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Original Article

Surgical resection of bilateral ankylosed hips caused by heterotopic ossification after complete spinal cord injury

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Abstract: Although heterotopic ossification (HO) is a known complication after spinal cord injury (SCI), HO due to complete SCI is not common, and severe neurogenic heterotopic ossification (NHO) resulting in totally fixed hips is rare. Paraplegic patients are commonly treated with medication and radiotherapy rather than surgical resection. To the best of our knowledge, this is the first case report describing the surgical treatment of bilateral ankylosed hips caused by HO after complete SCI (American Spinal Injury Association (ASIA) scale A). Here we report a case of a 36-year-old Chinese male who sustained a T10 fracture with complete SCI (ASIA scale A) in a gas explosion accident that resulted in paraplegia 5 years previously. Physical examination revealed that the patient had normal consciousness and upper extremities. However, he could not sit up or stay in the wheelchair. There was no sensation or active movement below the umbilical plane. The passive movement of the bilateral hips was zero. Radiologic investigations showed massive HO from the iliac fossa down to the proximal third of both thighs; the Brooker grade was 4 and the DeLee grade was 3. After comprehensive treatment including arterial embolization, radiotherapy, surgical resection, diclofenac, and exercise, the short-term effect was satisfactory.

Keywords: Neurogenic heterotopic ossification, surgical resection, bilateral ankylosed hips, complete spinal cord injury

Introduction

Although heterotopic ossification (HO) is a known complication after spinal cord injury (SCI), HO due to complete SCI is not common, and severe neurogenic heterotopic ossification (NHO) resulting in totally fixed hips is rare. Paraplegic patients are commonly treated with medication and radiotherapy rather than surgical resection. To the best of our knowledge, this is the first case report describing the surgical treatment of bilateral ankylosed hips caused by HO after complete SCI (American Spinal Injury Association (ASIA) scale A).

Case presentation

A Chinese male (36 years old on admission to our department) had sustained a T10 fracture with complete SCI (ASIA scale A) in a gas explosion accident that resulted in paraplegia 5 years previously. He also fractured three ribs in the accident. The local hospital gave him decompression fusion and internal fixation surgery followed by regular physiotherapy. One year later, he had developed pressure sores in both hips; because of a bad outcome with conservative treatment, he then had three skin flap transplantation surgeries. After years of remaining in a prone position, the passive movements of both hips had gradually decreased to zero. There was no family history of abnormal ossification.

Physical examination revealed that the patient had normal consciousness and upper extremities. However, he could not sit up or stay in the wheelchair. There was no sensation or active movement below the umbilical plane. The passive movement of the bilateral hips was zero.

Radiologic investigations showed massive HO from the iliac fossa down to the proximal third of both thighs. The Brooker grade was 4 and the DeLee grade was 3 (**Figures 1** and **2**).

Bone scintigraphy was negative. Serum alkaline phosphatase (ALP), erythrocyte sedimentation

Figure 1. Preoperative posterior-anterior radiograph of the bilateral hips.

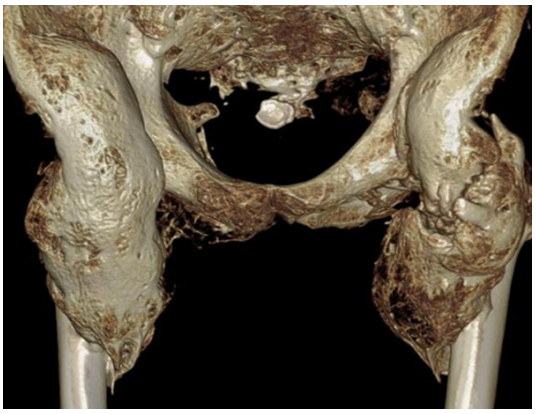

Figure 2. Preoperative 3-dimensional computed tomography reconstruction of the bilateral hips.

rate(ESR) and C-reaction protein(CRP) were all normal.

After admission to our department, the patient and his family received preoperative counseling, including an explanation of the available management options, expected outcome, and possible complications. After extensive discussion, the patient and his family agreed to our comprehensive treatment.

Before the surgical resection, surgeons from the Vascular Surgery Department gave the patient super-selective arterial embolization (**Figures 3** and **4**).

