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CLINICAL ARTICLE

The Treatment of Subtrochanteric Fracture with Reversed Contralateral Distal Femoral Locking Compression Plate (DF-LCP) Using a Progressive and Intermittent Drilling Procedure in Three Osteopetrosis Patients

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Objective: To describe the application of reversed contralateral distal femoral locking compression plate (DF-LCP) inserted through a progressive and intermittent drilling procedure in the treatment of osteopetrotic subtrochanteric fracture (OSF).

Methods: Three patients (one male and two females with an average age of 45.33 ± 11.09 years) with OSF hospitalized between September 2015 and September 2020, were included in this present study. Lateral approach was applied in all patients who accepted open reduction and internal fixation (ORIF) with a reversed contralateral DF-LCP inserted through a progressive and intermittent drilling procedure. The operation time and intraoperative blood loss were recorded to evaluate the efficiency of this surgical method. Physical examination and imaging examination of the fracture site were used to evaluate the fracture union status, the position and stability of the implant, and the alignment of the injured limb at 1, 3, 6, and 12 months after operation, then a subsequent visit was conducted at least once a year. Harris Hip Score (HHS) was used to evaluate the hip joint function at 6 and 12 months after operation.

Results: The average operation time was 140 ± 21.60 min (110, 160, and 150 min); The average intraoperative blood loss was about 333.33 ± 23.57 ml (300, 350, and 350 ml). The average follow-up time was 22.33 ± 7.41 months (29, 26, and 12 months). All patients achieved bone union with an average time of 6.67 ± 0.94 months (6, 8, and 6 months). At the time of 6 months after operation, case 1 and 3 were almost pain-free and could walk with full weight bearing while case 2 could walk only with partial weight bearing using a crutch. The HHS scores of cases 1, 2, and 3 were 84/100, 74/100, and 92/100, respectively. At the follow-up at 12 months after operation, the HHS score improved to 91/100, 81/100, and 96/100, respectively. The contralateral incomplete old subtrochanteric fracture was deteriorated in case 1 at 26 months after operation. After 3 months of limited weight bearing using a crutch, bone union was verified in radiograph imaging. Fresh contralateral subtrochanteric fracture occurred in case 2 at 26 months after operation, which was treated using a similar surgical approach, and its clinical outcome is under follow-up. Moreover, no perioperative complications including operation-related death, vascular/nerve injury, deep venous thrombosis, pulmonary embolism, and incision infection, and long-term complications

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involving malunion, nonunion, implant failure, ankylosis, heterotopic ossification, osteonecrosis, and osteomyelitis were identified.

Conclusion: The application of reversed contralateral DF-LCP in OSF is practicable and reliable. Progressive and intermittent drilling is a safe and efficient method for implant insertion in this complicated situation.

Key words: Albers-Schönberg disease; Locking compression plate; Osteopetrosis; Subtrochanteric fracture

Introduction

Osteopetrosis (OPT), also named Albers-Schönberg disease or marble bone disease, was first described by Heinrich Albers-Schönberg in 1904[1]. It refers to a group of descendible illnesses with increased bone mass density (BMD) and bone fragility, which was reported to be associated with impaired bone absorption caused by osteoclast dysplasia and dysfunction[2]. Historically, due to the pathogenic mechanism being unclear, descriptive classification such as malignant, intermediate, and benign based on the severity of clinical manifestation was used widely. According to its inheritance patterns, osteopetrosis was differentiated as autosomal dominant osteopetrosis (ADO), autosomal recessive osteopetrosis (ARO), and X chromosome-linked osteopetrosis (XLO)[3]. Generally, ARO refers to the malignant type, which is usually early onset and lethal in the first decade. ADO refers to the benign type which is generally asymptomatic and hardly impair life expectancy[3]. Nowadays, the genetic bases of osteopetrosis have been largely clarified. Biallelic mutations in TCIRG1, CLCN7, OSTM1, SNX10, and PLEKHM1, associated with acidification of the resorption lacuna or vesicular transport, are responsible for osteoclast-rich ARO, while mutations in TNFSF11 (RANKL) and TNFRSF11A (RANK), related to osteoclastogenesis, contribute to osteoclast-poor ARO. Mutations in CAII cause impaired acidification in resorption lacuna, leading to ARO with renal tubular acidosis, but its manifestation is milder than typical ARO. Besides, mutations associated with ARO in FERMT3, SLC29A3, LRRK1, CSF1R, and CTSK were reported. Mutations in LRP5 and CLCN7 lead to ADOI and ADOII, respectively. But ADOI was no longer regarded as OPT since the mutation affects osteoblast instead of osteoclast. Hypomorphic mutation in IKBKG leads to XLO but it was limitedly reported[4,5]. Altogether, 17 types of OP and related disorders were recorded[6]. The total incidence of OP is hard to estimate. According to limited studies, it's about 5.5/100,000 for ADO and 1/250,000 for ARO[7,8]. The diagnosis is mainly based on the characterized radiological signs, which consist of diffusely increased BMD, obstructed bone marrow cavity, endobone, rugger-jersey vertebrae, and erlenmeyer deformity. The clinical presentation of osteopetrosis varies extremely. Frequently occurred fragility fracture is typical. Besides, short stature, delayed tooth eruption, dental caries, osteomyelitis, renal tubular acidosis, bone marrow failure, compensatory extramedullary hematopoiesis, hepatosplenomegaly, pancytopenia, cerebral calcification, and neurological defects such as optic atrophy, blindness, deafness, and facial paralysis were also reported[3,4,9,10].

