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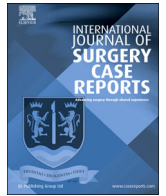
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# Locking plate osteosynthesis for a femoral fracture and subsequent nonunion in a patient with osteopetrosis

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## ABSTRACT

**INTRODUCTION:** Osteopetrosis is a skeletal disorder characterized by increased osteodensity and a remodeling defect. The fragility of dense sclerotic bones may lead to an increased incidence of fractures. Although internal fixation can be performed, technical challenges may be experienced because of the increased bone density. Complications such as delayed union, nonunion, or implant failure may occur postoperatively.

**PRESENTATION OF CASE:** We describe a patient with autosomal-dominant osteopetrosis type 2 who suffered a shaft fracture below a plate of his right femur. We performed osteosynthesis with a single locking plate. Union was delayed, and plate breakage occurred along with nonunion of the fracture. The nonunion was addressed using double locking plates, which secured fixation and allowed complete fracture healing.

**DISCUSSION:** There were three reasons of nonunion in our case. First, we left gaps between the fragments. Second, we used mainly cerclage wires, rather than screws, for plate fixation, which led to inadequate stability. Third, the patient was large (height 167 cm, weight 93.1 kg), so the single plate provided insufficient fixing force. We then used double locking plates and attained stronger internal fixation with complete fracture healing.

**CONCLUSION:** Double plating with locking plates may be an effective treatment option for femoral fractures in patients with osteopetrosis.

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## 1. Introduction

Osteopetrosis is a rare hereditary skeletal disorder characterized by increased osteodensity and a remodeling defect, where bone resorption is impaired due to the osteoclast dysfunction [1]. The fragility of dense sclerotic bones can lead to an increased incidence of fractures. Operative treatment of osteopetrotic fractures is technically challenging. Because of the increased bone density, drilling and insertion of the screws is often difficult. In children, an intramedullary nail is used only when there is some evidence of an open medullary canal [2]. Impaired bone remodeling prolongs bone union.

In our patient, a femoral fracture was initially treated with one locking plate, but plate breakage occurred along with nonunion of the fracture. Complete fracture healing was ultimately achieved by secure fixation with double locking plates. Few case reports have described the use of a locking plate to address an osteopetrotic femoral fracture, and even fewer have reported nonunion. This work has been reported in line with the SCARE criteria [3].

## 2. Presentation of case

A 27-year-old man who presented with a femoral shaft fracture had a history of several previous fractures. He had been diagnosed with osteopetrosis at age 7 years. At age 15 years, his left femoral trochanter was broken as a result of the impact of jumping. At age 16 years, he was admitted to another hospital with complaints of right hip pain and inability to walk. He was diagnosed with an intertrochanteric fracture of the right femur, for which he underwent skeletal traction followed by surgical open reduction with a plate and wiring. The skeletal traction caused osteomyelitis of the femoral epicondyles for which he was treated at ages 18 and 21 years.

