

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/mjafi

Case Report

A rare case of intra-articular heterotopic ossification of knee following intra-medullary nailing of fracture tibia in a patient with fat embolism

Col B. Hari Krishnan ^{a,*}, Brig Yogesh Sharma, vsm^b, Maj Anjan Prabhakara^c^a Associate Professor, Department of Orthopaedics, Armed Forces Medical College, Pune 411 040, India^b Professor & Head, Department of Orthopaedics, Armed Forces Medical College, Pune 411 040, India^c Clinical Tutor, Department of Orthopaedics, Armed Forces Medical College, Pune 411 040, India

ARTICLE INFO

Article history:

Received 21 September 2015

Accepted 7 January 2016

Available online 2 March 2016

Keywords:

Heterotopic ossification

Intramedullary nailing

Fat embolism

Introduction

Heterotopic ossification (HO) is the formation of bone in non-skeletal soft tissues where it is normally not expected to form. Development of HO was observed and reported by Reidel and Ceilliar among patients with spinal cord injury in World War I.² Gerhard Küntscher, the pioneer of intramedullary nailing, reported 'callus caps' in the soft tissues around the hip after femoral nailing in the 1960s.³

HO is known to occur following a variety of situations such as severe muscular injury, following total hip and knee replacement surgery, severe head and spinal cord injury, prolonged assisted ventilation, prolonged immobilisation, ARDS, burns and pancreatitis.^{1,4–7}

The pathogenesis of HO is unknown. An imbalance between certain forms of bone morphogenetic protein and their antagonists has been suggested as likely precipitating factor in development of HO. Mesenchymal stem cells have been noticed to differentiate into bone via the endochondral pathway due to over-expression of BMP in the traumatised soft tissues.^{1,6}

Majority of orthopaedic literature have focused on HO in the hip subsequent to operative fixation of hip and femur pathology.^{8,9} Few cases of HO of the ligamentum patellae after intramedullary nailing of the tibia have been reported in literature.^{10–12} However, till date there have been no reported cases of intra-articular HO in the knee involving the entire retropatellar fat pad and ligamentum patellae following intramedullary nailing of a tibial shaft fracture in the setting

* Corresponding author. Tel.: +91 8390489661.

E-mail address: hari_os@yahoo.co.in (B. Hari Krishnan).<http://dx.doi.org/10.1016/j.mjafi.2016.01.004>

0377-1237/© 2016 Published by Elsevier B.V. on behalf of Director General, Armed Forces Medical Services.

of fat embolism. In symptomatic HO of knee, excision of HO mass may be required to improve joint mobility. Here, we present a case of a young individual with symptomatic intra-articular HO in the knee following closed intramedullary nailing of a tibial shaft fracture in the setting fat embolism managed successfully by excision of the heterotopic bone mass.

Case report

This case is of a 24-year-old male patient who suffered a fracture of distal third shaft of Tibia and Fibula on the left side with no other injuries following a two wheeler accident on 10 May 2014. He was managed at a nearby private hospital with initial resuscitation and first aid. However, within the first 24 h of injury, the patient developed features of fat embolism in the form of altered sensorium, blurring of vision associated with respiratory distress warranting the need for mechanical ventilation. After haemodynamic stabilisation, the patient was transferred to our hospital for further management on 12 May 2014.

On arrival at our centre, the patient was on mechanical ventilator with a GCS of $E_2V_T M_4$. However, rest of the vital parameters were normal. Systemic examination revealed decreased air entry in right infra axillary and infra-scapular region. Clinico-radiologically, he had a closed fracture distal third shaft of Tibia and Fibula on the left side without any distal neurovascular deficits. Fundoscopy was normal. MRI brain done on the day of injury was unremarkable, D-dimer level was 834 ng/ml (normal <200 ng/ml) and haematological and biochemical parameters showed leucocytosis with raised serum creatinine and liver enzymes.

