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ATRAUMATIC HETEROTOPIC OSSIFICATION OF THE SUPERIOR GAMELLUS MUSCLE : A CASE REPORT.

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ABSTRACT

Heterotopic ossification is defined as the formation of normal mature lamellar bone in atypical, extra skeletal tissues, commonly around the hip joint involving the abductor muscles and occasional iliopsoas muscles. Among the known causative factors are traumatic brain injury, spinal cord injury, and other trauma including iatrogenic causes such as surgery. A less common cause is exposure to pharmacologically induced paralysis involving prolonged intubation. We present a case of heterotopic ossification involving the Superior Gamellus muscle without antecedent trauma but with history of prolonged intubation who functionally improved after surgical resection.

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ABSTRACT

Heterotopic ossification is defined as the formation of normal mature lamellar bone in atypical, extra skeletal tissues, commonly around the hip joint involving the abductor muscles and occasional iliopsoas muscles. Among the known causative factors are traumatic brain injury, spinal cord injury, and other trauma including iatrogenic causes such as surgery. A less common cause is exposure to pharmacologically induced paralysis involving prolonged intubation. We present a case of heterotopic ossification involving the Superior Gamellus muscle without antecedent trauma but with history of prolonged intubation who functionally improved after surgical resection.

Keywords: Heterotopic Ossification, Hip joint, Intubation, Pharmacological, Skeletal muscle.

INTRODUCTION

Heterotopic ossification (HO) is the formation of normal mature lamellar bone in atypical, extra skeletal tissues. While the exact pathogenesis of HO remains elusive, some causative factors to this detrimental disorder have been identified. Trauma (from stab wound and blunt trauma), repetitive mechanical stress, arthroplasty, fractures, injury to spinal cord and thermal injury are the common causes for HO.[1] Apart from the listed common causes, recent literature has described the occurrence of HO in patients exposed to pharmacologically induced paralysis; such as in prolonged intubation.[2,3]

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Even though HO is a self-limiting condition, it may cause significant morbidity. It may lead to notable limitation in movements especially when it occurs close to joints. This may eventually leads to impairment of activities of daily living. HO frequently occurs around the hip joint, mainly involving the abductor muscles and least commonly the iliopsoas.

The following case highlights a unique case of HO, in which the hip joint was affected with the involvement of the superior gamellus muscle in the absence of antecedent trauma or intramuscular haemorrhage but associated with a history of prolonged intubation five years ago. The HO of the superior gamellus muscle of the left hip resulted in a fixed flexion deformity of her hip with reduced hip rotation and persistent stiffness which was sucessfully resolved with surgical excision of

the calcified mass.

CASE REPORT

A 46-year-old lady with SLE presented to our orthopedic clinic with a history of painless, stiff left hip for the past five years. She also complained of difficulty in walking due to the hip stiffness and shortening of her left lower limb. In order for her to walk, she had to plantar flex her ankle. There was no significant history of trauma, surgery or bleeding disorders.

However, she had a history of prolonged intubation of 10 weeks duration in another health facility about five years ago. She suffered cardiopulmonary collapse as a result of severe acute hemolytic anemia which required intubation and intensive care. Her Hb on admission was dangerously low at only 2.9g/dL. She was eventually diagnosed as having Autoimmune Hemolytic Anemia (AIHA), which is a hematological manifestation of lupus. Since her discharge from Intensive Care Unit, she noticed that she had stiffness over multiple joints including her hips, knees and ankles. She had undergone soft tissue releases over both ankles and knees to address her joints stiffness, but no procedure was done for her hip stiffness.

Physical examination revealed a 45^{o} fixed flexion deformity of the left hip with an ipsilateral ankle equinus which was possibly due to the mechanical block of HO at the superior gamellus. There was also significant restriction in internal and external rotation of the left hip. She had a hyperlordotic spine and the left lower limb appeared to be short. Plain radiograph showed a vague ossified mass traversing the left neck of femur (Figure 1a). Further assessment of the bony mass with a computed tomogram scan showed a well-defined, mature trabeculated mass extending from the greater trochanter to the posterior column of the left acetabulum (Figures 1b and 1c). The ossified mass measured approximately 8 cm in length.