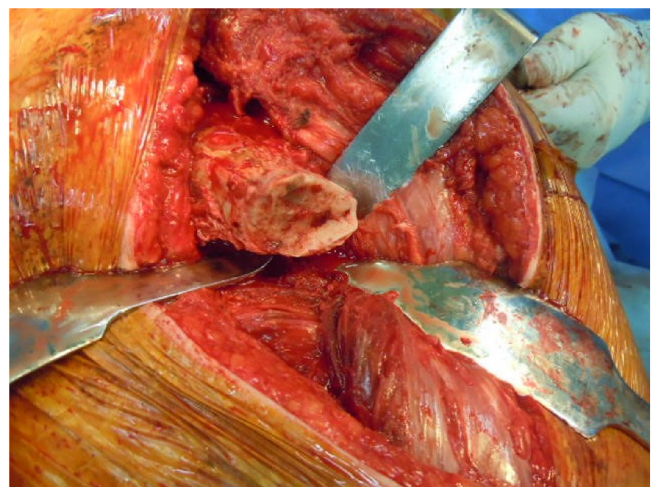
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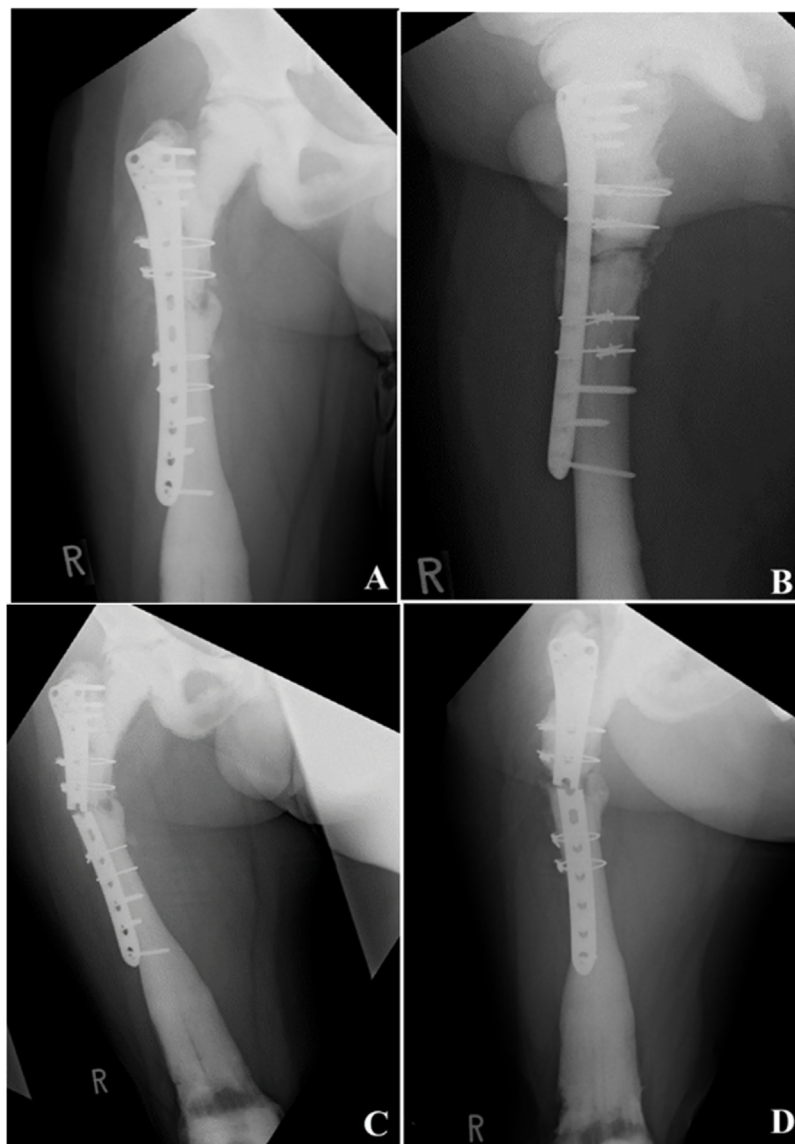


**Fig. 1.** Anteroposterior(AP) (A) and lateral (B) radiographs of the right femur show the fracture below the plate. Postoperative AP (C) and lateral (D) radiographs show internal fixation using a reversed distal femoral locking compression plate on the contralateral side with cerclage wirings and screws.

At 26 years of age, a fall resulted in a shaft fracture below the plate of his right femur. At our institution, anteroposterior and lateral plain radiographs of the right femur showed the fracture with apparent varus deformity. We performed open reduction and internal fixation using a reversed distal femoral locking compression plate (LCP-DF; Synthes, Obodors, Switzerland) on the contralateral side and cerclage wiring (Fig. 1). The intramedullary canal was closed (Fig. 2). Postoperatively, consolidation was delayed, and 14 months after the surgery he experienced sudden pain. Plain radiography showed plate breakage and right femoral nonunion with displacement (Fig. 3). Bridge plating was performed using two locking compression plates. A revised LCP-DF was applied on the lateral side of the right femur and a broad LCP on the anterior side. Anteroposterior and lateral radiographs obtained at the latest follow-up (2 years 8 months after the last operation) demonstrated bone union (Fig. 4). Breakage of the most distal screw is apparent, reflecting relative stability of the fracture. He is currently pain-free and walks without support.



