

An Extremely Rare Case of Extensive Myositis Ossification: Complete Extra-Articular Ankylosis of Hip Joint



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ABSTRACT

Myositis ossificans, in its literal translation, is defined as an inflammatory ossification of muscle and the neighboring soft tissues. A rare case of post-brain injury ossification of the Gluteus maximus muscle extending from the greater trochanter of the femur to the right wing of the iliac bone, leading to extra-articular bony ankylosis of the hip is reported. A 40-year-old male presented to the Outpatient department with a stiff right hip and restriction of its movements. The patient had a history of cerebral hemorrhage 18 months back and subsequently developed stiffness in the right hip since the last 3 months. X-ray and CT scan revealed the extension of an ossified bar extending between the lateral aspect of the right iliac wing above the right acetabulum and upper 1/3 of the anterior lateral aspect of the femur near the greater trochanter, measuring 1003240 mm. The patient was then allowed to go for rehabilitation with appropriate physiotherapy and anti-inflammatory drugs. Extensive hypertrophic ossification can occur at a totally unrelated site as a consequence of cerebral insult/head injury.

Introduction

M

yositis ossificans (MO), in its literal translation, is defined as an inflammatory ossification of muscle and the neighboring soft tissues. In 1923, Lewis [1] classified MO into three forms: traumatic, nontraumatic, and neurotic. MO was further classified into

myositis (fibrous) ossificans progressiva, traumatic MO circumscripta, and MO circumscripta without a history of trauma. Most commonly, Myositis Ossificans is seen with Brain Injuries, spinal cord deficits, burns or polytrauma.

A rare case of post-brain injury ossification of the Gluteus maximus muscle extending from the greater trochanter of the femur to the right wing of the iliac

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bone, leading to extra-articular bony ankylosis of the hip is reported.

Case Presentation

A 40-year-old male presented to the Outpatient department with a stiff right hip and restriction of its movements. The patient had a history of cerebral hemorrhage 18 months back and subsequently developed stiffness in the right hip since the last 3 months. The patient also presented with an enlarging mass near the hip joint. On examination, the limb was fixed in 30 degrees flexion, 20 degrees of external rotation, and 15 degrees abduction. The ipsilateral knee also had a fixed flexion deformity of 20 degrees. The patient had weakness in bilateral upper and lower limbs and was unable to walk without support due to incoordination and cerebral deficits post brain insult. The swelling was non-tender, the local temperature was not raised, and it was fixed to the underlying bone. The patient was then screened with hip radiographs. The x-ray showed the presence of a growth of a bony ridge between the greater trochanter of the femur and iliac bone, leading to extra-articular ankylosis of the hip [Figure 1]. No accompanying periosteal reaction, zones of sclerosis, or any changes in the opposite hip could be identified. No similar mass was seen at any other site in the body.

The patient was then screened with 3D-CT and MRI scans. The 3D-CT scan revealed the extension of an

ossified bar extending between the lateral aspect of the right iliac wing above the right acetabulum and upper 1/3 of the anterior lateral aspect of the femur near the greater trochanter, measuring 100x32x40 mm [Figure 2]. MRI also revealed similar reports with the diagnosis of the appearance of a bar along the Right gluteal Maximus muscle and that it is situated superficial to the right hip joint capsule. The patient had normal serum alkaline phosphatase and S calcium, S. phosphorous, and S. LDH levels. Other hematological investigations were unremarkable. A true-cut biopsy was taken to rule out any tumor/other pathologies. The patient was then allowed to go for rehabilitation with appropriate physiotherapy and splints. Anti-inflammatory drugs and indomethacin were also added to stop further increase in heterotrophic ossification.

Discussion

Myositis Ossification is a well-differentiated ossification of soft tissues accompanying the bone, mainly involving the muscles. It can also be termed as Heterotrophic Ossification as here, the ossification extends the original boundaries of bone and can lead to various deformities and other complaints. Mainly three requirements are registered to develop myositis, including Inducing agents, Permissive environment, and the precursor cells (osteogenic). Trauma leads to the development of a hematoma that can lead to fibro and osteogenic activities. These, in turn, can lead to the generation of osteogenic precursor cells



