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Case report Stress fracture of proximal tibia after proximal fibula osteotomy: A case report $\stackrel{\star}{\sim}$

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A R T I C L E I N F O	A B S T R A C T	
Keywords: Proximal fibula osteotomy (PFO) Stress fracture Fatigue fracture Proximal tibia fracture Rheumatoid arthritis (RA)	Introduction: Proximal fibula osteotomy (PFO) is a new method for treating medial compartment osteoarthritis of the knee, which is based on the theory of differential settlement (nonuniform settlement). This procedure has been widely recognized for its advantages of relative simplicity, low rate of postoperative complications, and low postoperative costs. Stress fracture of the proximal tibia after PFO has not been previously reported. <i>Case presentation:</i> We report a 62-year-old woman with chronic rheumatoid arthritis (RA) underwent left PFO for chronic knee pain, who developed a stress fracture of the proximal tibia more than 1 year after PFO. <i>Clinical discussion:</i> In the early stage of proximal tibia stress fracture, due to the concealment of radiography manifestations, doctors from another hospital performed total knee arthroplasty (TKA) for the patient. They ignored the treatment of stress fracture of the proximal tibia, and the stress fracture was further aggravated after surgery. Six months later, the patient underwent open reduction and internal fixation with a plate and screw in the left proximal tibia fracture at our hospital. The patient was followed up at the hospital three months after open reduction, and the proximal tibia stress fracture began to heal. <i>Conclusion:</i> RA is usually not confined to the medial compartment and its pathogenesis is different from that of osteoarthritis. Therefore, PFO is not an appropriate procedure for this type of patient.	

1. Introduction

Stress fractures result from long-term repetitive stresses that are below the bone strength limit, causing cumulative local microdamage in the bone. Stress fractures can be categorized as insufficiency or fatigue fractures [1]. Young athletes and military personnel have an increased risk of stress fractures of the tibia [2,3]. Stress fractures in elderly patients are associated with underlying bone abnormalities, such as osteoporosis. Other reported risk factors for stress fracture include hyperparathyroidism, RA, Paget's disease, pyrophosphate arthropathy, and TKA [4–8].

PFO is a new method for treating medial compartment osteoarthritis of the knee, which is based on the theory of differential settlement (nonuniform settlement) [9]. The lateral part of the tibial plateau is additionally supported by the fibula and surrounding soft tissues, whereas the medial side is supported only by the medial tibial cortex. This, in addition to the fact that the medial side is subjected to more axial loading leads to non-uniform settlement and degeneration of the tibial plateau. This procedure has been widely recognized for its advantages of relative simplicity, low rate of postoperative complications, and low postoperative costs because it does not require hospitalization [10]. Stress fracture of the proximal tibia after PFO has not been previously reported.

2. Case-report

2.1. Case presentation

Written informed consent was obtained from the patient for publication of this case report and accompanying images and her case is presented through this article and is adhere to SCARE guidelines [11]. A 62-year-old woman with chronic RA underwent left PFO with concurrent arthroscopic debridement for chronic knee pain. Postoperative symptom improvement was not obvious, and the patient experienced

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Fig. 1. Radiographs of left knee after proximal fibula osteotomy. (A) Anteroposterior view. (B) Lateral view.



Fig. 2. Radiographs taken at onset of postoperative pain. (A) Anteroposterior view of left knee. (B) Lateral view of left knee. (C) Full-length radiograph of both lower limbs in standing position.

progressive left knee varus accompanied by walking pain. Eleven months after the operation, the patient had worsening left knee pain, needed crutches when walking, and could not squat. The pain was obvious when going up and down stairs, and the patient could walk only approximately 500 m on flat ground. The patient had regularly taken antirheumatic drugs in the previous 21 years. Her drug dosages in the preceding year were as follows: leflunomide, 10 mg daily; hydroxychloroquine sulfate, 0.2 g daily; methotrexate (MTX), 10 mg weekly; methylprednisolone, 4 mg every 2 days, alfacalcidol, 0.5 μ g daily, caltrate, 1 pill daily, and alendronate sodium tablets weekly.

2.2. Investigations

Physical examination revealed claudication, left lower extremity varus deformity, and surgical scars on the left anterolateral knee and lateral upper leg. The left knee joint space showed obvious tenderness. The left leg was approximately 1 cm shorter than the right and had a varus deformity of approximately 10° . The range of movement of the left knee joint was 0 to 90 degrees. The patient was positive for sensation of joint friction and negative for internal and external stress testing and left knee activity-induced pain. Radiographs taken immediately after PFO

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Table 1

Patient's laboratory examination.

