

World Journal of *Clinical Cases*

World J Clin Cases 2023 October 16; 11(29): 6974-7260



MINIREVIEWS

- 6974** Applications of time series analysis in epidemiology: Literature review and our experience during COVID-19 pandemic

Tomov L, Chervenkov L, Miteva DG, Batselova H, Velikova T

ORIGINAL ARTICLE**Retrospective Cohort Study**

- 6984** Acute cholangitis: Does malignant biliary obstruction *vs* choledocholithiasis etiology change the clinical presentation and outcomes?

Tsou YK, Su YT, Lin CH, Liu NJ

Retrospective Study

- 6995** Usefulness of analyzing endoscopic features in identifying the colorectal serrated sessile lesions with and without dysplasia

Wang RG, Ren YT, Jiang X, Wei L, Zhang XF, Liu H, Jiang B

- 7004** Roles of biochemistry data, lifestyle, and inflammation in identifying abnormal renal function in old Chinese

Chen CH, Wang CK, Wang CY, Chang CF, Chu TW

- 7017** Clinical efficacy and safety of Guipi decoction combined with escitalopram oxalate tablets in patients with depression

Yu J, Xu FQ

- 7026** Artificial intelligence technology and ultrasound-guided nerve block for analgesia in total knee arthroplasty

Tong SX, Li RS, Wang D, Xie XM, Ruan Y, Huang L

- 7034** Axenfeld-Reiger syndrome: A search for the missing links

Morya AK, Ramesh PV, Sinha S, Nishant P, Nain N, Ramavath RN, Gone C, Prasad R

Observational Study

- 7043** Self-management of osteoarthritis while waiting for total knee arthroplasty during the COVID-19 pandemic among older Malaysians

Mahdzir ANK, Mat S, Seow SR, Abdul Rani R, Che Hasan MK, Mohamad Yahaya NH

- 7053** "In situ bone flap" combined with vascular pedicled mucous flap to reconstruction of skull base defect

Qian M, Chen X, Zhang LY, Wang ZF, Zhang Y, Wang XJ

- 7061** Reference values of gait parameters in healthy Chinese university students: A cross-sectional observational study

Yu JS, Zhuang C, Guo WX, Chen JJ, Wu XK, Xie W, Zhou X, Su H, Chen YX, Wang LK, Li WK, Tian K, Zhuang RJ

- 7075** Effect of T-regulatory cells and interleukin-35, interleukin-10, and transforming growth factor-beta on diffuse large B-cell lymphoma

Wu H, Sun HC, Ouyang GF

META-ANALYSIS

- 7082** Meta-analysis on the effectiveness of parent education for children with disabilities

Jang J, Kim G, Jeong H, Lee N, Oh S

- 7091** Meta-analysis of the efficacy and safety of daratumumab in the treatment of multiple myeloma

Wang P, Jin SY

CASE REPORT

- 7101** Varicella-zoster virus meningitis with hypoglycorrhachia: A case report

Cao LJ, Zheng YM, Li F, Hao HJ, Gao F

- 7107** Unusual presentation of penile giant condyloma acuminatum with spontaneous prepuce perforation: A case report

Hsu FC, Yu DS, Pu TW, Wu MJ, Meng E

- 7113** Primary renal lymphoma presenting as renal failure: A case report and review of literature from 1989

Lee SB, Yoon YM, Hong R

- 7127** Intravascular ultrasonography assisted carotid artery stenting for treatment of carotid stenosis: Two case reports

Fu PC, Wang JY, Su Y, Liao YQ, Li SL, Xu GL, Huang YJ, Hu MH, Cao LM

- 7136** Mucoepidermoid carcinoma of the lung with hemoptysis as initial symptom: A case report

Xie WX, Liu R, Li Z, Zhou PL, Duan LN, Fu DD

- 7144** Co-infection of *Chlamydia psittaci* and *Tropheryma whippelii*: A case report

Du ZM, Chen P

- 7150** Surgical treatment of severe anterior capsular organized hard core cataract: A case report

Wang LW, Fang SF

- 7156** First platelet transfusion refractoriness in a patient with acute myelocytic leukemia: A case report

