

Management of heterotopic ossification affecting both hips and knees

Devnani A S

ABSTRACT

The management of heterotopic ossification is controversial. This is a rare case affecting both hips and knees of a 22-year-old woman that prevented her from walking. The management and the outcome, three years after excision of the ossific masses, as well as the measures to prevent recurrence after excision of the heterotopic ossification, are discussed.

Keywords: heterotopic ossification, excision of ossific mass, prevention of recurrence

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INTRODUCTION

Heterotopic ossification (HO) occurs in 5%–10% of patients following brain and spinal cord injuries, and can severely restrict the movements of the major joints.⁽¹⁻⁵⁾ Its causes are still not well understood.^(4,6) The effectiveness of pharmacological prophylaxis is not yet irrefutably proven.^(1,2) Prophylactic irradiation has a potential risk of malignancy.^(1,7) There is lack of consensus regarding the timing of the excision of the ossific mass.^(2,5,7) We report a 22-year-old woman with HO of both hips and knees that prevented her from walking.

CASE REPORT

A 22-year-old woman was seen with complaints of inability to walk for the last six years. In April 1998, at the age of 16 years, she was admitted to a hospital for observation of abdominal pain. Two days later, she underwent a laparotomy. Information pertaining to the indications for surgery and operative findings were not available. She did not regain consciousness from anaesthesia and was kept in the intensive care unit for over a month. Two months later, after she had regained consciousness, she was discharged to go home and was given a follow-up appointment. At the time of discharge, she could not walk and was wheelchair-bound. Radiographs showed HO affecting both the knees and hips. She consulted with several doctors regarding the stiffness of both the hips and knees, but her condition did not improve and she could not continue with her schooling. In January 2004, she went abroad for medical consultation and was seen by an orthopaedic surgeon, who advised radiotherapy to each joint followed by joint replacements. As her parents could not afford the cost of the treatment, they returned home and she was subsequently seen in our



Fig. 1a Preoperative radiograph of the pelvis shows extensive heterotopic ossification on the posterolateral aspect of both hips.

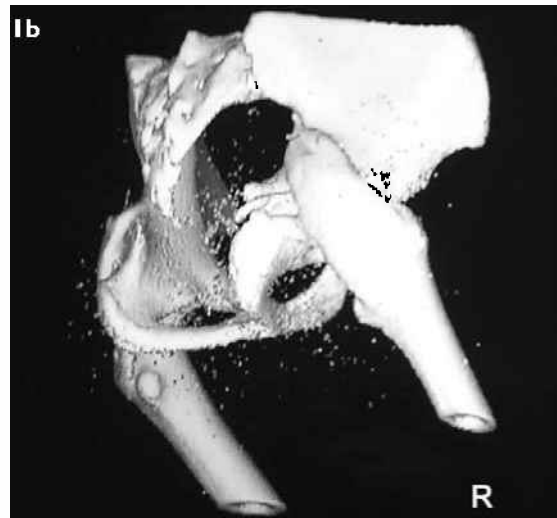


Fig. 1b 3D reconstructed CT image of the right hip shows a large ossific mass on the posterior aspect.



Fig. 1c Postoperative radiograph of pelvis shows no recurrence 18 months after excision of the ossific mass on the right hip, and two years and two months on the left hip. The right hip had 90° arc of flexion and the left hip had 70° arc of flexion at the last follow-up.

orthopaedic clinic.

She attended our clinic in March 2004 in a wheelchair. She had normal higher functions, normal bladder and bowel control, and normal menstrual periods. After the

Department of
Orthopaedics,
Faculty of Medicine
and Health Sciences,
University Putra
Malaysia,
Clinical Campus
at Hospital Kuala
Lumpur,
Kuala Lumpur
50586,
Malaysia

Devnani AS, MS
Associate Professor

Correspondence to:
Dr Devnani AS
Tel: (60) 3 2050 1000
ext 322
Fax: (60) 3 2050 1001
Email:
anandsdevnani@
yahoo.com



Fig. 2a Preoperative radiographs of the left knee show heterotopic ossification along the distal femur.



