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Case report

Myositis ossificans of the hip joint causing sciatic nerve palsy following Guillain-barré syndrome: A case report

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ABSTRACT

Introduction and importance: Myositis ossificans of the hip is a rare entity caused by trauma and neurological conditions which lead to lamellar bone formation around the joint.
Case presentation: We present a 47 years old patient with myositis ossificans of the left and the piriformis muscle following Guillain Barré syndrome causing sciatic nerve palsy (Piriformis syndrome).
Clinical discussion: Clinical assessment revealed global limitation of the left hip movements and palpable bony mass on the lateral aspect of the left hip. Radiographs and computed tomography showed extensive myositis ossificans of the left hip and non-bridging calcification noted on the asymptomatic right side. During initial medical management and physiotherapy patient developed sciatic nerve palsy due to piriformis syndrome and ankylosis of the hip on the left side. Surgical exploration of the sciatic nerve and debridement of the hip. The left hip was found ankylosed and not salvageable. Uncemented total hip arthroplasty was carried out in the session. The patient recovered completely from sciatic nerve palsy and regained the range of motion of the hip. He is under follow-up for the recurrence of myositis ossificans.
Conclusion: Guillain-barré syndrome causing piriformis syndrome is a rare entity. Extensive myositis ossificans causing sciatic nerve palsy is even rarer. An awareness of this entity, early detection, and intervention of this condition may help to preserve the native hip.

1. Introduction

Myositis ossificans, a self-limiting pseudotumor condition, that results in new lamellar bone formation outside the skeletal system mostly in skeletal muscles and soft tissues near the joints due to unknown reasons [1].

The incidence of Myositis ossification is around 10–20% in the world and 10% of them are clinically significant with massive and cause restriction of joint movements or ankylosis [2]. It is seen commonly after traumatic and neurological injuries. The incidence of Myositis ossification in Guillain Barre Syndrome has been reported in the literature and it is a rare association. Likewise, myositis ossificans causing piriformis syndrome are also rare. We have reported a Sri Lankan experience in piriformis syndrome due to extensive myositis ossificans causing sciatic nerve palsy in a patient who had Guillain-Barré syndrome.

This case has been reported according to the SCARE guideline [3].

2. Case report

47 years old, farmer was referred from the neurology unit at Teaching Hospital Jaffna, Sri Lanka, with left inguinal region pain and global restriction of movements of the ipsilateral hip for two weeks. He also reported a palpable painless mass on his left hip. He denied any history of trauma or surgery in the recent past. On clinical assessment, he had a bony hard painless mass palpable in the lateral aspect of the left hip with mild global restriction of movements.

He also stated sudden onset of muscle weakness which commenced in both lower limbs and gradually ascend to bilateral upper limbs three months back. His medical records revealed that he had a flaccid type of paralysis which made him an intensive care admission and mechanical ventilation for three weeks. The diagnosis of Guillain-Barré Syndrome was made with clinical assessment, investigation, such as cerebrospinal fluid analysis, haematological evaluation, and imaging scan of the brain. He was treated with intravenous Immunoglobulin and Respiratory

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support during this phase of treatment.

The pelvic radiograph with both hips (Fig. 1) revealed circumferential calcification in the left hip with radiolucent centre and cleft suggestive of myositis ossificans.

The computed tomography demonstrated extensive myositis ossificans involving the left hip and mild non-bridging bone formation on the other side (Fig. 2).

The patient was managed initially conservatively with indomethacin and range of motion exercises. He was on regular follow-up at the Orthopaedic clinic at the same institution and planned for excision of the lesion and the hip joint debridement once the lesion matured fully or he developed ankylosis.

He had lost the follow-up, physiotherapy, and non-steroidal anti-inflammatory drugs for around ten months due to the COVID-19 pandemic and again presented with a left side foot drop and ankylosed hip. The sciatic nerve palsy was gradual in onset and it is involving both compounds of the sciatic nerve. The Nerve conduction study revealed an absent H wave on the left side.

Exploration of the left sciatic nerve and debridement of the hip joint were planned. The patient was explained about the surgery and possible complications especially bleeding and sciatic nerve injury apart from other complications. The surgery was performed by an experienced Orthopaedic surgeon (The guarantor of this study) at Teaching hospital -Jaffna (a Tertiary care hospital in this region), Sri Lanka. The patient was placed right lateral position and the hip approached through the Kocher-Langenbech approach.

