

World Journal of *Clinical Cases*

World J Clin Cases 2022 September 6; 10(25): 8808-9179



Contents

Thrice Monthly Volume 10 Number 25 September 6, 2022

MINIREVIEWS

- 8808** Ear, nose, and throat manifestations of COVID-19 and its vaccines
Al-Ani RM
- 8816** Potential influences of religiosity and religious coping strategies on people with diabetes
Onyishi CN, Eseadi C, Ilechukwu LC, Okoro KN, Okolie CN, Egbule E, Asogwa E

ORIGINAL ARTICLE

Case Control Study

- 8827** Effectiveness of six-step complex decongestive therapy for treating upper limb lymphedema after breast cancer surgery
Zhang HZ, Zhong QL, Zhang HT, Luo QH, Tang HL, Zhang LJ

Retrospective Study

- 8837** Hospital admissions from alcohol-related acute pancreatitis during the COVID-19 pandemic: A single-centre study
Mak WK, Di Mauro D, Pearce E, Karran L, Myintmo A, Duckworth J, Orabi A, Lane R, Holloway S, Manzelli A, Mossadegh S
- 8844** Indocyanine green plasma clearance rate and 99mTc-galactosyl human serum albumin single-photon emission computed tomography evaluated preoperative remnant liver
Iwaki K, Kaihara S, Kita R, Kitamura K, Hashida H, Uryuhara K
- 8854** Arthroscopy with subscapularis upper one-third tenodesis for treatment of recurrent anterior shoulder instability independent of glenoid bone loss
An BJ, Wang FL, Wang YT, Zhao Z, Wang MX, Xing GY
- 8863** Evaluation of the prognostic nutritional index for the prognosis of Chinese patients with high/extremely high-risk prostate cancer after radical prostatectomy
Yang F, Pan M, Nie J, Xiao F, Zhang Y

Observational Study

- 8872** Chlorine poisoning caused by improper mixing of household disinfectants during the COVID-19 pandemic: Case series
Lin GD, Wu JY, Peng XB, Lu XX, Liu ZY, Pan ZG, Qiu ZW, Dong JG
- 8880** Mental health of the Slovak population during COVID-19 pandemic: A cross-sectional survey
Kralova M, Brazinova A, Sivcova V, Izakova L

Prospective Study

- 8893** Arthroscopic anatomical reconstruction of lateral collateral ligaments with ligament advanced reinforcement system artificial ligament for chronic ankle instability
Wang Y, Zhu JX

SYSTEMATIC REVIEWS

- 8906** How to select the quantitative magnetic resonance technique for subjects with fatty liver: A systematic review
Li YW, Jiao Y, Chen N, Gao Q, Chen YK, Zhang YF, Wen QP, Zhang ZM
- 8922** Lymphocytic choriomeningitis virus: An under-recognized congenital teratogen
Ferenc T, Vujica M, Mrzljak A, Vilibic-Cavlek T

CASE REPORT

- 8932** Alagille syndrome associated with total anomalous pulmonary venous connection and severe xanthomas: A case report
Zeng HS, Zhang ZH, Hu Y, Zheng GL, Wang J, Zhang JW, Guo YX
- 8939** Colo-colonic intussusception with post-polypectomy electrocoagulation syndrome: A case report
Moon JY, Lee MR, Yim SK, Ha GW
- 8945** Portal vein gas combined with pneumatosis intestinalis and emphysematous cystitis: A case report and literature review
Hu SF, Liu HB, Hao YY
- 8954** Quadricuspid aortic valve and right ventricular type of myocardial bridging in an asymptomatic middle-aged woman: A case report
Sopek Merkaš I, Lakušić N, Paar MH
- 8962** Treatment of gastric carcinoma with lymphoid stroma by immunotherapy: A case report
Cui YJ, Ren YY, Zhang HZ
- 8968** Gallstone associated celiac trunk thromboembolisms complicated with splenic infarction: A case report
Wu CY, Su CC, Huang HH, Wang YT, Wang CC
- 8974** Extracorporeal membrane oxygenation for lung cancer-related life-threatening hypoxia: A case report
Yoo SS, Lee SY, Choi SH
- 8980** Multi-disciplinary treatment of maxillofacial skeletal deformities by orthognathic surgery combined with periodontal phenotype modification: A case report
Liu JY, Li GF, Tang Y, Yan FH, Tan BC
- 8990** X-linked recessive Kallmann syndrome: A case report
Zhang P, Fu JY
- 8998** Delayed complications of intradural cement leakage after percutaneous vertebroplasty: A case report
Ma QH, Liu GP, Sun Q, Li JG