Tu SK, Fan HJ, Shi ZW, Li XL, Li M, Song K

- 7162** Rare finding of primary aortoduodenal fistula on single-photon emission computed tomography/computed tomography of gastrointestinal bleeding: A case report

Kuo CL, Chen CF, Su WK, Yang RH, Chang YH

- 7170** Rituximab combined with Bruton tyrosine kinase inhibitor to treat elderly diffuse large B-cell lymphoma patients: Two case reports

Zhang CJ, Zhao ML

- 7179** Use of Ilizarov technique for bilateral knees flexion contracture in Juvenile-onset ankylosing spondylitis: A case report
Xia LW, Xu C, Huang JH
- 7187** Case of takotsubo cardiomyopathy after surgical treatment of liver hydatid cyst: A case report
Altaş Y, Abdullayeva Ü
- 7193** Laparoscopic choledocholithotomy and transductal T-tube insertion with indocyanine green fluorescence imaging and laparoscopic ultrasound: A case report
Yoo D
- 7200** Hematopoietic stem cell transplantation of aplastic anemia by relative with mutations and normal telomere length: A case report
Yan J, Jin T, Wang L
- 7207** Emphysematous thrombophlebitis caused by a misplaced central venous catheter: A case report
Chen N, Chen HJ, Chen T, Zhang W, Fu XY, Xing ZX
- 7214** Aggressive angiomyxoma of the epididymis: A case report
Liu XJ, Su JH, Fu QZ, Liu Y
- 7221** Gastric and intestinal ectopic pancreas: Two case reports
Zhang H, Zhao HY, Zhang FH, Liang W
- 7227** Congenital leukemia: A case report and review of literature
Yang CX, Yang Y, Zhang FL, Wang DH, Bian QH, Zhou M, Zhou MX, Yang XY
- 7234** Imaging misdiagnosis and clinical analysis of significant hepatic atrophy after bilioenteric anastomosis: A case report
Liang SY, Lu JG, Wang ZD
- 7242** Surgical treatment of mixed cervical spondylosis with spontaneous cerebrospinal fluid leakage: A case report
Yu Z, Zhang HFZ, Wang YJ
- 7248** Simultaneous thyroglossal duct cyst with parathyroid cyst: A case report
Chen GY, Li T
- 7253** Submandibular solid-cystic mass as the first and sole manifestation of occult thyroid papillary carcinoma: A case report
Chen GY, Li T

LETTER TO THE EDITOR

- 7258** Artificial intelligence and machine learning in motor recovery: A rehabilitation medicine perspective
Swarnakar R, Yadav SL

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Use of Ilizarov technique for bilateral knees flexion contracture in Juvenile-onset ankylosing spondylitis: A case report

Li-Wei Xia, Cheng Xu, Jian-Han Huang

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Abstract

BACKGROUND

Ankylosing spondylitis (AS) is a chronic rheumatic disease that primarily affects the spine and the sacroiliac and peripheral joints. Juvenile-onset AS (JoAS) patients will likely present with peripheral joint symptoms. Knee flexion contracture (KFC) and hip flexion contracture (HFC) are common in these patients due to subchondral bone inflammation. The Ilizarov technique is the most commonly used technique for treating KFC. However, its use to treat JoAS-associated KFC has not been reported.

CASE SUMMARY

This report presents a case study of a 31-year-old male patient with a squatting gait due to severe bilateral KFC and HFC. The patient had a normal walking pattern until the age of eight, after which he experienced knee and hip pain, leading to the gradual development of KFC and HFC. The patient's primary complaint was an inability to walk upright. The patient was diagnosed with JoAS and underwent hip dissection and release, limited soft tissue release of the hamstring, and gradual traction using the Ilizarov method. Ultimately, the patient was able to walk upright.

CONCLUSION

The incidence of squatting gait due to KFC in individuals diagnosed with JoAS was low. Utilizing the Ilizarov technique has proven to be a secure and effective method for managing KFC in JoAS patients. Although the Ilizarov technique cannot substitute for total knee arthroplasty (TKA), its application can delay the need for primary TKA in JoAS patients and alleviate the intricacy and potential complications associated with the procedure.

