
CASE REPORT

Extensive Myositis Ossificans with Extra-articular Ankylosis of the Hip Joint

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ABSTRACT

Myositis ossificans, also known as heterotrophic ossification, is characterised pathologically by the presence of lamellar bone at sites where bone normally does not exist. Its aetiology is unknown but is known to occur following damage to the muscle. The resultant proliferative connective tissue later differentiates into the mature bone mass. Extensive myositis ossificans leading to a complete extra-articular bony ankylosis is rare. We report a case of post-traumatic myositis ossificans in a 27-year-old female following a head injury. The iliopsoas was involved in this case, and lead to extra-articular bony ankylosis of the hip causing severe functional impairment. Surgical excision of the mass as described resulted in a marked functional improvement.

Key Words: Craniocerebral trauma; Myositis ossificans

中文摘要

大範圍骨化性肌炎伴發髖關節外骨性僵硬

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骨化性肌炎又稱為異位性骨化症，從病理學的角度來說，這種病是指在不應該長有骨的組織內出現了板狀骨。發病原因目前仍然不明確，但一般在肌肉受損後才發生。增生性結締組織分化至成熟的骨質。因大範圍骨化性肌炎而引致關節外的骨性僵直很罕見。本文報告一名27歲女性頭部受傷後引發創傷後骨化性肌炎。這病例牽涉髂腰肌而導致髖關節外的骨性僵直。病人髖關節活動度變小而導致功能嚴重受損。患者接受上述骨質切除手術後功能活動明顯恢復。

INTRODUCTION

Myositis ossificans was first reported in 1749¹ and was considered to be of unknown aetiology. It has been classified later into three types: myositis ossificans circumscripta, myositis ossificans progressiva, and myositis ossificans traumatica. Post-traumatic myositis

ossificans is the commonest, and usually presents as well-circumscribed lesion complicating a muscle haematoma. It is frequently seen following central nervous system disorders (brain injuries, tumours, encephalitis, spinal cord injury), multiple injuries, hip surgery, and burns.

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We report a case of post-traumatic myositis ossificans involving the iliopsoas muscle leading to extra-articular bony ankylosis of hip. To the best of our knowledge, our patient had the largest size of myositis ossificans ever reported (approximately 25 cm in length), and presented with almost complete ankylosis of the hip joint.

CASE REPORT

A 27-year-old woman presented with significant restriction of movements at the left hip joint. The patient had sustained a head injury following a motor vehicle accident four years earlier, at which time she had been in a comatose state for a month. Subsequently, she developed stiffness in the left hip joint with an enlarging mass. She was unable to squat or sit cross-legged.

On examination, the limb was in an attitude of fixed flexion and external rotation. Palpation revealed a well-defined bony swelling in the Scarpa's triangle that extended above the inguinal ligament. The swelling was non-tender, the local temperature was not raised, and it was fixed to the underlying bone. The hip was ankylosed in 30 degrees of flexion, 15 degrees of external rotation, and 20 degrees of abduction. The ipsilateral knee also had a postural flexion deformity of 20 degrees.

The hip radiograph showed a bony mass consisting of a mature bone resembling a bar. The bony bar was extended from iliac region to anterior aspect of femur up to the level of the lesser trochanter, and had caused a fixed deformity at the left hip (Figure 1). There was no bone destruction or periosteal reaction, and no associated soft issue mass was evident. The patient's alkaline phosphatase level was normal and other haematological investigations were unremarkable.

Three-dimensional (3D) reconstruction computed tomographic images showed the complete extent of the mass (Figure 2). Excision of the bony mass was performed using an extended Smith Peterson approach. The mass was detached from iliac bone using a sharp osteotome, and then exposed through to its distal attachment at the lesser trochanter, before being detached. The excised mass measured 19.4 cm in length. Complete excision of the mass resulted in a full range of hip movement, intraoperatively. Histopathology confirmed the diagnosis of myositis



Figure 1. (a) Anteroposterior radiograph of left hip showing extensive bony mass (arrow) extending from ilium to the lesser trochanter leading to ankylosis of the hip joint. There is no bone destruction or periosteal reaction. No associated soft issue mass is evident. (b) Lateral radiograph of left hip shows the similar bony mass (arrow).