It is a challenge for orthopaedists to treat osteopetrotic fracture (OPF) since the combined hardness and brittleness makes it difficult to drill screw canal and insert screw but easy to cause iatrogenic fracture and instrument breakage. Moreover, there are increased risks of thermal necrosis, malunion, nonunion, subsequent implant loosening, and postoperative infection. In 2010, Amit *et al.*[11] reviewed the available literature (four femoral neck fractures and 21 peritrochanteric fractures) on the surgical treatment of OPF in adults; they reported a non-union rate of 12%, infection rate of 12%. And in peritrochanteric group, both the hardware failure rate and reoperation rate were 29% and the periprosthetic fracture rate was 14%. Therefore, conservative treatment in certain situation is reasonable. Surgical management is recommended in situation involving femoral head/shaft fracture, coxa vara, and failure of conservative treatment[12,13]. A few studies introduced the application of intramedullary (IM) nail[14–18], dynamic condylar screw (DCS)[19,20], dynamic hip screw (DHS)[21–23], proximal femoral locking compression plate (PF-LCP)[24,25], and reversed contralateral distal femoral locking compression plate (DF-LCP)[26–28] in osteopetrotic subtrochanteric fracture (OSF) and the rate of complication was high. The most optimal implant and a proper surgical intervention remain controversial.

We retrospectively studied three patients with OSF who accepted open reduction and internal fixation (ORIF) with reversed contralateral DF-LCP. They recovered without evident complication during the follow-up period. Although osteopetrotic proximal femoral fracture is a rare injury, we hope that these three cases will benefit trauma surgeons in other institutions.

The purpose of this study is as follows: (i) to summarize the clinical features of osteopetrotic fracture (OPF); (ii) to report the clinical outcome of patients with OSF treated with ORIF with a contralateral DF-LCP inserted through a progressive and intermittent drilling procedure; (iii) to discuss the choice of conservative treatment and surgical treatment for OSF; and (iv) discuss the proper implant of OSF and the efficient method of implant insertion.

Method and Materials

Inclusion and Exclusion Criteria

The inclusion criteria in this retrospective study were as follows: (i) patients diagnosed with osteopetrotic femoral subtrochanteric fracture based on the classic radiological manifestations; (ii) underwent open reduction and internal fixation (ORIF); (iii) the follow-up duration was more than 12 months. Clinical outcomes were assessed by using X-ray for bone union and Harris Hip Score (HHS) for hip joint function.

Exclusion criteria were as follows: (i) the patients were below 14 years old; (ii) open fracture; (iii) clinical data was not sufficient or the outcome was not verified; (iv) the patients had severe medical comorbidities and inability to accept surgery treatment.

General Information

From September 2015 to September 2020, a total of three patients with OSF were included in the present study according to the inclusion and exclusion criteria listed above. The basic information of the three cases included in this study is shown in Table 1. There was one male and two females with an average age of 45.33 ± 11.09 years (38, 61, and 37 years old). All patients underwent a trivial fall in stand position, which caused two right subtrochanteric fractures and one left subtrochanteric fracture (Classification, AO 32-A3). Case 1 had left humeral fracture about 30 years ago and recovered well with plaster immobilization. The fracture line was still recognizable (Fig. 2A). Incomplete old fracture was discovered in left subtrochanteric region of case 1 (Fig. 2B) and left tibia and fibula of case 2 (Fig. 3B). Pancytopenia and anemia of case 1 was verified by his laboratory examination results (Table 2). Case 2 had hypertension, which was controlled with reserpine.

Preoperative Preparation

Routine laboratory examination was implemented. All patients underwent X-ray and 3D-CT examination for the evaluation of the type, location, and displacement of the fracture. Ultrasound examination was used for evaluating cardiac function and the condition of vein in lower limbs. Conventional treatment of immobilization, detumescence, analgesia, and anticoagulation were used. Case 1 underwent blood transfusion therapy to treat his anemia. Three sets of tungsten steel drill bits (12 in total) with diameter of 3.0, 3.5, 4.0, 4.5 mm were prepared preoperatively for progressive drilling procedure (Fig. 5B). Time from fracture to surgical treatment was 5–35 days, with an average of 16.0 ± 13.49 days (35, 8, and 5 days; case 1 had undergone conservative treatment of Chinese traditional plaster for 30 days before his hospitalization).

