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Case Report

A rare repeated recurrence of myositis ossificans in the lateral right knee

Rui-Lan Lin, Zhen-Jiang Liu, Li-Jun Zhang

Department of Pediatric Orthopedics, The Shengjing Hospital, China Medical University, Heping District, Shenyang, Liaoning Province, People's Republic of China

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Abstract: Myositis ossificans (MO) is a benign condition characterized by abnormal heterotopic bone formation, typically involving the striated muscle and soft tissue. MO has an excellent prognosis but may appear clinically and radiologically as a malignant neoplasm. Sometimes the diagnosis of MO is very difficult to establish, even delayed. In the particular cases, when the calcific lesion appears in an atypical location in the body, MO is more likely to be misdiagnosed. A rare case of a MO in the lateral right knee was reported in this article, and surgical resection of the mass was performed. The final pathological diagnosis confirmed MO. Five months after the resective surgery a recurrence of MO took place and the patient underwent another surgical resection of the lesion. Three months later, the lesion reappeared for the third time in the same location. After a seven-year-four-month follow-up, the latest plain radiographs showed the MO to be still present in the lateral right knee. The range of motion of the affected knee and the neurologic examination of this patient were both normal. The cause of the multiple recurrence of MO was unknown. The biopsy of MO is the key factor to avoiding misdiagnosis.

Keywords: Myositis ossificans, knee, treatment, recurrence

Introduction

Myositis ossificans (MO) refers to the formation of ossifications in the muscles, ligaments and fascias [1, 2]. It is typically described as a reactive, self-limiting process characterized by prominent heterotopic ossification [3, 4]. MO is not malignant [1, 2]. When it happens in rare part, MO is more likely to be misdiagnosed [5, 6]. A rare case of MO in the lateral right knee right knee was reported in this article, and twice surgeries were performed, however, the new mass reappeared repeatedly.

Case report

On March 8, 2005, a 13-year-old boy presented for evaluation of a mass in the right lateral knee with mild pain over the past 10 days. The mass was first noticed 45 days prior to this presentation and increased gradually since then. Physical examination revealed a 6 cm × 10 cm, hard and painful mass in the lateral right knee, attached to deeper soft tissues. The pain was not severe as to disturb sleep or hinder physical activities but worsened after muscular contraction and mobilization of neighboring joints. There were no motor deficits of the knee and joint amplitudes were normal. There were no neurologic symptoms. Plain radiographs initially revealed an irregular shape, unevenly dense calcified mass in the lateral right knee with a cloud-like calcification shadow around the bone cortex and unclear boundaries, which occured in the soft-tissues without any involvement of the surrounding bone (**Figure 1**). Computed Tomography (CT) showed a neoplasm located in the lateral right knee, with the center remaining isodense to normal muscle, and the periphery developing a calcified rim of variable thickness (**Figure 2A**, **2B**). Initial magnetic resonance imaging (MRI) revealed a heterogeneous soft-tissue signal in the lateral right knee. The lesion displayed an isointense signal to normal muscle on the horizontal and coronary view T1-weighted images (**Figure 3A**, **3B**). MRI showed hyperintense signal to normal muscle on the coronary view T2-weighted image (**Figure 3C**). The gadolinium enhanced MRI showed a

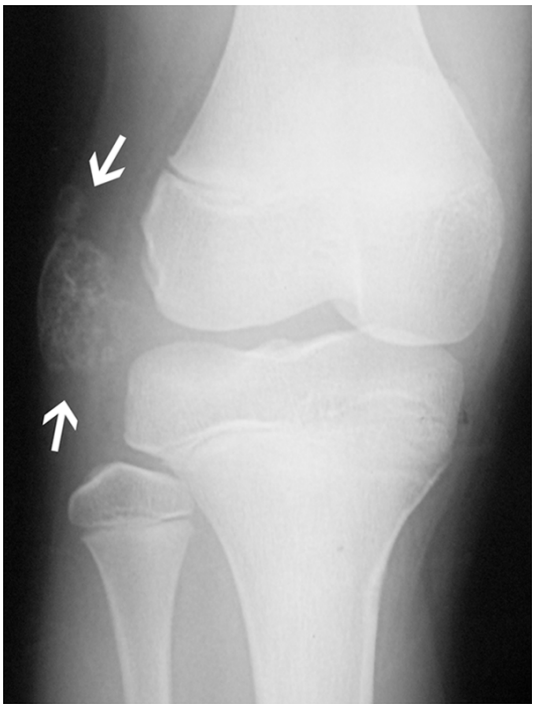

Figure 1. The initial plain radiograph of right knee. Anteroposterior view of plain radiogragh revealed an irregular shape, uneven density calcified mass (*arrows*) in the right lateral knee, a cloud-like calcification shadow around the bone cortex, the boundary was not clear, it occured in the soft-tissues without any involvement of the surrounding bone.

hyperintense mass to normal muscle mixed with some hypointense signals on the horizontal view T1-weighted image (**Figure 3D**).