A diagnosis of HO of the left hip was made and treatment options were discussed with the patient. Eventually, patient consented for open surgical excision of the ossified mass. During the surgery, the patient was positioned in right lateral decubitus with anterior and posterior pelvic support. The skin incision was made at the center of the left greater trochanter and extended slightly curved backward. After incising the fascia lata and splitting the gluteus maximus with removal of some adipose tissues; the bony mass attached to the greater trochanter was felt and identified (Figure 2a). The sciatic nerve was carefully isolated; starting from beneath the piriformis, to avoid iatrogenic injury. The dissection was performed meticulously as the hip could not be extended to protect the sciatic nerve during the surgery

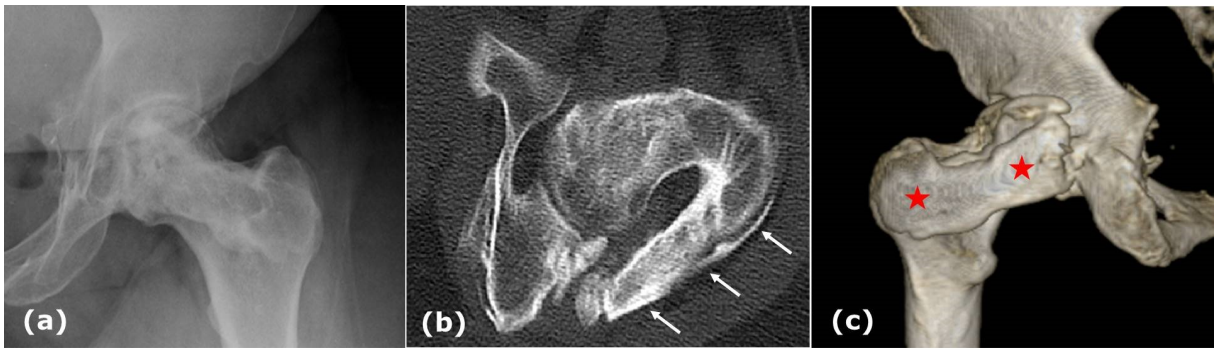

Figure 1: (a) Patient's left Hip AP radiograph suggesting a vague rectangular ossified mass traversing the left neck of femur with reduction of superior hip joint space, (b) computed tomogram scan showed a well-defined, mature trabeculated ossified mass extending from the greater trochanter to the posterior column of the left acetabulum (White arrows) and (c) which measured approximately 8 cm in length (red stars). [Click on image].

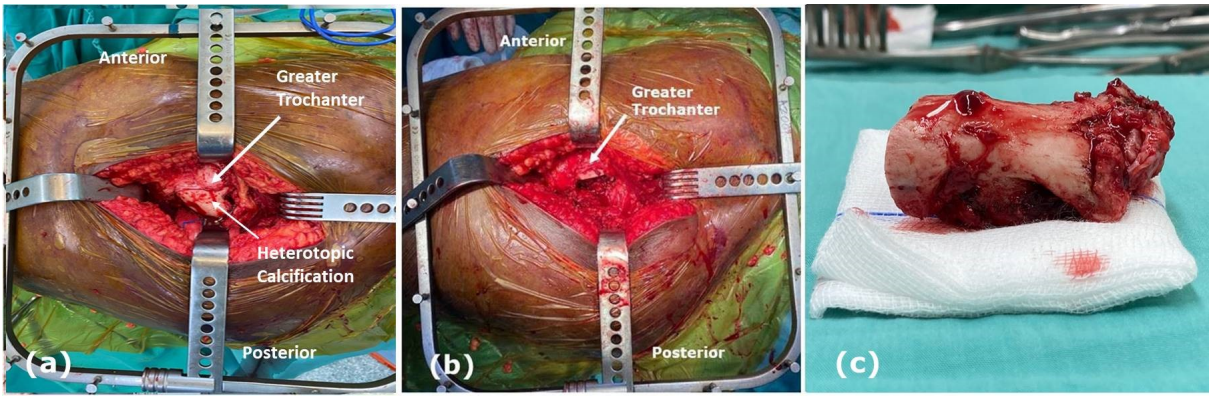

Figure 2: Intraoperative pictures showing before (a), and after (b,c) excision of heterotopic ossification. [Click on image to enlarge].

due to the hip deformity. The sciatic nerve was at the center of the bony bar, covered with some fatty tissues. The long bony bar which measured 8 cm was carefully separated with minimal bleeding, osteotomized at the lateral end, detached from the greater trochanter, stripped from the adherent soft tissue at the medial end and successfully removed (Figure 2b, c, d). Intraoperatively, a full passive range of motion of the left hip was tested and the haemostasis was secured before closing the wound. Prophylactic celecoxib was started postoperatively and continued for six weeks to prevent recurrence of HO. In view of the risk of wound healing delay, prophylactic irradiation was not given. Histopathological examination of the excised bony mass further confirmed the diagnosis of HO.