The Surgical Method

Anesthesia and Position

The patient was placed in the lateral decubitus position under general anesthesia.

Approach and Exposure

A straight incision originating from 3 cm above the top of the greater trochanter, passed through the midpoint of the

TABLE 1 The basic information of three cases included in this study

	Age /Gender	Fracture site	Admitted date	Management (ORIF)	Operation time (minutes)	Outcome
Case 1	38/M	RS; LS (insufficient),	Feb. 22, 2018	Reversed DF-LCP (R); immobilization (L)	110	Union, painless, full weight bearing
Case 2	61/F	RS; L (insufficient), tibia & fibula	Jun. 17, 2018	Reversed DF-LCP (R); immobilization (L)	160	Union, pain released, partial weight bearing
Case 3	37/F	LS	Sep. 4, 2019	Reversed DF-LCP	150	Union, painless, full weight bearing

DF-LCP, distal femoral locking compression plate; F, female; L, left; M, male; ORIF, open reduction and internal fixation; R, right; S, subtrochanteric

TABLE 2 The results of laboratory examination

	RBC (10^{12} /l)	WBC (10^9 /l)	PLT (10^9 /l)	Hb (g/l)	CA (mmol/l)	PHOS (mmol/l)	ALP (U/l)
Case 1	2.24	1.72	68	59	2.18	1.54	96
Case 2	3.86	7.10	229	118	2.4	1.32	114
Case 3	3.96	8.38	223	116	2.26	1.16	-

Part of related laboratory examination results is listed above. Pancytopenia and anemia occurred in case 1. Serum Calcium and phosphorus level in three cases was almost normal.; ALP, serum alkaline phosphatase; CA, serum calcium; Hb, hemoglobin; PHOS, serum phosphorus; PLT, blood platelet count; RBC, red blood cell count; WBC, white blood cell count.

Fig. 1 Schematic diagram of surgical incision.

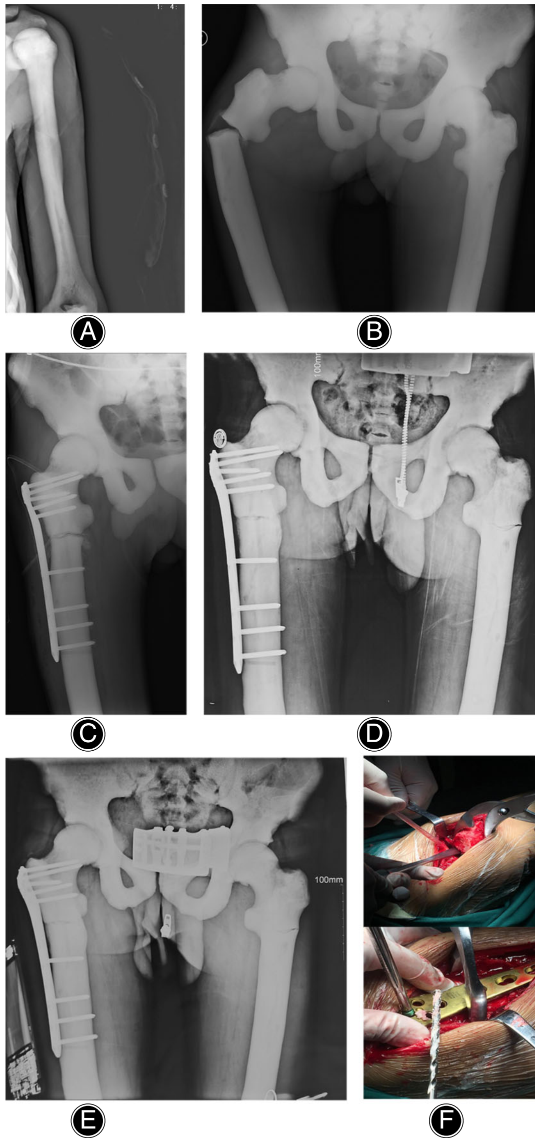

Fig. 2 Radiographs of case 1. (A) The fracture line of left humeral fracture was still recognizable. (B) Preoperative X-ray shows the right subtrochanteric fracture and the insufficient left subtrochanteric fracture in the osteopetrotic bone. The medullary cavity was obstructed. (C) Postoperative X-ray shows the fixation with DF-LCP. (D) X-ray image at the follow-up of 26 months after operation shows fracture union on the right side and the deteriorated insufficient left subtrochanteric fracture. (E) X-ray image at the follow-up of 29 months after operation shows the insufficient fracture healed. (F) Obstructed medullary cavity and stop to clean the drill bit.

greater trochanter and the fracture site, and extended properly towards the distal femoral end was made (Fig. 1). Dissection was carried out layer by layer, stopping the bleeding immediately. The fascia lata was exposed and dissected along its posteroinferior part. The fascia lata and tensor fasciae latae were pulled anteriorly. The vastus lateralis was exposed, dissected gently along its muscle fiber, and pulled to both anterior and posterior side. The femur was exposed.