The patient was managed by closed reduction and internal fixation of left Tibia by unreamed intramedullary interlocking nail on 14 May 2014. Surgery was performed using a patellar tendon splitting approach. Post-operatively, the patient

showed a steady recovery in his haematological and biochemical parameters. He was weaned off mechanical ventilator on the 8th post-op day and sutures were removed on 14th post-op day. The patient was rehabilitated with physiotherapy, touchdown weight bearing ambulation and discharged for home based convalescence with a knee range of motion of 5–120°.

On review after 6 weeks, the patient complained of restricted range of motion left knee. Clinically, he had 2 cm of thigh wasting, well healed operative wounds and a ROM of 20–45° in left knee without any distal neurovascular deficits. He had a palpable non-tender mass in the infra patellar region of left knee anteriorly. Neither erythema nor swelling was noted. AP and lateral radiographs and CT scan of the left knee revealed HO in the retropatellar fat pad involving the ligamentum patellae (Figs. 1 and 2a, b). The patient was diagnosed with symptomatic HO within the left knee and offered surgical excision after ascertaining normal serum alkaline phosphatase levels at a week's interval (ALP – 79 IU/L and 51 IU/L). Within 2 weeks, he underwent excision of the HO mass adherent to ligamentum patellae. Per-operatively, the entire retropatellar fat pad was an irregular bony mass severely restricting flexion of his knee (Figs. 3 and 4). Post-operative radiographs confirmed complete excision of heterotopic bone mass (Fig. 5). Pathology report confirmed HO. Post-operatively, the patient was put on oral indomethacin and physiotherapy. Following up at six months, the individual had a healing fracture shaft tibia with normal range of motion in the knee and no recurrence of HO.

Discussion

HO is the formation of trabecular bone in extra osseo-articular soft tissues where it does not normally exist. It is a known complication arising in critical care patients, causing significant long-term morbidity. The condition results in progressive

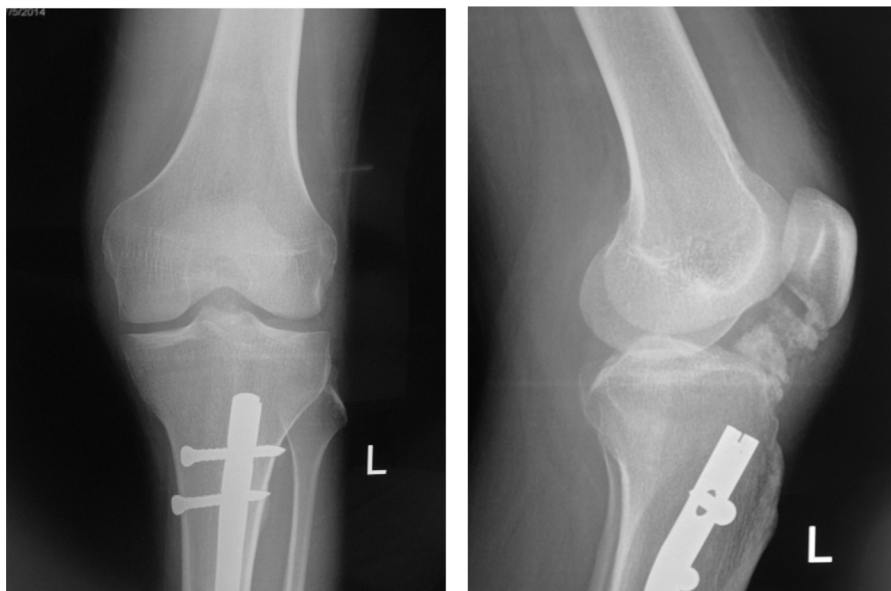


Fig. 1 – AP and lateral radiographs of the left knee showing heterotopic ossification in the retropatellar fat pad involving the patellar tendon.