After 1 week, we gave the patient radiotherapy about 4 hours before each surgery. In view of redundant bleeding and surgical stress, the hips were approached in two stages, with the left hip operated on first. We used the Smith-Peterson approach. Surgery included resection

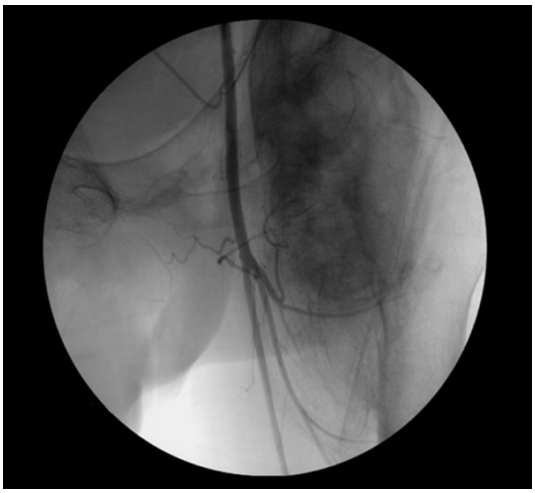

Figure 3. Left femoral angiogram showed the feeding vessels of the HO.

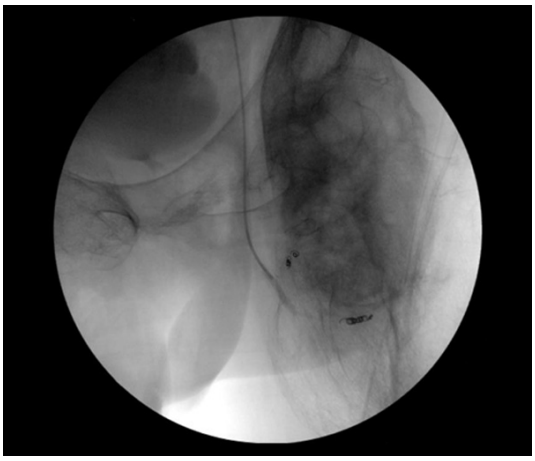

Figure 4. After super-selective arterial embolization, the feeding vessels were blocked.

of the bone masses, with osteotomy conducted to free the joint (**Figure 5**).

The extent of resection was determined by the adequacy of joint movements achieved by manipulation during surgery. The bone masses that did not hinder the joint movements were not removed, in order to shorten the surgical time and minimize the bleeding and infection risk. After the first surgery on the left hip, the patient was given diclofenac 25 mg three times daily; this was continued for 6 weeks after the first surgery until the second surgery on the right hip, and then for another 4 weeks after the second surgery. After each surgery, the patient was exercised via continuous passive movement within the range of pain-free move-

Figure 5. After the surgery, the patient could sit up in bed.

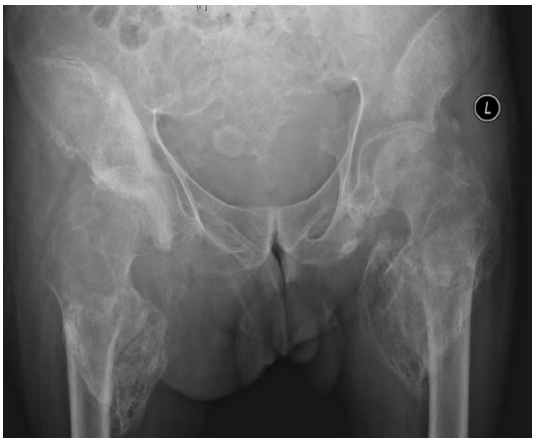

Figure 6. Postoperative posterior-anterior radiograph of the bilateral hips.

ments. At the last follow-up, which was 12 months after the second surgery, both hip movements were acceptable, with flexion of 0-80°. The patient could sit up and stay in the wheelchair. No recurrence was observed on radiography (**Figure 6**).

Discussion

NHO is a known complication after SCI. Regarding complete SCI (ASIA scale A), there are few reports describing surgical resection of bilateral ankylosed hips caused by NHO [1, 2] (**Table 1**).

Our patient is unique in that both of his hips were ankylosed without any motion, he had paraplegia due to complete SCI (ASIA scale A), and he was treated with surgical resection. No previous studies have compared the difference between the management protocols for HO associated with complete and incomplete SCI; hence, our management of this HO induced by complete SCI may provide valuable insights.

