Chinese Journal of Traumatology 20 (2017) 173–176

Contents lists available at ScienceDirect

Chinese Journal of Traumatology

journal homepage: http://www.elsevier.com/locate/CJTEE

Case Report

Case report: A 10 years follow-up of periprosthetic femoral fracture after total hip arthroplasty in osteopetrosis

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ARTICLE INFO

Article history:
Received 30 May 2016
Received in revised form
2 September 2016
Accepted 5 April 2017
Available online 10 May 2017

Keywords:
Osteopetrosis
Arthroplasty, replacement, hip
Periprosthetic femoral fractures

ABSTRACT

Osteopetrosis is an inherited disorder characterized by increased bone density and brittle bone quality. Degenerative changes often occur after the age of 40 in patients with osteopetrosis. Operative intervention is the primary treatment option if the clinical manifestation of secondary osteoarthritis is severe. A 44-year-old male suffering autosomal dominant osteopetrosis and progressive unilateral hip osteoarthritis required a total hip arthroplasty. However, there were several technical challenges associated with this procedure including creating a femoral medullary canal and developing a Vancouver type B2 periprosthetic femoral fracture postoperatively. To afford some experience for the management of similar cases, we here present our technical solutions to these problems.

Introduction

Osteopetrosis is a rare condition characterized by increased bone density as a result of osteoclast dysfunction, often demonstrating generalized osteosclerosis on radiographs.[1,2] Later, three types of osteopetrosis were reported by Shapiro[3]: a malignant fetal form inherited as an autosomal recessive condition, an intermediate autosomal recessive form, and a benign autosomal dominant form. Gwynne et al[4] subdivided the autosomal dominant osteopetrosis into two types: type 1 showing increased thickness of the cranial vault, diffuse osteosclerosis of the lumbar spine, pelvis and symmetrical long-bone; type 2 presenting more basal skull involvement. The latter type suffers a high risk of fracture, osteomyelitis, and early-onset osteoarthritis.

Reported by many authors, fracture is the most common complication of osteopetrosis. The osteopetrotic bone is consolidated by pathological callus without Haversian organisation. As Girard et al[5] reported, femoral neck and subtrochanteric fracture often lead to coxa vara which may contribute to the degenerative process and secondary osteoarthritis. In these situations, operative intervention is the primary treatment option, and thus we resort to operation, especially total joint arthroplasty for patients even without fracture if the clinical manifestation of secondary osteoarthritis proves severe.

Periprosthetic femoral fracture remains a severe postoperative complication subsequent to total hip arthroplasty (THA).[6] Because of the high morbidity, treatment has evolved from traction and bracing in a cast to open reduction and internal fixation (ORIF), a revision procedure or even a combination of both.[7] There are no reports, with follow-up study, of periprosthetic fracture in osteopetrotic patients who received cementless total hip arthroplasty. Therefore, we report an osteopetrotic patient who developed a periprosthetic femoral fracture after THA was managed nonoperatively.

Case report

A 44-year-old male, 167 cm in height, 50 kg in weight, with a type 2 benign osteopetrosis, presented a 2-year history of pain located in the left hip which progressively worsened in 2 months. His past orthopedic history included a fracture of the left distal femur when he was 12-year-old, which was healed by nonoperative treatment, and another left humeral fracture at the age of 15 treated again nonoperatively with a satisfactory outcome. His family history is noncontributory.

On physical examination, he had a marked antalgic gait to the left with a Harris hip score (HHS) of 42 out of 100. The initial laboratory investigations revealed no special findings. Radiographs

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Peer review under responsibility of Daping Hospital and the Research Institute of Surgery of the Third Military Medical University.

http://dx.doi.org/10.1016/j.cjtee.2017.02.001

showed extremely high density in bilateral femoral head, ilium, acetabulum and pubic symphysis. The femur was uniformly dense with narrow medullary canal, and the hip joint-clearance was narrow with some cystoid changes in the femoral head and acetabulum, indicating osteopetrosis and advanced osteoarthritis. The bone density of the left femur and the spine was remarkably higher than normal according to the bone density analysis. The preoperative pelvic X-ray film is shown in Fig. 1.

The patient was treated with left total hip arthroplasty in May 2004 via a posterolateral approach. The femoral neck was cut with an air-pressure oscillating saw at the level about 1 cm above the lesser trochanter. It was very hard to cut the femoral neck because of the dense sclerotic bone, after which the cut surface showed no medullary canal. When the femoral head and neck were removed, the articular cartilage of the acetabulum showed marked degenerative changes.

Fig. 1. Anteroposterior (AP) radiograph of pelvis showing signs of osteopetrotic bone, osteoarthritis in the left hip.