Key Words: Juvenile-onset ankylosing spondylitis; Knee flexion contracture; Squatting gait; Ilizarov ring external fixator; Total knee arthroplasty; Case report

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Core Tip: In Juvenile-onset ankylosing spondylitis (JoAS) patients, knee flexion contracture (KFC) is uncommon, with sparse literature on arthroplasty for ankylosed knees. We share a case where a JoAS patient's severe bilateral KFC was effectively treated using the Ilizarov method.

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INTRODUCTION

Juvenile-onset ankylosing spondylitis (JoAS) can cause deformities and disabilities of the hip and knee joints. The American College of Rheumatology recommends arthroplasty for AS patients with end-stage hip arthritis[1,2]. Total knee arthroplasty (TKA) is the most commonly performed surgical intervention for AS patients, aimed at reducing pain and improving knee mobility[3,4]. The incidences of total hip arthroplasty and TKA have recently increased in older AS patients[5]. Young and middle-aged AS patients also have an elevated risk of undergoing TKA[6]. However, performing TKA on patients with AS presents orthopedic surgeons with challenges, including surgical field exposure, soft tissue balance, and prosthesis selection[7].

Furthermore, AS patients have higher rates of infection, systemic complications, revision, and readmission following TKA than osteoarthritis patients[8]. Therefore, explore alternative options for delaying primary TKA in young and middle-aged individuals, especially those with partial hip and knee function and no joint rigidity. The Ilizarov technique is a skeletal traction method that has been widely used for fracture management, limb salvage, and deformity correction, including knee flexion contracture (KFC)[9-11]. However, there are few reports on managing severe KFC of JoAS with Ilizarov technique. The present study hypothesizes that the Ilizarov technique can safely and effectively correct KFC in JoAS.

CASE PRESENTATION

Chief complaints

A 31-year-old male patient presented to our orthopedic clinic with a squatting gait and the desire to walk upright (Figures 1A-C).

History of present illness

The patient could walk normally until the age of eight.

History of past illness

The patient, at the age of 8, experienced pain in various peripheral joints, including the knees and hips. Regrettably, the family failed to acknowledge these symptoms, resulting in a delayed diagnosis and subsequent treatment.

Personal and family history

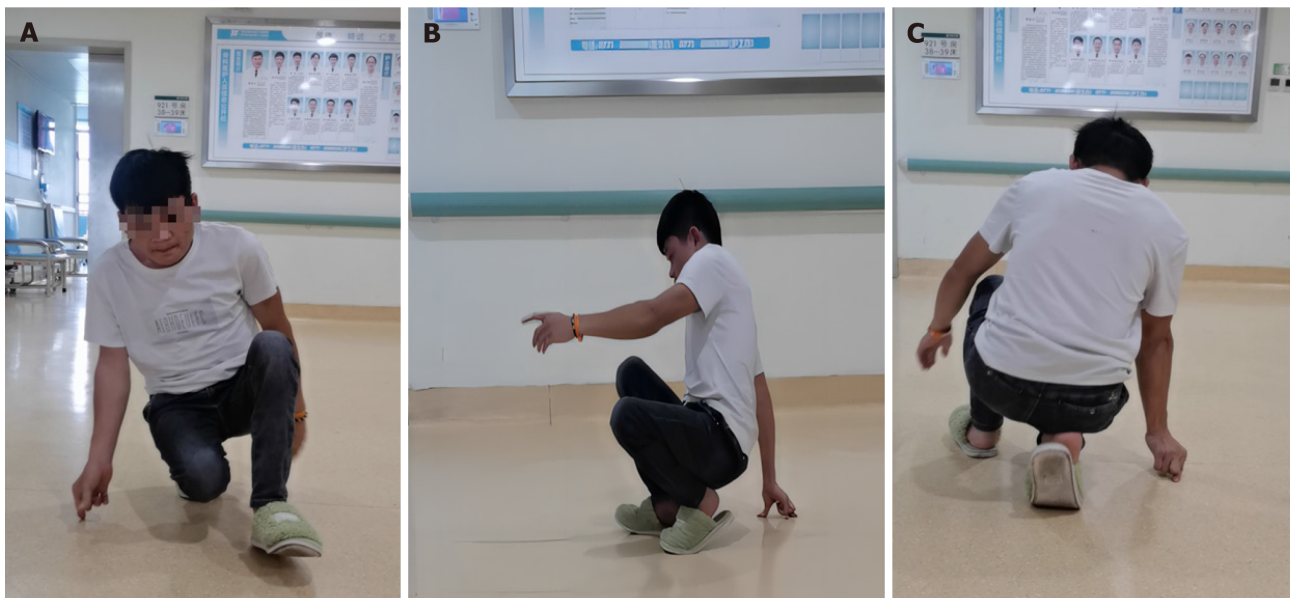
The patient had no significant medical history and denied any family history of AS.

