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Journal of Interdisciplinary Medicine

CASE REPORT

ORTHOPEDICS // NEUROLOGY

Giant, ankylosing ilio-femoral heterotopic ossification in a 50-year-old male patient with paraplegia after myelic fracture of the T12 vertebra. Case Report

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ARTICLE HISTORY

Received: June 18, 2024
Accepted: August 30, 2024

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ABSTRACT

Introduction: Neurogenic heterotopic ossification (HO) is the abnormal formation of bone in non- skeletal tissues, typically near major joints, following traumatic or nontraumatic spinal cord or brain injuries. This condition significantly affects the patients' quality of life, often leading to joint ankylosis and severe limitations of mobility. **Case presentation:** In June 2023, a 50-year-old construction worker fell from a height of 3 m and suffered multiple severe injuries, including an occipito-parietal cranium fracture with subsequent epidural and subarachnoid hematomas, a myelic fracture of the T11 and T12 vertebrae, traumatic anterolisthesis of the T11 vertebral body, and paraplegia. Following prehospital stabilization, he underwent lifesaving procedures, including a craniectomy for evacuation of the epidural hematoma and posterior stabilization of the thoracolumbar spine with decompressive laminectomy, at a trauma center in Budapest. By October 2023, rehabilitation efforts were hampered by extensive HO in the right hip, as confirmed by plain radiography. The patient was discharged in December 2023, with rehabilitation impossible due to the ankylosis of the right hip. At this advanced stage, surgical resection was identified as the only viable therapeutic option. **Conclusion:** Mature HO can only be effectively treated through surgical excision. Early diagnosis, prevention, and medical management of immature lesions are essential to avoid severe complications and preserve joint function.

Keywords: heterotopic ossification, neurogenic, spinal trauma, paraplegia, resection

INTRODUCTION

Heterotopic ossification (HO) is the development of lamellar bone in non-skeletal tissues that contain undifferentiated mesenchymal cells. Its precise etiopathogenesis is unknown.[1] Many factors, such as vascular stasis (edema), microtraumas, or altered tissue oxygenation, contribute to the activation of osteoblast and chondroblast progenitor cells, with the consequent precipitation of calcium salts within the periarticular soft tissues.[2] HO usually occurs

in the following five clinical settings: in rare congenital disorders, postsurgical, posttraumatic, neurogenic and as reactive lesions.[3,4] Neurogenic HO appears generally near the greater articulations of the skeleton, following traumatic or nontraumatic spinal cord or brain injury.[5] The incidence of HO in patients with spinal cord injury is 20%, with most occurring in the hip region.[6] The cause is uncertain. Tissue denervation with local edema, vascular stasis, microtrauma from patient manipulation are all considered contributing factors, without the presence of precipitating local trauma.[1] The ossifying process has three distinctive phases: amorphous calcification of soft tissues, immature ossification, mature ossification. Clinical findings in the early stage are confusing and are related to the inflammatory process: local swelling, pain with muscle spasms, redness. In this early clinical setting, HO may be confused with infection or neoplasia. In a later stage, when connective tissue mass is formed, range of motion decreases progressively in the adjacent joint and new, ectopic bone formation appears. In the mature phase, HO can cause motion restriction, joint ankylosis that greatly affects patient handling, local hygiene, and quality of life. In cases where quality of life is severely affected, as in our patient, surgical excision is the only option to restore mobility (in a wheelchair) and to prevent decubitus complications.[7,8]

CASE PRESENTATION

In June 2023, a 50-year-old construction worker fell from a height of 3 m and suffered the following injuries: an occipito-parietal cranium fracture with subsequent epidural and subarachnoid hematomas; a myelic fracture of the T11 and T12 vertebrae with traumatic anterolisthesis of the T11 vertebral body and consequent paraplegia; right tympanic hemorrhage; right facial paresis; multiple right-sided rib fractures with massive pulmonary contusion.

Following prehospital management and stabilization of vital parameters, the comatose patient underwent an initial assessment and a whole-body computed tomography (CT) scan, which confirmed the polytrauma and described the aforementioned lesions. Because of the life-threatening, massive epidural hematoma, an emergency craniectomy with evacuation of the hematoma was performed, followed by posterior stabilization of the thoracolumbar spine using transpedicular instrumentation between T10 and L2 with a decompressive laminectomy at the T12 level. These procedures were carried out at the South-Pest Central Hospital's Traumatology department in Budapest, Hungary (Figure 1).