The acetabulum was reamed till uniform surface bleeding, and then a 50 mm acetabular component measured 28 mm, size E microstable HMWPE liner was inserted (Reflection, Smith & Nephew, Menphis, USA). The cup was press-fitted and fixed by 2 screws (2.5 cm in length, 6.5 mm in diameter, Smith & Nephew, Menphis, USA). The preparation of femoral canal was so difficult that the extended trochanter osteotomy (ETO) was used to help create a bony cavity for the femoral stem. The femur was then sequentially reamed with power reamer, oscillating saw and hand-held rasp. A size 8 uncemented femoral component (Synergy, Smith & Nephew, Menphis, USA) was inserted. The osteotomic part was secured by multiple cerclage wires. A standard 28 mm femoral head was chosen, and a reasonable arc of movement was established. During the 3.5 h operation, the estimated blood loss was 1000 ml with no intraoperative complications.

Four weeks after operation, the patient developed a femoral pain caused by a minor fall. Radiographs indicated a periprosthetic femoral fracture in distal osteotomy site with slight migration, but the stem seemed stable as no subsidence was found at that time, leading us to categorizing it into a Vancouver B1 fracture (Fig. 2). After a discussion with the patient about the treatment options, he refused to accept the operation of internal fixation considering the surgical complications and his previous fracture history. A conservative treatment was thus performed and the patient was required to have no weight bearing for at least 12 weeks.

After 8 weeks the X-ray film showed callus formation around the fracture, while the prosthesis subsided 5 mm by measuring the distance between the top of great trochanter and the center of femoral head, indicating potential stem loosening (Fig. 3). Since the patient admitted partial weight bearing on crutches for 4 weeks already, we recategorized the fracture into a Vancouver B2 fracture and suggested a revision. However, the patient declined it and insisted on conservative treatment. The patient was advised to keep bedridden for another 4 weeks and the follow-up X-ray film showed good callus formation and no further subsidence in 12

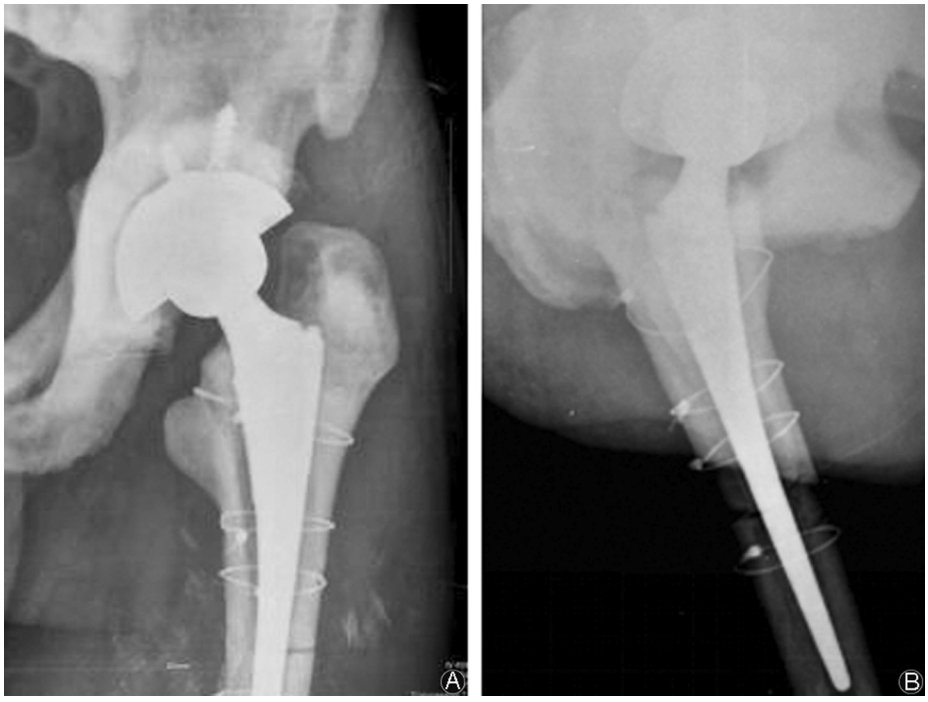

Fig. 2. A: AP radiograph of pelvis showing no subsidence of the prosthesis. B: Lateral radiograph of pelvis showing a periprosthetic fracture in distal osteotomy site with slight migration.

