CASE REPORT

Combined Proximal Tibial Osteotomy for Adult Kashin–Beck Disease with Severe Varus Knee Osteoarthritis: Case Report and Literature Review

Yunfei Liu, MD[1], Ruiyang Li, MD[2], Yu Zhan, MD[2], Xuetao Xie, MD, PhD[2], Congfeng Luo, MD, PhD[2]

[1]Department of Orthopaedics, The Affiliated Suzhou Science & Technology Town Hospital of Nanjing Medical University, Suzhou and [2]Department of Orthopaedic Surgery, Shanghai Jiao Tong University Affiliated Sixth People's Hospital, Shanghai, China

Background: Kashin–Beck disease (KBD) is an endemic, chronic osteoarthropathy that seriously affects joint function and can lead to severe knee deformity. Osteotomy is considered to be one of the effective methods for the treatment of this disease. Therefore, we designed a novel type of osteotomy named combined proximal tibial osteotomy (CPTO), which combines the characteristics of opening-wedge high tibial osteotomy and tibial condylar valgus osteotomy.

Case presentation: We report the case of a 48-year-old male with knee pain and varus deformity who was diagnosed with KBD and varus knee osteoarthritis (Kellgren–Lawrence stage IV). Considering the patient's relatively young age, a varus deformity of the right knee of 16.79°, and an intra-articular instability, we performed a CPTO treatment. In this procedure, we performed an L-shaped osteotomy from the medial edge of the proximal tibia to the intercondylar eminence and an osteotomy from the medial side of the proximal tibia to the lateral side through the same incision, to adjust the leg alignment and the congruity of the joint by valgus correction. At 29 months follow-up, this patient achieved satisfactory results, with a varus right knee of 2.87°. There was significant improvement in his right knee function, pain, and joint stability.

Conclusions: CPTO may be an acceptable treatment for KBD patients with severe knee varus deformity and intra-articular instability. It can be considered as an alternative treatment, especially for patients with advanced osteoarthritis needing knee preservation.

Key words: Kashin–Beck disease; osteoarthritis; osteotomy; varus

Introduction

Kashin–Beck disease (KBD), which was first described in Russia by Kashin in 1848 and then Eugene Beck in 1906[1], is an endemic, chronic, and deformed osteoarthropathy. Pathogenesis of the KBD is characterized by the degeneration and deep cell necrosis of epiphyseal cartilage, epiphyseal plate cartilage, and articular cartilage of the extremities in childhood[1]. Clinically, patients present with multiple symmetrical joints involvement, including knee joints, with joint pain, swelling, deformation, muscle atrophy, and even joint deformity[2].

The medial opening-wedge high tibial osteotomy (OWHTO) is increasingly recognized as an effective surgical procedure for the treatment of osteoarthritis with varus knee, especially in young, active patients[3]. OWHTO is an extra-articular osteotomy of the proximal tibial to correct the mechanical leg axis and shift the weight-bearing from the medial compartment of the knee joint to the lateral, thus relieving pain and delaying the progression of osteoarthritis[4]. Nevertheless, patients with advanced varus knee osteoarthritis frequently consist of both intra-articular and extra-articular deformities[5]. It was reported that when the joint line convergence angle (JLCA) of the knee joint was more than 6° or a tibial plateau shape was the pagoda-type, OWHTO could not achieve satisfactory results[6]. Tibial

Address for correspondence Congfeng Luo, MD, PhD, and Xuetao Xie, MD, PhD Department of Orthopaedic Surgery, Shanghai Jiao Tong University Affiliated Sixth People's Hospital, Shanghai, China 200233 Tel: +86 021-64369181; Fax: +86 021-64701361; Email: congfengl@outlook.com; xuetaoxie@163.com

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condylar valgus osteotomy (TCVO), a technique described by Chiba in the 1990s in Japan, is an L-shaped osteotomy from the medial margin of the proximal tibia to the intercondylar eminence to adjust the mechanical axis of the leg and the knee congruity[7]. TCVO is an intra-articular osteotomy altering the shape of the tibial plateau. This procedure has shown promising clinical and radiographic results for advanced medial knee osteoarthritis with subluxated lateral joint[8]. However, the ability of TCVO to correct severe varus deformity of the knee may be insufficient. Hence, a novel osteotomy method named combined proximal tibial osteotomy (CPTO) was designed, which combines the characteristics of OWHTO and TCVO[9].

