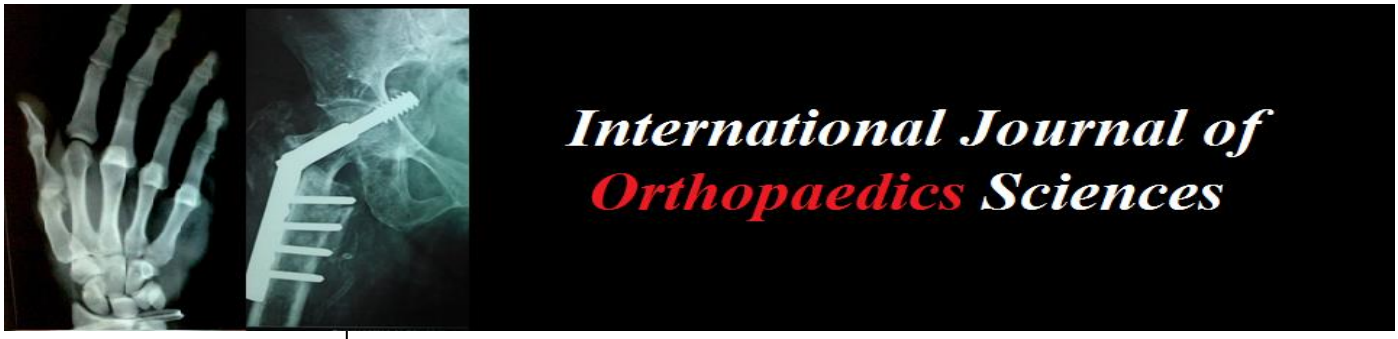

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Case report: Surgical management of a rare colossally sized neurogenic heterotrophic ossificans in the hip

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Abstract
A rare case of Neurogenic heterotrophic ossification in Hip region presented to our institute with gross clinical features involving global restriction of movement in the hip joint. The patient had sustained a head injury 9 months back due to road traffic accident. Initial radiographs revealed a bony mass in the left hip region from Anterior superior iliac spine to proximal shaft of femur. Later CT scan of hip revealed various facets of the condition, the expanse of the entire bony lesion was traced. A 10 x 10 x 4cm irregular bony mass was visualized in the left hip spanning the entire left hip joint. The extent of involvement of vastus muscles and gluteal muscles was still uncertain. After thorough pre operative planning the mass was excised and sent for histopathological examination. Complete range of movements were checked perioperatively after excision involving flexion, adduction abduction and rotations. The biopsy findings suggested that the specimen contained large osteoid seams with fibrocollagenous bands and striated muscle tissue suggestive of heterotrophic ossificans.

Keywords: Heterotrophic ossificans, neurogenic heterotrophic ossificans, bony mass, excision biopsy, pelvis, hip

Introduction
Neurogenic heterotopic ossification is a potential sequelae of spinal cord injury or head injury. It is a heterotopic bone formation in soft tissues, usually post traumatic brain injury [TBI]. Commonly seen among in active young men between 2nd and 3rd decades. NHO is more common in adults with an incidence generally reported between 3 ± 10% [1-2]. The clinical symptoms in children do not differ from adults, but are less severe compared to adult patients. Moreover, spontaneous regression is more often seen in children and young adults.
The clinical spectrum of Neurogenic heterotopic ossification ranges from an incidental finding on X-rays to severe restriction of the range of motion [20 -30%] and even complete ankylosis of joints [3-8%]. Neurogenic heterotopic ossification always occurs below the level of the spine injury, most commonly at the hip (70-97%) [3, 4, 5, 6, 8, 9, 11, 12, 14, 16, 17, 18].
Although the etiology remains unclear, it is supposed to be due to osteoblast metaplasia with benign self limiting course. Remarkably, at onset it can be clinically, radiologically as well as histologically indistinguishable to soft tissue malignancies, leading to misdiagnoses and improper treatments. The natural history of neurogenic heterotrophic ossficans consists of a rapid overgrowth in the first 4 weeks, when osteoblasts and chondrocytes produce new osteoid matrix in middle of the lesion. Other clinical factors associated with Neurogenic heterotrophic ossification are the presence of pressure sores, urinary tract infections or renal stones, deep venous thrombosis (DVT) and severe spasticity

Case Report
A 30 year old male presented to our institute with complaints of pain, swelling and restriction of movements at the left hip joint since 9 months. Patient gives history of head injury 9 months back due to road traffic accident, he also gives history of loss of consciousness. Initial diagnostic evaluation revealed a contusion in the fronto temporal region of brain and was bedridden for 1 month. Since then he had reduced speech, difficulty in standing, reduced power in both upper and lower limbs. However the pain in left hip joint along with the swelling was increasing progressively.