- 9004** Coexistent Kaposi sarcoma and post-transplant lymphoproliferative disorder in the same lymph nodes after pediatric liver transplantation: A case report
Zhang SH, Chen GY, Zhu ZJ, Wei L, Liu Y, Liu JY
- 9012** Misdiagnosis of pancreatic metastasis from renal cell carcinoma: A case report
Liang XK, Li LJ, He YM, Xu ZF
- 9020** Discoid medial meniscus of both knees: A case report
Zheng ZR, Ma H, Yang F, Yuan L, Wang GD, Zhao XW, Ma LF
- 9028** Simultaneous laparoscopic and arthroscopic excision of a huge juxta-articular ganglionic cyst compressing the sciatic nerve: A case report
Choi WK, Oh JS, Yoon SJ
- 9036** One-stage revision arthroplasty in a patient with ochronotic arthropathy accompanied by joint infection: A case report
Wang XC, Zhang XM, Cai WL, Li Z, Ma C, Liu YH, He QL, Yan TS, Cao XW
- 9044** Bladder paraganglioma after kidney transplantation: A case report
Wang L, Zhang YN, Chen GY
- 9050** Total spinal anesthesia caused by lidocaine during unilateral percutaneous vertebroplasty performed under local anesthesia: A case report
Wang YF, Bian ZY, Li XX, Hu YX, Jiang L
- 9057** Ruptured splenic artery aneurysms in pregnancy and usefulness of endovascular treatment in selective patients: A case report and review of literature
Lee SH, Yang S, Park I, Im YC, Kim GY
- 9064** Gastrointestinal metastasis secondary to invasive lobular carcinoma of the breast: A case report
Li LX, Zhang D, Ma F
- 9071** Post-bulbar duodenal ulcer with anterior perforation with kissing ulcer and duodenocaval fistula: A case report and review of literature
Alzerwi N
- 9078** Modified orthodontic treatment of substitution of canines by first premolars: A case report
Li FF, Li M, Li M, Yang X
- 9087** Renal cell carcinoma presented with a rare case of icteric Stauffer syndrome: A case report
Popov DR, Antonov KA, Atanasova EG, Pentchev CP, Milatchkov LM, Petkova MD, Neykov KG, Nikolov RK
- 9096** Successful resection of a huge retroperitoneal venous hemangioma: A case report
Qin Y, Qiao P, Guan X, Zeng S, Hu XP, Wang B
- 9104** Malignant transformation of biliary adenofibroma combined with benign lymphadenopathy mimicking advanced liver carcinoma: A case report
Wang SC, Chen YY, Cheng F, Wang HY, Wu FS, Teng LS

