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Fracture of tibial tuberosity in an adult with Paget's disease of the bone – An interesting case and review of literature

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ABSTRACT

Introduction: Paget's disease of the bone is a non-malignant skeletal disorder characterized by focal abnormalities in bone remodelling at one or more skeletal sites. Pathological fractures occurring from trivial injuries are a well-known clinical presentation in patients with Paget's disease. An avulsion fracture of the tibial tuberosity is an infrequent injury and has an extremely low occurrence in adults, with only a few cases reported in literature. We describe a case of a patient with undiagnosed Paget's Disease of the bone, sustaining a pathological avulsion fracture of the tibial tuberosity.

Case report: A 54-year-old male presented with right knee pain after his knee gave way whilst standing in the goal area during a game of football, twisting his right ankle and falling. Plain radiographs of the knee revealed an avulsion fracture of the tibial tuberosity with abnormal modelling of the proximal half of the tibia. An MRI confirmed a diagnosis of Paget's disease of the bone. The patient underwent open reduction internal fixation. At 3 months follow up, the patient had good knee range of motion from 0 to 100 degrees and by 6 months he had returned to his usual activities.

Conclusion: We describe a unique case of tibial tuberosity avulsion fracture in an adult with PDB. Treatment was successful with cannulated screws and tension band wiring. Patients with PDB who fracture present with diagnostic and operative challenges, it is vital to progress with caution in the postoperative rehabilitation phase.

Introduction

Avulsion fractures of the tibial tuberosity are an extremely infrequent injury in adults, with only a few case reports described in literature. Fractures of the tibial tuberosity constitute only 3% of proximal tibial fractures [1]. The mechanism of injury in avulsion fractures of the tibial tuberosity in adults has not been fully elucidated, however it has been attributed to direct trauma to the tubercle [2], as opposed to the classical injury due to eccentric contraction of the quadriceps during pushing off or landing whilst jumping seen

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in adolescents [3]. We describe a case of a 54-year-old male patient with undiagnosed Paget's Disease of the bone (PDB), who sustained a pathological tibial tuberosity avulsion fracture during low impact activity, case management and outcomes.

Case report

A 54-year-old male presented with right knee and right ankle pain after his knee gave way whilst standing in the goal area during a game of football, resulting in a fall and twisting of his right ankle. He was unable to weight-bear afterwards. On examination, he had significant swelling and tenderness anteriorly along the proximal tibial shaft with a moderate effusion in the knee. He also had a swollen right ankle with tenderness over the lateral malleolus and could not actively straight leg raise or extend the right knee. The patient had a past medical history of asthma, vitamin B12 deficiency and mobilised independently.

Plain radiographs of the knee and ankle revealed an avulsion fracture of the tibial tuberosity with antero-superior displacement and overlying soft tissue swelling. It was noted that there was abnormal modelling of the proximal half of the tibia [Figs. 1 and 2]. Furthermore, there was an oblique fracture of the distal fibula at the level of the syndesmosis. An MRI scan was performed to obtain a better understanding of the underlying pathology and this revealed trabecular signal irregularities consistent with a diagnosis of Paget's Disease involving the tibia [Figs. 3 and 4]. Previous radiographs which had been taken in 2015 also confirmed Paget's disease [Fig. 5].

On the second day of admission the patient developed shortness of breath and had a syncopal episode on the ward; an urgent Computerised Tomography pulmonary angiogram (CTPA) showed multiple filling defects in the right middle and lower lobe pulmonary arteries in keeping with pulmonary embolism. However bilateral Doppler ultrasound scans of his legs were normal. On the advice of the Haematologist, the patient was started on treatment dose Dalteparin, and surgery was delayed for 2 weeks, with further advice to use subcutaneous unfractionated heparin infusion during the peri-operative period.

Intraoperatively, the synovial sheath of the knee was found open and the bone quality around the tibial tubercle was very poor, with an extremely soft consistency. The fracture fragment was reduced to its footprint and fixed with a single AP 4 mm cannulated screw with a washer and supplemented with a tension loop through another cannulated screw distally. Additionally, a robust retinacula repair was performed. An attempt to place an intraosseous suture was shelved as this could have easily cut through the bone. The reconstruction was stable despite the deficient bone integrity even at 90 degrees of flexion. An open reduction internal fixation of the ankle was performed at the same sitting. Postoperatively, the patient was immobilised in an above knee backslab.

We followed-up the patient three weeks postoperatively; his wounds were clean and dry and postoperative radiographs were satisfactory [Figs. 6 and 7]. The backslab was converted to an above knee full plaster cast, and the patient remained non-weight bearing. At the subsequent follow-up 6 weeks post-operatively, the full plaster cast was removed, and the knee was fitted into a

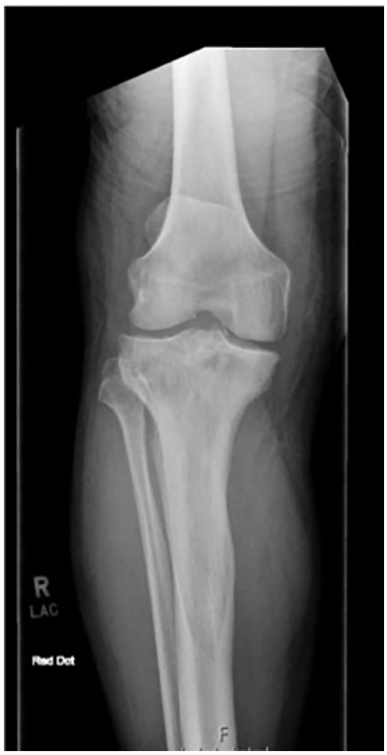

Fig. 1.. Right knee AP radiograph showing avulsion fracture of the tibial tuberosity.