To our knowledge, clinical cases of CPTO have not been reported in the previous literature. The purpose of this study is to introduce the clinical application of this innovative osteotomy and report its clinical outcome in a KBD patient.

Case Report

A 48-year-old male with a height of 165 cm and a weight of 75 kg visited our clinic for the first time due to both knee pain for several years. X-rays of the right knee showed hyperplasia and degeneration (Figure 1A). One year after conservative treatment, the patient complained increasing pain in the right knee, and was unable to walk more than 200 m every day. The standing full-length leg X-ray showed medial tibial articular surface collapse, lateral plateau gap widening, and both knees varus (Figure 1B). We measured the following parameters of the right leg: mechanical lateral distal femoral angle (mLDFA): 87.82°, mechanical medial proximal tibial angle (mMPTA): 76.85°, JLCA: 5.82°, mechanical femorotibial angle (mFTA): 196.79°. On physical examination, both knees were swollen and varus, both knee active range of motion (ROM) of 0°–100°, passive ROM of −10° to 100° (Figure 1C,D), and varus–valgus stress test was positive. Magnetic resonance imaging (MRI) and computed tomography (CT) of the right knee indicated knee osteoarthritis, wear of the medial tibiofemoral articular surface, collapse of the anteromedial tibial plateau, medial meniscus injury, periarticular ligament degeneration, knee effusion, synovial hyperplasia, and formation of synovial osteochondroma (Figure 2).

Tracing back the patient's medical history, we found that he was born in an epidemic area of KBD in northeastern part of China. But he had been living in our city with his

Fig. 1 Preoperative photographs and radiography. (A) The X-rays of the right knee in the non-weight-bearing position. (B) The standing full-length leg X-ray. (C) Standing with weight-bearing showed the right knee joint was significantly varus. (D) Incomplete knee flexion while squatting