Physical examination

Upon examination, the right and left knees had a limited range of motion (ROM) of 75°-135° and 55°-135°, respectively (Figures 2A and B). The ROM of the right hip was limited to 50°-135°, while the left hip had a range of 45°-135°. Posterior extension of both the hips was limited.

Laboratory examinations

The patient tested positive for HLA-B27, whereas negative for rheumatoid factor.



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Figure 1 Preoperative squatting gait. A: Frontal view; B: Lateral view; C: Posterior view.



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Figure 2 Range of motion of bilateral knees. A: Range of motion of right knee was 75°-135°; B: Range of motion of left knee was 55°-135°.

Imaging examinations

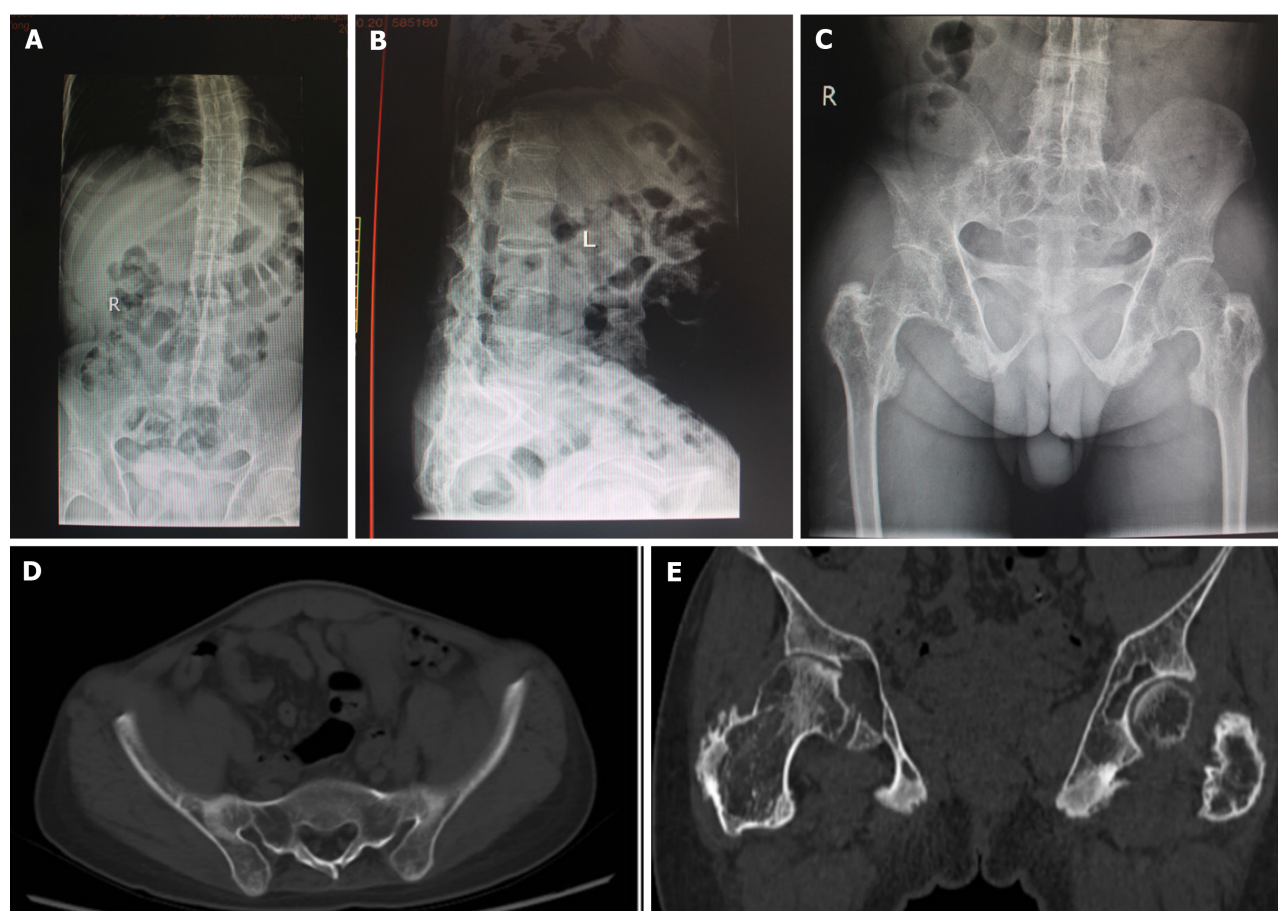
Plain radiographs of the spine exhibited characteristic bamboo-like alterations (Figures 3A and B). Plain radiography of Pelvis (Figure 3C) and computed tomography (CT) of Pelvis (Figure 3D) revealed sacroiliac joint fusion. Pelvis CT plain scans showed osteoarthritic changes in both hips, such as narrowing of the hip gaps, cystic changes beneath the articular cartilage, and significant bone hyperplasia around the greater and lesser trochanters (Figure 3E). Plain radiography of bilateral knees revealed a narrowing of the knee gaps and bilateral KFC (Figures 4A and B). Plain radiography of feet displayed ankylosing tarsitis (Figures 4C and D).