At the time of presentation, the patient was able to stand with help of support and bear weight on affected limb but was unable to walk. On examination of left hip joint showed a 10 cm tender bony hard irregular swelling over postero lateral aspect of thigh with raised local temperature and complete restriction of movement of left hip joint. After initial blood investigations and X ray imaging were done, they revealed a massive bony mass extending from anterior superior iliac spine to the level of lesser trochanter of proximal femur distally along with raised S. Alkaline phosphatase.

Further CT revealed hidden facets of this injury, an Osteid lesion involving left iliacus, gluteus group of muscles, obturator internus, proximal vastus and adductor group of muscles with no bony injury or fractures and normal articular surface without any destruction. After proper counselling and thorough planning of the surgical management, the bony mass was excised and biopsy was sent. Upon exposing the lesion, it showed irregular pattern of osteoid matrix suggestive of malignant lesion. The mass was approximately 10 x 10 cms and had good vascular supply. Intra operatively, 3 planes of movements i.e. flexion, extension, abduction, adduction and rotation movements were performed to full extent.

The excisional biopsy showed large osteoid seams along with fibrocollagen bundles and striated muscle tissue indicative of Neurogenic Heterotrophic Ossificans.

Post operatively he was started on oral Indomethacin and NSAID's, he was started with supervised physiotherapy for left hip joint movements along with quadriceps strengthening exercises for the first few weeks and later with proper motivation he was able to perform the complete range of movements on his own.

Such severe massive and grotesque swelling in the hip region is rare occurrence, hence reporting the case to orthopaedic fraternity.

Discussion:

Neurogenic heterotrophic ossificans is a benign self-limiting condition of non neoplastic heterotopic bone formation in the muscle or soft tissue. A genetic predisposition for Neurogenic heterotrophic ossification based on human lymphocyte antigen (HLA) [22, 23] typing has also been suggested. Surgery is deserved to persistently symptomatic cases and is preferably delayed until the complete maturation of the lesion has been reached and ossification has stopped to reduce relapses [19].

Pathophysiology Neurogenic heterotrophic ossification originates in the connective tissues and may be contiguous with the skeleton, but doesn't involve the periosteum. Near a joint, it leaves the joint capsule and space well preserved. Muscle fibres are not primarily involved, they can be incorporated or compressed by the fibrosis and calcifying soft tissues [19].

The centripetal pattern of development and maturation that is seen in the following weeks is the basis of zone phenomenon explained by Ackerman [20].

- It is characterized by a thin outer zone in the surrounding muscle that encloses a broad intermediate zone.
- In the intermediate zone has areas of immature bone that are lined by osteoblasts, while in the outer part of this intermediate zone mature bone forms a well-demarcated outer trabecular rim.
- The central zone consists of an undifferentiated highly cellular proliferation of fibroblasts with haemorrhage and muscle necrosis [20].

Mature Neurogenic heterotrophic ossification resembles normal bone, both histologically and radiologically. Histologically like presence of Cortex, Haversian canals, marrow and blood vessels [19, 24, 26, 29, 30].

According to Chalmers *et al*. [21] three conditions must be present for the formation of ectopic ossification:

- The presence of osteogenic percursor cells,
- An inducing agent
- Permissive environment.

Even if Neurogenic heterotrophic ossification displays the typical radiological and histological features in the course of disease, it can represent a diagnostic challenge during the initial weeks from onset, requiring a differential diagnosis with malignancy.

Radiographic imaging typically show a lesion made up of a central radiolucent area indicating immature bone formation with a calcified peripheral rim of mature ossification [29-34]. A thin radiolucent cleft separates the ossified mass from the adjacent cortex, which is intact, thus guiding the differential diagnosis with bone malignancies [29-34].

The biopsy is used to differentiate lesions, but if performed too early or within the core lesion, the presence of pleomorphic osteoblasts with atypical nuclei and mitosis can be misleading. It is likely to run into the misdiagnosis of soft tissue sarcoma, due to the presence of isolated mitotic fibroblast-like cells.

In our case, before contemplating for surgery we had counselled the patient as well as the attenders regarding the chances of recurrence, fracture neck of femur during the surgery and other surgical complications.

Operatively, the patient was placed on a radiolucent operating table and kept in supine position. Lateral approach to the hip joint was chosen, soft tissue dissection was done meticulously to not disturb the already interposed tissues which was a challenge. Heterotrophic mass was identified and excision was carried out along the surface of mass. Hip joint was not opened and complete mass was excised. Intraoperatively complete range of movements were demonstrated. The patient was started on Oral Indomethacin tab and gentle physiotherapy exercises along with protected weight bearing.