The patient was initially unconscious (GCS 3), requiring endotracheal intubation and intermittent positive pressure ventilation. On postoperative day 7, a tracheostomy was performed due to the anticipated prolonged need for mechanical ventilation, and gradual weaning from mechanical ventilation was initiated. By day 13, the patient was able to breathe independently. Postoperative CT scans showed favorable results with evacuation of the hematoma and resolution of the mass effect. The patient's general neurological status improved rapidly; he regained consciousness, although sensitivity loss and paraplegia persisted without improvement, with a positive Babinski sign in the right lower extremity. By day 24, the tracheostoma was removed, and the patient regained the ability to speak. He was transferred to the Early Rehabilitation for Spinal Injury Patients department at the National Medical Rehabilitation Center to begin a comprehensive rehabilitation program.

After 6 days of rehabilitation, the patient began experiencing low back pain and intermittent fever. Blood cultures were positive for methicillin-susceptible Staphylococcus aureus, and inflammatory markers were elevated (C-reactive protein (CRP) 156.06 mg/l, procalcitonin 0,41 ng/ml). He was transferred back to the Traumatology Department for further evaluation of a potential infection. Surgical wounds appeared clean, and CT scans of these areas showed no signs of local infection. Pulmonary and urinary tract infections were ruled out, and no other infection source was found. Based on the blood culture results, a combination of ceftriaxone and clindamycin was initiated, leading to a good clinical response. Once his symptoms resolved and inflammatory markers normalized, the patient returned to the Rehabilitation Center to resume his rehabilitation program. Two days after resuming rehabilitation, his left lower extremity became swollen. Venous Doppler ultrasound revealed thrombosis in the popliteal and femoral veins. He was started on therapeutic-dose low-molecular-weight heparin, which was later transitioned to rivaroxaban (20 mg daily) for one year as recommended by a cardiologist.

After one month of rehabilitation, in early September, the therapist noted a limitation in the passive range of motion (ROM) of the right hip. Radiography revealed incipient periarticular HO, with an otherwise intact hip joint. At this time, serum alkaline phosphatase (ALP) levels were within the normal range (63 U/l). However, the passive ROM of the right hip progressively worsened, and follow-up radiographs showed further development of the HO. By mid-September, ALP levels had risen above the normal range (288 U/l), indicating active HO growth. By early No-

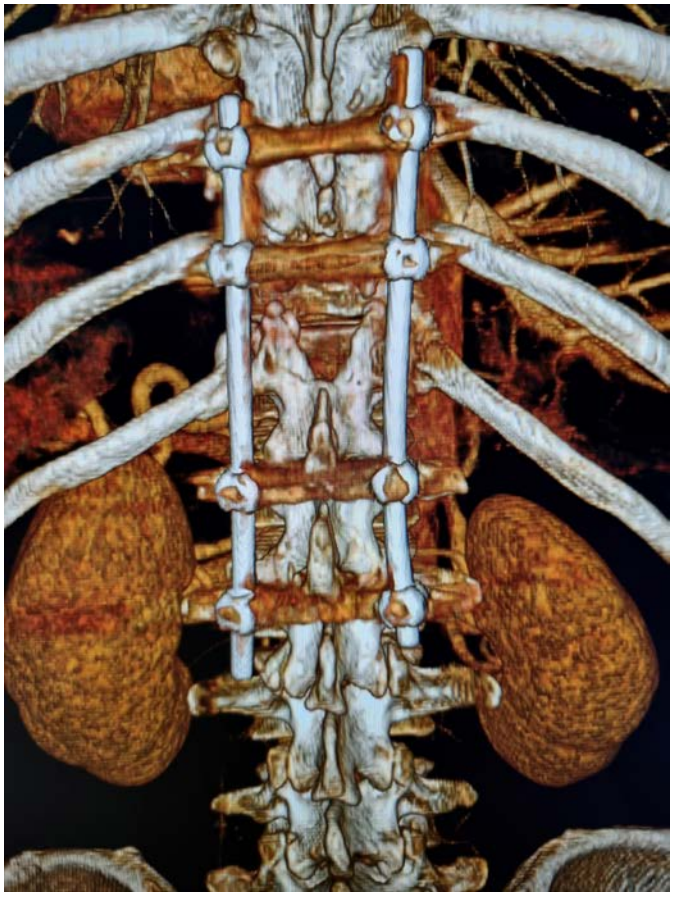

FIGURE 1. Posterior transpedicular stabilization of the thoracolumbar spine. CT reconstruction.

