Neurogenic Heterotopic Ossification of the Hip after Brain Injury. Function Recovery Preserving Hip Joint: A Case Report

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Learning Point of the Article:
The pathogenesis and the proper management of neurogenic heterotopic ossification after brain injuries. NHO Surgeons should be aware of the condition and refer soon patients to specialized centers, cause “functional resection” could restore a satisfying patients’ quality of life preserving at the same time the hip joint.

Abstract

Introduction: Up to 30% of patients with spinal cord injury and to 20% of patients with traumatic brain injury develop neurogenic heterotopic ossification (NHO). Patients develop restriction in joint range of motion (ROM) and impairment in activities of daily life. When neurological recovery occurs, joints dysfunction represents the cause for patients’ autonomy loss.

Case Report: We present the case of a 39-year-old Caucasian male involved in a car accident and experienced 14 days of post-traumatic coma. After rehabilitation, no residual motor or sensory neurological deficit was present, but bilateral NHOs surrounding hip joints developed. Seventeen months after trauma, the patient was admitted to our institute. He was confined to bed, absolutely unable to walk, stand or sit. Radiological evaluation consisted in Antero-Posterior X-ray view only, due to the inability to open up his hips for lateral views, and 3D computed tomography scan. “Functional resection” of the ossifications was performed and rehabilitation started from day 1 after surgery. At the final follow-up 24 months from the second operation high grade of clinical satisfaction was reported. The patients were able to walk independently, to put on his socks, and to drive a car and bike. Painless right and left hip ROM was, respectively, 100° and 90° for flexion, 10° for extension, 35° and 30° for abduction, and near normal internal and external rotation. The patient referred that further improvement in mobility and fluency was still present day by day.

Conclusion: The rarity of the condition frequently bring to a delay in treatment and the absence of specific guidelines made treatment still dependent on surgeon experiences. A multidisciplinary approach is essential for success. Surgeons should be aware that it is important to refer patients to specialized center, because early resection could provide excellent results, preserving hip, and restoring function and patient independency.

Keywords: hip, ossification, injury, resection, neurogenic heterotopic ossification.

Introduction

Neurogenic heterotopic ossification (NHO) consists in an unregulated differentiation of soft tissues to endochondral bone, which occurs after a traumatic neurological lesion. Up to 30% of patients with spinal cord injury (SCI) and to 20% of patients with traumatic brain injury (TBI) develop NHO [1, 2, 3]. Although proper pathogenetic mechanism is unclear, prolonged coma, tissue hypoxia, mechanical ventilation, spasticity and lack of extremity movements have been supposed as possible trigger of the disease [4, 5]. Proximal and grater joints are most commonly affected, with the hip accounting for about half of the cases [6]. Patients develop restriction in joint range of motion (ROM), besides various grade of neurological sequelae. Thus, normal activities of daily living (ADLs) and autonomy are severely hindered.
Case Report

We present the case of a 39-year-old Caucasian male involved in a car accident in February 2017. The patient sustained a severe TBI requiring surgical intervention for subdural hematoma drainage, followed by post-traumatic coma. Awakened from the coma after 14 days he gradually regained his mental functions. He was then transferred to a rehabilitation center specialized in neurological injuries. In 5 months memory and other cognitive abilities were completely restored as well as body functions except for hip mobility. In fact, no residual motor or sensory neurological deficit remained, but bilateral NHOs surrounding hip joints and entirely limiting hip motion developed.

In June 2018, the patient was admitted to our Institute. He was confined to bed, absolutely unable to walk, stand or sit, although normal neurological functions of the lower limbs and normal power and mobility of the upper limbs were present. No pressure sores were present. Radiological evaluation consisted in Antero-Posterior X-ray view only, due to the inability to open up his hips for lateral views, and 3D computed tomography (CT) scan (Fig. 1). Size, location, relation with neurovascular bundles, and intra-articular involvement were investigated. Thorough counseling about treatment and related risk were provided and patient informed consensus was obtained.