Fig. 1. X-rays_Bony Bar showing extra-articular ankylosis of Right Hip

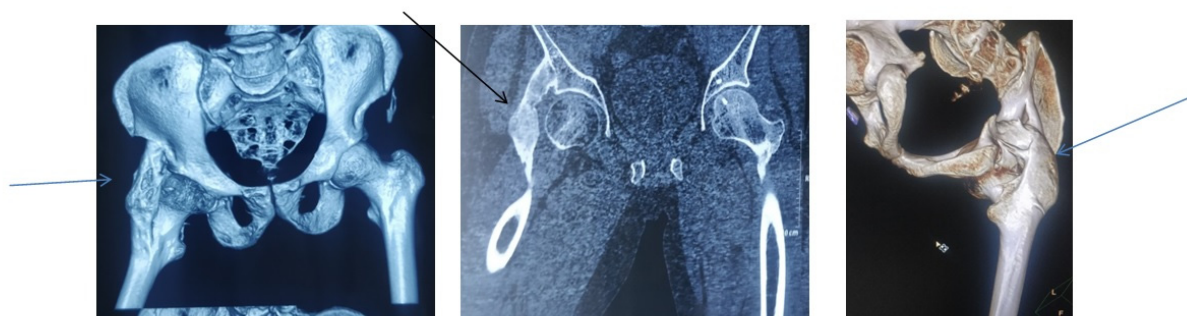


Fig. 2. 3D-CT sections showing extent of ankylosis anteriorly, Cut sections and posteriorly.

and improperly differentiated mesenchymal cells [2]. Bone Morphogenetic Protein (BMP-4) is released by the insult to the brain, as in head injuries that can also lead to the generation of osteogenic progenitor cells [3]. Histopathologically, myositis ossificans has 3 well-defined areas: a central undifferentiated area, a surrounding/middle area of immature osteoid, and a peripheral area of mature osteoid.

It is important to differentiate it from any bony malignancy. The presence of mass dense calcification in the lesion, with no bony destruction, no sclerosis within the lesion, and aggressive periosteal reaction points towards a benign heterotrophic ossified mass [4].

Very few cases of extra-articular ankylosis of the hip have been reported in the past [4,5]. But such extensive involvement of heterotrophic ossification has not been reported earlier along the gluteus maximus muscle distribution. The case in our study had a history of spontaneous cerebral hemorrhage, unlike other case reports where head injury due to trauma was reported.

Myositis Ossificans is mostly self-limiting and there are various reports suggesting it may lead to spontaneous regression. Surgical intervention is not the primary choice of treatment by most surgeons [6]. The most important indications for surgery are pain and neurovascular compromise by the ossified mass. This patient had none of these complaints. Some surgeons also warrant surgery for restriction of movements and stiffness. This patient had some stiffness and deformity of the hip joint but was not a surgical candidate due to residual cerebral injury weakness, including lack of coordination and spasticity of muscles. Also, there is a high incidence of recurrence reported even after excision of myositis ossificans masses. Conservative management and rehabilitation using appropriate physiotherapy and required braces were chosen.

Conclusion

An extremely rare case of extensive hypertrophic ossification of the hip leading to complete extra-

articular ankylosis of the hip is reported. The patient had a history of cerebral insult 18 months back and this ankylosis is believed to be a consequence of the same.

Ethical Considerations

Compliance with ethical guidelines

There were no ethical considerations to be considered in this article.

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Conflict of Interests

The authors have no conflict of interest to declare.

References

- [1] Lewis D. Myositis ossificans. JAMA. 1923;80:1281. <https://doi.org/10.1001/jama.1923.02640450001001>
- [2] Nishio J, Nabeshima K, Iwasaki H, Naito M. Non-traumatic myositis ossificans mimicking a malignant neoplasm in an 83-year-old woman: A case report. J Med Case Rep. 2010;4:270. <https://doi.org/10.1186/1752-1947-4-270>
- [3] Chalmers J, Gray DH, Rush J. Observations on the induction of bone in soft tissues. J Bone Joint Surg Br. 1975;57:36-45. <https://doi.org/10.1302/0301-620X.57B1.36>
- [4] Bagaria V, Rasalkar D. Extensive myositis ossificans with extra-articular ankylosis of the hip joint. Hong Kong J Radiol. 2011;14:35-8.
- [5] Shah KA, Desai MM, Kantanavar R, Shah S. Complete Extra-articular Ankylosis of Hip and Stiff Elbow in a Case of Extensive Myositis Ossificans: A Rare Presentation. J Orthop Case Rep. 2020 Jan-Feb;10(1):74-77.
- [6] Aneiros-Fernandez J, Caba-Molina M, Arias-Santiago S, Ovalle F, Hernandez-Cortes P, Aneiros-Cachaza J. Myositis ossificans circumscripta without history of trauma. J Clin Med Res. 2010;2:142-4. <https://doi.org/10.4021/jocmr2010.05.364w>