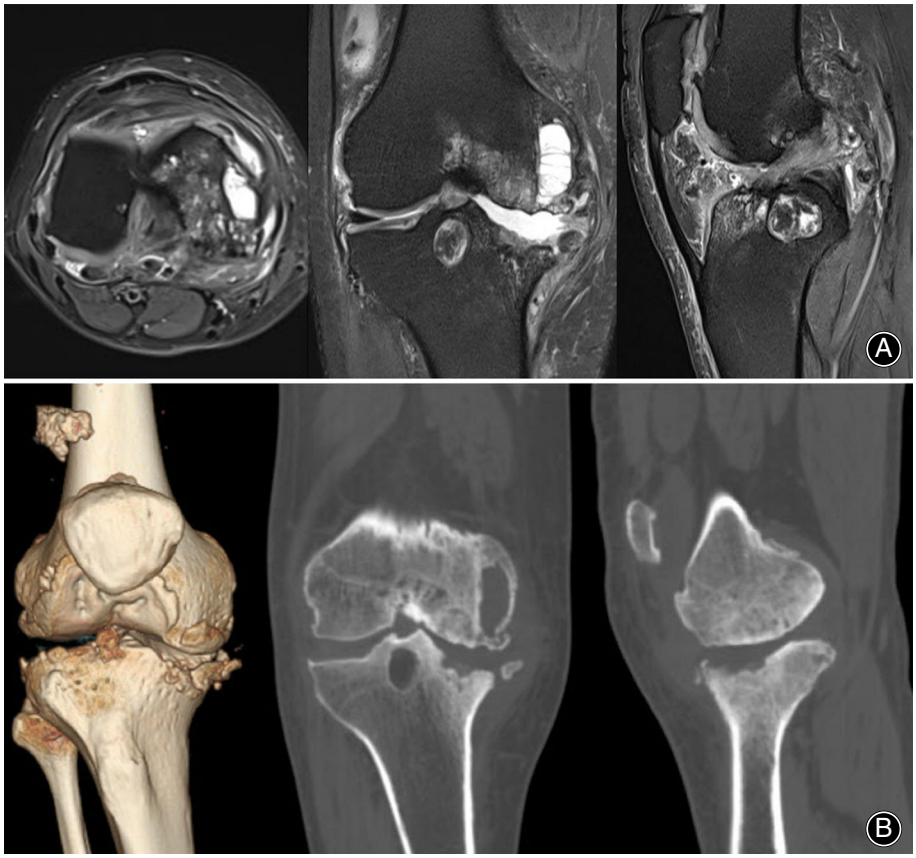

Fig. 2 The preoperative MRI images (A) and CT images (B) of the right knee joint. CT, computed tomography; MRI, magnetic resonance imaging

parents since he was 7 years old. Several years ago, he gradually developed swelling and pain in some joints of his body, but he never went to the hospital. His siblings also had similar symptoms while his parents and children did not. Thus, primary diagnosis was given: KBD, varus knee osteoarthritis (Kellgren–Lawrence stage IV). The results of X-ray examination of both the hands and elbows could further confirm the diagnosis of KBD.

Preoperative examinations, such as chest radiograph and electrocardiogram, showed no abnormalities. Due to limited financial conditions, the patient requested only right knee surgery. Considering the patient's relatively young age, a varus deformity of the right knee of 16.79°, and intact lateral compartment, osteotomy was feasible. We planned to restore intra-articular stability and then adjust the lower limb alignment. CPTO has the ability to correct severe intra- and extra-articular deformities, so we performed a CPTO treatment.

Operative Procedure

With the patient in a supine position, a longitudinal skin incision was made from the medial patellar ligament to the medial tibial crest (Figure 3A). The superficial insertion of the pes anserinus and the medial collateral ligament were dissected medially, and the tibial tubercle was exposed laterally. The anteromedial periosteum of the proximal tibia was exposed. After locating the insertion point using TomoFix Medial High Tibial Plate (DePuy Synthes, Solothurn, Switzerland), one Kirschner wire was placed to design the OWHTO plane, then the other two Kirschner wires were placed to design the TCVO plane (Figure 3B). After the satisfactory orientation and depth of Kirschner wires under the fluoroscopic guidance, the osteotomy was performed below the tibial tubercle. Then, the L-shaped osteotomy was made from the medial margin of the proximal tibia to the intercondylar eminence. The medial condyle of the tibia was elevated with the spreaders (Figure 3C,D). After completing the TCVO, two Kirschner wires were used to temporarily fix the medial tibial condyle to the lateral plateau. The next step was to perform the wedge-shaped osteotomy from the medial to the lateral tibia along the direction of the Kirschner wire in the previous OWHTO plane (Figure 3E,F). Keep the lateral hinge as intact as possible. Then the entire proximal tibia was slowly elevated with the osteotomes (Figure 3G). The osteotomes were replaced with the spreader, and the leg alignment was corrected to the destinated mechanical axis (Figure 3H). After placing the plate, the fluoroscopic image and the opening gap of the osteotomy (18 mm anteriorly and 14 mm posteriorly) were satisfactory (Figure 3I,J). The TomoFix plate system were installed, and one additional