Reduction and Fixation

Under the assistance of manual traction, the fracture reduction was achieved and then temporarily maintained with two reduction clamps controlled manually. A reversed contralateral titanium DF-LCP (DePuy Synthes, Obedors, Switzerland) was placed under the two clamps on the fracture site to be used in a bridge manner. Then progressive and intermittent drilling procedure was implemented carefully under C-arm fluoroscopy with continuous saline irrigation for cooling.

Two high-speed drill motors were used alternately. One stopped to clean or replace the drill bit, the other kept working. We started with 3.0 mm tungsten steel drill bit, stopping and cleaning frequently (Fig. 2F), changing to a new one when it got dull, and replacing a larger one after the penetration of the contralateral cortex. After the diameter of the screw canal was drilled up to 4.5 mm, 5.0 mm self-tapping locking screws (DePuy Synthes, Obedors, Switzerland) were inserted manually. The reduction quality of the fracture and the position of the DF-LCP and screws were verified by intraoperative anterior and lateral fluoroscopy (Fig. 2C,3B,4C-D).

Postoperative Management

A drainage tube was placed in the incision and the sutured incision was dressed with multiple layers of gauze. Conventional postoperative symptomatic treatment of detumescence, analgesia, infection prevention, and anticoagulation (subcutaneous injection of low molecular weight heparin or oral medication of rivaroxaban) were used. After the anesthesia subsided, patients were encouraged to do active joint movement of the unimpaired limbs and isometric muscle contraction exercise of the injured lower limb. The drainage

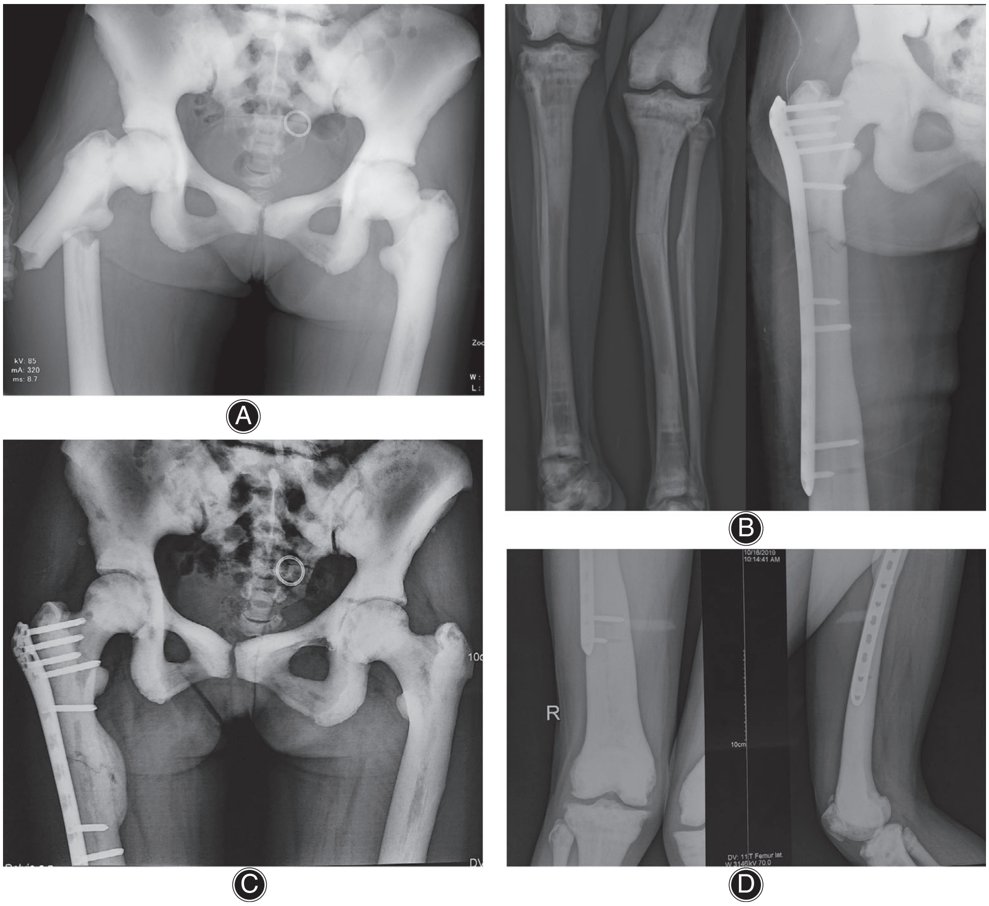

Fig. 3 Radiographs of case 2. (A) Preoperative X-ray shows the right subtrochanteric fracture; (B) The recovered insufficient left tibial and fibular fracture in osteopetrotic bone and the postoperative X-ray shows the fixation with DF-LCP. (C, D) At the follow-up of 16 months after operation, the fracture line was still clear after fracture union.

tube was removed after the drainage volume was less than 50 ml. Weight bearing exercise was not commenced until 3 months after operation to prevent the deterioration of the occurred incomplete fracture, the occurrence of fresh fracture, or implant breakage. Then, progressive weight bearing exercise with a crutch was advised. The duration of transition to full weight bearing was individualized.

