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Myositis ossificans: A case treated with radiotherapy and literature review

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Abstract

Introduction: Myositis Ossificans is a benign, self-limiting process characterised by heterotopic ossification (HO) in extraskeletal sites. Radiotherapy (RT) seems to have a therapeutic effect in this disease and the aim of this article is to present a case of HO successfully treated with radiotherapy and review the literature.

Case Presentation: A 42-year-old woman presenting severe left shoulder pain of several months evolution with progressive reduction of joint movements without significant pain relief on analgesic and corticosteroid drugs. Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) showed a significant intramuscular calcification in infraspinous fossa. Biopsy was required and the histological diagnosis was MO. The patient was treated with RT with a total dose of 6 Grays in six fractions. The one year follow-up showed full function of the left harm and complete pain relief, therefore surgery was avoided and patient follows up with no need of analgesics treatment.

Discussion: The treatment of HO is complex and often needs a multidisciplinary approach. Conservative treatment's goal with rehabilitation therapy and non-steroidal anti-inflammatory drugs (NSAIDs) is to reduce symptoms and enhance mobility while surgical excision is performed for symptomatic and big lesions that are not controlled with conservative management.

Conclusion: This case shows that RT is helpful to control this disease and improve patient's symptoms with relatively low side effects, in order to avoid a surgery intervention and adverse effects of NSAIDs. There are only few articles in literature about the use of RTin HO treatment and further research is needed.

Keywords

heterotopic ossification; myositis ossificans; radiotherapy; humerus

Abbreviations

MO: Myositis Ossificans; MOP: Myositis Ossificans progressive; MOT: Myositis Ossificans Traumatica; CT: Computed Tomography; MRI: Magnetic Resonance Imaging; NSAID's: Non-Steroidal Anti-Inflammatory Drugs; HO: Heterotopic ossification; Gy: Grays; 3D-CRT: Three Dimensional Conformal Radiotherapy; IMRT: Intensity Modulated Radiotherapy

Introduction

MO is a self-limiting benign disease characterized by heterotopic bone formation in extraskeletal sites such as muscle, ligaments, tendons and nerves. This disease was described for the first time in 1918 by Dejerine and Ceillier with the term Myositis Ossificans (MO). More recently three categories of HO have been described: MO Progressiva (MOP), MO Traumatica (MOT) and MO circumscripta without trauma [1]. These older terms for Heterotopic Ossification have been replaced, but the term Myositis Ossificans and Heterotopic Ossification are interchangeable. MOP, also known as fibrodysplasia ossificans progressiva, is a rare hereditary autosomal dominant disease with multiple heterotopic ossifications in soft tissues associated with skeletal malformations that usually affect young people, leading to a progressive and functional limitation. MOT is the more common presentation and it seems related to repeated injuries, traumatism and medical or surgical procedures [2]. A non-traumatic type of MO also exist because a traumatic event is not always clearly described. RT seems to have a therapeutic effect in this disease and in literature there is evidence that suggest the efficacy of RT in the prevention of HO [3]. Although there is no general consensus about nonsurgical therapy for MO and there are only few articles in the literature about conservative treatment by using RT [4] .Therefore, the purpose of this article is to present a case of MO of left humerus successfully treated with radiotherapy and to review the literature of MO.

Case Presentation

A 42-year-old Caucasian woman was referred to our Radiotherapy Unit on June 2016 complaining of severe left shoulder pain of three months evolution with a progressive limitation in arm movements. She was on corticosteroids (Methylprednisolone 60 mg per day orally) and Tramadol 37,5 mg/Paracetamol 325 mg with partially pain relief. Clinical examination revealed a palpable, not well delimited painful mass with associated edema in left scapula spine and in posterior aspect of proximal humerus, without skin lesions, neither hematoma or distal neurovascular compromise. Shoulder's range of motion was limited because of severe pain with objective weakness on her left arm without anesthesia or paresthesia. Serum laboratory tests and complete hemogram values were within normal ranges. Radiographs of the affected shoulder showed a soft-tissue flocculent calcified mass extending laterally and posteriorly around the proximal humerus (Figure 1). CT scan demonstrated infraspinatus muscle swelling with low-attenuation and a not-well delimited calcification, making the diagnosis challenging and requiring additional evaluation with MRI. MRI revealed a flocculent mass in the thickness of the infraspinatus and teres minor muscle, slightly hyperintense on T1, with hyperintense foci in T2 and an irregular, heterogeneous peripheral line hypointense on T1 related with peripheral calcification. Moreover, anterior displacement of the humeral head with glenohumeral joint congruence lost and significant edema in supra and infraspinatus fossa was observed (Figure 2). Since MRI findings were nonspecific, patient's case was presented in Musculoskeletal Committee. Differential diagnosis included soft-tissue sarcoma and neuroendocrine tumor's metastasis; therefore biopsy of the mass was performed. Histopathological examination revealed peripheral and central lamellar bone with a definitive diagnosis of MO. Committee's decision was conservative management with radiotherapy. The patient received a total dose of 6 Grays (Gy) in six fractions with Three-Dimensional Conformal Radiotherapy (3D-CRT) on pathologic ossification with no side-effects (Figure 3). At 2 months follow-up

our patient reported almost complete relief of pain with partially restore of left arm movements. The MRI at 3-months follow-up, revealed a decrease of previous calcifications, less edema, improvement of glenohumeral joint congruence and a slightly reduction of the mass with intralesional necrosis suggesting response to the treatment (Figure 4). At one year follow-up the patient had complete pain relief and she was not taking analgesics, therefore she was not treated surgery and she follows-up regularly.