Fig. 2b Postoperative radiographs of the left knee shows no recurrence 36 months after the excision of the ossific mass. The knee had 100° arc of flexion.

laparotomy and the hospital episode, she developed epilepsy and was on medication for it. Both upper limbs were normal. There was no pain, spasticity, clonus or sensory loss in both lower limbs. There were no ulcers or bedsores. Both hips were stiff; the left was fixed in 30° of flexion and the right in 40° of flexion. There was bilateral wasting of the thigh muscles. The range of motion was 20°–80° in the left knee, and 30°–80° in the right knee. Both ankles and feet were fully mobile. She could take a step or two with great difficulty. The power of hip and knee muscles could not be ascertained as there were contractures. There was no family history of myositis ossificans.⁽²⁾ The blood investigations, including the erythrocyte sedimentation rate and alkaline phosphatase level, were within normal limits.⁽²⁾

Radiographs of the pelvis showed para-articular HO on the posterolateral aspect of both the hips (Fig. 1a). Computed tomography (CT) delineated the extent and location of HO (Fig. 1b). The left knee had HO on the anterior aspect of the distal femur and the lateral femoral condyle (Fig. 2a), and the right knee had HO on the anteromedial aspect of the medial femoral condyle and extended towards the patella (Fig. 3a). The joint spaces were preserved. Both the tibiae were not involved. The patient fell into class III of Garland's classification—severe physical disability with minimal or no cognitive deficit.⁽⁸⁾

Between April 2004 and September 2005, she underwent four operations, one joint at a time with an interval of a few months between the operations, to excise the ossific mass and to regain mobility of the joints. Both knees were done before the hips. The left knee was approached anterolaterally and the right knee anteromedially. Both hips were approached posteriorly.⁽⁹⁾ In each joint, the ossific mass was mature and the periosteum was elevated cleanly. An osteotome was

used to excise the ossific mass in sufficient amounts to 'free' the joints. The articular surfaces of both the knees were well-preserved. The hip joints were not dislocated to inspect the articular surfaces. After each operation, the postoperative recovery was uneventful. All wounds healed primarily. Postoperatively, after about a week, she was encouraged to move the operated joint actively, within tolerable pain limits, under the care of a physiotherapist. Forcible and passive movements were avoided. Pharmacological or local radiotherapy prophylaxis was not used. Histopathological examination of the excised ossific mass showed normal lamellar bone and no abnormality.

At the last follow-up visit, 18 months after the last operation and three years after the first operation, she had travelled by public transport to attend the clinic. She had no pain and could walk independently albeit slowly. The range of flexion was 10°–100° in the right hip, and 10°–90° in the left hip. The range of abduction and adduction was 0°–20° each, in both the hips. The right knee had a range of flexion of 0°–120°, and the left knee from 0°–100°. Motor power was grade 5 in both quadriceps. Both ankles and feet remained mobile and painless. The radiographs of both the hips and the knees showed no recurrence of HO (Figs. 1c, 2b & 3b).

DISCUSSION

HO may occur after brain or spinal cord injury, or after direct trauma to the joints. Its mechanism of formation is not well established.^(4,6) After the HO has begun, it cannot be stopped,⁽⁷⁾ nor does it resolve spontaneously. The emphasis should be on prevention.⁽¹⁾ Unfortunately, it is not easy to predict which patients are likely to develop HO,⁽¹⁾ so that preventive measures could be taken. Serum alkaline phosphatase level is a poor indicator.^(2,3,8,10) Radioisotope scans do not help to predict the recurrence.^(4,9)



Fig. 3a Preoperative radiographs of the right knee show heterotopic ossification involving the anteromedial aspect of the distal femur and the medial femoral condyle.



Fig. 3b Radiographs of the right knee 31 months after excision of the ossific mass show no recurrence. The knee had 120° arc of flexion.

Radiographs are helpful in evaluating the maturity of HO, especially when coupled with clinical examination.⁽²⁾

There are no completely effective prophylactic measures.^(7,8) Oral indomethacin, and recently, cyclooxygenase-2 inhibitors, have been used for prevention of HO in patients undergoing elective total hip replacement or internal fixation of acetabular fractures, although it has not yet been universally proven to be effective.^(4,11) Some of these medications have various side effects, such as gastrointestinal intolerance and bleeding, antiplatelet activity that may interfere with anticoagulation therapy. This interference may adversely affect fracture healing and impaired bone ingrowth, which may in turn affect the long-term fixation of uncemented prosthesis.^(7,11) Local radiotherapy, up to 24 hours before, or 24–48 hours after, the surgical excision of the HO, has been advocated to prevent recurrences after total hip arthroplasty.^(1,4,7,12,13) The logistics of transferring a recently-operated patient to the radiotherapy centre can be challenging. Radiotherapy also increases the risk of infertility and sarcomatous change, particularly in those with multiple joint involvement with a high dose requirement.^(1,7)