- 9112** Congenital hepatic cyst: Eleven case reports
Du CX, Lu CG, Li W, Tang WB
- 9121** Endovascular treatment of a ruptured pseudoaneurysm of the internal carotid artery in a patient with nasopharyngeal cancer: A case report
Park JS, Jang HG
- 9127** Varicella-zoster virus meningitis after spinal anesthesia: A case report
Lee YW, Yoo B, Lim YH
- 9132** Chondrosarcoma of the toe: A case report and literature review
Zhou LB, Zhang HC, Dong ZG, Wang CC
- 9142** Tamsulosin-induced life-threatening hypotension in a patient with spinal cord injury: A case report
Lee JY, Lee HS, Park SB, Lee KH
- 9148** CCNO mutation as a cause of primary ciliary dyskinesia: A case report
Zhang YY, Lou Y, Yan H, Tang H
- 9156** Repeated bacteremia and hepatic cyst infection lasting 3 years following pancreatoduodenectomy: A case report
Zhang K, Zhang HL, Guo JQ, Tu CY, Lv XL, Zhu JD
- 9162** Idiopathic cholesterol crystal embolism with atheroembolic renal disease and blue toes syndrome: A case report
Cheng DJ, Li L, Zheng XY, Tang SF
- 9168** Systemic lupus erythematosus with visceral varicella: A case report
Zhao J, Tian M

LETTER TO THE EDITOR

- 9176** Imaging of fibroadenoma: Be careful with imaging follow-up
Ece B, Aydın S

ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Mohsen Khosravi, MD, Assistant Professor, Department of Psychiatry and Clinical Psychology, Zahedan University of Medical Sciences, Zahedan 9819713955, Iran. m.khosravi@zaums.ac.ir

AIMS AND SCOPE

The primary aim of *World Journal of Clinical Cases* (WJCC, *World J Clin Cases*) is to provide scholars and readers from various fields of clinical medicine with a platform to publish high-quality clinical research articles and communicate their research findings online.

WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

INDEXING/ABSTRACTING

The WJCC is now abstracted and indexed in Science Citation Index Expanded (SCIE, also known as SciSearch®), Journal Citation Reports/Science Edition, Current Contents®/Clinical Medicine, PubMed, PubMed Central, Scopus, Reference Citation Analysis, China National Knowledge Infrastructure, China Science and Technology Journal Database, and Superstar Journals Database. The 2022 Edition of Journal Citation Reports® cites the 2021 impact factor (IF) for WJCC as 1.534; IF without journal self cites: 1.491; 5-year IF: 1.599; Journal Citation Indicator: 0.28; Ranking: 135 among 172 journals in medicine, general and internal; and Quartile category: Q4. The WJCC's CiteScore for 2021 is 1.2 and Scopus CiteScore rank 2021: General Medicine is 443/826.

RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Xu Guo*; Production Department Director: *Xiang Li*; Editorial Office Director: *Jin-Lei Wang*.

NAME OF JOURNAL

World Journal of Clinical Cases

ISSN

ISSN 2307-8960 (online)

LAUNCH DATE

April 16, 2013

FREQUENCY

Thrice Monthly

EDITORS-IN-CHIEF

Bao-Gan Peng, Jerzy Tadeusz Chudek, George Kontogeorgos, Maurizio Serati, Ja Hyeon Ku

EDITORIAL BOARD MEMBERS

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

PUBLICATION DATE

September 6, 2022

COPYRIGHT

© 2022 Baishideng Publishing Group Inc

INSTRUCTIONS TO AUTHORS

<https://www.wjgnet.com/bpg/gerinfo/204>

GUIDELINES FOR ETHICS DOCUMENTS

<https://www.wjgnet.com/bpg/GerInfo/287>

GUIDELINES FOR NON-NATIVE SPEAKERS OF ENGLISH

<https://www.wjgnet.com/bpg/gerinfo/240>

PUBLICATION ETHICS

<https://www.wjgnet.com/bpg/GerInfo/288>

PUBLICATION MISCONDUCT

<https://www.wjgnet.com/bpg/gerinfo/208>

ARTICLE PROCESSING CHARGE

<https://www.wjgnet.com/bpg/gerinfo/242>

STEPS FOR SUBMITTING MANUSCRIPTS

<https://www.wjgnet.com/bpg/GerInfo/239>

ONLINE SUBMISSION

<https://www.f6publishing.com>



One-stage revision arthroplasty in a patient with ochronotic arthropathy accompanied by joint infection: A case report