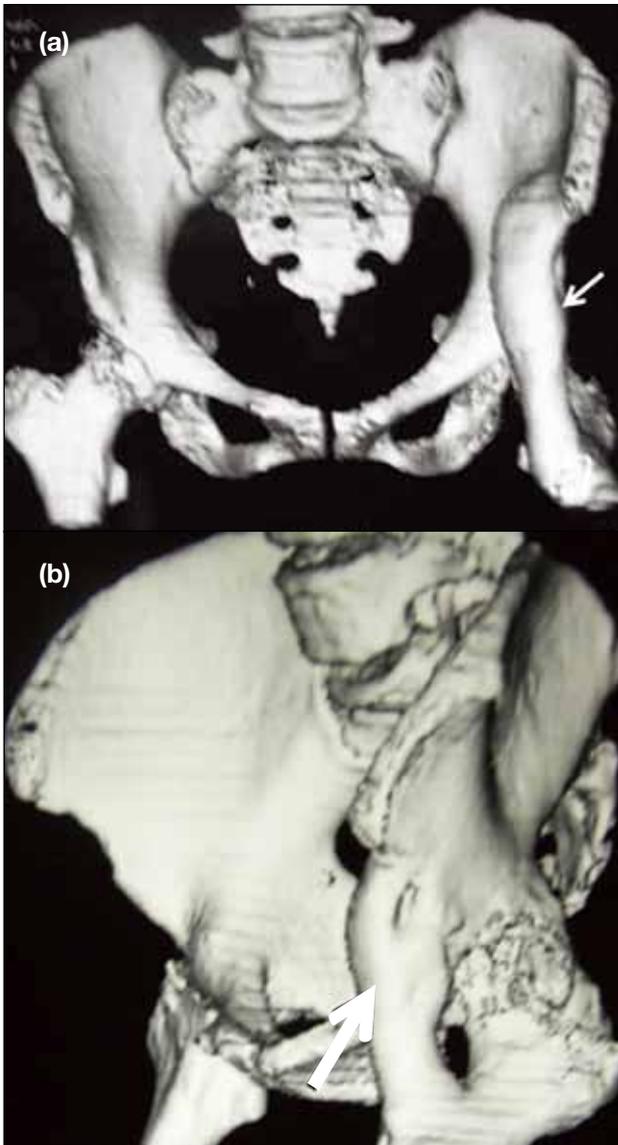


Figure 2. (a) Coronal, and (b) oblique three-dimensional reconstruction computed tomographic images showing the complete extent of the mass (arrows). These images also help in the preoperative planning of surgical intervention.

ossificans. Skin traction was applied to the left lower limb and intermittent hip range of movement exercises were encouraged. The patient was treated with anti-inflammatory drugs for six weeks. After two months, the patient had 90 degrees of flexion, 30 degrees of abduction-adduction, and 20 degrees of external and internal rotations. The range of hip movement at the two-month follow-up was adequate for performing activities of daily living. The follow-up imaging showed no further recurrence (Figure 3).

DISCUSSION

Following head injury or spinal cord injury, post-

traumatic heterotopic ossification most commonly occurs at the hip joint.² Though believed to be of unknown aetiology, myositis ossificans is known to occur after damage to muscle, which results in proliferation of the connective tissue and differentiation into the mature bone. Chalmers et al³ described three conditions necessary for its formation:

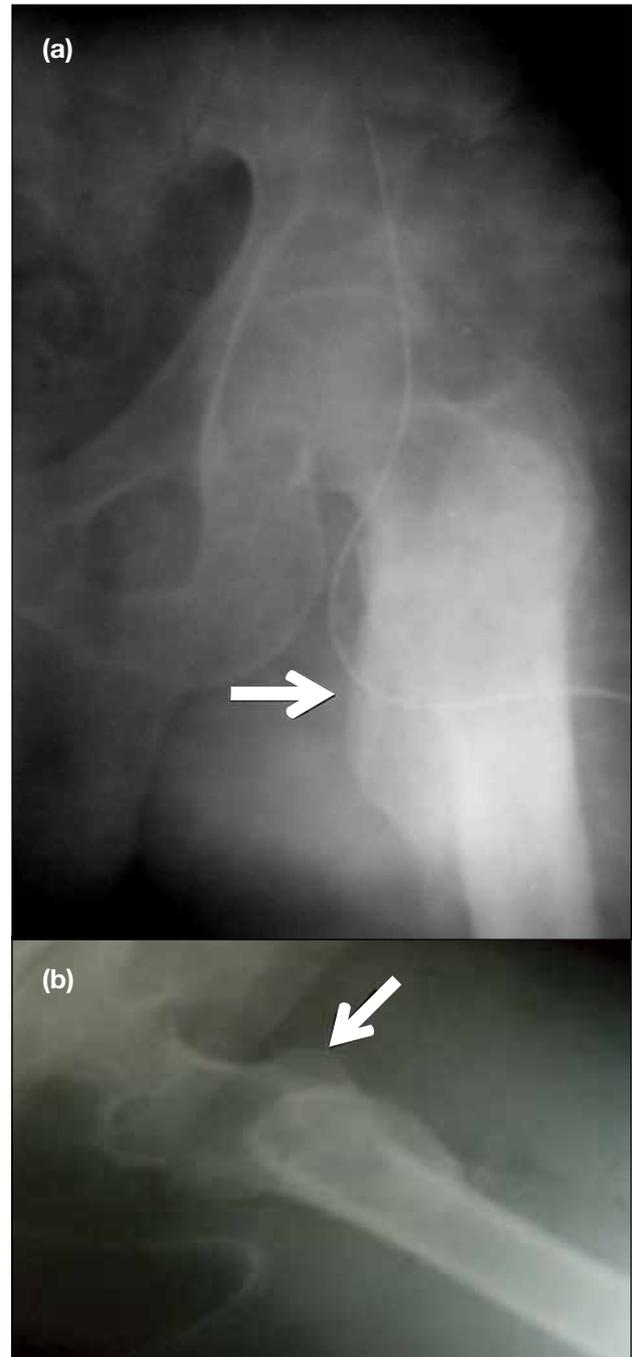


Figure 3. Follow-up radiographs: (a) anteroposterior and (b) lateral views after 6 weeks of operation showing the unremarkable left hip with no evidence of recurrence or residual bony mass. The left lesser trochanter (insertion site of the psoas muscle) has been shown by the arrows.

presence of (1) osteogenic precursor cells, (2) inducing agents, and (3) a permissive environment. It is believed that head injury provides a permissive environment that permits inducing agents like BMP 4 to act on osteogenic precursor cells released as a result of muscle trauma and periosteal stripping. The latter cells may then differentiate into osteoblasts. In a large series described by Auerbach et al,⁴ there were 115 cases with sizes ranging from 1.0 to 20.0 cm.

Usually, the initial presentation of the myositis ossificans entails pain, tenderness, and local muscle atrophy. Rarely it can be asymptomatic and may be diagnosed after radiography for other problems. In 10 to 20% of patients, it presents with functionally significant restriction of the range of movement at a specific joint.⁵ Plain radiographs show a floccular calcified density in the early stages (2-6 weeks from the onset), and over time these mature into sharply circumscribed areas of calcification. In half of all instances, myositis ossificans has a certain degree of adherence to periosteum. Radiopathological differentiation from bone malignancy (mainly osteogenic sarcoma) may be difficult at times but is often crucial and mandatory in view of differing prognoses and management strategies. A lucent zone between the lesion and adjacent bone, an intact underlying cortex, a diaphyseal location, dense calcification in the periphery, and loss of volume on serial films are the key radiological features favouring a diagnosis of non-neoplastic heterotopic bone formation (as in myositis ossificans). In contrast, associated bone destruction, an aggressive periosteal reaction, and a soft tissue mass are features of bone malignancies. They were all absent in our patient. Reconstructed 3D images of the lesion are useful in determining the true extent of the mass and planning the surgical approach.

Histopathologically, myositis ossificans displays three well-distinct zones: a central undifferentiated zone, a surrounding zone of immature osteoid, and a peripheral zone of mature osteoid. In contrast to osteogenic sarcoma, myositis ossificans exhibits a zonal pattern, viable muscle fibres in the lesion, and does not invade surrounding tissues.

Surgery is warranted when the lesion mechanically interferes with joint movement or causes neurovascular compromise. The patient in this report was unique, because of its large size and presentation with bony extra-articular ankylosis of the hip joint. The Smith Peterson approach we used allowed an extensive exposure of the proximal and distal extent of the lesion and permitted complete excision of the mass. Following its surgical excision, the patient benefited by regaining a functional range of hip joint motion with the ability to squat and sit cross-legged.

CONCLUSION

Extensive myositis ossificans leading to complete extra-articular ankylosis is rare. Surgical excision of the mass results in a significant functional improvement.

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