Observation Indicators

The Operation Time

The operation time was recorded from the beginning of skin incision until surgical incision closure, which could reflect the efficiency of the surgical method in this complex situation.

The Amount of Intraoperative Blood Loss

The amount of operative blood loss was the sum of the amount of blood from the suction device and the amount of blood on the gauze.

Radiographic Evaluation

Osteopetrosis was diagnosed by classic radiological signs of the trunk and limbs, such as diffusely increased BMD, obstructed bone marrow cavity, endobone, rugger-jersey vertebrae, and erlenmeyer deformity. Subtrochanteric fracture was diagnosed by anteroposterior view X-ray radiographs and it was further verified by CT scan with 3D reconstruction.

Follow-Up

The patients were followed up at 1, 3, 6, and 12 months postoperatively and then at least once a year. Physical examination and anteroposterior view X-ray of the fracture site were used to evaluate the fracture union status, the position and stability of the implant, and the alignment of the injured limb.

Clinical Evaluation

Harris Hip Score (HHS). Harris Hip Score (HHS) was used to evaluate the function of the hip joint at 6 and 12 months after operation. The HHS was developed for the assessment of the results of hip surgery and evaluation of various hip disabilities and methods of treatment in an adult population. The domains covered are pain, function, absence of deformity, and range of motion. The score has a maximum of 100 points (best possible outcome) covering pain (1 item, 0–44 points), function (7 items, 0–47 points), absence of

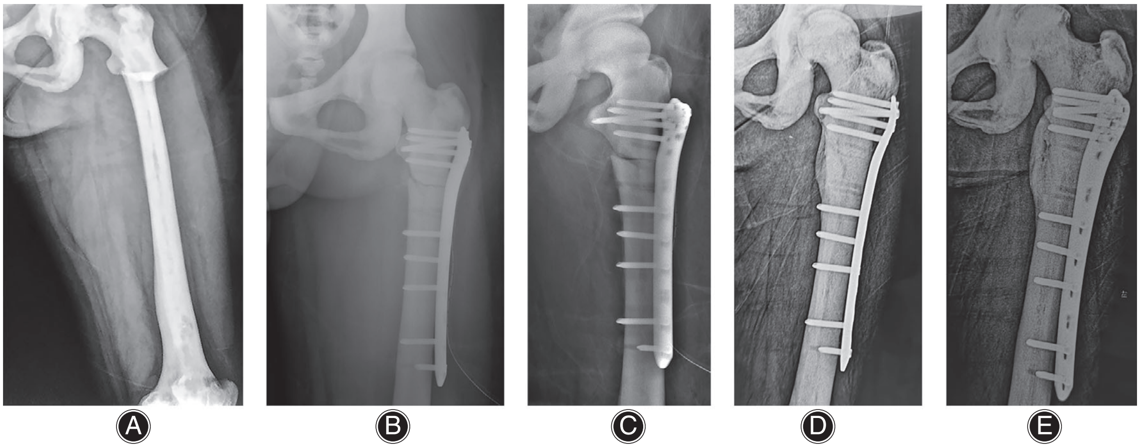

Fig. 4 Radiographs of case 3. (A) Preoperative X-ray shows the left subtrochanteric fracture in osteopetrotic bone. (B, C) Postoperative X-ray shows the fixation with DF-LCP. (D, E) At the follow-up of 12 months after operation and bone union.

deformity (1 item, 4 points), and range of motion (2 items, 5 points)[29].

Results

Intraoperative Results

The chalk-like bone was fragile but hard to drill. Krischner wire was not suitable for temporary fixation in osteopetrotic bone. It was not easy to maintain the reduction. The medullary cavity was significantly obstructed. For case 1, the osteopetrotic bone callus around the fracture site was removed, cut into small cubes, and grafted back. Partial recreation of the medullary canal and limited osteotomy to freshen the fracture surface were tried but there wasn't satisfactory punctate bleeding, which indicated the blood supply of the fracture site was poor, and the time of bone union might be longer than normal. In the drilling procedure, bone debris would adhere to the flute of drill bit and needed to be removed frequently. Four units of erythrocyte and 400ml plasma were used intraoperatively in cases 1 and 2 (Figs 3 and 4).