On March 14, 2005, the patient underwent a surgical resection of the mass (**Figure 4**). The final pathological diagnosis was MO. Unfortunately, there was a recurrence of this MO five months later. The parents of the patient did not hesitate to agree with a second surgical resection of the lesion. However, a new recurrence of this MO happened again three months later. The patient's parents could not have their son endure another surgery and refused the third resection of the mass. Preoperative or postoperative administration of NSAIDs or radiation therapy was never used. On July 17th, 2012, the latest seven-year-four-month follow-up, the patient returned to the hospital for a routine review. The physical examination showed a firm mass in the lateral right knee and a normal range of motion of the knee. There were no neurologic deficits. The plain radiographs revealed that MO was still present in the lateral right knee (**Figure 5**). From July 2012 up to December 2015, the doctor called the patient's parents annually to schedule a clinical and radiographic follow-up appointment but the patient's mother always refused. However on December 15, 2015, the patient's mother called the doctor to say that the mass was still in the lateral right knee.

Discussion

MO predominantly affects adolescents and active adults [4, 7]. It can be located throughout the body, especially in thighs, buttocks or elbows after muscles repeated microtrauma [1, 2, 8]. The etiology of MO remains unclear [9, 10]. MO is related with the injuries resulting from hematomas, passive movements, burns, infections, or illicit drug abuse [1, 5, 6, 11]. The presentation of MO is variable. According to the course of time, MO can be divided into three periods: the early or acute stage (from 2 weeks to 4 weeks), the subacute or intermediate stage (from 4 weeks to 6 months) and the mature stage (6 months or more) [12]. The present case was in the intermediate stage before the first surgical resection.

The diagnosis of MO is clinical and mainly radiological [12]. The classic symptoms are pain, joint stiffness, paresthesia, weakness, lymphedema and venous thrombus [13, 14]. Symptoms often abate as the lesion matures [13]. When MO happens in rare locations, the clinical diagnosis may be more difficult and often raises concern for malignancy [13].

Plain radiographs reveal a soft tissue mass with faint peripheral calcification visible. Further, the most peripheral calcification becomes coarser and denser and lesions gradually become mature [12]. However, The ossifications are often missed on radiographs when these are performed 2-3 weeks after MO onset [9]. Moreover, mature lesions are sometimes adherent to the adjacent bone, and differentiation from parosteal osteosarcoma may thus require CT or MRI [13]. CT scan optimally demonstrates the cross sectional zonal pattern, with excellent definition of both soft tissues and calcification [12]. In the intermediate stage of MO, CT shows a well defined lesion with a thick peripheral calcific rim [15]. A typical zonal phenomenon is noted [12]. MRI is the best single modality for imaging soft-tissue mass-

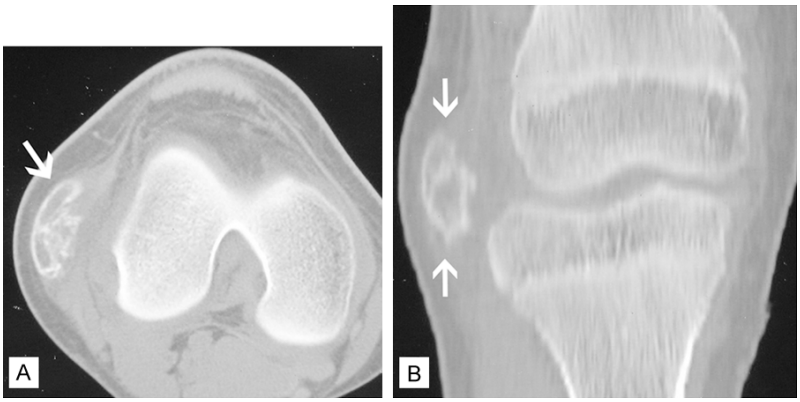

Figure 2. The preoperative CT. On horizontal (A) and coronary (B) view, CT showed a mass (*arrows*) located in the right knee lateral, with the center remaining isodense to normal muscle, and the periphery developing a calcified rim of variable thickness.