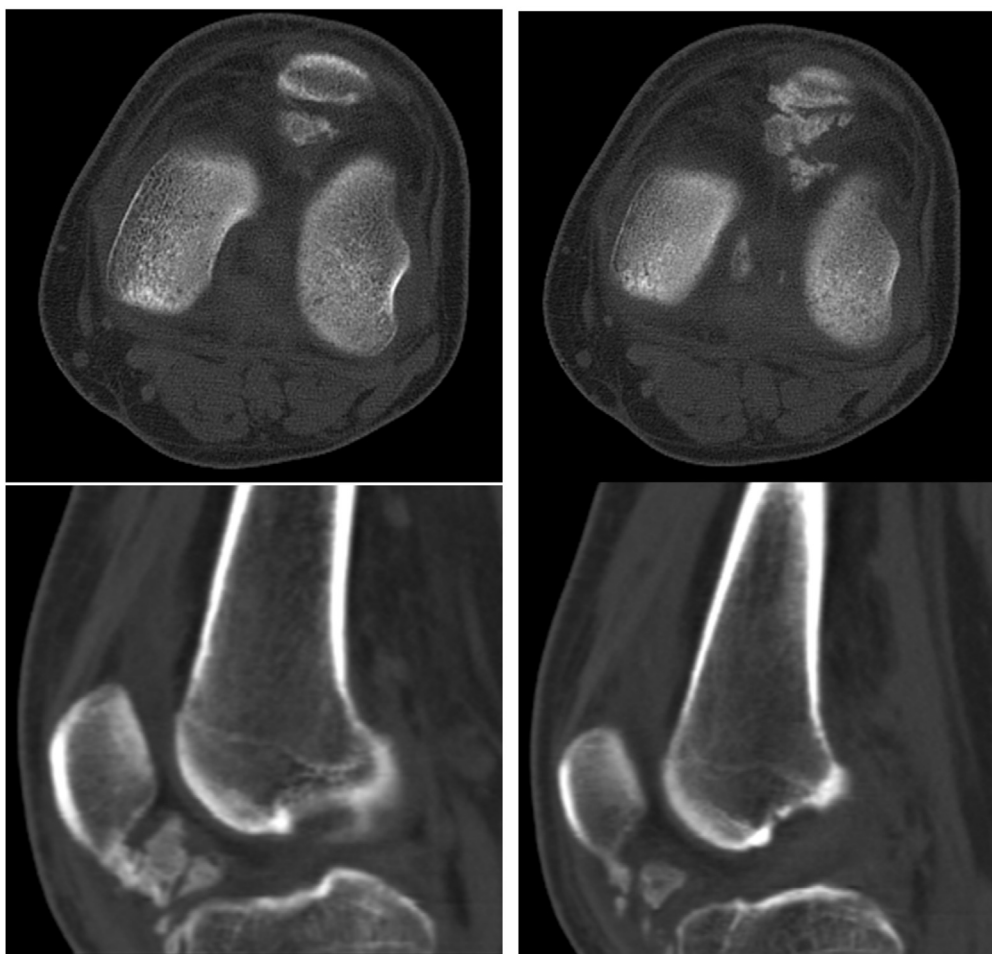


Fig. 2 – Axial and sagittal section computed tomography images (bone window) of left knee showing heterotopic ossification mass in the retropatellar fat pad.

restriction of movements and pain in the adjacent joints in the months after discharge. The condition is also a common complication in patients with head or spinal cord injury, pancreatitis and burns.⁶ Our patient had fat embolism for

which he was on ventilator support for a week following the injury.

To date, there has been no reported case of intra-articular HO of knee following nailing of tibia in a patient of fat

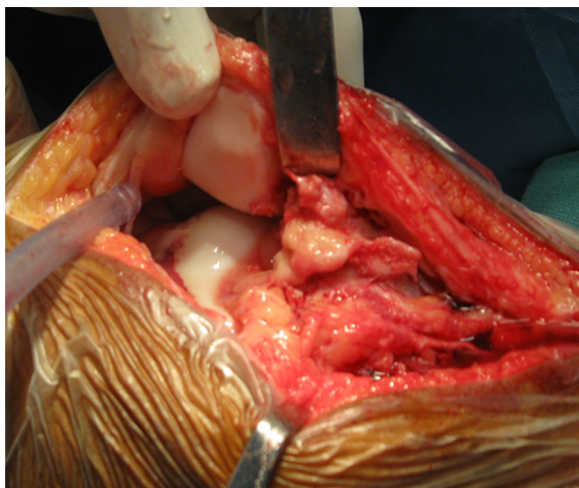


Fig. 3 – Per-operative photographs of the heterotopic ossification mass seen in retropatellar fat pad.