In July 2018 and September 2018, the left and right hips were treated, respectively. In the left hip NHO was predominantly anterior and ileoinguinal approach was used. For the right hip a circumferential pattern of NHO was present and a combined triradiate and ilioinguinal approaches were used. The covering muscles and soft tissues were progressively released, thus exposing proximal and distal NHO implantsations. “Functional resection” was performed to avoid risk of post-surgical fractures (Fig. 2). Tendon insertions were good landmarks, as well as capsule and periosteum planes were useful for guiding resection. In the left hip, excessive release of rectus femoris occurred and a 5-mm corkscrew was needed to reattach the released part of the tendon. Neurovascular bundles were identified and isolated. Electric cautery was thoroughly used to reduce bleeding. In both hips, capsule was not open and articular procedures were not performed, being joint space not involved. Drainage was not applied. No post-surgical

Figure 1: Pre-operative antero-posterior X-ray (a), axial computed tomography (CT) (b) and 3D-CT scan (c) showing bilateral neurogenic heterotopic ossification of the hips. Circumferential and predominantly anterior patterns were present into the right and left hip, respectively. No articular involvement was noted. Three cancellous screws are present into the right hip for a previous femoral neck fracture.

Figure 2: Twenty-three months after surgery Antero-Posterior X-ray showing functional resection and no recurrence of the ossifications. Note the 5-mm corkscrew used into the left hip to reinsert part of the rectus femoris tendon.

Figure 3: Twenty-four months follow-up clinical evaluation. Abduction and Flexion hip ROM restore reflects the patient’s activities daily life recovery.
hematoma, infection, deep venous thrombosis, or fracture occurred.

In Our Institute of a dedicated Department of Physiatry and Physiotherapy allowed for a multidisciplinary management which is essential for successful treatment of such patients. Thus, rehabilitation protocol started from day 1 after surgery with short sessions of supervised passive pain-free mobilizations twice a day and continuous Passive Motion device during night for the 1st week. Progressively, longer sessions, and active pain-free motion were introduced. Weight bearing on the operated limbs was permitted after 15 days. Indometacin (100 mg) once daily for 6 weeks was administered postoperatively to reduce recurrence risk.

The patients was discharged from our institute 4 weeks after the second operation when he was able to pass from supine to standing and sitting position independently and to ambulate with a walking aid.

At the final follow-up 24 months from the second operation high grade of clinical satisfaction was reported. The patients were able to walk independently, to put on his socks, to drive a car, and bike. He did not refer impairment in the most of the ADLs. Right and left hip ROM was, respectively, 100° and 90° for flexion, 10° for extension, 35° and 30° for abduction, and near normal internal and external rotation (Fig. 3). Motion in all planes was painless and patient referred that further little improvement in mobility and fluency were still present day by day. No recurrence of NHO was observed radiologically.

Discussion

NHO is a common complication after traumatic neurological injuries. The exact mechanism is still not clear. Pluripotent mesenchymal cells of skeletal muscles have been postulate to differentiate into osteoprogenitor cells producing ectopic bone that leads to NHO [4, 7]. However, the exact stimuli of the process are investigational. Prolonged coma, tissue hypoxia, mechanical ventilation, subsequent spasticity, and lack of extremity movements have been supposed as triggers [5]. Regardless of the etiology, the process develops in 4–12 weeks and completely mature within 2 years [8]. Once instituted, regression is not feasible.

Not many papers are present in literature about this topic and no guidelines on the proper treatment can be found. Thus, management of NHO could be seriously arduous for orthopedic surgeons facing with this uncommon condition. The mainstay consists in a multidisciplinary management [1, 2]. Patient neurological status and NHO features must be evaluated simultaneously [1]. Patients most commonly present with various grade of sensitive or motor deficit. In those cases, treatment aims to improve quality of life avoiding patients’ bedding and related complications (e.g., decubit ulcers, infections, or deep vein thrombosis) [6]. Our patient had completely restored his functions after 5 months of rehabilitation and did not present residual neurological deficits at the time of surgery. Sensory and muscle tone were normal. Thus, the expected functional gain was exciting. According to the recent literature, radiological evaluation was based on X-ray and 3D CT scan. Scintigraphy was considered not useful to define proper timing of surgery, which is independent from NHO “maturation” [2, 9]. We agree that surgery must be considered when NHO reduce ROM or produce pain due to neural/vascular compression [1]. In this setting, early surgical resection represents the best treatment choice, reducing the rate of intra-articular complications which rises over time, without increasing recurrence risk.