Xiao-Chao Wang, Xiao-Min Zhang, Wan-Ling Cai, Zhen Li, Chao Ma, Yi-Hai Liu, Qi-Lian He, Tian-Sheng Yan, Xue-Wei Cao

Specialty type: Orthopedics

Provenance and peer review:

Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's scientific quality classification

Grade A (Excellent): 0
Grade B (Very good): B
Grade C (Good): C, C
Grade D (Fair): D
Grade E (Poor): 0

P-Reviewer: Al-Ani RM, Iraq; Tajiri K, Japan; Vyshka G, Albania

Received: March 26, 2022

Peer-review started: March 26, 2022

First decision: April 13, 2022

Revised: April 22, 2022

Accepted: July 29, 2022

Article in press: July 29, 2022

Published online: September 6, 2022



Xiao-Chao Wang, Zhen Li, Chao Ma, The Second Clinical Medical College, Guangzhou University of Chinese Medicine, Guangzhou 510000, Guangdong Province, China

Xiao-Min Zhang, Yi-Hai Liu, Qi-Lian He, Tian-Sheng Yan, Department of Orthopaedics, Xi'an International Medical Center Hospital, Xi'an 710100, Shaanxi Province, China

Wan-Ling Cai, Department of Surgery, Baoji Hospital of Traditional Chinese Medicine, Baoji 721000, Shaanxi Province, China

Xue-Wei Cao, Department of Orthopaedic Surgery, Guangdong Provincial Hospital of Traditional Chinese Medicine, Guangzhou 510120, Guangdong Province, China

Corresponding author: Xue-Wei Cao, MD, Chief Doctor, Department of Orthopaedic Surgery, Guangdong Provincial Hospital of Traditional Chinese Medicine, No. 111 Dade Road, Guangzhou 510120, Guangdong Province, China. wscxw123@163.com

Abstract

BACKGROUND

Ochronotic arthropathy (OcA) is a rare disease, which is caused by the accumulation of homogentisic acid in the joint. Patients with OcA have obvious joint pain and the disease progresses rapidly, eventually resulting in disability. Arthroplasty is an efficacious treatment in patients with OcA. However, when OcA patients have joint infection, is joint replacement an option? In the present report, we performed total knee arthroplasty in a patient with OcA and knee infection under the guidance of one-stage revision theory.

CASE SUMMARY

A 64-year-old male was referred to our hospital due to severe left knee pain with limited mobility for 2 years. On physical examination, the patient was found to have dark brown pigmentation of the sclera and auricle. Laboratory test results showed elevations in C-reactive protein level (65.79 mg/L) and erythrocyte sedimentation rate (90.00 mm/h). The patient underwent debridement of the left knee joint, during which the cartilage surface of the knee joint was found to be black-brown in color. Bacterial culture of synovial fluid revealed *Achromobacter xylosoxidans*. We then carried out arthroplasty under the guidance of the theory of one-stage revision. After surgery, the patient's left knee joint pain disappeared and function recovered without joint infection.

CONCLUSION

OcA accompanied by joint infection is rare. One-stage revision arthroplasty may be a treatment option for this disease.

Key Words: Ochronotic arthropathy; Arthroplasty; One-stage revision; Alkaptonuria; Homogentisic acid; Case report

©The Author(s) 2022. Published by Baishideng Publishing Group Inc. All rights reserved.

Core Tip: Ochronotic arthropathy (OcA) is a rare disease caused by the accumulation of homogentisic acid in the joint. Compared with general arthritis patients, patients with OcA have severe pain, poor function, and high disability rate. Arthroplasty is an effective treatment for patients with end-stage OcA. However, it is unknown how to select treatment when the OcA patient has joint infection. Under guidance of the one-stage revision theory, we treated a patient with OcA accompanied by knee infection with arthroplasty. Postoperation, the patient's knee joint pain disappeared and function recovered without joint infection. One-stage revision can be a treatment for this disease.