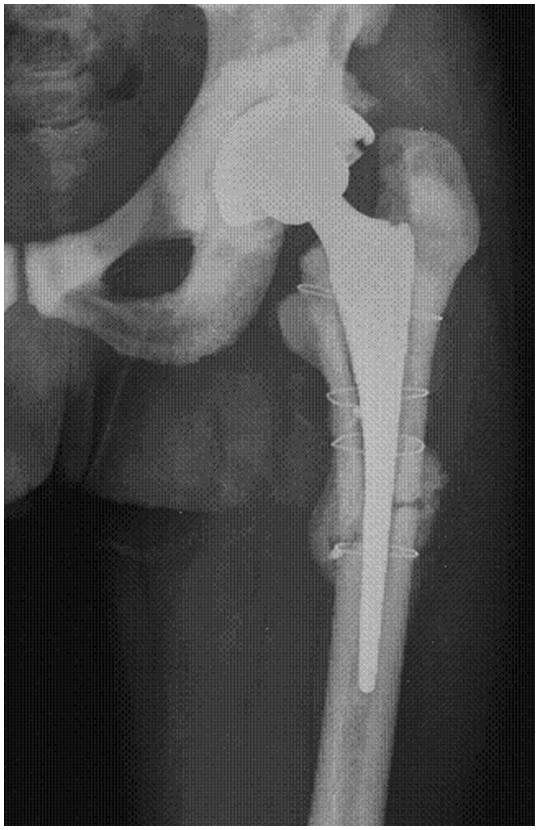

Fig. 3. AP radiograph of pelvis showing an approximately 5 mm subsidence of the prosthesis and some callus formation at 8 weeks after the periprosthetic fracture.

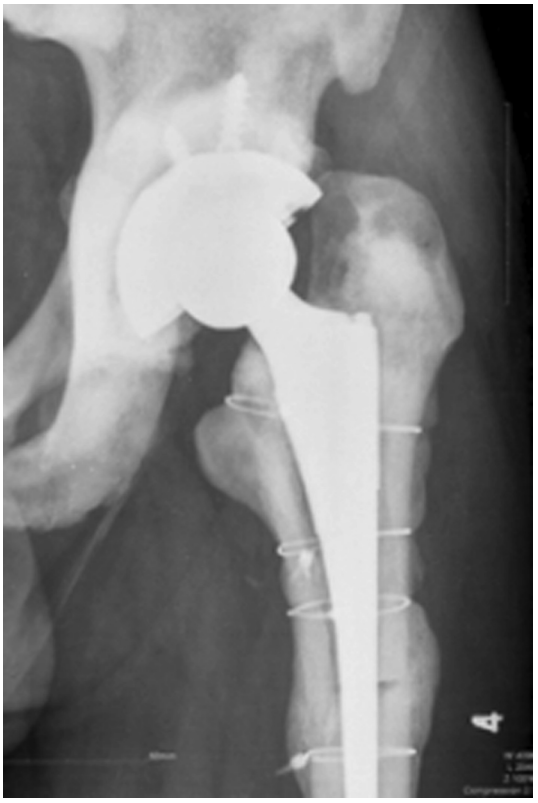

Fig. 4. AP radiograph of pelvis showing no subsidence of the prosthesis and good callus formation at 12 weeks after the periprosthetic fracture.

weeks after the fracture (Fig. 4). Then the patient was required to start partial weight bearing on crutches and have careful movement.

At the latest follow-up which was 10 years after the fracture, the patient's condition was satisfactory. He could walk without support and achieve a 90-degree arc of flexion in left hip with a recent HHS of 86. Although he bears occasional thigh pain after prolonged activity, a slight limp, and limited walking ability, the follow-up X-ray film demonstrated a stable stem (Fig. 5).

Discussion

Degenerative changes often occur after the age of 40 in patients suffering osteopetrosis without deformity.[4,8] Operation is the most effective treatment for degenerative osteoarthritis caused by autosomal dominant osteopetrosis. According to reported cases, THA is a good option despite technical difficulties recognized by most authors. The abnormal osteoclastic function results in very hard bone with thickened trabeculae, and femoral cavity preparation is much harder than the acetabular preparation.

Matsuno and Katayama[9] suggested using a high-speed bur to create a femoral cavity and implant a cemented stem because of the difficulty of placing press-fit cementless stem and the possibility of developing a femoral shaft fracture. Strickland and Berry[10] used a cannulated reaming system under fluoroscopic guidance to create a femoral canal. Ramiah et al[11] reported that they used tungsten carbide drill bits over a guide wire under radiologic control. Egawa et al[12] and Benum et al[13] also performed THA in patients suffering osteopetrosis with computer-assisted fluoroscopic navigation system. Despite the prevalence and effectiveness of the fluoroscopy used by orthopedic surgeons during operation, several problems exist as this process can be incorrect, time-consuming, and ergonomically hard for the surgeons and radiograph technologists.[12] In this case, we used ETO without any radiologic guidance to help create a femoral cavity but gained a satisfactory outcome. The depth of femoral longitudinal cutting was shorter than the length of the femoral component since it could be modified by power reamer and hand-held rasp. Though osteotomy is helpful for creating the femoral canal, it could in turn become a risk factor in periprosthetic fracture which would be mentioned later.