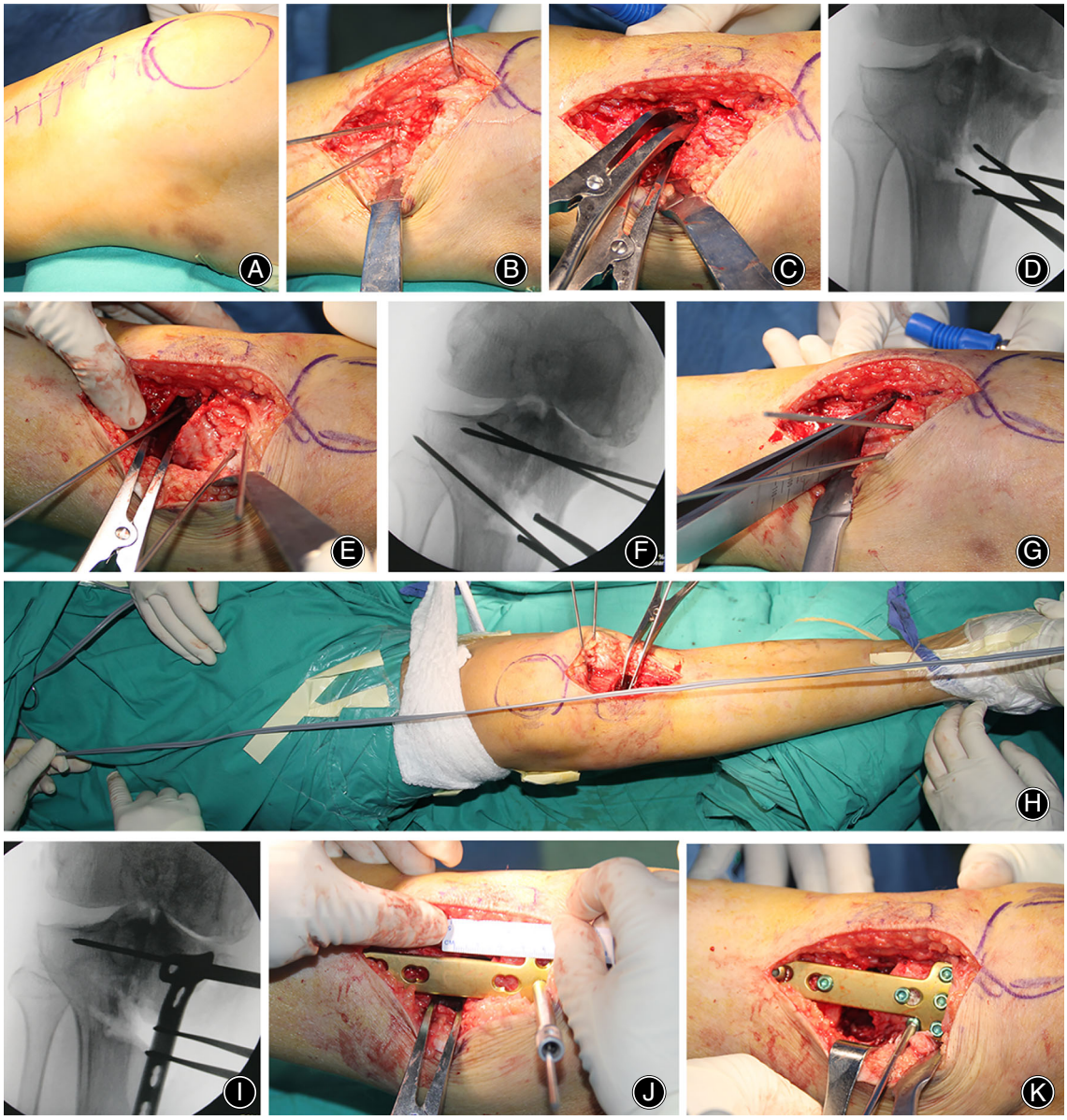

Fig. 3 Intraoperative photographs and fluoroscopy. (A) The incision and anatomical landmarks. (B) Location of three Kirschner wires. (C, D) The L-shaped osteotomy was completed. (E, F) The wedge-shaped osteotomy was performed. (G, H) The leg alignment was corrected. (I-K) Osteotomy fixation

locking screw next to the plate was inserted to reach the lateral cortex of the tibia (Figure 3K). The surgical duration was 120 min, and the intraoperative blood loss was about 200 ml.