Citation: Wang XC, Zhang XM, Cai WL, Li Z, Ma C, Liu YH, He QL, Yan TS, Cao XW. One-stage revision arthroplasty in a patient with ochronotic arthropathy accompanied by joint infection: A case report. *World J Clin Cases* 2022; 10(25): 9036-9043

URL: <https://www.wjgnet.com/2307-8960/full/v10/i25/9036.htm>

DOI: <https://dx.doi.org/10.12998/wjcc.v10.i25.9036>

INTRODUCTION

Alkaptonuria (AKU) is a rare autosomal recessive hereditary metabolic disease[1], resulting from the absence of homogentisate 1,2-dioxygenase (HGD). HGD, an intermediate in the tyrosine degradation pathway, is responsible for the breakdown of homogentisic acid (HGA). As a result of this defect, a large amount of HGA cannot be metabolized normally and accumulates in the body over a long period, resulting in the formation of pigment-like polymers, which are deposited in connective tissue and cause ochronosis[2]. In patients with AKU, some HGA is excreted in the urine. Characteristically, excess HGA causes sufferers to pass dark urine, which upon standing turns black. Some HGA is also deposited in a variety of connective tissues, such as cartilage, sclera, and tendons, resulting in blue-black tissue changes, causing local lesions. Typically, the pigment is clearly seen in the sclera and auricle of patients. The disease usually progresses from simple AKU to alkaptonuric ochronosis and finally to ochronotic arthropathy (OcA)[3]. With advancing age, renal excretion of HGA decreases, and the accumulation of HGA gradually increases in the body. HGA can damage articular cartilage and cause joint degeneration, leading to OcA. The disease first affects the spine and then the peripheral joints. The patient's symptoms usually begin around the third or fourth decade with low back pain and stiffness[4]. In the peripheral joints, the knee is most commonly involved, followed by the hip and shoulder[5]. OcA develops rapidly and tends to result in disability within a short time.

A literature review showed that arthroplasty in patients with OcA can be a good treatment choice[6]. However, arthroplasty for the treatment of OcA with joint infection has not been reported in the literature. Periprosthetic infection (PJI) is a catastrophic complication after arthroplasty, and two-stage exchange arthroplasty is the most common operation for patients with PJI[7]. However, the cost of this treatment is high[8,9], the mortality and morbidity rates are high, and the long-term function is poor [10]. A study found that by selecting appropriate patients and optimizing surgical procedures, one-stage exchange arthroplasty has significant benefits for patients with joint infection[11]. Zahar *et al*[12] found that the success rate of one-stage exchange arthroplasty was approximately 85%-90%. Based on the theory of one-stage revision, we treated a patient with OcA accompanied by knee infection with arthroplasty, and achieved satisfactory clinical results.

CASE PRESENTATION

Chief complaints

A 64-year-old man was admitted due to left knee pain and limited mobility.

History of present illness

The patient presented with left knee pain and limited mobility 2 years ago. He visited a local hospital several times and was diagnosed with left knee osteoarthritis. His left knee pain was only relieved by oral analgesics. His symptoms improved with medication, and relapsed when the analgesics were stopped. During the past 2 years, his symptoms gradually progressed. The patient was admitted to our department 4 mo prior with severe pain in the left knee and limited knee motion.

History of past illness

The patient had hypertension, but he did not take medicine regularly in the last month.

Personal and family history

The patient had no relevant personal or family history.

Physical examination

Dark-brown pigment was clearly seen in the sclera and ears of the patient (Figure 1). The patient's spine was stiff, and a major reduction in movement of the vertebral column was noted. Atrophy of the left quadriceps femoris and slight swelling of the left knee were observed. The left knee had a flexion contracture of 10°, and the range of motion of the left knee was 10° (extension) to 90° (flexion). The left knee patella grinding test and McMurray sign were positive.

