Case Report

Embolization followed by resection of the heterotopic hip joint ossification with spinal cord injury

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ABSTRACT

Background: Heterotopic ossification of large joints, such as knees and hips, has been reported after spinal cord injury, possibly leading to decreased activity of daily living due to a limited range of motion of the affected joint. Therefore, heterotopic ossification resection is performed to improve the range of motion, but it might cause massive bleeding as a complication.

Methods: In this case, the patient had a history of spinal cord injury and developed heterotopic ossification after the left hip injury. He had left hip ankylosis and could not transfer to a wheelchair by himself; therefore, heterotopic ossification resection was planned. On conducting contrast-enhanced computed tomography, the supplying arteries extending to the heterotopic ossification could be identified. A day before the surgery, embolization of the branches by interventional radiology was performed.

Results: Heterotopic ossification resection was performed with an 820-ml blood loss. Postoperative rehabilitation was continued, and range of motion continued to improve without heterotopic ossification recurrence 2 years post-surgery.

Conclusions: The combination of preoperative contrast-enhanced computed tomography and embolization was useful in treating heterotopic ossification.

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1. Introduction

The cause of heterotopic ossification (HO) remains unclear. However, HO often occurs after total hip arthroplasty, brain or spinal cord injury, and acetabulum and elbow fractures [1]. Although the pathogenesis of HO is not elucidated completely, it is known that the local inflammation, hypoxic environment, change in pH, and mechanical stress cause cytokine release and cellular interactions, resulting in progenitor cell transformation from mesenchymal to osteogenic cells and leading to HO [2]. HO is frequently seen in cervical and thoracic spinal injuries and mainly affects the hip joint [3].

HO of the hip joint leads to immobility and a significant decrease in the activity of daily living (ADL) as it progresses. Although surgical resection improves ADL, it can cause massive bleeding due to the proximity of the supplying arteries [4–6]. Recently, with the widespread use of computed tomography (CT), preoperative contrast-enhanced CT has made it easier to evaluate blood vessels [7]. Here we report a case of a patient who underwent contrast-enhanced CT and embolization of supplying arteries followed by HO resection.

2. Report of the case

A 56-year-old male was diagnosed with cervical cord injury (C5, Frankel classification A, Zancolli classification C6A) 6 years ago due to a fall after drinking alcohol. After rehabilitation, he could independently transfer to a wheelchair. Past medical history included neuropathic bladder and angina. He fell from the wheelchair, injured his left hip 9 months ago, and visited a nearby clinic. There were no obvious fractures on radiographs (Fig. 1). In addition, based on the magnetic resonance imaging (short inversion-time inversion recovery), he was diagnosed with muscles (left iliacus, abductors and rectus femoris) and bone (left lesser trochanter) bruise and treated conservatively (Fig. 2a and b). After that, left groin pain and left hip flexion contracture appeared, increasing the burden of care. On return to the clinic 7 months later, the left hip HO was identified on radiographs (Fig. 3a and b), and he was referred to our
department for further examination and treatment. At his first visit to our hospital, he presented with ankylosis of the left hip (fused at 25° on flexion, 20° on abduction, and 10° on external rotation) and could not transfer to a wheelchair by himself. Due to ankylosis of the left hip and resulting limitation of life activity, surgical resection was planned. Preoperative contrast-enhanced three-dimensional CT showed large HO anteriorly at the hip joint (Fig. 4a), and the supplying arteries extending to the HO were detected as the branches of the deep femoral artery (Fig. 4b). After consultation with a radiologist, embolization of the branches by interventional radiology was performed the day before surgery (Fig. 4c). Surgical resection was performed in a supine position under general anesthesia. The HO of the anterior hip was resected piece by piece with a chisel through

Fig. 1. Initial radiograph revealing no obvious fractures of the left hip.

Fig. 2. Initial magnetic resonance imaging (short inversion-time inversion recovery) revealed wide extent of muscle bruise around left iliacus, abductors and rectus femoris. And also revealed bone bruise around left lesser trochanter (a, b).

Fig. 3. Radiographs after seven months revealing heterotopic ossification of the left hip (a, b).
the direct anterior approach. Intraoperatively, a C-arm image intensifier was used to confirm the ossification lesion. The estimated blood loss was 820 mL during the surgery. The range of motion on the left hip improved to 90° on flexion and −10° on extension.

After the surgery, oral administration of bisphosphonate (Etidronate 800 mg/day) and cyclooxygenase (COX)-2 inhibitor (Celecoxib 200 mg/day) was started to prevent an HO recurrence. Prophylactic radiation was not performed after the surgery. Physical therapy was also started. Three weeks later, the patient could transfer to the wheelchair with light caregiving, and he was discharged from the hospital. Fourteen months after the surgery, there was no radiographic evidence of HO (Fig. 5a and b), and the range of motion of the left hip was 90° on flexion, −10° on extension, 10° on abduction, 0° on adduction, 30° on external rotation and 10° on internal rotation. His ADL improved, and he could independently transfer to a wheelchair.