The surgical indications for HO associated with SCI include pain, pressure sores, loss of joint function, and loss of the ability to sit [3]. However, for ASIA scale A SCI patients, there have been no definite surgical indications reported. Although we did not have to take walking problems and pain into account for this patient, ankylosed hips may cause many other problems such as hygiene problems around the perineum and resulting increased urinary infection risk. With no flexion of both hips, the patient could only be positioned in bed with the sacrococcygeal region weighted, which increases the risk of pressure sores. The patient complained of an inability to sit up and stay in a wheelchair, remarkably reducing his life quality. Therefore, we considered that our patient had surgical indications and recommended a surgical resection.

The timing of the surgery is very important. Traditionally, the optimal timing of surgery is thought to be after the maturation of HO, as early removal of immature HO causes bleeding and recurrence. Garland put forward a timetable of the timing of the surgical intervention according to the causes of HO: 6 months after trauma, 12 months after SCI, and 18 months after brain injury [4]. It is important that the results of ALP, ESR, CRP and bone scintigraphy are normal, which indicates the maturation of HO. However, delayed surgical resection of HO may lead to irreversible joint injury. Some authors have suggested that the timing of surgery should be based more on the loss of range of movement before ankylosis rather than the maturity of the HO, as early surgical intervention minimizes the development of intra-articular pathology, osteoporosis, and resultant complications, without increasing the risk of recurrence of HO [2, 5, 6]. Others have reported that the neurological status of the patients should be considered in determining the timing of the surgical resection. Sarafis et al. considered poor neurologic condition to be probably the main reason for unsatisfactory outcome after the surgical treatment of hip HO after brain injury [7]; the maturity of HO and the time interval between the brain injury and surgery had little influence in predicting the outcome [7].

Table 1. Previous studies regarding bilateral ankylosed hips due to neurogenic heterotrophic ossification

Authors	Number of cases	Description	Unclear
Garland and Orwin [1]	3	Bilateral ankylosed hips due to SCI	Whether the SCI was complete or incomplete
Genêt et al. [5]	17	Bilateral NHO ankylosed hips	Whether there was brain injury or SCI, whether the SCI was complete or incomplete

SCI: spinal cord injury, NHO: neurogenic heterotrophic ossification.

Because of the pressure sores and skin flap transplantation surgeries that were present when the patient was referred to our hospital, the hips were already ankylosed and the HO was already mature. In our patient, the HO had resulted from complete SCI; hence, we were more concerned with passive movement of the joint rather than intra-articular deterioration. Even if this patient had been referred to us earlier, we might still have delayed the surgical timing until the HO was mature in order to decrease intraoperative blood loss.

The operation is mainly done via open resection, and in order to reduce the operation time, bleeding and infection risk, it is not necessary to remove HO that is not associated with joint movement. Han et al. reported that superselective arterial embolization before surgical resection could notably reduce bleeding in the operation and provide a more clear operative field [8]; it could also reduce the recurrence rate by destroying the blood supply of the HO around the soft tissue [8]. We successfully completed arterial embolization of the left side, however, because of technique problems and the lack of arterial blood supply in the right side HO, the right side was not given embolization. However, the blood loss associated with the left side was significantly larger than that of the right side. This maybe because the right side HO had a lack of arterial blood supply as seen in the arterial angiography. In addition, blood loss during the surgery may result from vein errhysis, which cannot be embolized during arterial embolization. Previous studies on arthroscopic resection and total hip arthroplasty (THA) in treating HO have reported good outcomes [9, 10], but none of these previously reported patients suffered from complete SCI. In our patient with complete SCI, gaining better passive movement was more important than gaining better intra-articular function. Hence, we chose not to perform THA as this would have caused a longer surgical time, higher risk of blood loss and infection, higher surgical difficulties, and higher cost. Arthroscopy was not suitable because of the large size of the bone mass.

To the best of our knowledge, this is the first case report describing the surgical treatment of bilateral ankylosed hips caused by HO after complete (ASIA scale A) SCI. Few previous studies have discussed the management protocol of HO associated with complete SCI. We believe that the appropriate surgical timing should be further elucidated in the complete SCI patient. Surgical resection for complete SCI paraplegic HO patients would improve their quality of life and should be done more actively than it is currently.

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Disclosure of conflict of interest

None.

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