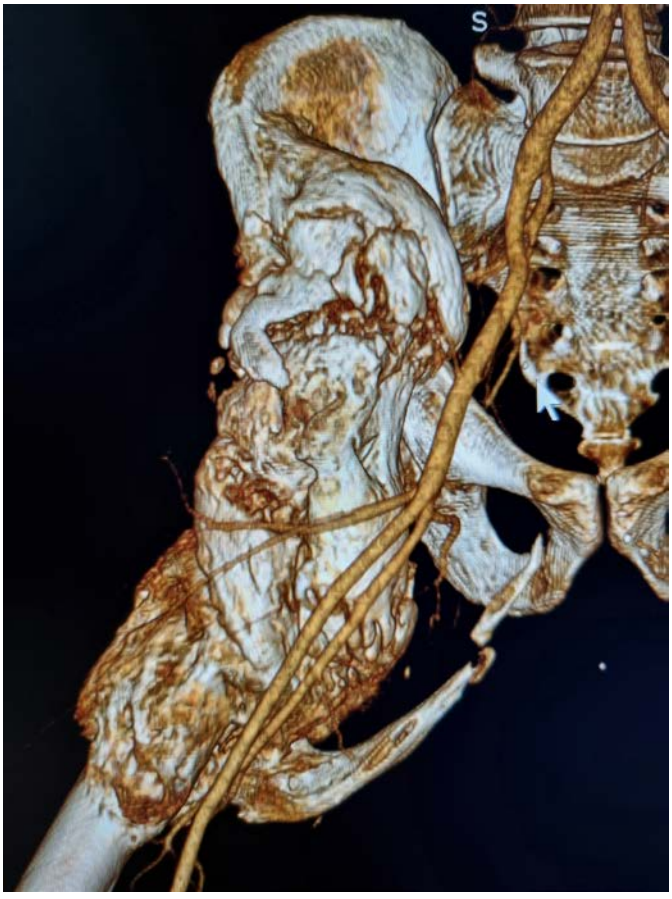

FIGURE 2. 3D CT reconstruction of the extensive HO of the right hip. The articulation was not involved.

vember, the ALP level peaked at 515 U/l, and the hip had developed ankylosis, with a negative effect on the patient's rehabilitation. On 27 November 2023, the patient received a single, low-dose radiation therapy session, after which ALP levels began to decline, allowing the rehabilitation protocol to resume, albeit with difficulties.

On 21 December 2023, the patient was discharged from the hospital with a recommendation to continue physical therapy. However, at the end of January 2024, he returned to our hospital for a second opinion due to his inability to perform the rehabilitation program, along with difficulties in transportation and maintaining local hygiene. Upon examination, the patient presented severe ankylosis of the right hip, which was fixed in external rotation, 30o of flexion, and abduction. Paraplegia was present, with sensory loss extending approximately 5 cm below the umbilicus. A CT scan revealed an extensive, 26 cm-long bony bridge with a maximum width of 8.56 cm between the internal surface of the iliac bone, passing beneath the inguinal ligament and lateral to the neurovascular bundle, to the proximal third of the femur. The structure consisted of mature bone with an irregular shape and surface, including medial spike-like extensions. CT angiography confirmed that the femoral neurovascular bundle remained outside the bony formation but was displaced anteromedially (Figure 2).

Although HO is primarily a musculoskeletal disorder, because of its large size and complex anatomic relationships, we decided that the case needs an interdisciplinary approach. With assistance from the 3D Reconstruction and Printing Department of the "George Emil Palade" University of Medicine, Pharmacy, Science and Technology of Targu Mures, we created a 3D reconstruction model of the HO based on CT scans to precisely assess its shape and dimensions. After consulting with general and vascular surgeons, we decided to proceed with surgical removal of the ossification. Preoperative evaluations for anesthesia revealed no contraindications.