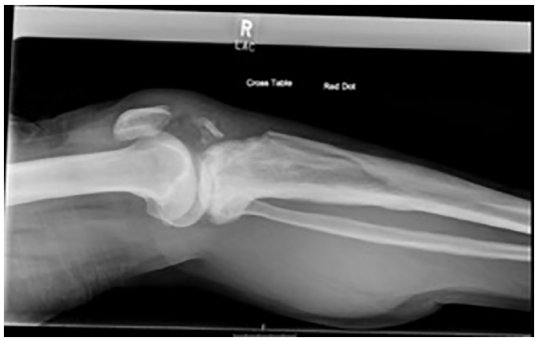

Fig. 2.. Right knee lateral radiograph showing avulsion fracture of the tibial tuberosity.

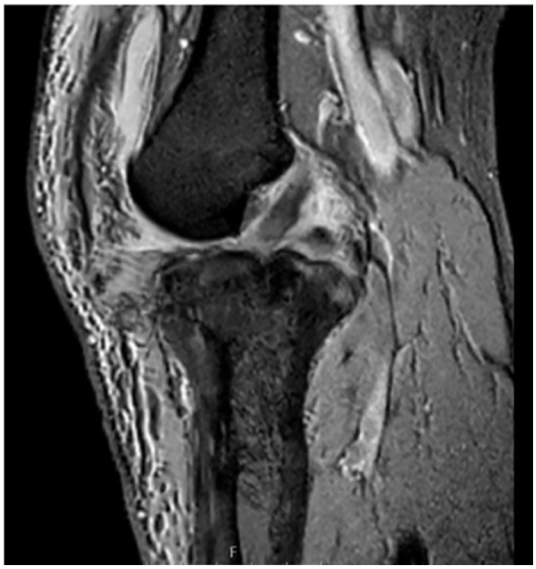

Fig. 3.. MRI scan revealing marked Paget's disease of the proximal tibia.

range of motion brace limiting flexion from 0 to 45 degrees with progressive weight-bearing. The range of flexion was increased by 20 degrees every 2 weeks. At 12 weeks postoperatively, the patient was able to mobilise unaided without the brace, he could extend the knee with no extension lag and flex to 100 degrees. Unfortunately, he developed a pyogenic granuloma on the surgical site which was successfully treated with cautery and a course of oral antibiotics. Repeat radiographs showed radiological signs of union and no evidence of failure of the fixation construct.

Discussion

Avulsion fractures of the tibial tuberosity are an extremely uncommon injury in the adult population [4]. In contrast, such injuries are more common in athletic male adolescents during physiologic physiodesis of the proximal tibial tubercle [5]. The mechanism of injury in adults has not been clearly described, however it has been attributed to direct trauma as previously stated. A sudden eccentric contraction of the quadriceps in adults is more likely to disrupt the more proximal and relatively weaker structures of the extensor apparatus as opposed to avulsing the tibial tubercle. Nevertheless, in this case the 'diseased, fragile bone' at the attachment of the patella tendon on the tibial tuberosity due to Paget's disease, ostensibly presented the weakest link in his extensor mechanism. Compartment syndrome, loss of range of motion and bursitis are some of the complications that can occur after an avulsion injury of the tibial tuberosity [6,7].

PDB is a benign skeletal disorder illustrated by focal abnormalities in bone remodelling at one (monostotic) or more (polyostotic) skeletal sites. The condition is rare in people under the age of 50 years but becomes more common with age [8]. PDB can affect any skeletal site, but it is mostly seen in the pelvis, spine, femur, tibia, and skull [9]. The cause of Paget's disease is unknown, however there is a strong genetic component with 15%–20% of those affected having first-degree relative with PDB [10,11]. Risk factors for PDB include male sex, increasing age, and ethnic background [12].

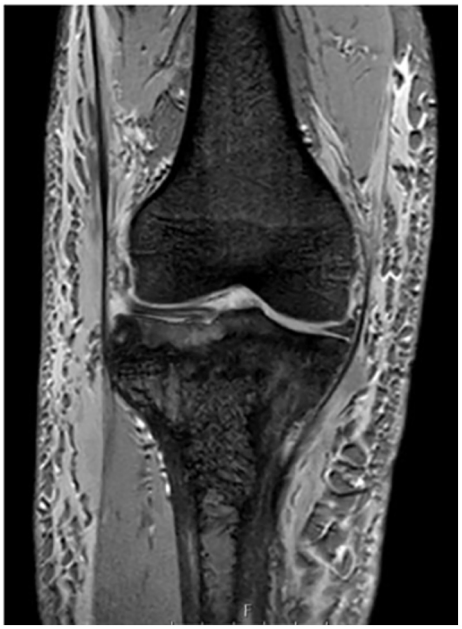

Fig. 4.. MRI scan revealing marked Paget's disease of the proximal tibia.