Postoperative Management and Follow-Up

On the first postoperative day, the patient underwent X-ray and CT examination (Figure 4) and was instructed to exercise the flexion and extension of the right knee. Then, he was followed up at regular intervals. The patient had partial weight-bearing activity of the right knee in the fourth week and full weight bearing activity in the eighth week after surgery.

At 29 months follow-up, there was significant improvement in his right knee function, pain, and joint stability. The hospital for special surgery knee score (HSS) was 94 (pain, 30; function, 22; ROM, 12; muscle strength, 10; flexion deformity, 10 and stability, 10). The active and passive ROM of the right knee joint was 0°–100°. The right knee varus-valgus stress test was negative. Standing full-length leg X-ray indicated that the mFTA was 182.87°, the mMPTA was 88.40°, and the JLCA was 2.38° (Figure 5A). CT and MRI showed the osteotomy area was healed and improvement in osteoarthritis of the right knee (Figure 5B, C). The patient was satisfied with the status of his right knee (Figure 5D,E).

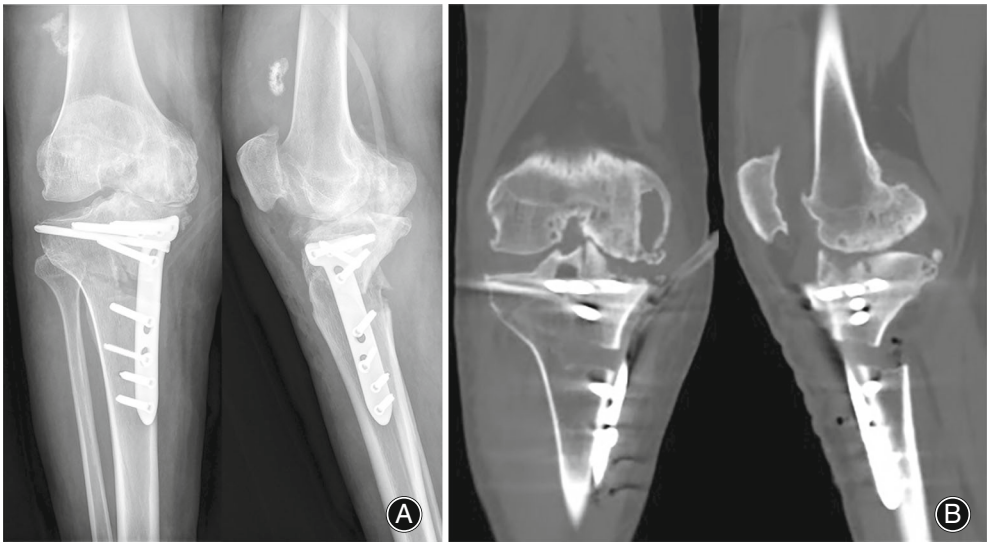

Fig. 4 The imaging appearances of the right knee after the CPTO. The postoperative X-rays (A), coronal and sagittal CT scans (B) showed that the medial tibial plateau had been elevated and had an adjusted posterior slope. CPTO, combined proximal tibial osteotomy; CT, computed tomography