We performed the surgical intervention on 18 March 2024, under general anesthesia. A proximally and distally extended right iliofemoral incision (right pararectal) was made. Major vessels, including the right common, internal, and external iliac arteries, were identified in the retroperitoneal space, isolated, and protected with elastic loops. The iliac portion of the HO was dissected from the internal

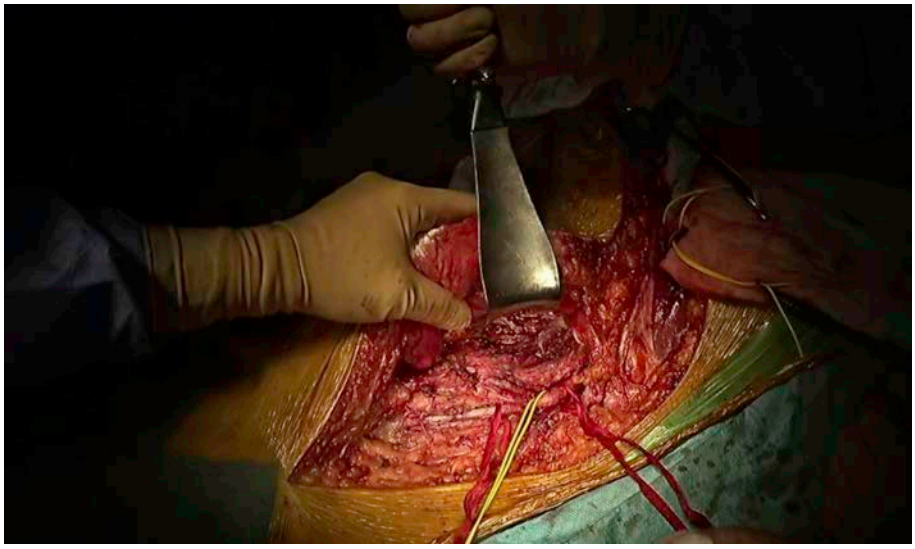

FIGURE 3. Intraoperative view of the dissected external iliac, deep femoral, and femoral artery with vessel loops, inguinal ligament, and the subjacent ossification

surface of the iliac bone. Distal dissection involved the inguinal ligament, the femoral neurovascular bundle in the Scarpa triangle, and the ossification itself (Figure 3). The bifurcation of the femoral artery was identified and protected, but the medial portion of the deep femoral artery, encased in extensive scar tissue, was sacrificed. Due to the size and extent of the HO, en bloc resection was not feasible. Despite meticulous preoperative planning, intraoperative orientation was difficult. Constant capillary bleeding and scarred, delicate soft tissues hindered our progress. Using a chisel, we fragmented the bony mass, allowing for step-by-step dissection and removal of smaller pieces (Figure 4). Post surgery, the right hip joint was found to be intact, and satisfactory movement was achieved, including 105° flexion, full extension, and rotation. Two drains were placed for 48 h, one in the lower abdomen and one in the femoral wound.

After the surgical intervention, no major complications occurred. Postoperative care was provided in the Orthopedics and Traumatology Department of the hospital. The patient received prophylactic antibiotics for 48 h following the procedure (cefazoline 1 g preoperatively, intraoperatively, and three times daily for 48 h postoperatively) and underwent transfusion of 2 units of red blood cells per day for 2 days. Wound care was performed every 2 days, and the drains were removed on day 2. Early postoperative physical therapy, immediately after surgery, was encouraged to maintain the passive ROM achieved.

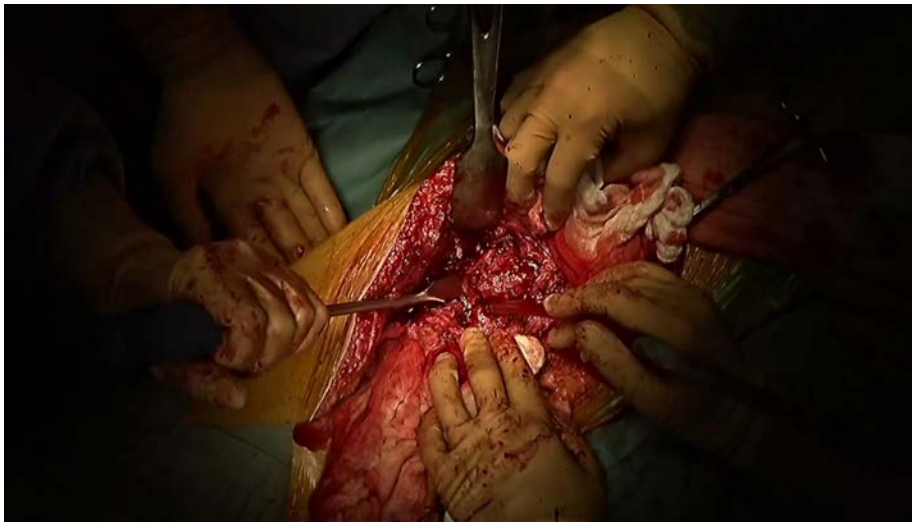

FIGURE 4. Step-by-step resection of the ossification mass in front of the right hip joint