The sciatic nerve found entrapped within the myositis ossificans (Fig. 3), released completely and the continuation of the nerve confirmed.

Following the complete release of the sciatic nerve, the hip joint was debrided. The extensive nature of the myositis ossificans and the cartilaginous damage caused by the debridement rendered the hip unsalvageable. An uncemented total hip arthroplasty (Zimmer CLS® stem, Trilogy® acetabular component, Longevity® crosslinked polyethylene liner and VerSys® Hip system femoral head) was performed. His postoperative period was uneventful, was encouraged for active range of motion exercises to prevent the recurrence of myositis, and was prescribed with Indomethacin. Post-operative radiograph ensured a good alignment of implants and complete excision of myositis (Fig. 4).

The patient was discharged with a foot drop splint, oral Cefuroxime 500 mg twice daily, Aspirin 150 mg daily, and Indomethacin 25 mg thrice daily. He followed up at the same clinic. Indomethacin and Aspirin were continued post-operatively for six weeks. Ultrasonic nerve stimulation, hip range motion exercises, and walking exercises have been provided in the physiotherapy department. Foot drop has improved (in weeks) and the patient is now pain-free and walking without support. He is under follow-up for recurrence of myositis.

The following videos describe the recovered nerve function following the intervention.

https://youtu.be/ofcwzf5OPqo

https://youtu.be/jswRCNUfRN8

3. Discussion

Many extrinsic causes for sciatic nerve compression have been described in the literature. Those are tumours, muscles, tendons, haematomas, abscesses, and endometriosis. The most frequent cause among them is piriformis syndrome [4]. The anatomical relationship between the sciatic nerve and piriformis renders the nerve suffers compression on certain occasions [5]. Aetiologies of piriformis syndrome are trauma, infection, spasm, anatomical variations of the muscles, dystonic conditions, pseudoaneurysm of the inferior gluteal artery [5], and heterotrophic ossification. Heterotrophic calcifications causing sciatic nerve compression are very rare. There are traumatic causes (Following penetrating injuries, Acetabular fractures, and dislocations), iatrogenic causes (Following extensive surgeries), mechanical causes (One case of ossification of the proximal part of the biceps femoris), idiopathic causes, and neurogenic causes (Spinal cord injuries and Guillain-barré syndrome) [4].

Guillain-Barre syndrome is a progressive acute flaccid paralysis inflammatory polyneuropathy followed by an infection usually. The patient develops progressive ascending and symmetrical muscle weakness and areflexia. Nearly 15% of the patient have neurological or functional deficits following recovery [6].

The pathophysiology of myositis ossificans is incompletely understood. Recent studies confirmed myositis ossificans as a benign, solitary, ossifying soft tissue mass typically occurring within skeletal muscles. The endothelial-mesenchymal plays a vital role in the pathophysiology of myositis ossificans. The local inflammatory reaction caused by the skeletal muscle injury releases cytokines such as bone morphogenic proteins 2 and 4, Transforming growth factors induces the endothelial – mesenchymal transition. In the presence of inflammatory mediators, these endothelial mesenchymal cells may differentiate into chondrocytes or osteoblasts. Osteoblasts and chondrocytes will then undergo endochondral bone formation in extra-skeletal tissues [1].