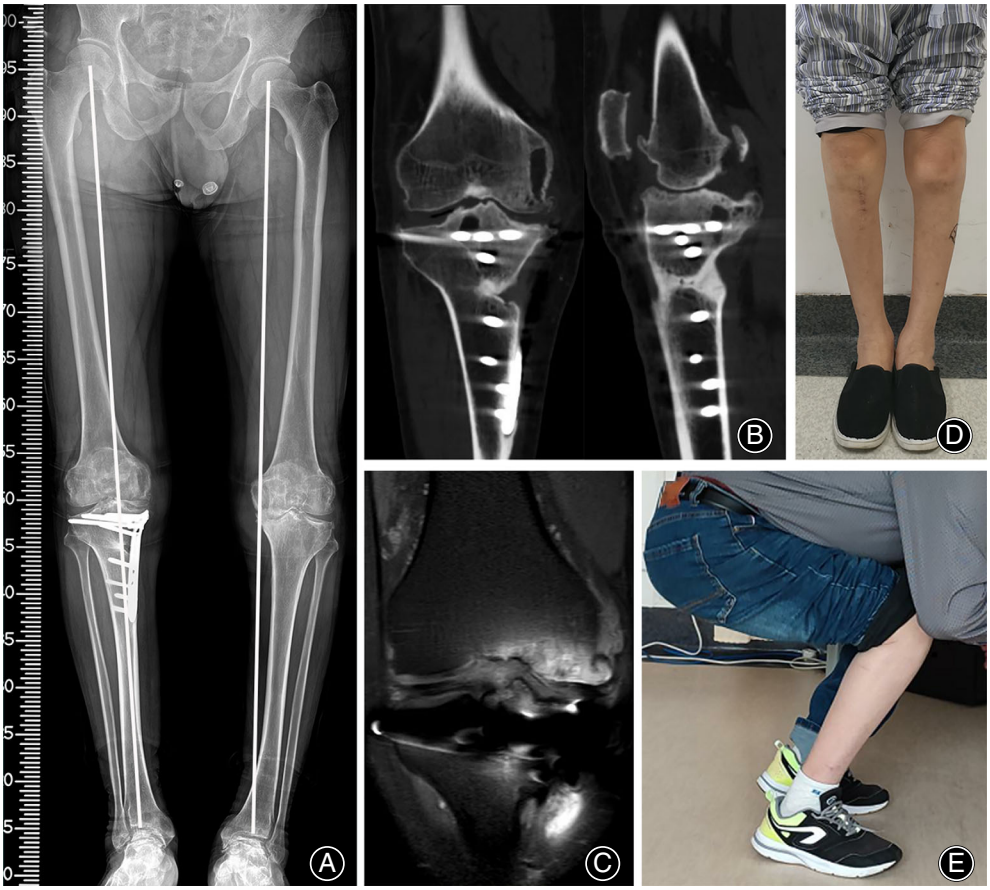

Fig. 5 The clinical outcome and imaging appearances at 29 months follow-up. (A) The standing full-length leg X-ray. The osteoarthritic presentations were improved in the views of CT (B) and MRI (C). (D, E) Good functions while standing and squatting. CT, computed tomography; MRI, magnetic resonance imaging

Discussion

In non-epidemic areas, it is not easy to detect and confirm KBD. Although there were about 645,000 KBD patients in China by the end of 2012, they were mainly distributed in economically poor mountainous areas[10]. The age of onset in adolescents is generally before the age of 15[2]. With the national prevention and control of endemic diseases in recent decades, the emerging trend of pediatric KBD in China has been significantly controlled[11]. In most cities far from the affected area, this disease is rare, especially with

TABLE 1 Surgical treatment for KBD with knee deformity: Review of the English literature

Report	Gender (n)	Age (years)	Varus/ valgus deformities (n)	Treatment	Follow-up (months)	Outcome	Complications	Refs.
Jin et al.	M, 7; F, 15	57.55 ± 7.49	Varus 21; Valgus 4	TKA	44.7 ± 30.3	Good	None	13
Tang et al.	M, 2; F, 13	66.3	Varus 18	TKA	32.47 ± 10.05	Good	None	16
Liu et al.	M, 121; F, 74	12–41	Varus 37; Valgus 210	Osteotomy with impaction	74.4	Good	Infection (6.4%)	18

Abbreviations: F, female; KBD, Kashin–Beck disease; M, male; Ref., references; TKA, total knee arthroplasty.

onset in adulthood. Therefore, many doctors know little about the disease, which may easily cause clinical misdiagnosis and missed diagnosis. Currently, there are no specialized biochemical tests to confirm the diagnosis of KBD. The diagnosis needs to be satisfied by (i) living in endemic areas for at least 6 months, and (ii) characteristic changes of hand X-ray image[12]. This requires doctors to carefully analyze the patient's medical history, perform physical examination, and combine specific X-ray and MRI examinations to make the correct diagnosis. This patient was born in the epidemic area and migrated to another city more than 1000 km away from his hometown at the age of 7 years. The change in the living environment during childhood may be responsible for his late onset.