NHOs typically develop around the joints, sparing the articular surfaces [1]. Stiffness is only extra-articular and surgical resection allows for excellent results. The proper approach depends on the location of the ossifications [2, 10]. Resection does not need to be demolitive like in oncologic surgery. “Functional resection” aims to remove only the ossification causing problems. The bone at the implantation base should be respected to avoid iatrogenic fracture. Tendon, capsule, and vessel may be useful to guide resection and attention must be paid to avoid soft tissues damage. Drainage is commonly used because resection leaves a large void and hematoma formation represents a well-known complication of the procedure. Furthermore, it is fundamental for success to start the rehabilitation program after the first post-operative day. Neurological status permitting, resumption of weight bearing depends on the bone density and the risk of femoral neck fracture. In case of bone fragility, it needs to be postponed for 3–4 weeks and should progress gradually [1].

In our case in the left hip an excessive release of rectus femoris occurred and a 5-mm corkscrew was needed to reattach the released part of the tendon. At the final follow-up, the muscle function was comparable to the contralateral side. No drainage was used. Hemostasis was carefully performed and no severe hematoma was noted in both the sides. Furthermore, rehabilitation program started the day after surgery with gentle mobilization of the hip and weight bearing was allowed 2 weeks after surgery.

Besides hematoma formation, the fearsome complication consists in the risk of infection. Gatlin et al. [11] recently reported a 10% incidence of post-operative infection. The risk was associated with higher ASA score, younger age, and SCI. In our case none of these factors was present and the patient did not experience any deep or superficial finding of infection.

In case of intra-articular involvement, total hip arthroplasty
(THA) represents an effective solution providing good clinical results [7]. However, this option is confined to those cases left untreated for years, in which intra-articular complication occurred. Thus, early diagnosis and treatment allow to preserve the hip joint, restoring function, and avoiding THA in the affected patients which are commonly young.

Extra-corporeal shock wave therapy (ESWT) has been proposed as symptomatic treatment in patients with NHO [12]. Regardless the time after injuries, ESWT showed to reduce patients’ referred pain, without improving function or NHO size. Because it is not a permanent treatment, ESWT was not commonly taken into account in our center.

Garland et al. [13] reported that the number of joints involved and the level of neural deficit were the main risk factors for recurrence. NHOs typically occur after traumatic SCI or TBI and neurologic sequelae are not uncommon. In our patient NHOs only affect right and left hip and no residual neurological deficit was present. Thus, removing the ossifications, the cause for mobility and autonomy loss was eliminated and the patient was permitted to go back to his independency and satisfying quality of life. At the final follow-up of 24 months, no recurrence of NHO was observed.

**Conclusion**

The absence of specific guidelines made treatment still dependent on surgeon experiences. A multidisciplinary approach is essential for success. It is important to refer patients to specialized center, because early resection could provide excellent results, preserving hip and restoring function and patient independency.

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**Clinical Message**

The rarity of the condition frequently brings to a delay in treatment. Patients usually received several unfavorable clinical opinions. Besides various grade of patient’s frustration, it could lead to local complications and joint ankylosis. Surgeons should be aware of the condition to refer soon patients to specialized center, cause “functional resection” could provide excellent results, preserving the joint and restoring a satisfying patients’ quality of life and independency.
| **Conflict of Interest:** Nil |
| **Source of Support:** Nil |

**Consent:** The authors confirm that informed consent was obtained from the patient for publication of this case report.

**How to Cite this Article**

Iorio R, Viglietta E, Massafra C, Ferretti A. Neurogenic Heterotopic Ossification of the Hip after Brain Injury. Function Recovery Preserving Hip Joint: A Case Report. Journal of Orthopaedic Case Reports 2021 June; 11(6): 97-101.