Laboratory examinations

Routine blood tests were normal and showed the following results: White blood cell count: $6.14 \times 10^9/L$; neutrophils: 63.9%; hemoglobin: 104 g/L; platelet count: $310 \times 10^9/L$. C-reactive protein (CRP) and erythrocyte sedimentation rate (ESR) were elevated at 65.79 mg/L and 90.00 mm/h, respectively.

Imaging examinations

Radiographic evaluation revealed that the patient's left knee joint showed a similar appearance to that of osteoarthritis, such as joint space stenosis and osteophyte formation (Figure 2). Magnetic resonance imaging of the knee showed bone marrow edema (Figure 3).

Further diagnostic work-up

We first carried out debridement of the left knee. During surgery, it was found that the cartilage of the knee joint was black-brown in color and there was a lot of synovial fluid in the joint cavity. We collected the synovial fluid for bacterial culture and the results showed the presence of *Achromobacter xylosoxidans* (*A. xylosoxidans*) in synovial fluid.

FINAL DIAGNOSIS

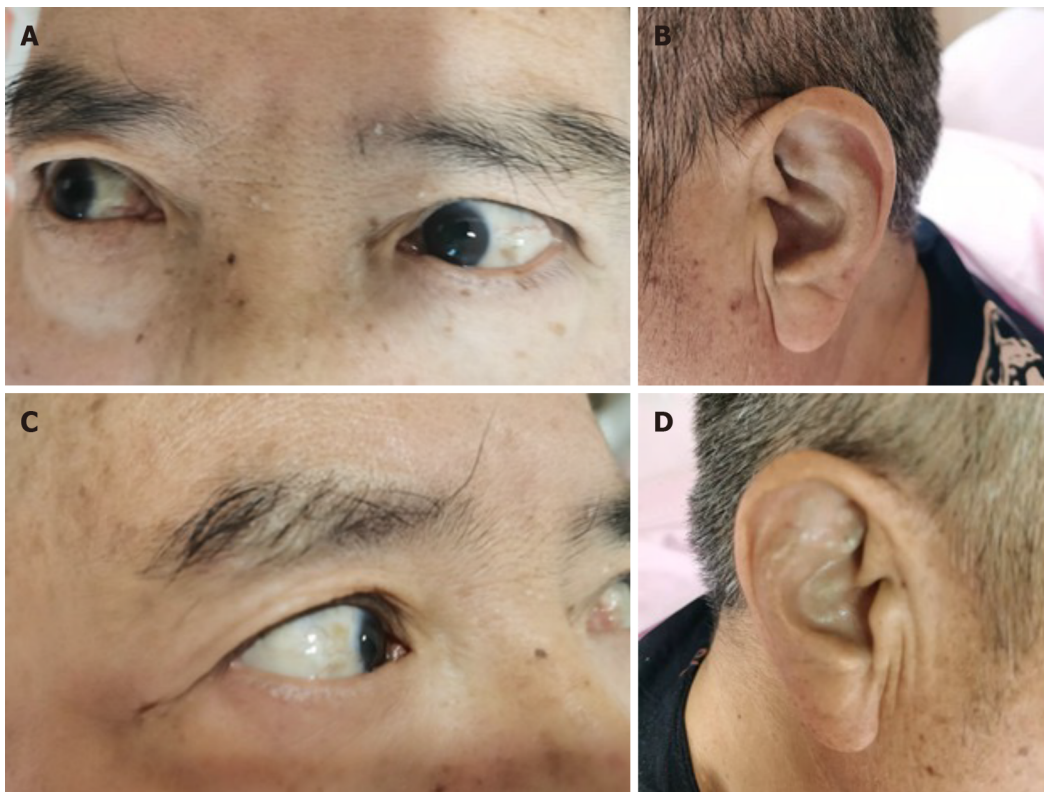
The final diagnosis in this patient was OcA and left knee infection.