Fig. 4 – Photographs of the heterotopic ossification mass excised from retropatellar fat pad.

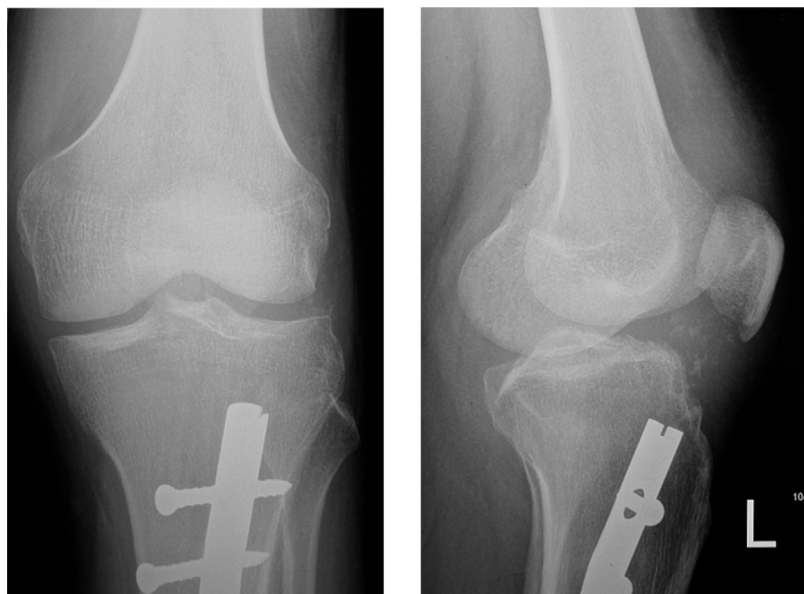


Fig. 5 – AP and lateral radiograph of the left knee following excision of heterotopic ossification mass.

embolism with no evidence of primary neuro trauma such as brain or spinal cord injury. We present a case of symptomatic HO of retro patellar fat pad following intramedullary nailing of an open fracture shaft tibia with fat embolism and no evidence of primary neuro trauma. In previously reported cases of HO in the knee joint following either trauma or neurogenic causes, the lesion was located in vastus medialis muscle.^{4,13,14,16} In our patient, almost the entire retropatellar fat pad had become a HO mass requiring excision.

Severe head or spinal cord injury, muscle injuries and burns are well-recognised predisposing conditions for the development of HO.^{1,4-6} Sedation and mechanical ventilation may also play a role in the development of HO by inducing an environment similar to neurogenic HO arising out of fluctuations in local tissue partial pressure of oxygen and pH.^{4,5,15} Bone morphogenetic protein released from mesenchymal cells in para articular tissue has also been implicated in the pathogenesis of HO.^{16,17}

There are limited safe treatment options for established HO. NSAIDs and irradiation are common preventive measures prescribed for HO occurring after total hip replacement.¹⁰ Excision of heterotopic bone is an option, but only after full maturation has occurred or else recurrence is inevitable.¹¹ After surgery, bisphosphonates may be prescribed to prevent further recurrence.¹² However, no therapies have been proven to be of benefit in prevention or arrest of development of HO in critical care patients. Prophylactic radiation therapy and NSAIDs following surgery are associated with many hazards and shortcomings. They are known to result in nonunion, gastrointestinal disturbances, and in the case of radiation therapy, carcinogenesis. Keeping in view the hazards and shortcomings of various known modalities of treatment, we managed our patient by excision of the HO mass.

HO is generally excised after it matures to prevent recurrence. Delaying surgical excision of intra-articular HO of knee to prevent a recurrence may be more debilitating and

make future surgical treatment outcome unpredictable and difficult. Hence, early surgery with prophylaxis against recurrence of HO may be required in certain situations and the exact timing of surgical intervention for an immature HO causing progressive ankylosis of a joint is still controversial.