Laboratory examination	Result	Normal range
Rheumatoid factor	33.1 IU/ml	0-20.0
Anti-cyclic citrullinated peptide antibody	382.0 U/ml	0-17.0
C-reactive protein	23.2 mg/L	0-5.0
Interleukin-6	12.7 pg/ml	0–7.0
Erythrocyte sedimentation rate	7.0 mm/h	0-38.0
Serum vitamin D (25-OH)	61.1 nmol/L	47.7-144.0
Alkaline phosphatase	28.2 μg/L	11.4–24.6

showed no fracture lines in the proximal tibia (Fig. 1). Radiographs taken at the onset of postoperative left knee pain showed dislocation of the upper segment of the left fibula, postoperative changes in the proximal fibula, and suspicious fracture lines in the proximal tibia (Fig. 2). Bone mineral density (BMD) testing showed a T-score of -1.2 for the hip and -0.2 for the lumbar spine, which indicates the patient with mild BMD loss but no osteoporosis. Table 1 shows the Patient's laboratory examination.

2.3. Treatment

In the early stage of proximal tibia stress fracture, due to the concealment of radiography manifestations (Fig. 3), and the pain of knee masked the symptoms of the tibial fracture, doctors from another hospital performed total knee arthroplasty(TKA) for the patient, but ignored the treatment of stress fracture of the proximal tibia(Fig. 4). Three months after TKA, the patient experienced worsening pain in the left knee joint and gradually progressive lameness. Six months after surgery, the patient was admitted to our hospital for radiography (Fig. 5). The patient underwent open reduction and internal fixation with a plate and screw in the proximal left tibia fracture and autologous iliac bone grafting. The postoperative radiographs are shown (Fig. 6). Pathological biopsy of the fractured bone revealed scant fibrous and vascular tissue, which excluded pathological fractures caused by bone tumors. At

discharge, the patient's left knee joint function had recovered well, that range of movement was 0 to 110 degrees. The patient had mild pain during movement, which was tolerable. The patient continued to receive RA treatment after discharge.

2.4. Outcome and follow-up

The patient was followed up at the hospital three months after the open reduction (Fig. 7). The proximal tibia stress fracture began to heal after open reduction and internal fixation with autologous iliac bone grafting and other supportive treatments. We will continue to monitor the patient until the proximal tibia fracture has healed and then will decide whether to remove the plate, depending on the patient's condition and willingness.

3. Discussion and conclusion

3.1. Discussion

Stress fractures are most common among athletes and military recruits [12,13,14]. Stress fractures that occur in normal bone are often called fatigue fractures, whereas those that occur in abnormal bone (such as osteoporotic bone) are called deficiency fractures [15]. Most stress fractures occur in the lower limb bones, including the tibia (23.6%), navicular bone and metatarsals (17.6%), fibula (approximately 16%), and femur (6.6%), and the pelvic bones (1.6%) and spine (0.6%)may also develop stress fractures [16]. Stress fractures were first described by Breithaupt [17] in soldiers in the 19th century. In 1980, Rand and Coventry [18] reported 15 cases of stress fracture after TKA. They concluded that these fractures resulted from axial misalignment of the components. Post-knee-replacement stress fractures resulting from the use of trackers and navigation pins have also been reported [19]. In 2016, Vaish et al. [20] reported a single case of proximal fibular stress fracture that caused rapid progression of knee eversion 14 years after TKA. There have been no previous reports of stress fractures of the



Fig. 3. Radiographs taken before total knee arthroplasty. (A) Anteroposterior view. (B) Lateral view.



Fig. 4. Radiographs taken after total knee arthroplasty. (A) Anteroposterior view. (B) Lateral view. (C) Full-length radiograph of both lower limbs in standing position.

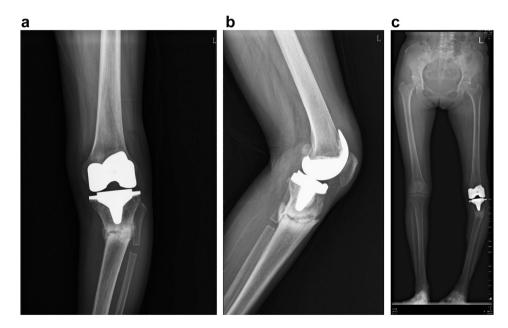


Fig. 5. Radiographs taken 6 months after TKA. (A) Anteroposterior view. (B) Lateral view. (C) Full-length radiograph of both lower limbs in standing position.

proximal tibia after PFO.