General Results

All patients accepted ORIF using a reversed contralateral DF-LCP inserted through the progressive and intermittent drilling procedure. The average operation time was 140 ± 21.60 min (110, 160, and 150 min, respectively); the average intraoperative blood loss was about 333.33 ± 23.57 ml (300, 350, and 350 ml respectively). Because of pancytopenia and anemia (Table 2), case 1 underwent several times of blood transfusion therapy with a total of 16-unit erythrocyte and 1800 ml plasma in perioperative period. His blood routine examination showed hemoglobin of 96 g/l, white blood cell count of 2.52×10^9 /l, red blood cell count of 3.43×10^{12} /l, and platelet of 67×10^9 /l at 3 days after operation.

Follow-Up

The patients were followed up with physical examination and anteroposterior views X-ray at the time of 1, 3, 6, and 12 months postoperatively, and then at least once a year. The average follow-up period was 22.33 ± 7.41 months (29, 26, and 12 months respectively). At the time of 6 months after operation, case 1 and 3 were almost pain-free and could walk with full weight bearing while case 2 could walk only with partial weight bearing using a crutch. The HHS score of case 1, case 2, and case 3 was 84/100, 74/100, and 92/100, respectively. At the follow-up of 12 months after operation, the HHS score improved to 91/100, 81/100, and 96/100, respectively. Bone union was achieved in all patients with an average time of 6.67 ± 0.94 months (6, 8, and 6 months, respectively). At the latest follow-up, the stability and position of the implant and the alignment of the impaired lower limb were good (Figs 2-4). The contralateral incomplete old subtrochanteric fracture was deteriorated in case 1 at 26 months after operation (Fig. 2D). Three months of crutch assistance and limited weight bearing was advised, and bone union was verified 3 months later (Fig. 2E) and then he was back to full weight bearing. Fresh contralateral subtrochanteric fracture occurred in case 2 at 26 months after operation. A similar surgical approach was implemented. The outcome of her fresh fracture is under follow-up.

Complications

No perioperative complications such as operation-related death, vascular/nerve injury, deep venous thrombosis, fat embolism, pulmonary embolism, and incision-related superficial or deep tissue infections were identified. No late complications including malunion, nonunion, ankylosis, heterotopic ossification, osteonecrosis, and infection were discovered.

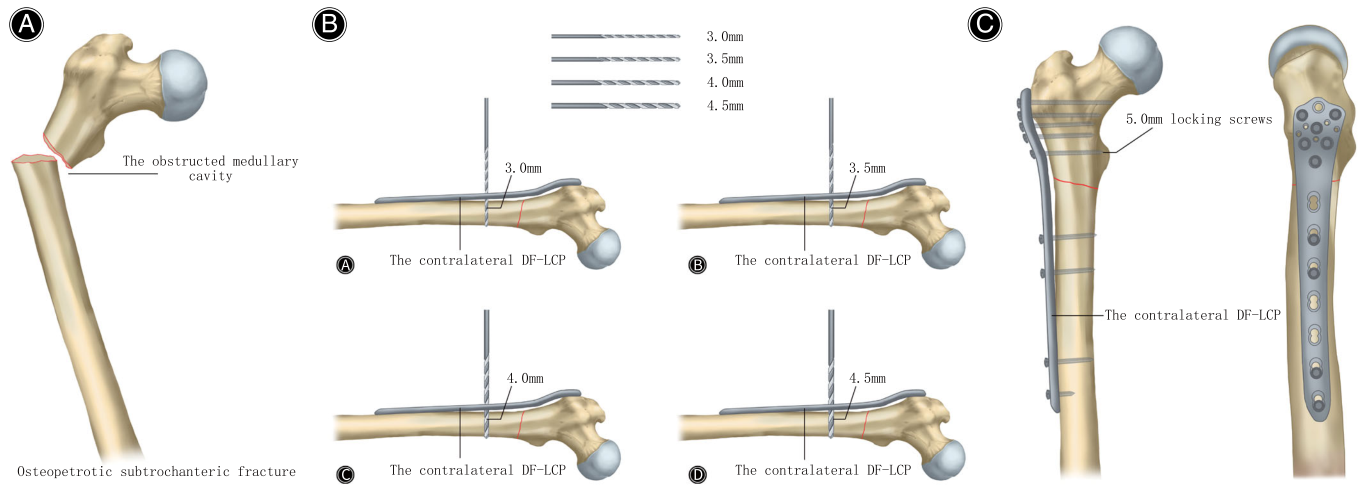

Fig. 5 (A) Osteopetrotic subtrochanteric fracture and obstructed medullary cavity (B) Progressive drilling procedure, each screw canal was formed by drill bits with sequentially increased diameter (3.0, 3.5, 4.0, and 4.5 mm). (C) Anteroposterior view and lateral view of osteopetrotic subtrochanteric fracture after the fixation with contralateral distal femoral locking compression plate. The diameter of those locking screws was 5.0 mm.