FINAL DIAGNOSIS

The patient was diagnosed with neglected JoAS with bilateral KFC and hip flexion contracture (HFC).

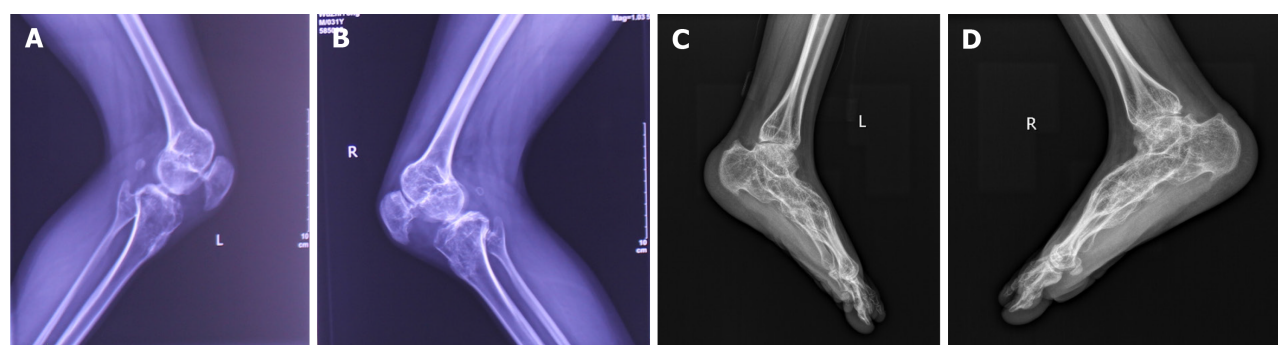
TREATMENT

In the present study, the patient did not exhibit knee or hip joint ankylosis. Although X-rays displayed knee and hip joints destruction, the patient did not complain of significant pain in these areas. Considering the relatively young age of



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Figure 3 Radiographic changes in the spine, hip joints and sacroiliac joints. A and B: Plain radiographies of spine showed bamboo spine; C and D: Plain radiography of pelvis and computed tomography scan of pelvis showed sacroiliac joint fusion; E: Computed tomography scan of hips revealed osteoarthritic changes.