In this case, stress fracture of the proximal tibia occurred 1 year after PFO. In a meta-analysis, Wright et al. [21] found that female sex was a risk factor for stress fractures. Studies have shown that osteoporosis and low bone density are additional risk factors for stress fracture of the lower extremities, and that female athletes have a higher risk than other individuals [22]. Our patient did not have osteoporosis and was not an athlete or soldier. However, she worked in the clothing industry, which required considerable travel and walking. This employment could have increased the risk of stress fractures. In addition, the loss of support from the fibula after proximal osteotomy may have further promoted the occurrence of stress fractures of the tibia. In a 2015 case report, Tan et al. [23] concluded that long-term use of MTX and glucocorticoids predisposed patients to stress fractures. Several Studies have shown that MTX is acting by inhibiting dihydrofolate reductase, which is an essential factor in deoxyribonucleic acid (DNA) and ribonucleic acid (RNA) synthesis, but also has far-reaching anti-inflammatory and immunoregulatory effects [24]. MTX is postulated to affect osteoblastic activity and may enhance osteoclastic bone resorption. Prolonged usage of MTX can result in MTX osteopathy, which was defined by a clinical triad: pain, osteoporosis, and stress fractures [25]. In addition, application of glucocorticoids (GC) is the most frequent cause of osteoporosis provoked by medicaments. Its effect on bone BMD is manifested through the increase of bone resorption and decrease of bone formation which all lead to the increased risk of the occurrence of fractures [26]. The patient we reported presented with mild BMD loss but no osteoporosis, despite a clear history of MTX and glucocorticoid use. This may suggest that a history of glucocorticoid and MTX use was not the primary cause of the stress fracture in this case.

Several studies have been published in recent years on PFO, which is

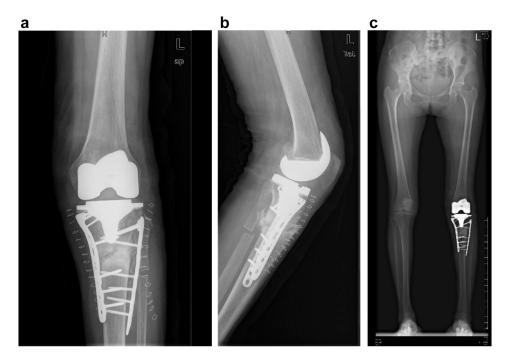


Fig. 6. Radiographs taken 1 day after open reduction. (A) Anteroposterior view. (B) Lateral view. (C) Full-length radiograph of both lower limbs in standing position.

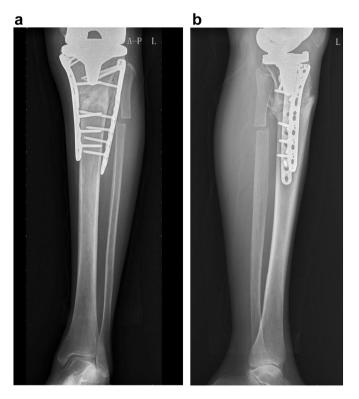


Fig. 7. Radiographs taken 3 months after open reduction. (A) Anteroposterior view. (B) Lateral view.

a relatively new treatment for medial interventricular osteoarthritis of the knee [27,28,29]. The procedure has been shown to significantly improve pain severity, increase medial joint space, and reduce varus deformity of the knee [29,30]. A clinical study concludes that the fibula supports one sixth of the body weight [27]. PFO mainly helps rebalance or redistribute the load on the lateral and medial tibia plateau after surgery. The patient in this case was diagnosed with RA with varus deformity before undergoing PFO. She did not experience significant improvement in pain and varus deformity after proximal fibular osteotomy and developed a proximal tibial stress fracture 1 year later. We believe that one of the important reasons may be the improper selection of surgical indications. The patient's arthritis of the knee was caused by RA without confined to the medial compartment. So this patient is not suitable for PFO. This, in turn, increases the burden on the tibia due to the loss of fibula support.

In this case, chronic knee pain masked the symptoms of stress fracture of the proximal tibia. Moreover, because the initial radiographic signs were relatively subtle, the attending doctors ignored the tibial stress fracture, thus missing the best treatment opportunity. The radiograph taken before scheduled TKA revealed that the patient required conservative treatment for the tibial stress fracture before surgery, including rest, use of a knee brace, and non-weight bearing for 3 to 6 months [31]. However, the patient underwent TKA, continued to work after surgery, and continued frequent walking, which further worsened the stress fracture and fracture displacement. The sensitivity of X-rays in diagnosing stress fracture of the tibia is very low [31]. Magnetic resonance imaging has the greatest sensitivity, specificity, and accuracy in the diagnosis of the condition [32]. Therefore, the possibility of stress fracture in addition to primary knee disease should be considered in patients with insidious tibial pain. X-ray examination is generally recommended. If negative, magnetic resonance imaging may be performed to assist in the diagnosis of early stress fractures.

3.2. Conclusion

RA is usually not confined to the medial compartment and its pathogenesis is different from that of osteoarthritis. Therefore, PFO is not an appropriate procedure for this type of patient.

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Ethical approval

This is a report of a case under the waiver of approval of the ethics committee or institutional review board.

Consent

Written informed consent was obtained from the patient for the publication of this report.

CRediT authorship contribution statement

Wang Xing managed the patient and wrote the first draft. Huang Qiang, Pei Fuxing helped in editing and reviewing the paper. All authors read and approved the final version to be published.

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Guarantor

Huang Qiang.

Declaration of competing interest

No conflict of interest exits in the submission of this manuscript.

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