Discussion

Osteopetrotic bone is characterized by increased bone deposition on unresolved calcified cartilage or primary spongiosa. The majority of OPF were fractures around trochanteric region, such as femoral neck fractures, trochanteric and subtrochanteric fractures[15], which may be on the account of concentrated stress. OPF in other bones such as tibia[30–32], fibula[30,31], humerus[16,33], vertebrae[34,35] were also reported but relatively infrequent. In present study, case 1 had left humeral fracture about 30 years ago and recovered well with plaster immobilization. Case 2 and 3 denied previous fracture history but old fracture in left fibula and tibia was verified in radiograph of case 2.

The Choice of Conservative Treatment or Surgical Treatment

While several cases underwent conservative therapy obtained fracture union without evident complication[30,36–38], the complication rate of conservative management was high in other reports. Relatively, fewer studies support conservative treatment of OSF. Hasenhuttl *et al.*[39] reported bone union at 10 weeks following the use of Russell traction to treat a peritrochanteric fracture in a 27-year-old male with osteopetrosis. In addition, some researchers have reported successful bone union in patients with osteopetrotic femur fractures who were conservatively treated with plaster and traction[30,36,37]. Birmingham *et al.*[40] described coxa vara and external rotation deformity in a patient with OSF and ipsilateral femoral neck fracture treated with spica cast. Two studies[21,41] revealed that there were fracture malunion and coxa vara left in their patients treated nonoperatively. In our study, case 1 had accepted conservative treatment of immobilization with Chinese traditional plaster to treat his right OSF for a month before he was transported to us. The outcome was not satisfactory because the reduction cannot be maintained well. Then he turned to us for a choice of surgery. Anatomical reduction was difficult to maintain and longer recovery periods were required following conservative treatment. Weight bearing was restricted for an average of 3 months in those case reports. Long-term immobility and bed rest may lead to deep venous thrombosis and amyotrophy.

For surgical treatment of OSF, the largest challenge is the combined brittleness and hardness of osteopetrotic bone, which makes it easy to cause instruments (the drill bit and reamer) and screw breakage[11,16,18,33,42], iatrogenic fracture[11,43], thermal osteonecrosis, prolonged operation time, increased blood loss, and increased risk of infection[25]. Amit *et al.*[11] reviewed the available literature and reported a non-union rate of 12%, infection rate of 12%, and hardware failure rate of 29% on the surgical treatment of osteopetrotic fractures in adults. According to our literature review on osteopetrotic fracture around femoral trochanter[13–28,31,36–41,43–52], the non-union rate of the surgical treatment was about 15.28% (11/72), infection rate was about 9.72% (7/72), hardware failure rate was about 11.11% (8/72), and periprosthetic fracture incidence was about 8.33% (6/72). It is a serious challenge for orthopaedic surgeons to choose the appropriate implants and surgical procedures for osteopetrotic fracture.

The choice of conservative treatment or surgical treatment depends on the medical support available. Surgical treatment could offer better reduction and better condition to maintain it. When the condition of operation is sufficient, surgical treatment should be implemented after proper preparation. If not, conservative treatment under frequent follow-up should be implemented.

The Choice of Implant

The frequently used implant for subtrochanteric fractures is intramedullary (IM) nail, which minimizes blood loss and fracture exposure, and enables early weight bearing. Chhabra *et al.*[15] reported internal fixation with IM nails in two cases and observed the generally increased time to union in their case series. They found relatively more failure of load-bearing implants and summarized two primary factors attributed to failure regardless of treatment approach: one is the increased mechanical demands placed on implants because of the prolonged time to union, the other is the biochemical inability of osteopetrotic bone to hold the screws securely. Thus, they believed load-sharing IM implant had superiority compared with extramedullary (EM) load-bearing implant. They also pointed out the extraordinary difficulty in opening marrow canal and introduced the use of a series of progressively larger drill bits. Inserting an intramedullary nail into the narrow or obstructed medullary canal of osteopetrotic bone was difficult. They experienced the breakage of several drill bits and at least 2-hour longer operation time compared with standard IM nailing. Kumbaraci *et al.*[16] reported the application of proximal femur nail antirotation (PFNA) in one case, emphasized the difficulty of recreating the medullary canal and the risk of iatrogenic fracture of nail insertion, and stated that the reversed less invasive sterilization system (LISS) provided less fixation strength than PFNA. If reversed LISS was applied, stress concentration and locking screws loosening may be induced by early weight bearing.

Obviously, in osteopetrotic bone, inserting screws and nails with larger diameter mean that the drilling procedure takes a longer time. This might answer why femoral component with shorter length and smaller diameter was applied or recommended in total hip arthroplasty (THA) in osteopetrosis[46,53]. The complications, such as delayed fracture union and infection, were found in at least three case reports in the treatment of osteopetrotic fracture with IM nail. To address this issue, we chose EM implants.