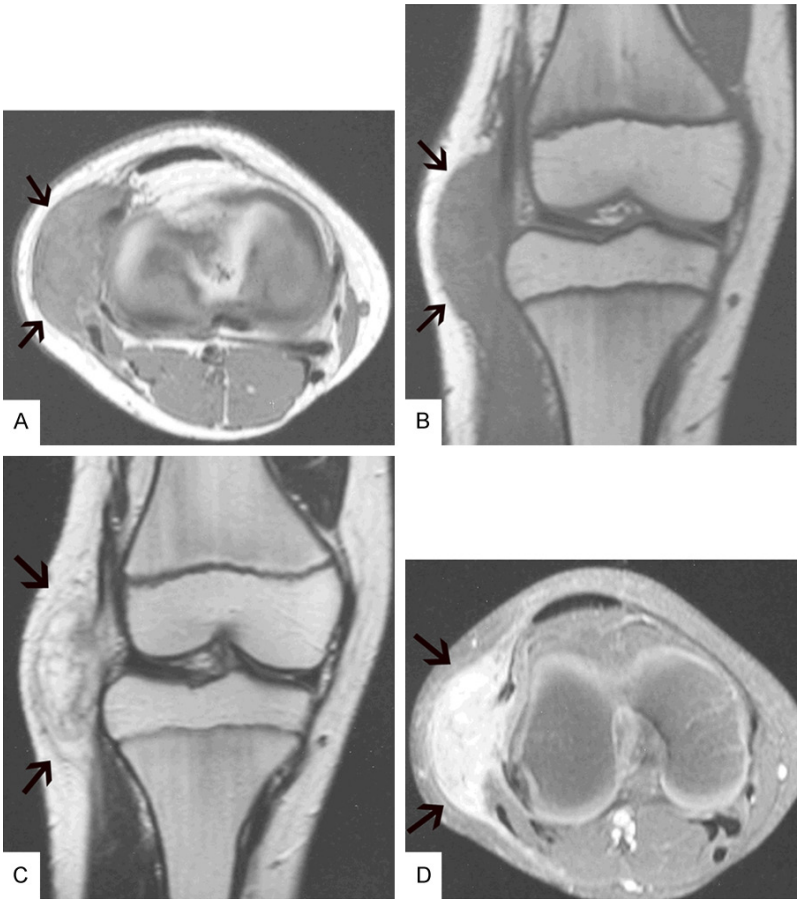

Figure 3. Preoperative MRI. MRI revealed a heterogeneous soft-tissue signal in the right lateral knee. The lesion was isointense signal to normal muscle on the horizontal (A) and coronary view (B) T1-weighted images (*arrows*). MRI showed a hyperintense signal to normal muscle on the coronary view T2-weighted image (C) (*arrows*). The gadolinium enhanced MRI showed a hyperintense signal to normal muscle mixed with some hypointense signal on the horizontal view T1-weighted image (D) (*arrows*).

es [13]. In the intermediate stage, MO demonstrates a variable appearance on MRI. The center is isointense or hyperintense to normal muscle on T1-weighted images. On T2-weighted images, lesions tend to be inhomogeneous, with a variable, but predominantly high signal center, and irregular focal areas of intralesional decreased signal intensity [12, 15].

A biopsy is required for the final diagnosis and is the key factor avoiding of misdiagnosis [5, 6, 16]. But if premature biopsy is done at the early stage of MO, it may lead to a misdiagnosis of sarcoma, or if biopsy is delayed, a true sarcoma may be missed [9].

MO is a self-limiting condition that may eventually regress spontaneously [12]. This process can take up to 6 months or more [13]. Treatment is mostly conservative because of the characteristic of spontaneous regression [15]. Lesion should be managed conservatively with compressive dressing, rest, avoidance of additional injury. Drug therapy includes the use of nonsteroidal anti-inflammatory drugs (NSAIDs), such as indomethacin and bisphosphonates, to relieve pain [11]. However, Tyler et al. [12] thought there was currently no effective treatment to prevent or halt progression of MO lesions. Radiation therapy is associated with fewer side effects and is preferred [17], and it is com-

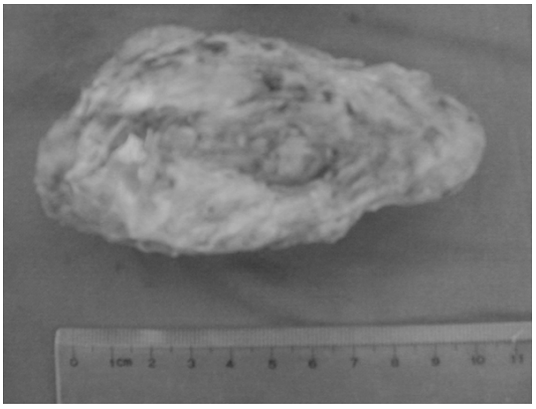

Figure 4. The clinical appearance of the removed mass. The size was about 10 cm × 6 cm.