**Fig. 2.** Intraoperative photograph shows a closed intramedullary canal.



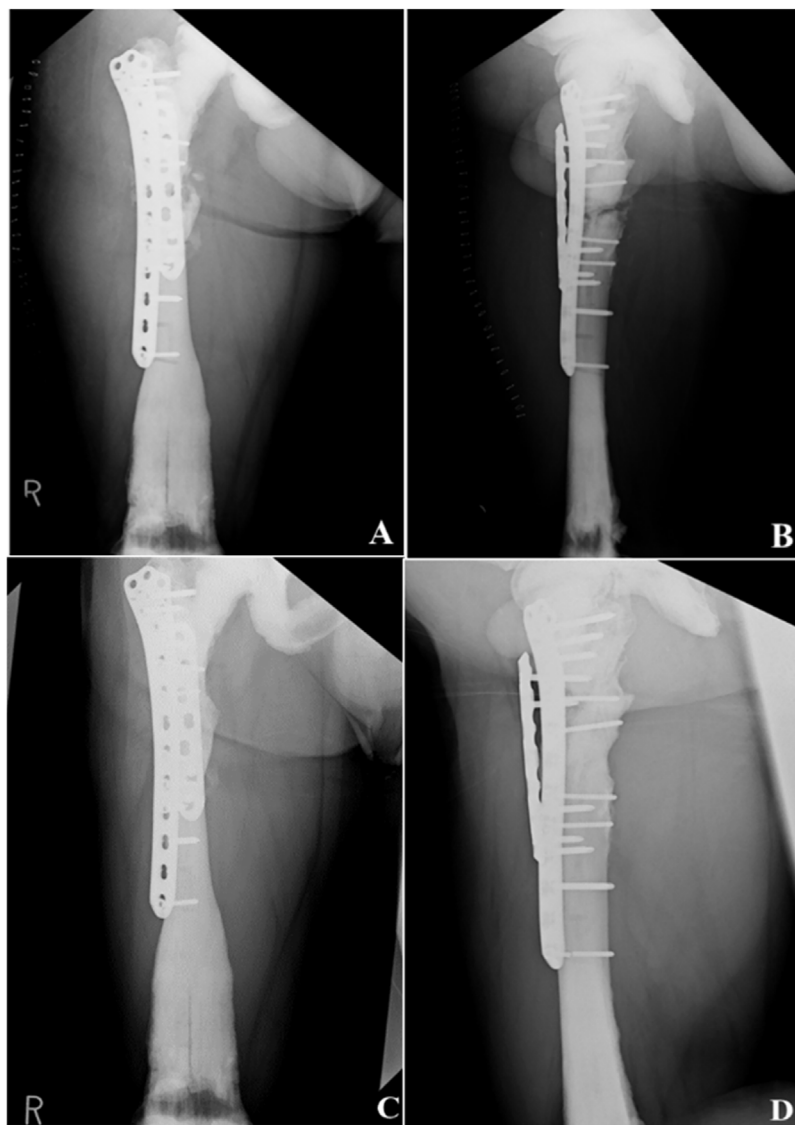
**Fig. 3.** AP (A) and lateral (B) radiographs 12 months after osteosynthesis show delayed consolidation. AP (C) and lateral (D) radiographs 14 months after osteosynthesis show plate breakage and right femoral nonunion with displacement.

### 3. Discussion

Osteopetrosis, which comprises a group of rare bone dysplasias [4], is categorized into three primary types: infantile or malignant, intermediate autosomal recessive (ARO), and autosomal dominant (ADO) osteopetrosis [5,6]. ADO is classified into ADO1 and ADO2 because of different underlying genetic mutations. ADO2 patients have increased numbers of nonfunctional osteoclasts and elevated TRACP 5b, which is an enzyme derived from osteoclasts [7]. Thus, in an ADO2 patient, highly increased serum TRACP 5b activity may be attributable to increased numbers of inactive osteoclasts [8]. ADO2 is usually diagnosed based on a history of multiple nontraumatic fractures, radiographic evidence of widespread osteosclerosis, and the presence of endobones (bone-within-a-bone appearance), most commonly in the vertebrae (sandwich or rugger-jersey vertebrae), pelvis, and at the ends of long bones [9]. Our patient was diagnosed with ADO2 based on his past history of several fractures, typical radiographic findings, and high serum TRACP 5b activity (10,403 U/L; normal 170–590 U/L).