Discussion

Pathophysiology of MO seems to involve a deregulation of local mesenchymal stem cells in response to inflammation process secondary to a tissue injury, leading to a wrong differentiation of fibroblasts into osteogenic cells [5,6]. MO can occur in any age group; clinical presentation is usually characterized by pain and joint stiffness with a history of previous traumatism and the most common sites are flexor muscles of arm and extensor muscles of lower limbs. When neurovascular structures are compressed patients can also develop weakness and paresthesia. MO diagnosis is not always easy, especially when is not associated with a typical presentation and imaging findings can be nonspecific, miming other diseases such as tumors [7]. Diagnosis is based on history, physical examination and imaging such as radiographs, CT and MRI. MRI is the best technique to diagnose lesions of soft-tissue mass [8] and often can give the definitive diagnosis but sometimes biopsy is needed to confirm the nature of indeterminate lesions in order to rule out other diagnostics, especially malignancies. Differential diagnosis include osteosarcoma and soft-tissue sarcoma and benign diseases such as abscess or periosteal reaction [9,10]. Histopathology of a mature MO usually shows mature lamellar peripheral bone with proliferating fibroblasts in the central area but in early stage it may be difficult to differentiate MO from sarcoma due to the presence of immature and undifferentiated mixed tissue [6]. A multidisciplinary approach is helpful to make a correct diagnosis and decide the best therapeutic options since MO treatment is complex and need a careful evaluation to decide the better approach. Conservative treatments are focused on improve function and reduce the pain ad are usually successful also because MO is a self-limiting disease. Rehabilitation therapy, NSAIDs and cryotherapy help to attenuate patient's symptoms. Surgery is usually performed when conservative treatments failed, especially for patients with intractable pain, neurovascular structures compression and loss of motion that compromise normal activities [1].

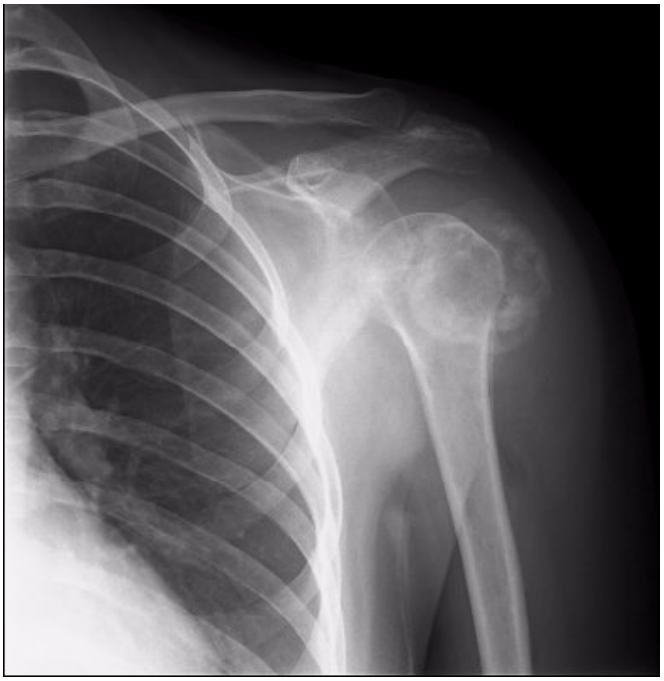
Surgery alone is associated with high incidence of recurrence. Therefore, surgical excision is combined with preventive treatments such as NDAIDs, which reduce MO recurrence in 30-45% at the expense of increase in gastrointestinal adverse effects and can determine therapeutic failure [3,11]. Historically radiotherapy has been used not only for cancer treatment but also for benign diseases and benign tumor with low doses prescription. Techniques such as Intensity Modulated Radiotherapy (IMRT) achieve a better dose conformation and low radiation doses are relatively safe, therefore radiotherapy can be offered as effective treatment in selected patients [12]. The rationale is that mesenchymal stem cells are highly radiosensitive13 and the use of RT can prevent the proliferation of pluripotent mesenchymal cells that may differentiate into bone-forming cells. In literature, RT has been studied especially for the prevention of HO in high-risk patients following hip surgery. A meta-analysis reported that postoperative RT is more effective NSAIDs in HO prevention after major hip procedures

and another multicenter study revealed the effectiveness of prophylactic RT in preventing HO in hip surgery[11]. Although, there are only few cases in literature about the use of RT for MO treatment and the most effective doses, delivering schedule and timing is not well established [4] . A study revealed that a single irradiation of 7 Gy can be given in patients that develop MO after a previous surgery or when indomethacin was contraindicated [14]. Moreover, another experimental study with rats founds that alternative fractionated RT schemes of 5 Gy in 2 fractions, compared to a single doses of 7 Gy, may be associated with a better suppression of bone formation and authors recommended fractionated irradiation to reduce radiation side effects [15]. A multidisciplinary team approach is useful for the diagnosis and treatment of MO and one therapeutic option is low dose radiotherapy that alone or combined with other therapeutic strategies could reduce pain and improve patient's symptoms without severe side effects.