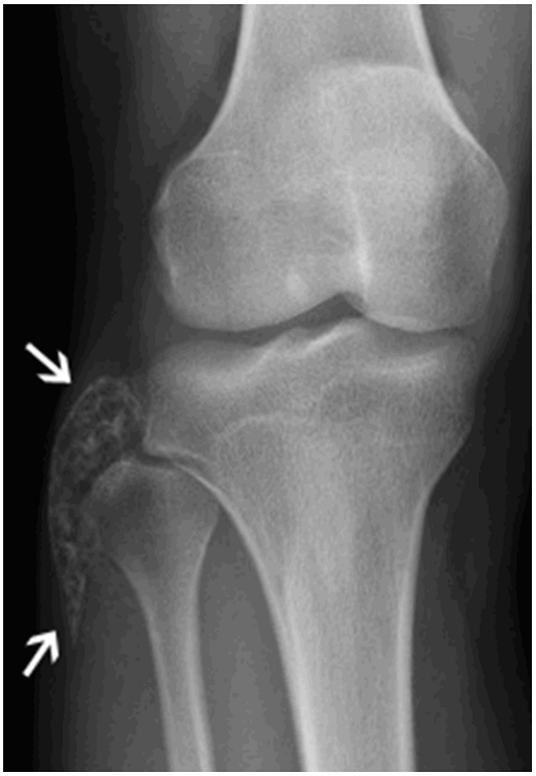

Figure 5. The latest plain radiograph of right knee. The mass was still in the lateral right knee but migrated to a lower position than the initial one.

monly used to prevent MO [18]. Generally, surgical intervention is recommended if MO interferes with joint motion, functional limitation or if it generates a significant pain due to the compression of nerve trunk [19] or if the symptoms do not improve after the conservative treatment [20] and the diagnosis is uncertain [21].

Recurrence of MO following excision may happen [13, 22] and is extremely rare [22]. In the present case, the rapid recurrences were remarkable, and the cause of the recurrence was unknown. Generally, the optimal timing of surgery is when lesion has reached maturity [13, 15, 20, 21, 23, 24]. The removal of immature lesion may result in local recurrence and possibly progress [13, 15, 20, 21, 23]. MO in this case was more than six weeks old therefore was in the intermediate stage. The resection of the immature lesion may be the primary cause of repeated postoperative recurrence. Sodemann et al. [25] reported that treatment with NSAIDs following resection of periarticular MO could prevent recurrence. Pape et al. [26] and Biering-Sørensen et al. [27] reported that administration of indomethacin was a useful adjunct to reduce the incidence of recurrent ossification. Preoperative or postoperative administration of NSAIDs had never been used in the present case, and could be the second cause of recurrence. Some reports showed the preoperative or postoperative radiation therapy especially low-dose radiation therapy [28] was safe and efficacious in prevention of MO recurrence [18, 29-33]. Ayers et al. [28] reported radiation therapy was the only treatment used to prevent MO that was delivered locally and not systemically. Radiation therapy had never been used in the present case and could be the third cause of recurrence. The present MO has at least existed for seven-year-four-month or more postoperatively. This MO is very different from the previous reports [10, 11, 15, 16, 19, 20, 24] because of the repeated postoperative recurrence and long-term existence. Another interesting phenomenon was noticed. Although the recurrent lesion was located in the lateral right knee, the lesion had migrated to a lower position than the initial one. The definite cause of this phenomenon is unknown. The stimulation from surgical resection may be responsible for the phenomenon.

This rare case, to the best of our knowledge, is the first reported case of repeated recurrence of MO in the lateral right knee. The resection of immature lesion, not using NSAIDs or radiation therapy could be the cause of the multiple recurrence of MO in this case.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Zhen-Jiang Liu, Department of Pediatric Orthopedics, The Shengjing Hospital, China Medical University, Heping District, Shenyang, Liaoning Province, People's Republic of China. Tel: +86 024-96615-57511; E-mail: lzjsdd@163.com

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