Uncemented femoral implant was used in this case. Strickland and Berry[10] considered treating patients suffering osteopetrosis with uncemented femoral implant would be difficult during THA unless a system that allows bone preparation without broaches is used. As Gwynne et al reported,[4] uncemented femoral component was successfully implanted by opening a femoral canal with drills and reamer. In this case, we managed to implant an uncemented femoral component and gained satisfactory initial stability.

Patients with osteopetrosis are at a high risk of periprosthetic fracture, most of which are iatrogenic.[14] Prosthesis loosening, infection, especially osteomyelitis and failed rehabilitation could also occur subsequently. The treatment of periprosthetic femoral fracture includes revision, revision with bone grafting, ORIF[7] and conservative treatment. The Vancouver classification is regarded as a reliable guide for treatment of periprosthetic femoral fractures.[15] Traditionally, among the type B fractures, B1 fractures with a well-fixed stem are recommended to be treated by ORIF, while the B2 fractures with a loose stem are indications of a revision. However, Spina et al[16] and Moloney et al[17] found a high rate of loosening and refracture in patients with B1 fractures treated by ORIF. It can be partially attributed to surgeon's misinterpretation of the stability of the stem and a mistaken classification of type B2 fractures as type B1 fractures, under which condition, a plate fixation without

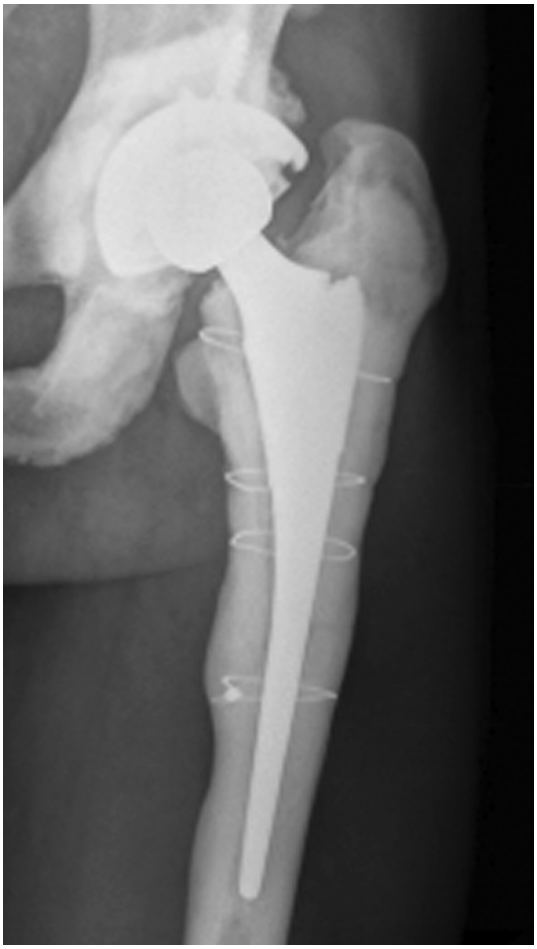

Fig. 5. AP radiograph of pelvis showing a satisfactory fracture union and stable prosthesis at 10 years follow-up after the periprosthetic fracture.

revision was taken by surgeons. Meanwhile, some alternative treatments for B2 fractures are reported recently.[16,18,19] Joestl et al[18] reported good results of ORIF for B2 fractures. Niikura et al[19] treated one B2 fracture conservatively and gained a satisfying outcome. Therefore a customized treatment of B2 fracture seems to be valid.

In this case, the fracture of the patient was at first considered to be a Vancouver type B1 fracture as no obvious subsidence was found in X-ray film. According to the fracture location, we considered that osteotomy during THA may play a role in the fracture in addition to the brittle bone quality as the level of the fracture was right at the level of ETO, 13 cm from the top of the great trochanter. This was treated conservatively considering the slight fracture migration and the patient's strong personal concern, which was also referred by Ramiah et al[11] in their treatment of periprosthetic femoral fracture for patients with osteopetrosis. Nonoperative treatment for this kind of fracture proves to be a reasonable choice since fewer complications were observed. The most common complication following the nonoperative treatment may be coxa vara or fracture union with deformity.[20] We corrected the fracture diagnosis when a radiological 5 mm subsidence appeared 8 weeks later, and prudently employed a conservative treatment when the patient refused to accept any other invasive procedure. Our patient's latest X-ray showed well healed fracture without any deformity. This case tested the idea that the conservative treatment works in patients with specific indications like low-grade pain, previous wiring around the fracture, and light weight when faced with the type B2 fractures.[19]

In summary, this is a case of periprosthetic fracture following ETO and cementless THA in an osteopetrosis patient. Though the fracture was managed by nonoperative treatment, the clinic and radiological results demonstrated a good recovery after 10 year's follow-up study.

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