Inflammatory symptoms such as pain, swelling, erythema, and

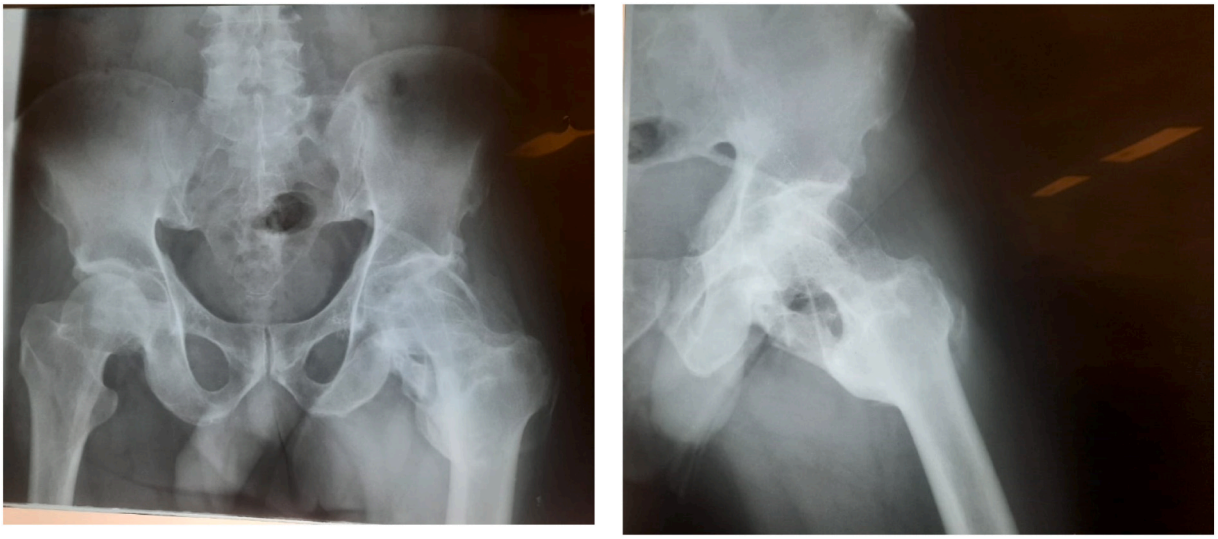

Fig. 1. Antero posterior view of pelvis and the hip shows circumferential bridging calcification of the left hip with radiolucent centre

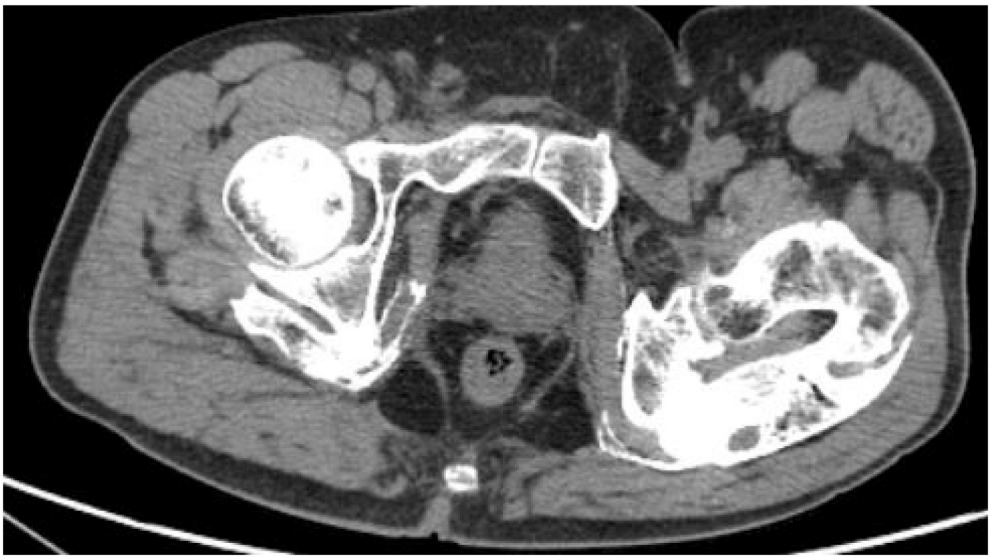

Fig. 2. Axial view of bilateral hips shows bridging calcification of left side and non-bridging bone formation on the other side.