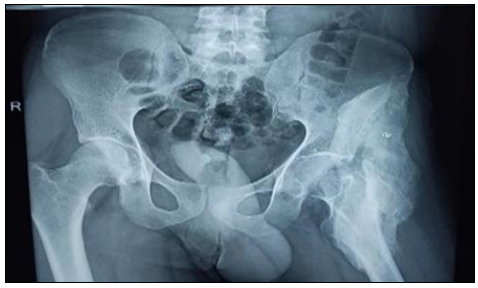

Fig 1: Pre op X-ray

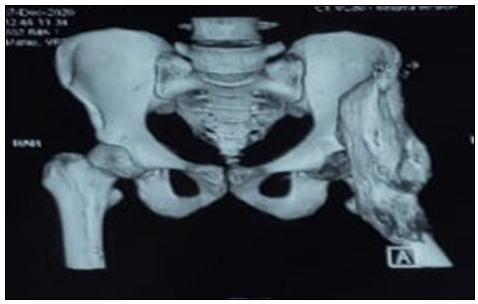

Fig 2: CT scan

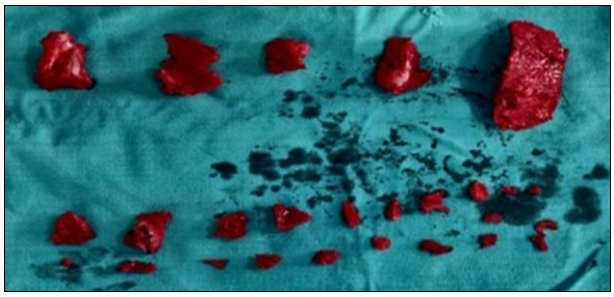

Fig 3: Excised bony mass

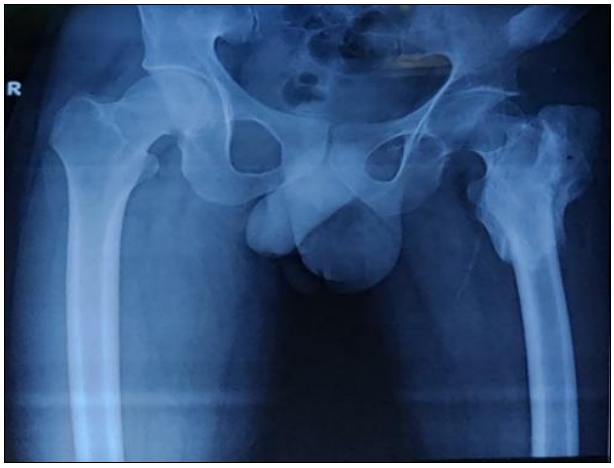

Fig 4: Post op X-ray

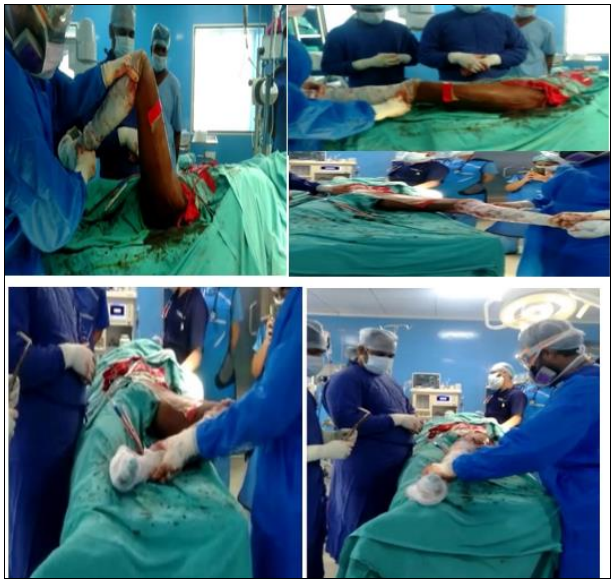

Fig 5: Perioperatively performing movements at hip joint

Conclusion

Neurogenic heterotrophic ossificans should be considered in the differential diagnosis of bone and soft tissue sarcomas in front of painful soft tissue swelling, valuing the radiological pattern and benign course of the disease, even in the absence of a clear history of trauma, to avoid unnecessary treatments and to maximize functional outcomes. Radiographic signs of Neurogenic heterotrophic ossification recurrence have been mentioned in 82-100% of the operated patients [35-41], although clinically significant NHO would develop in only 17-58% of the cases [35-41]. However such complex cases of Neurogenic heterotrophic ossification with massive lesions can be effectively treated with patience and adequate understanding of local anatomy for productive outcome.

In our case, the outcome was exceptionally good as complete range of movements could be achieved without causing any iatrogenic fractures such as neck of femur or at subtrochanteric region being susceptible. At 4 weeks post operatively, the patient was able to walk with the help walker and was satisfied with the outcome. Further improvement in movements and walker free gait can be achieved with the help of time, confidence and adequate support by the team especially involving physiotherapist. Long term results and probability of recurrence was not documented as patient is still in follow up.

However according to the literature, to date there is no satisfactory prevention of Neurogenic heterotrophic ossification and its prophylaxis is mainly based on prompt diagnosis and adequate treatment. However there is evidence that regular and cautious mobilization of the large peripheral joints should be recommended, from the day of the injury, to keep the joint capsules away from stiffness as possible and to maintain adequate muscle length. With such a methodical approach contractures and spastic sequelae will can be delayed and Neurogenic heterotrophic ossification related to traumatizing mobilization might be prevented.

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