Recurrence after surgical excision is worrying. A delay of up to 18 months before surgical excision has been advocated to allow for maturation of the HO and neurological recovery, thereby reducing the risk of recurrence.^(4,5,7,13) The other prophylactic measure is to keep the soft tissue dissection and periosteal stripping to a minimum during excision.⁽¹⁾ These preventive measures were preferred for this patient. She had no spasticity in both lower limbs. The joints were quiescent with no local signs of inflammation; plain radiographs showed mature bone formation. The erythrocyte sedimentation rate, leucocyte count, serum calcium and serum alkaline

phosphatase were repeatedly within normal limits. There was no difficulty in deciding the timing of the operation as the patient presented after six years. All these are good prognostic indicators for nonrecurrence of HO.^(2-4,8,9) Drugs were not used as their efficacy has been reported mostly in patients undergoing elective joint replacement,⁽¹¹⁾ and not in patients with neurogenic HO.⁽²⁾ Radiotherapy was not used as this patient had four large joints affected. The total dose required may engender a risk of developing malignancy or infertility.^(1,7)

The aim of surgery for this patient was to increase the range of movements at the joints, and thereby improve function and quality of life. It was decided to excise the ossific masses to free the joints rather than do joint replacements as the patient was very young. The ossific masses were extra-articular in both the hips, and the joint spaces were well preserved. The risk of recurrence is greater with joint replacements as this involves more tissue dissection,⁽⁴⁾ and with recurrence, subsequent salvage would be difficult. The costs involved are also higher than with simple excision of the ossific mass. The risk of recurrence is also higher in patients undergoing multiple operations simultaneously.^(6,8) Therefore, in the present case, we operated on one joint at a time as a precautionary measure. We decided to do the knees before the hips as the ossific masses were smaller and the procedure on the knees was easier. Tissues were cut neatly and not torn.⁽¹⁾ Muscles were separated along anatomical planes. The periosteum was elevated as a continuous layer rather than torn or shredded. A sufficient amount of bone was excised to free the joints. It was not necessary to excise all the heterotopic bone. The wound was washed thoroughly with saline to remove all the bone dust and debris, which may act as a seed for recurrence. Suction drains were used to prevent

haematoma formation. A week after the operation, when the wound pain had settled, the patient was encouraged to move the joints as actively as pain tolerance can allow. Passive mobilisation exercises were avoided for the fear of inciting tissue reaction and consequent recurrence of HO.⁽⁴⁾ The range of movement in all the joints improved gradually with use and exercise, and at the last follow-up, it was more than what was obtained intraoperatively.

This patient had certain uncharacteristic features. Males are affected more often than females with HO.⁽⁶⁾ Elbows are involved more frequently, when the duration of coma is more than a month.^(4,5) Knees are rarely affected after brain injury, but more commonly after spinal cord injury.^(4,9,10) The ossific mass is more likely to be on the anterior aspect of the hips rather than on the posterior aspect.^(3,6,8) Recurrences are more common when three or more joints are involved.⁽⁸⁾ Results are poor, with little or no improvement, when four major joints are involved.⁽⁸⁾

In conclusion, a 22-year-old woman underwent laparotomy under general anaesthesia at 16 years of age, but did not regain consciousness for over a month. She developed HO in her hips and knees that prevented her from walking for six years. The HO was excised to free the joints in a staged manner, one joint at a time. She had no recurrence three years after excision. Aside from medication or local radiotherapy, we have discussed patient selection factors and other favourable factors which may reduce the recurrence rate of HO. Favourable prognostic indicators for low recurrence include: adult patients with good neurological recovery following brain injury, radiographical evidence of a mature HO lesion, and a duration of at least 18 months following HO onset. Dissection should be kept to a minimum and soft tissues must be handled with care. The periosteum should be elevated as a continuous, rather than torn, layer. Sufficient ossific mass should be removed to free the joint. Complete excision is not necessary. The wound should be washed thoroughly and a suction drain should be used. Active

exercises can begin after the first postoperative week, but on no account should the joint be manipulated or put through a passive range of motion exercises.

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