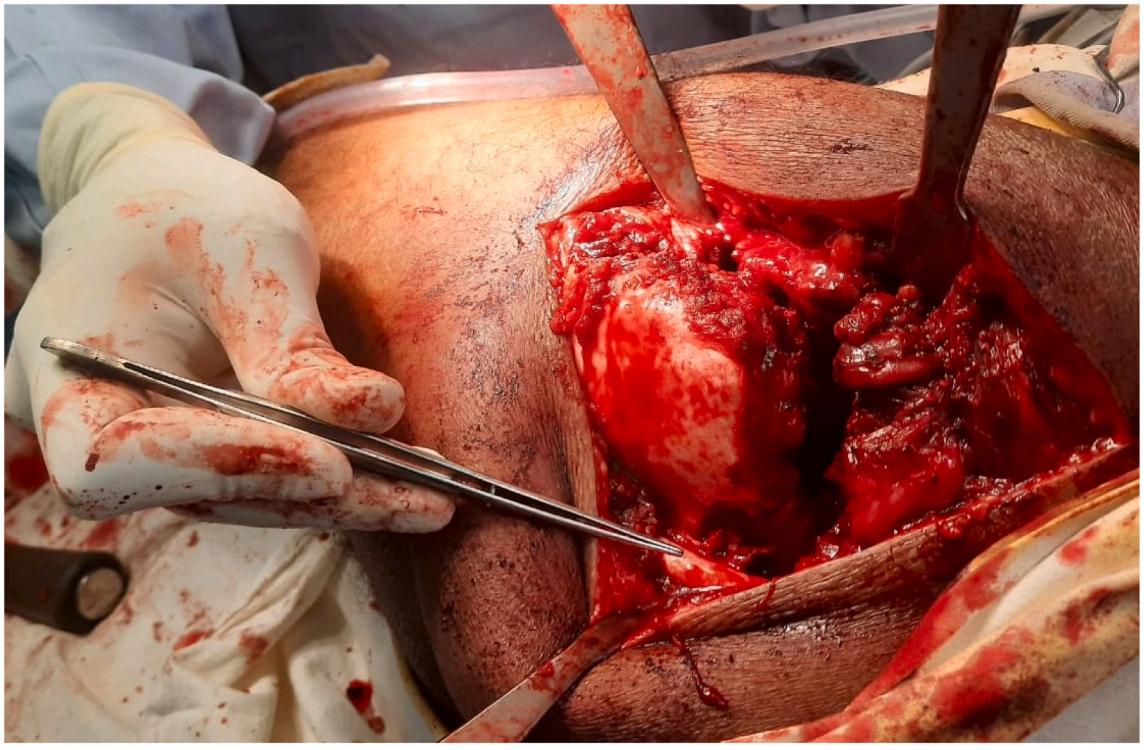

Fig. 3. The sciatic nerve entrapped within the Myositis ossificans.

warmness along with restriction of joint mobility will be presenting complaints that can appear anytime from three weeks to three months [7].

The risk factors for heterotopic ossification are any form of soft tissue trauma, neurological injury, joint arthroplasty, hypoxic conditions, immobilization for a prolonged duration, mechanical ventilation, hypermetabolic status. In our patient neurological injury, mechanical ventilation and prolonged immobilization are found to have as risk factors [8].

Brookers introduced a classification for myositis ossificans to classify the lesion around 1973 using an anteroposterior view of the pelvis [9].

Type 1

Formation of a bony island within the musculature around the hip.

Type 2

Presence of bony spurs from the pelvis or proximal end of the femur, separated at least 1 cm between them.

Type 3

Presence of bony spurs from the pelvis or proximal end of the femur, with less than 1 cm gap in-between.

Type 4

Apparent bony ankylosis of the hip.

The common sites of occurrence of myositis ossificans in decreasing order are hip, knee, shoulder, and elbow. The gold standard method for diagnosing heterotrophic ossification is mainly with plain radiographs