3. Discussion

The patient in this case had a history of spinal cord injury and developed HO after the left hip injury. According to Garland et al., HO after spinal cord injury was almost always evident on

Fig. 4. Preoperative three-dimensional computed tomography (CT) (a) and contrast-enhanced CT (b). The arteries extending to heterotopic ossification are seen as the branches of the deep femoral artery. Preoperative embolization of the branches (c).

Fig. 5. Radiograph after 14 months of heterotopic ossification (a, b).
radiographs within 6 months of injury, and some patients tended to develop HO after 6 months [8]. Although the mechanism of HO after spinal cord injury remains unknown, it is possible that HO in this case was related to the history of spinal cord injury.

Shehab et al. [9] previously reported the criteria for resection of HO, and one of which was a significantly limited range of motion. Kim et al. [6] also reported that a grade III or IV HO, according to the Brooker classification [10], leads to limitations of range of motion, and surgical treatment is needed in order to improve the condition. In this case, owing to ankylosis of the left hip and the resulting limitation of life activities, surgical resection was planned.

One of the major complications of surgical resection of HO is intraoperative bleeding [11]. As increasing vascularization of the HO was reported [5] and HO of the hip joint was adjacent to the femoral arteries and veins, it was possible to lacerate vessels during HO resection and cause massive bleeding. Garland et al. [12] reported a case of a patient in whom surgical resection resulted in a blood loss of 5000 mL due to femoral vein laceration. Contrast-enhanced CT is commonly used to evaluate blood vessel, and can identify supplying vessels as well as the extent of heterotopic ossification. The images taken by contrast-enhanced CT can be discussed with the radiologist to plan for embolization, and can also help surgical planning by identifying the adjacent arteriovenous vessels. For these reasons, contrast-enhanced CT might be useful for HO resection. In our study, the arteries supplying the HO were identified as the branches of the deep femoral artery. Thus, preoperative embolization of these arteries was performed. Consequently, surgical resection could be performed with the 820-mL blood loss.

Verifying that preoperative embolization is useful [13], Kim et al. [6] reported a case of bilateral hip HO with the right side bleeding of 1500 mL whereas the left side underwent preoperative embolization of the superior gluteal and the lateral circumflex femoral arteries, limiting the side bleeding to 500 mL. Zielinski et al. [7] also reported a case of bilateral hip HO with preoperative embolization of both medial femoral circumflex arteries with a bleeding volume of 750 mL and 800 mL on the right and left sides, respectively. Embolization before HO resection might allow a safer surgery with less blood loss. As other reports included the intravenous administration of tranexamic acid during the hip surgery to prevent intraoperative bleeding [14], it might also be effective in hip HO resection.

Garland et al. [12] classified HO into five types according to the neural residua (cognitive and physical disability), and showed that the more severe type was associated with a worse outcome and a higher the recurrence rate. This case was classified as V type with severe physical disability, and was predicted to possibly have a high recurrence rate. For postoperative medications, a COX-2 inhibitor and a bisphosphonate were used. NON-steroidal anti-inflammatory medications are known to prevent HO by inhibiting COX function [15]. As COX-1 inhibitors have gastrointestinal side effects, the use of COX-2 inhibitors should be considered. In Japan, bisphosphonates are commonly indicated for the prevention of HO after spinal cord injury (800–100 mg/day for 3 months). These medications are helpful to prevent HO recurrence, and thus, they were used in this case. Radiation is also effective in preventing HO, but its indications are limited. It was reported that a single dose of 700–800 cGy administered from 24 h preoperatively to 48–72 h postoperatively could prevent HO [16]. However, radiation has serious side effects, including fibrosis, nonunion, and delayed wound healing [17]. HO-preventive radiation therapy is uncommon in our country and was not used in this case because of concerns about delayed wound healing. Although it has not yet been reported that embolization significantly reduces the postoperative recurrence of HO, there are some case reports in which concomitant embolization was useful for preventing HO recurrence [6,7,13]. Additionally several reports have implied the existence of arteries supplying HO when HO has formed [13,18]. Therefore, identification and embolization of the supply arteries may help prevent recurrence. Increasing the number of embolization cases before HO resection will reveal the effectiveness of embolization in preventing recurrence.

Informed consent

The patients and their families were informed that data from the research would be submitted for publication, and gave their consent.

Declaration of competing interest

None declared.

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