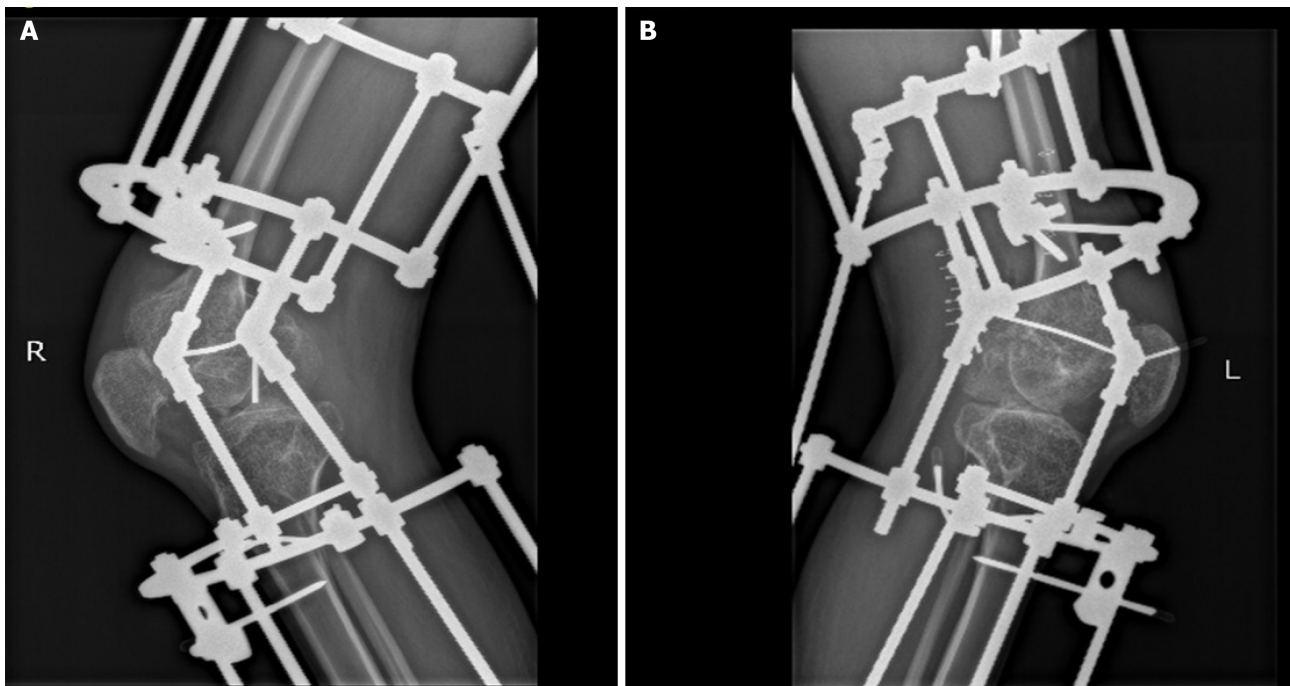


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Figure 4 Radiographic changes in the knee joints and feet. A and B: X-rays of the knees revealed osteophytes and bilateral knee flexion contracture; C and D: Plain radiography of ankles showed narrowing of ankle joints space and ankylosing tarsitis.

the patient, performing primary TKA early increases the likelihood of future knee revisions. Therefore, the patient underwent hip dissection and release, as well as limited soft tissue release of the hamstring, along with gradual traction using the Ilizarov method.

The patient's spinal vertebral fusion rendered epidural anesthesia impossible, necessitating general anesthesia despite the challenges associated with tracheal intubation. After administering anesthesia, the patient was positioned supine and a lateral incision measuring approximately 10 cm in length was made on the right hip to release the adherent hip joint. After the hamstring lengthening procedure, an Ilizarov ring external fixator was attached to the right thigh and proximal tibiofibular region. Neurolysis of the peroneal nerve was performed to prevent paralysis of the common peroneal nerve during knee distraction.