Conclusion

MO diagnosis can be challenging and requires a multidisciplinary team approach in order to make a correct diagnosis and decide on the best treatment. Literature data are focused especially on heterotopic ossification prevention and suggest that RT is effective in this clinical situation but there are limited data about its treatment with RT and no consensus about the total doses and fractionation, with a relatively short follow-up. Therefore, our case report supports that RT is an effective and relative safe alternative, with good symptomatic control compared to NSAIDs and less aggressive than surgery, so it must be considered in the approach and management of MO. In conclusion, low dose RT alone or combined with other therapeutic strategies can reduce pain and improve patient's symptoms without severe side effects and we strongly encourage its use in symptomatic patients not candidates for surgery with poor symptoms control. Further studies are required to confirm these findings in a larger number of patients with a longer follow-up, to better evaluate the role of RT in MO.

Figures



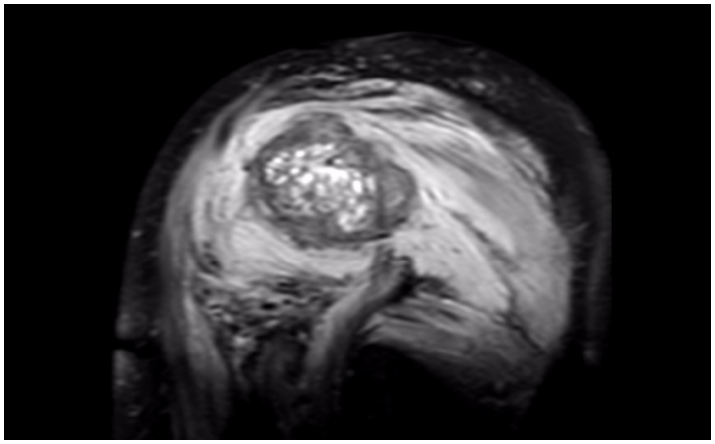


Figure 2: MRI T2-weighted image of left humerus. It shows hyperintense foci with significant muscle edema.

Figure 1: Anteroposterior radiography of left humerus. It shows a calcified mass around the proximal humerus.

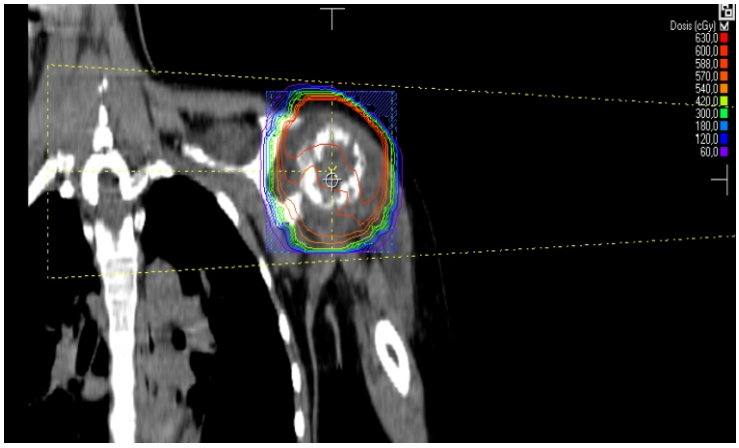


Figure 3: Radiotherapy planning. It shows delivering radiation dose of 6 Gy.

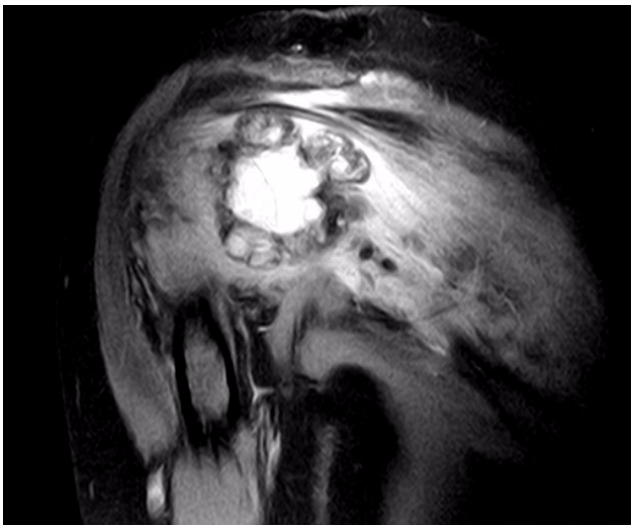


Figure 4: MRI T2-weighted image of left humerus at 3 months follow-up.

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