For KBD patients with knee deformity, few studies have been published to date on surgical treatment (Table 1), and the best surgical treatment has been controversial. Although the etiology of KBD is different from that of osteoarthritis, the symptoms of chronic osteoarthritis caused by KBD in adults are similar and common. The current treatment methods for KBD are based on the treatment experience of osteoarthritis[13]. Surgical methods include unicompartmental knee arthroplasty (UKA), total knee arthroplasty (TKA), and osteotomy. UKA is indicated for patients younger than 60 years with unicompartmental knee osteoarthritis who have low activity requirements and varus or valgus deformity under 15°[14]. Young age and varus deformity directly affect the postoperative satisfaction of patients[15]. Tan *et al.*[16] performed TKA treatment on 15 patients with KBD (18 knees). The patients' pain and knee function improved significantly 32 months after surgery. However, the ideal indication for TKA is patients older than 65 years with advanced knee osteoarthritis and with total knee injuries. Large sample statistics showed that the lifetime risk of revision was as high as 35% for men in their early 50s treated with TKA[17]. In this case, the patient was a 48-year-old employed in manual labor with significant varus deformity of the knee. Therefore, neither UKA nor TKA seemed to be the optimal option. Liu *et al.*[18] reported that they adopted a method of osteotomy with impaction and plaster immobilization in 195 underage KBD patients (247 knees) with severe knee deformities and achieved acceptable results. With the development of osteotomy, OWHTO is increasingly used for the treatment of young knee osteoarthritis patients who had high activity requirements and complete lateral genicular compartments[19]. But excessive correction angle leads to hinge fracture and patients with significant intra-articular deformity are prone to residual deformity[6]. For this patient, he had significant intra-articular instability because of severe intra-articular deformity. OWHTO might not improve his intra-articular instability. Compared with OWHTO, TCVO seemed to be more effective in patients with severe intra-articular instability[20]. However, the ability of valgus correction in TCVO is limited. In this case, we estimated that tibial valgus correction required at least 17°, whereas TCVO generally worked best when the varus deformity was less than 10°. At last, we decided to treat this patient with CPTO.

CPTO, as a novel osteotomy procedure, has several main characteristics. First, it combines the advantages of OWHTO and TCVO through the same surgical incision, which can not only correct large varus deformity but also deal with intra-articular instability in severe knee osteoarthritis. Second, CPTO uses the TomoFix plate system and requires only one additional screw compared with OWHTO. It does not require specially designed internal fixation. Both biomechanical tests and finite element analysis showed that there was no significant difference in stability between the CPTO construct and the OWHTO construct following fixation[9]. Third, CPTO consists of OWHTO and TCVO, both of which are familiar surgical procedures for orthopaedic surgeons. It may be easily mastered by surgeons who have already performed OWHTO or TCVO. We believe that the proper patient selection, the correct surgical method, the experience of the surgical team, and the reasonable rehabilitation are important reasons for the success of this case. Despite the successful case treated with CPTO, more studies and time are needed to evaluate its long-term efficacy and deficiencies in the future.

CPTO may be an acceptable treatment for KBD patients with severe varus knee osteoarthritis. It may also be considered as an alternative treatment for osteoarthritis patients with severe knee varus deformity and intra-articular instability, especially those needing knee preservation.

Author Contribution

Yunfei Liu wrote the manuscript. Ruiyang Li and Yu Zhan collected the data and reviewed the manuscript. Xuetao Xie participated in the surgery and revised the manuscript. Congfeng Luo was the lead surgeon and revised the manuscript.

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Conflict of Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Ethical Approval

The study was approved by the Ethics Committee of Shanghai Sixth People's Hospital (Approval Number: 2021–099). All authors listed meet the authorship criteria according to the latest guidelines of the International Committee of Medical Journal Editors. All authors were in agreement with the manuscript.

Informed Consent

The informed consent for publication was obtained from the patient.

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