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Figure 5 Postoperative X-rays of the bilateral knee joints. A: The right knee; B: The left knee.

The threaded rod could be rotated posteriorly to initiate distraction once local pain and swelling had diminished significantly, typically five to seven days after surgery. The knee joint gap can be increased by approximately 5 mm to prevent compression of the articular cartilage during distraction. Subsequently, the nut on the posterior popliteal distraction rod was rotated to gradually distract the posterior knee soft tissue, thereby correcting the residual knee flexion deformity (Figures 5A and B). During treatment, a sandbag was used to compress the patient's hip joint, and a physiotherapist was involved in providing bedside physiotherapy to address the residual hip flexion deformity.

Three weeks after the surgery, the same procedure was performed on the left lower extremity. Once the bilateral HFC and KFC were fully corrected, the patient was encouraged to stand and walk with the assistance of a walker. After 10 wk, the patient was readmitted to the hospital to remove the Ilizarov external fixation device (Figures 6A and B). During the procedure, a plaster model of the patient's lower extremities was obtained to facilitate the customization of a long-leg brace for both limbs. Subsequently, the patient was encouraged to walk on the ground wearing a bilateral lower limb long-leg brace. Radiographs of both knee joints showed the patient had full extension of both knees (Figures 7A and B). Following discharge from the hospital, the patient independently engaged in standing and upright walking exercises at home (Figure 7C).

OUTCOME AND FOLLOW-UP

Approximately three months later, the patient presented to the outpatient clinic with bilateral knee extension stiffness. After receiving physiotherapy, manipulation, and sodium hyaluronate injection into the knee joint by a rehabilitation therapist, the patient could actively flex both knees to 110°. Twelve months postoperatively, the patient achieved independent ambulation and discontinued using long-legged brace (Figure 8). The patient reported no hip pain but intermittent knee pain with joint effusion. The patient was prescribed long-term oral nonsteroidal and antirheumatic immune drugs, including methotrexate, as a treatment plan. Ultimately, the patient achieved the goal of walking upright.

DISCUSSION

KFC can arise from various etiologies, such as congenital diseases, inflammatory conditions, and miscellaneous reasons [12]. The Ilizarov technique is a safe and effective method that can be used to treat KFC caused by various pathologies. However, the purpose of using the Ilizarov technique differs depending on the etiology of the KFC.

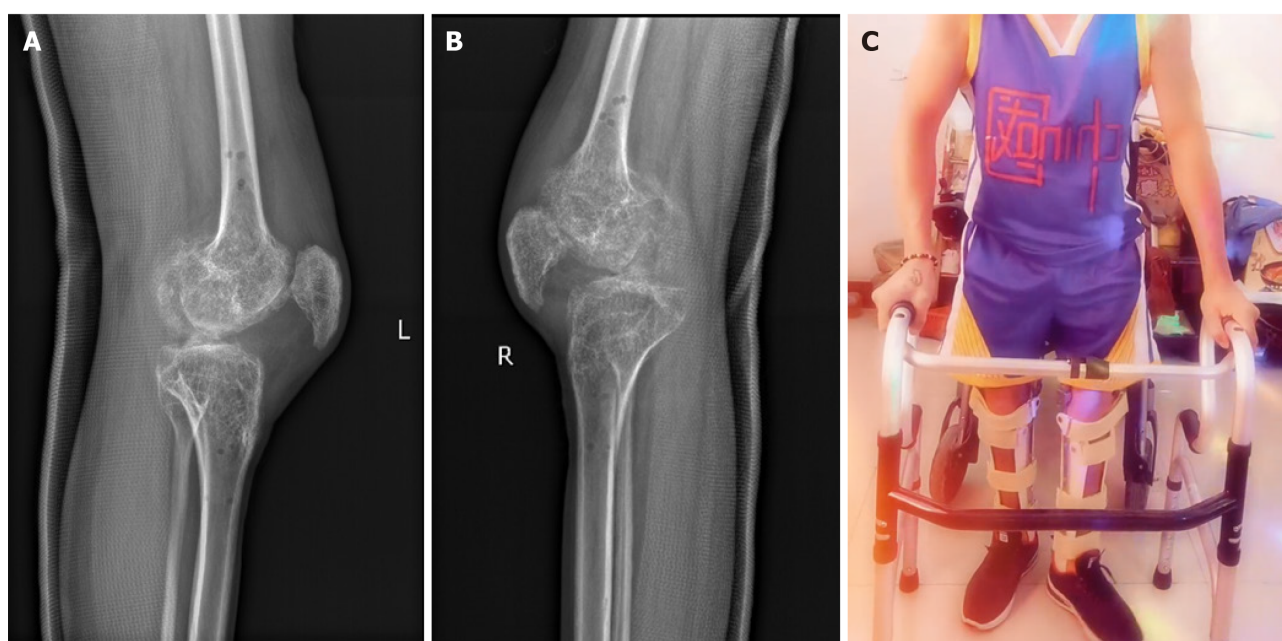
Common causes of KFC are cerebral palsy, poliomyelitis, congenital spina bifida, trauma, and burns. Limb length discrepancy affects patient aesthetics and ambulation. The knee is gradually straightened by the continuous and slow distraction of the Ilizarov ring external fixator, restoring symmetrical lower limb lengths.