After 8 days of hospitalization, the patient was discharged in good general condition. At the 1-week follow-up, we observed a seroma at the distal part of the wound, which was successfully evacuated through digital compression. No further surgical intervention was required. Currently, the patient reports feeling well, although his paraplegia remains unchanged. He continues physical therapy, with substantially improved results compared to before surgery. The passive ROM has been maintained, and transport in a wheelchair is possible, without limitations.

DISCUSSION

Neurogenic HO is difficult to diagnose in its early stages due to ambiguous imaging findings and potentially misleading patient histories from paraplegic individuals. In patients with neurological deficits, signs such as increased limb spasticity, reduced joint ROM, and localized inflammation near a joint strongly suggest the possibility of HO. Early-stage HO may be mistaken for infection and neoplasia.[9]

While magnetic resonance imaging (MRI) of mature HO shows cancellous bone (hyperintense on T1- and T2-weighted images) surrounded by hypointense cortical bone, amorphous calcification and immature HO exhibit nonspecific signal intensities and contrast enhancement patterns, resulting in a heterogeneous appearance.[10] The more immature the HO, the more pronounced these nonspecific features are.[1,10] Tumors, local infections, and abscesses can present with similar MRI characteristics. In such cases, CT imaging can aid in diagnosis by detecting amorphous calcification or immature ossification within the lesion and serves as a reference in the assessment of HO maturity.[1]

As a screening method, Ohlmeier *et al.* recommend biweekly ultrasound examinations conducted by trained radiologists, which, according to Rosteius *et al.*, have a sensitivity of 89%.[11] If HO is suspected, the diagnosis is confirmed through CT and/or MRI.[12] The average time from the traumatic event to HO diagnosis is 8–9 weeks.[12] Studies also highlight the hip as the most common site of HO among patients with spinal cord injury, with an occurrence rate of about 90%.[6,13]

Laboratory findings are not definitive for diagnosing HO. Creatin kinase (CK) is not specific but can indicate muscle involvement, with higher levels correlating with more severe HO, as described by Sherman *et al.*[14] CRP may be elevated but is also nonspecific; however, it correlates more strongly with HO-related inflammatory activity than the erythrocyte sedimentation rate.[15] CRP normalization often coincides with the resolution of soft tissue inflammation, thought to have a key role in the development of HO. Alkaline phosphatase (AP) is the most frequently used laboratory test, but early in HO formation, levels may not be elevated. By 10 weeks post injury, AP levels can increase to 3.5 times the normal value, with serum levels above 250 correlating strongly with HO.[16] However, AP levels can also be falsely elevated due to concurrent lung or bone injuries and are not reliable for assessing the maturity of HO prior to surgical removal. Surgical resection is only recommended once laboratory markers have normalized and the HO has reached its mature phase.

Several risk factors for HO are well documented. According to Citak *et al.*, spasticity, thoracic trauma, complete spinal lesions, pneumonia, tracheostomy, and urinary tract infections significantly increase the risk of developing HO.[13] Notably, our patient exhibited all these risk factors.

For prevention, the most commonly prescribed medication is indomethacin. Selective COX-2 inhibitors appear to be as effective as nonselective NSAIDs, having fewer side effects.[17] Additionally, early single-dose radiation therapy has proven to be a safe and effective prophylactic treatment for HO.[18]

CONCLUSION

HO in patients with spinal cord injuries is a challenging condition. Early diagnosis, prevention, and appropriate management of risk factors are critical to preventing the progression to mature HO and secondary hip ankylosis. Once mature ossification and severe ROM limitation occurs, surgical resection remains the only effective option to restore passive mobility and improve the patient's quality of life.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

ETHICS

This case report was conducted accordance with the principles outlined in the Declaration of Helsinki. Informed consent was obtained from the patient for the publication of this report and any accompanying images.

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