Several EM implants, such as dynamic hip screw (DHS), proximal femoral locking compression plate (PF-LCP), and DF-LCP, were reported to be used in treating OSF. For the use of DHS, Rysavy[23] described the prolonged operation time, which was up to 4 hours, and Kumar[22] emphasized the difficulty in the creation of the screw canal towards femoral head. Dawar *et al.*[24] reported four cases of osteopetrotic fracture managed with PF-LCP and obtained a good outcome, but they also faced the difficulty of inserting screws into femoral neck. Amit *et al.*[28] reported the application of reversed DF-LCP in two cases of OSF and fracture union was obtained without evident complication. They stated that the reversed DF-LCP matched well with the contralateral proximal femur and its mechanical quality had advantages. In our cases, we chose DF-LCP as there were no screws with large diameter (our locking screw is 5.0 mm in diameter) and long screws needed to be inserted towards femoral head. The requirement of screw canal drilling and screw insertion is more practicable. To avoid the problem of stress concentration, locking screws loosening, and implant failure, weight bearing exercise was prolonged in all our cases. The fracture union was verified with an average time of 6.67 ± 0.94 months (6, 8, and 6 months, respectively) after operation without evident complication. But the fracture line remained clear on X-rays of case 1 and 2 about 2 years after surgery, which was identical with the radiographic findings in a study by Hiyama *et al.*[26].

EM implant insertion was less time-consuming and more practicable than IM implant. When fixation was carried out with EM implant, weight bearing exercise should be postponed. From our experience, reversed contralateral DF-LCP fixation is suitable and reliable in treatment of OSF.

The Efficient Method of Implant Insertion

In regards to the application of the drill, Rafiq *et al.*[54] introduced the use of “high-speed steel drill bit” with “low-speed and high-torque drill motor” in an osteopetrosis patient with non-union humeral fracture; they described the use of continuous saline irrigation to avoid the problem of thermal necrosis. Four studies[25,51,53,55] reported the use of metal-cutting drill bit, diamond drill bit, and industrial grade tungsten carbide drill bit, indicating that reliable drill bits with enough intensity were necessary. Dawar *et al.*[24] reported the application of multiple special tungsten tipped drill bits with different sizes and introduced the use of high speed power system to avoid toggle while drilling and screw insertion. In our cases, we prepared several sets of 3.0, 3.5, 4.0, and 4.5 mm sterilized tungsten steel drill bits preoperatively. Progressive and intermittent drilling with continuous saline irrigation was implemented and the process of screw insertion was successful. The average operation time was 140 ± 21.60 min (110, 160, and 150 min, respectively), which was almost identical to operation in normal bone. From our experience, preparing several sets of drill bit with enough intensity preoperatively is important. Progressive and intermittent drilling is a safe and efficient method for implant insertion in this complicated situation.

Tapping Procedure is Not Necessary

For the tapping procedure, Yamane *et al.*[11] reported the implementing of anterior cervical arthrodesis for chronic hangman’s fracture in a case. They stated the hard bone stripped the thread of the tap and iatrogenic fracture occurred while tapping, which demonstrating that the intensity of the standard screw tap was insufficient for osteopetrotic bone. In normal procedure, the diameter of the screw canal will be drilled up to 4.3 mm and then a 5.0 mm locking screw was inserted. But it is difficult to insert 5.0 mm screw into the 4.3 mm bone canal in osteopetrotic bone without tapping procedure. Therefore, we chose to drill the diameter of screw canal up to 4.5 mm and then insert 5.0 self-tapping locking screws manually. It was practicable. No screw breakage or screw toggle occurred during operation and the stability of implant was satisfied.

According to our experiences, there were several key points for treatment of OSF. (i) Surgical treatment is recommended with sufficient medical support and proper preoperative preparation. (ii) Insertion of EM implant is more practicable than IM implant, and reversed contralateral DF-LCP is suitable and reliable in fixation of OSF. (iii) When fixation with EM implant, weight bearing exercise should be postponed to avoid stress concentration, locking screws loosening, and implant failure. (iv) Preparing several sets of drill bit with enough intensity preoperatively is important. (v) Intermittent and progressive drilling procedure with continuous saline irrigation is secure and efficient. (vi) Tapping procedure is not necessary if the screw canal is enlarged properly by drilling.

There were several limitations to our study. First, this is a retrospective clinical case analysis without control group. Second, the number of included cases is limited as OSF is a rare injury. Next, the comparison on application of different implant is descriptive and lacks statistical support. Lastly, further work of pedigree survey and gene analysis need to be conducted.

Conclusion

Application of reversed contralateral DF-LCP in OSF is practicable and reliable. Progressive and intermittent drilling is a safe and efficient method for implant insertion in treatment of OSF.

Disclosure

The authors declare that they have no competing interests. All authors are in agreement with the manuscript.

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Disclosure

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Authors' Contributions

HL J, JW W, YL Y, F W, and BM W implemented the operation. C L and XL L were responsible for collecting the patients' data. JJ Y and Y T were responsible for reviewing the previous reports. Y T and FX L wrote the paper. FX L and YL Y are responsible for the quality control of the article. All authors read and approved the final manuscript.

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