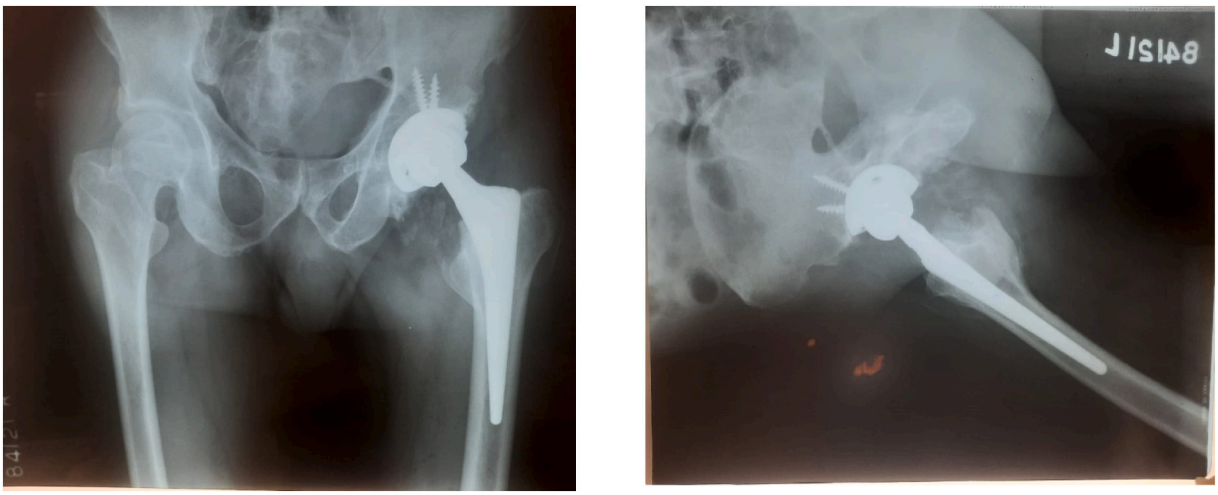

Fig. 4. Post-operative radiograph revealed good implant positions and complete excision of myositis ossificans.

and computed tomography [10]. Magnetic imaging will help to identify structures that are encircled by myositis.

Lab values of serum alkaline phosphate levels initially remain normal. It gradually increases with the new bone formation and reaches its peak when the patient is clinically symptomatic. In our patient serum, alkaline phosphate level was 247 IU/L, where the normal reference range in a healthy adult is 20–140 IU/L1.

In the initial phase of treatment, myositis ossificans well respond to conservative management as it is a self-limiting lesion. Indomethacin, a Non-steroidal anti-inflammatory drug, and bisphosphonates are commonly used as prophylaxis and its treatment. It interrupts the cascade of reactions by inhibiting prostaglandins-mediated (mainly through PGE-2) bone remodelling. Indomethacin also directly inhibiting the differentiation of osteoprogenitor cells. The effectiveness of indomethacin over other NSAIDs in the prevention and treatment of myositis ossificans has been proven in double-blind, randomized studies [11].

Bisphosphonate inhibits periods of mineralization by binding to calcium phosphate and by preventing hydroxyapatite crystallization. This therapy delays the formation of myositis ossificans but it is proven ineffective as prophylaxis treatment [11].

Single low-dose radiotherapy (700 Gy) in the treatment of myositis ossificans shows exemplary results. It is effective and safe with fewer observed serious adverse effects over long-term observation [12].

Surgical resection reserved for myositis ossificans significantly affecting the range of movements of the joint and entrapment syndrome [13]. In all cases, those have been described in the literature so far, neurolysis has been performed and the hip joint was preserved. Surgical treatment is rarely advocated and only after the myositis ossificans have fully matured [10]. It usually takes about a year. However, recent evidence emerging with the concept of early intervention [14]. In our case, the unsalvageable nature of the ankylosed hip joint with cartilage damage has turned us towards the replacement of the hip joint.

This case report revealed the possibility of this rare, delayed onset of sciatic nerve palsy. Even a slight delay in intervention may result in permanent damage to the nerve will results. Early detection not only saves the nerve but also may help to preserve the hip.

4. Conclusion

Myositis ossificans is a rare complication of Guillain-Barré Syndrome in which hips are affected more commonly. Hypo mobility with mechanical ventilation might be responsible for the development of myositis ossificans following Guillain Barre Syndrome in our patient. Early diagnosis of the progression and proper planning would have been allowed to preserve the native hip. A pre-operative magnetic resonance imaging would have been offered a good pre-operative idea of the site of the sciatic nerve compression. Anyhow in the experienced hand, the careful neurolysis and the hip replacement of an unsalvageable joint also give fairly good results.

Informed consent

Informed written consent was obtained from the patient for publication of the data and clinical images. A copy of the written consent is available for review by the Editor-In-Chief of this journal on request.

CRediT authorship contribution statement

Dishanth Sivakumaran, Kalaventhan Pathinathan, Joy Danicious, and Gobyshanger Thayasivam have equally contributed to the concept, design, data collection, and writing of this case report.

Declaration of competing interest

All authors disclose any financial and personal relationships with other people or organizations that could inappropriately influence their work.

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This case report is exempted from ethical approval.

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