TREATMENT

We decided to carry out arthroplasty immediately under the guidance of the theory of one-stage revision. The patient underwent left cementless total knee replacement. During the operation, we found that all cartilage surfaces of the patient's left knee joint were black-brown (Figure 4), and a large amount of normal saline and iodophor were used to wash the joint cavity. Postoperatively, based on the results of bacterial culture, the patient received intravenous injections of ceftazidime and intrarticular injections of meropenem for 2 wk. This was followed by oral administration of sulfamethoxazole tablets and rifampicin for 4 wk.

OUTCOME AND FOLLOW-UP

The patient's left knee pain was considerably relieved after surgery, and he was able to walk the day after surgery. One week postoperatively, the anteroposterior and lateral radiographs of the left knee joint showed that the prosthesis was in a good position (Figure 5). Routine blood tests, CRP and ESR were monitored after surgery. Both ESR and CRP decreased gradually, and eventually returned to normal. After 4 mo of postoperative follow-up, the patient's left knee joint function was normal without pain, and both ESR and CRP were normal.



DOI: 10.12998/wjcc.v10.i25.9036 Copyright ©The Author(s) 2022.

Figure 1 Brown-black pigment can be seen on both sides of the auricle and sclera. A: Left eye; B: Left ear; C: Right eye; D: Right ear.

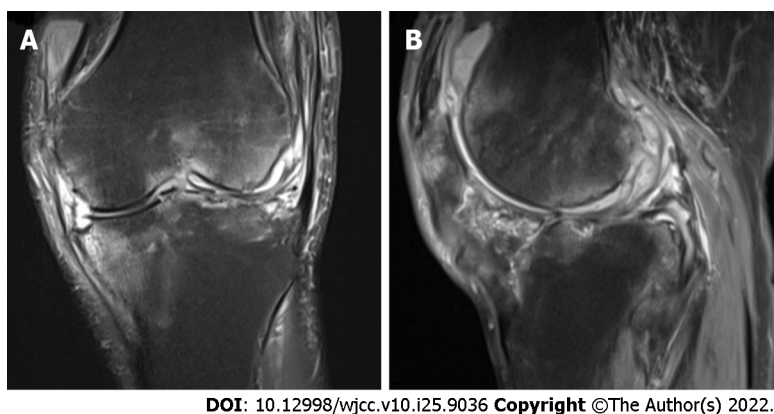


DOI: 10.12998/wjcc.v10.i25.9036 Copyright ©The Author(s) 2022.

Figure 2 Radiographs of the patient's knee showed joint space narrowing and osteophyte formation. A: Coronal plane; B: Sagittal plane.

DISCUSSION

AKU is an extremely rare disease that most clinicians have not seen before. The worldwide prevalence of AKU is 1 case in 250000-1000000 births[13]. It has three distinct clinical features: homogentisic aciduria, ochronosis, and ochronotic osteoarthropathy[14]. Patients initially have asymptomatic black urine, which has no significant impact on their life. They are usually over 40 years old when joint symptoms develop[15]. At this time, it is easy to misdiagnose OcA as osteoarthritis. The pathological changes due to knee osteoarthritis are characterized by degeneration of articular cartilage and periar-



DOI: 10.12998/wjcc.v10.i25.9036 Copyright ©The Author(s) 2022.

Figure 3 Magnetic resonance image of the patient's left knee revealed bone marrow edema. A: Coronal plane; B: Sagittal plane.



