections.<sup>21</sup>

Comprehensive clinical examination and radiological evaluation are of paramount importance for the accurate diagnosis of MO. At its early stages (7-10 days), MO manifests as an inflammatory rapidly growing mass, and ossification starts to develop between 11 days and 6 months from the onset of the lesion. From 10 weeks to 6 months, the lesion takes its "egg shell" appearance, which signifies its end stage of maturation. This late stage is characterized by a typical zonal pattern, which consists of a central area of proliferating fibroblasts and myofibroblasts with areas of necrosis, an intermediate zone of osteoblasts with immature osteoid formation, and a peripheral zone of mature bone.<sup>21</sup> During the initial phases of its development, bone scintigraphy is the imaging method of choice for its evaluation. On the lesion's evolution, plain radiographs may show de novo pathognomonic ossification surrounding a clear central area, as demonstrated in our case. MRI plays a key role in the evaluation of such lesions. In premature lesions, the T2-weighted images show a heterogeneous mass with high signal intensity in the central area, and on maturation, the hyperintense center becomes encircled by a hypointense rim.<sup>15</sup> The zonal pattern and the lack of surrounding tissue invasion are key diagnostic imaging characteristics of MO.

In our case, the patient had a mature lesion on presentation, and the diagnosis was established based on its radiological characteristics. In cases where the diagnosis remains indeterminate, a biopsy is required. However, the timing of the biopsy is critical, as it may not be informative if the lesion is at a rather early stage.<sup>15</sup>

In the majority of cases, particularly in early MO, the treatment is nonoperative and includes rest and antiinflammatory drugs such as indomethacin, ibuprofen, and bisphosphonates.<sup>24</sup> These have been used both for treatment and prophylaxis of HO. Radiotherapy has also been used to treat or prevent MO. Both radiation therapy and anti-inflammatory medications have demonstrated similar efficacy as means of postoperative prophylaxis for HO.<sup>4</sup> However, concerns have been raised about the use of radiation therapy in younger patients because of its potential carcinogenetic side effects, which may evolve later in the patient's life.<sup>4</sup> Surgical excision is the indicated treatment for patients with mature lesions, which compromise the range of motion of the involved joint. This applied to our case, as the patient presented with a fully matured HO significantly limiting the range of motion of his right elbow joint. Given his young age and the equivocal benefit of radiotherapy over nonsteroidal anti-inflammatory drugs, he received ibuprofen postoperatively and made a remarkable recovery with no recurrence at 5-year follow-up.

### CONCLUSION

This case illustrates that HO can stem from intense recreational workout if not done correctly. A history of trauma may be absent, however this should not preclude it as a possible differential diagnosis in cases of soft tissue tumors, particularly in young athletic patients. A thorough history and radiological investigation play a key role in establishing the diagnosis. Furthermore, it shows the good prognosis of this benign tumor confirming nonrecurrence at 5 years postexcision.

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