Symptomatic intra-articular HO after intramedullary nailing of long bone fractures represents a rare post-operative complication in most cases in our clinical practice. As outlined in the present case report, patients may rarely develop significant HO characterised by pain and compromised joint function, up to full joint stiffness. Osteogenic reaming debris and operative soft-tissue injury represent the main root causes of HO after nailing. Meticulous intraoperative care of soft tissues to avoid local seeding of reaming debris and copious fluid irrigation are parameters that can be influenced by the surgeon in prevention of development of HO. Further research is required to fully understand the pathogenesis of HO and to determine risk factors, root causes, and preventability of this potentially debilitating complication.

Conflicts of interest

All authors have none to declare.

REFERENCES

1. Kaplan FS, Glaser DL, Hebel N, Shore EM. Heterotopic ossification. *J Am Acad Orthop Surg.* 2004;12:116-125.
2. Dejerme A, Ceillier A. Para-osteo-arthropathies des paraplegiques par lesion medullaire; etude clinique et radiographique [Para-osteo-arthropathy in paraplegics due to medullar lesion; clinical and radiological study]. *Ann Med.* 1918;5:497.

3. Marks PH, Paley D, Kellam JF. Heterotopic ossification around the hip with intramedullary nailing of the femur. *J Trauma*. 1988;28:1207-1213.
4. Sugita A, Hashimoto J, Maeda A, et al. Heterotopic ossification in bilateral knee and hip joints after long-term sedation. *J Bone Miner Metab*. 2005;23:329-332.
5. Pape HC, Lehmann U, van Griensven M, Gansslen A, von Glinski S, Krettek C. Heterotopic ossifications in patients after severe blunt trauma with and without head trauma: incidence and patterns of distribution. *J Orthop Trauma*. 2001;15:229-237.
6. Shehab D, Elgazzar AH, Collier BD. Heterotopic ossification. *J Nucl Med*. 2002;43:346-353.
7. Jacobs JW, De Sonnaville PB, Hulsmans HM, van Rinsum AC, Bijlsma JW. Polyarticular heterotopic ossification complicating critical illness. *Rheumatology*. 1999;38:1145-1149.
8. Kaempffe FA, Bone LB, Border JR. Open reduction and internal fixation of acetabular fractures: heterotopic ossification and other complications of treatment. *J Orthop Trauma*. 1991;5(4):439-445.
9. Furlong AJ, Giannoudis PV, Smith RM. Heterotopic ossification: a comparison between reamed and unreamed femoral nailing. *Injury*. 1997;28(1):9-14.
10. Horne LT, Blue BA. Intraarticular heterotopic ossification in the knee following intramedullary nailing of the fractured femur using a retrograde method. *J Orthop Trauma*. 1999;13(5):385-388.
11. Gosselin RA, Belzer JP, Contreras DM. Heterotopic ossification of the patellar tendon following intramedullary nailing of the tibia: report on two cases. *J Trauma*. 1993;34(1):161-163.
12. Tornetta III P, Barbera C. Severe heterotopic bone formation in the knee after tibial intramedullary nailing. *J Orthop Trauma*. 1992;6(1):113-115.
13. Akgun I, Erdogan F, Aydingoz O, Kesmezacar H. Myositis ossificans in early childhood. *Arthroscopy*. 1998;14:522-526.
14. Saito N, Horiuchi H, Takahashi H. Heterotopic ossification in the knee following encephalitis: a case report with a 10-year follow-up. *Knee*. 2004;11:63-65.
15. Brighton CT, Schaffer JL, Shapiro DB, Tang JJ, Clark CC. Proliferation and macromolecular synthesis by rat calvarial bone cells grown in various oxygen tensions. *J Orthop Res*. 1991;9:847-854.
16. Chalmers J, Gray DH, Rush J. Observations on the induction of bone in soft tissues. *J Bone Joint Surg Br*. 1975;57:36-45.
17. Urist MR, Nakagawa M, Nakata N, Nogami H. Experimental myositis ossificans: cartilage and bone formation in muscle in response to a diffusible bone matrix-derived morphogen. *Arch Pathol Lab Med*. 1978;102:312-316.