Operative intervention, when needed, presents unique technical challenges. Although osteopetrotic hard bone may be penetrated

with a drill, high friction and prolonged drilling can blunt or break the drill bit. In addition, because of the difficulties encountered during the operation, the operative time may be prolonged, thereby increasing the risk of postoperative infection. There is also a risk of delayed consolidation and nonunion due to impaired bone remodeling [10]. In a case series study, Birmingham et al. reported a 12% nonunion rate and 12% infection rate. The rate of implant failure was 29% in their peritrochanteric group [11]. Kumbaraci et al. treated bilateral subtrochanteric femoral fractures with proximal femoral nail anti-rotation but used an intramedullary nail in children who showed evidence of a medullary canal [2]. Kumar et al. reported a case in which a femoral subtrochanteric fracture was treated with a dynamic hip screw [12]. Amit et al. treated subtrochanteric fractures with a less-invasive stabilization system (LISS) of plating [13]. Sen et al. [14] presented cases in which osteopetrotic femoral fractures were treated with locking plates. Bhargava et al. reported treating a periprosthetic fracture with a plate that led to delayed union and plate breakage. They revised the fixation using a LISS plate supplemented with cables of allograft, meshed fresh frozen femoral head, and bone morphogenetic protein (BMP). Bony union was achieved 6 months after the surgery



**Fig. 4.** AP (A) and lateral (B) radiographs show bridge plating using double locking plates. A distal femoral locking compression plate (LCP-DF) was applied on the lateral side and a broad LCP on the anterior side of the right femur. AP (C) and lateral (D) radiographs 2 years 8 months after the last operation show bone union.

[10]. Only in their study was a locking plate used to achieve osteosynthesis for a femoral fracture with subsequent nonunion in a patient with osteopetrosis. Some authors recommended BMP grafting as bone substitute, which stimulates mesenchymal cells to differentiate to osteoblasts via its osteoinductive nature, thereby exerting a positive effect on bone and callus formation and ultimately fracture healing [13].

Considering that the current case was a non-comminuted fracture, absolute stability with anatomical reduction was supposed to be required. We initially tried osteosynthesis with a single locking plate and cerclage wiring and screws, but union was delayed and the plate broke 14 months after the surgery. The demand for mechanical stability of the internal implants, used in osteopetrosis, is higher than usual because of the longer stay of these implants over the slowly healing osteopetrotic bone [15]. We then used double locking plates and attained stronger internal fixation with complete fracture healing. There were three reasons of nonunion in our case. First, we left gaps between the fragments. Second, we used mainly cerclage wires, rather than screws, for plate fixation, which led to inadequate stability. Third, the patient was large (height

167 cm, weight 93.1 kg), so the single plate provided insufficient fixing force.

Slow-speed, high-torque electric drills, frequent cooling with physiological saline, clearance of drill grooves, and the use of a staggered drill system have all been recommended [6,14,16]. We ultimately drilled the bone satisfactorily and created a favorable environment for osteosynthesis using plates and screws without the need for wiring.

#### 4. Conclusion

Double plating with locking plates may be an effective treatment option for femoral fractures in patients with osteopetrosis.

#### Conflicts of interest

None.

#### Funding source

None.



## Ethical approval

Approval to publish this case report was waived by the institution.

## Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

## Author contribution

All authors in this manuscript contributed to the interpretation of data, and drafting and writing of this manuscript. Tomoya Matsuo is first and Takahiro Niikura is corresponding author of this paper. They and San Yang Lee, Takashi Iwakura, Tomoaki Fukui, Keisuke Oe, Tomoyuki Matsumoto, Takehiko Matsushita, Kotaro Nishida conceived and designed the study and drafted the manuscript. Ryosuke Kuroda is a chief of orthopaedic surgery in our university hospital. All the authors read and approved the final manuscript.

## Registration of research studies

This is not a research study.

## Guarantor

Takahiro Niikura, MD.

## Provenance and peer review

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