KFC has also been observed in patients with brain injuries and stroke [13]. The Ilizarov technique can increase a patient's walking ability if they have the potential to walk. The Ilizarov technique has the potential to improve comfort and



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Figure 6 Clinical images before removal of bilateral lower limb ring external fixators. A: Standing with ring external fixator; B: Showing almost full extension of bilateral knees.



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Figure 7 Images after removal of bilateral lower limb ring external fixators. A and B: Lateral X-rays of bilateral knees; C: Standing up and walking with a walker.

hygiene and facilitate care for patients who are expected not to walk even after surgery[14].

Septic knee arthritis can also result in KFC[15]. The Ilizarov technique is a viable option to achieve knee arthrodesis in cases of septic failure following TKA, post-traumatic knee infection, and end-stage tuberculosis of the knee[16-18]. Junior rheumatoid arthritis (JRA) can destroy the knee cartilage and develop KFC. TKA is the most commonly reported surgical procedure, whereas the Ilizarov technique is rarely reported. Guo *et al*[19] reported nine patients diagnosed with severe KFC due to JRA. All the patients underwent gradual distraction using an Ilizarov ring external fixator. At the last follow-up, all patients completed knee extension without severe complications. The authors believe that Ilizarov technique corrected the knee flexion deformity, providing a foundation for subsequent TKA. Vecham *et al*[20] described a 22-year-old female patient with JRA and severe bilateral KFC who underwent a two-stage procedure. The initial stage involved using Ilizarov ring external fixators to completely correct the KFC. The patient underwent bilateral TKA during the



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Figure 8 Clinical photographs of the patient after removal of the long-legged brace (12 mo after the operation). A: Frontal view; B: Lateral view; C: Posterior view.

second stage. Since the patient's KFC was completely corrected in the first stage, second-stage TKA was relatively easy to perform, as it did not require excessive osteotomy and soft tissue balancing.

JoAS, like RA, is an inflammatory rheumatic disease that affects the joints, including the knee. However, clinically reported cases of KFC are less common due to JoAS. To our knowledge, there have been no reports of Ilizarov treatment of severe bilateral KFC caused by JoAS. This study presents a novel therapeutic approach for patients diagnosed with JoAS who exhibit severe KFC. In this case report, the patient realized his desire to walk upright and was satisfied with the results despite some complications, such as high-lever pain and sleep disturbances. However, It remains necessary to follow up for a longer period to determine potential long-term efficacy differences.

CONCLUSION

The presence of hip and knee arthritis in individuals diagnosed with JoAS can result in HFC and KFC, potentially inducing gait abnormalities in affected patients. A combination of limited soft tissue release from the knee joint and Ilizarov distraction therapy is a safe and effective procedure. Although this procedure is not a substitute for TKA, it can help avoid the need for early or delayed primary joint replacement, consequently mitigating the likelihood of future knee revision.

FOOTNOTES

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