Post-operative plain radiograph confirmed complete excision of the mass. Follow up and review at six months post-op revealed that patient had returned back to her normal activity of daily living with no clinical or radiological evidence of HO recurrence.

DISCUSSION

Despite the acknowledgement of the existence of HO for more than 400 years, its definite pathogenesis remains ambiguous. Less common causes included genetic predisposition such as fibrodysplasia ossificans progressive, in which a single heterozygous single nucleotide substitution of aginine to histidine (R206H) in the Activin A type 1 receptor (ACVR1) gene, leading to bone formation in ectopic sites.[4] However, the typical HO cases were usually due to significant trauma commonly associated with blast injury, high-energy transfer gunshot wounds, central nervous system injury, burns, hip replacement surgery, acetabular fractures, and elbow injuries.[4] There is significant evidence supporting the role of an exaggerated inflammatory response in the pathogenesis of HO and in major trauma situation as mentioned above, a multiphasic acute and chronic inflammatory dysregulation occurs which may be responsible for formation of HO.[4]

The present case is unusual in that there was the absence of trauma and neurological system compromise nor did she had any genetic predisposition and the location of HO was exclusively in the superior gamellus muscle. Her symptoms of joint stiffness dated back to 5 years ago and coincided with an acute ICU admission requiring prolonged intubation with pharmacological induced paralysis and immobilization. Patients requiring ICU admission and ventilation are generally very ill with severely acute inflammatory dysregulation.

The first evidence of the relationship between HO and neurologic injury was established by Reidel in 1883.[5] Subsequently, HO has been widely investigated in cases of individuals with central nervous insults. Clements in 1993 first documented the occurrence of HO in three patients subjected to prolonged mechanical ventilation with respiratory failure.[6] Subsequently, more authors reported the same phenomenon emphasizing the atraumatic nature of HO.[3,7,8] In essence, all the authors agree that atraumatic HO may be the result of pharmacologically induced paralysis in association with compromised neurological system. These could have been the causative factors leading to HO of the superior gamellus muscle and the left hip joint in our patient, which is considered rare.

Posterior ossification was the second most frequent pattern of HO, accounting for 29% of cases.[9] However in this pattern of HO, several muscles such as piriformis, obturator internus and externus, the superior and inferior gemelli and quadratus femoris and part of gluteus maximus are reportedly affected in various combinations but none of these muscles were affected in isolation.[7] In our patient, the ossification of the superior gamellus results in fixed flexion deformity with gross restriction of internal and external rotation of the hip. The AP hip radiograph (Figure 1a) can be misleading and confusing in view of the peculiar isolated superior.

Treatment options for HO can be group into 2 categories, preventive and symptom/functional relief once HO is established. In cases of major trauma or in patients on prolonged ventilation and paralysis, NSAID can be given, as in our case to reduce and regulate the inflammatory response set up by the initiating events.[4] In high risk cases, radiotherapy alone or in combination with NSAIDs have been advocated and reported to be successful, although there is a lack of consensus on the timing and dose of radiotherapy.[4] Similarly there is paucity of data on its use in trauma situations, not to mention the adverse effects of radiotherapy on bone and wound healing, which limits its use.[10] The use of bisphosphonates which is used for preventing and treating osteoporosis in preventing HO is controversial and current evidence does not support it.[4]

Surgery with excision of the ectopic calcification is recommended for symptoms/ functional relief as in our case.[4] However, such surgery can be challenging for the surgeon and patient. Similarly the timing of the surgical excision is still debatable. If too early, recurrence may occur but this could be offset with NSAIDs prophylaxs for 4 weeks duration after the surgical excision. Post excision, patients will required an extensive rehabilitation programme of physiotherapy and occupational health therapy.

Our case is distinguishable from the typical HO by the absence of the two usual causative factors; which are obvious compromise of neurological system and trauma. However, our patient experienced prolonged immobilization and intubation which are the less common causative factors for HO formation. Hence, this case denotes a distinctive clinical case in which HO may occur. This should forewarn clinicians to the necessity for raised awareness of the problem and the need to take further preventive measures such as passive physical therapy.

CONCLUSION

Heterotopic ossification of the superior gamellus may occur without a clear history of trauma. The plain radiograph findings can be confusing and might need further radiological imaging such as CT scan. Excision is feasible through posterior approach to the hip. A comprehensive surgical planning including adequate radiological images, accurate surgical timing and careful technique, added with

postoperative adjuvants such as celecoxib, ensure the best outcome for treating this mechanical block to motion.

CONFLICT OF INTEREST

The author reported no conflict of interest or financial liability.

INFORMED CONSENT

Consent has been obtained from the patient in regards to the pictures and details included in this report.

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