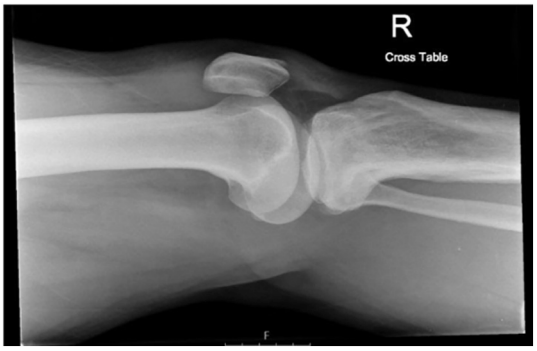

Fig. 5.. Right knee lateral radiograph from five years ago revealing similar picture to Figure 1.

At a cellular level, PDB is characterized by an increase in numbers and activity of osteoclasts combined with a complimentary increase in osteoblast activity [13]. This results in an increased and disordered formation of bone (woven bone) which is mechanically weak and prone to deformity and fractures. The focal increases in osteoclast and osteoblast activity are also coupled with marrow fibrosis and increased bone vascularity [14]. Bone marrow and circulating osteoclast precursors exhibit a heightened sensitivity to factors known to stimulate bone resorption such as 1,25 dihydroxy vitamin D and receptor activator of NF-kB ligand (RANKL) [15].

The bone may be hyperaemic, deformed, osteosclerotic or osteolytic [16], is less resilient to plastic deformation and at risk of crack propagation. PDB is also associated with poor fracture healing after surgical fixation leading to delayed union or non-union [17]. Metabolic control of the fracture and disease advancement is evolving and entails the use of calcitonin and bisphosphonates [18].

Conclusion

We describe a rare injury of the tibial tuberosity in an adult with PDB. Treatment was successful with cannulated screws and tension band wiring. To our knowledge this type of injury in a patient with concomitant PDB is unique and has not been reported in the past. Pathological fractures in patients with PDB present clinicians with diagnostic and operative challenges not only in terms of achieving a stable fixation, but also fracture union. A prompt diagnosis, early repair and cautious functional rehabilitation are pertinent goals of treatment to obtain the best functional outcome for the patient.

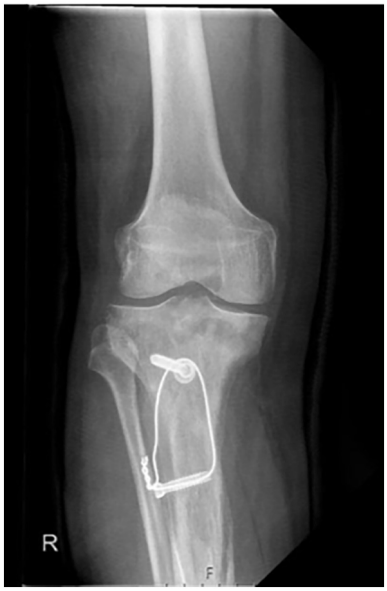

Fig. 6.. AP radiograph of the knee taken three weeks postoperatively showing fixation with cannulated screws and tension band wire.

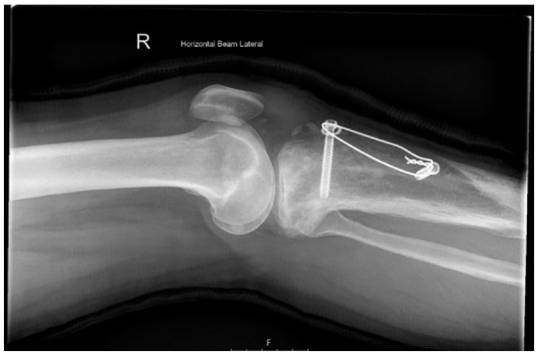

Fig. 7.. Lateral radiograph of the knee taken postoperatively.

Clinical message

Paget's disease of the tibia and tibial tuberosity fracture in an adult are both rare conditions. With an underlying bone disease such injuries can present after minimal trauma. When presented together the diagnosis and management can be challenging. A detailed evaluation of the patient with imaging such as MRI preoperatively is pertinent and postoperative rehabilitation should be monitored and undertaken with caution.

CRediT authorship contribution statement

Marjan Raad: Investigation, conceptualisation, methodology, Visualisation, Roles/Writing – original draft.
Sebastian Ndlovu – conceptualisation, review and editing.
Tord Hurtig Hogsand – review and editing.
Saif Ahmed: Supervision, writing – review and editing.
Mark Norris: Supervision, validation, writing – review and editing.

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Nil.

Declaration of competing interest

The authors declare that they have no competing interests.

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