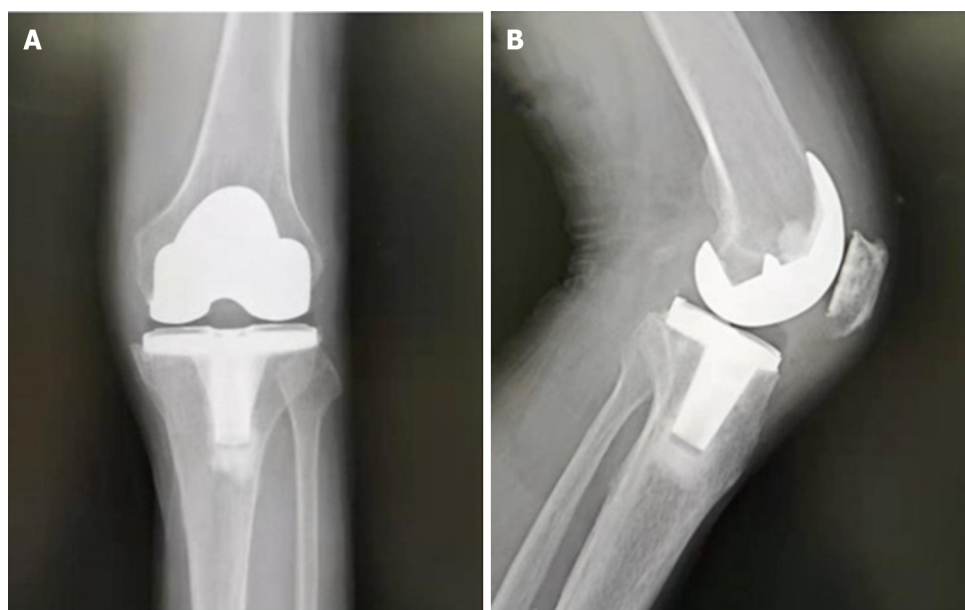
DOI: 10.12998/wjcc.v10.i25.9036 Copyright ©The Author(s) 2022.

Figure 4 Intraoperative images show that the surface of the knee cartilage was dark brown.

ticular hyperosteoegeny. Inflammatory indicators such as ESR and CRP are generally normal. The main symptoms are knee pain and limited mobility. The pain is often dull, involving the whole knee or more localized, increases with joint use, and abates with rest[16]. Rheumatoid arthritis, which is a chronic, systemic inflammatory disease, frequently causes knee joint lesions. The most prominent feature is symmetric pain and swelling of the hands, wrists, feet, and knees (polyarthritis)[17]. Knee joint lesions are often accompanied by systemic symptoms such as low-grade fever, anemia, and weight loss.

Arthroplasty is an effective treatment in patients with OcA[18]. Ozmanevra *et al*[6] reviewed 13 studies on OcA. In the study, 21 cases of OcA were treated with arthroplasty, and achieved excellent clinical efficacy. However, no study has reported the use of arthroplasty in the treatment of patients with OcA and knee infection. *A. xylosoxidans* is a gram-negative aerobic bacterium that was first described in 1971[19]. This bacterium has been identified as the cause of meningitis, pneumonia, central catheter infection, sepsis, mediastinitis, and other diseases[20,21]. It was found in our patient's knee joint, which is rare. This species is drug-resistant and widely distributed in both indoor and outdoor environments, especially in humid areas[22]. Elderly people living at home can be affected by the bacterium. The patient's infection with *A. xylosoxidans* may have been related to living in an old neighborhood.

PJI is a common complication after joint replacement, which is difficult to treat. There are many surgical strategies for the treatment of PJI, including surgical debridement and retention of a prosthesis, one-stage exchange arthroplasty or two-stage exchange arthroplasty. One-stage exchange arthroplasty is a method used to treat patients with a joint infection. This method resolves the patient's problem with only one operation, avoiding a second operation. This surgical method has many advantages for



DOI: 10.12998/wjcc.v10.i25.9036 Copyright ©The Author(s) 2022.

Figure 5 One week postoperatively, the knee X-ray showed that the prosthesis was in a good position. A: Coronal plane; B: Sagittal plane.

patients, such as early walking, avoiding multiple surgical trauma, cost savings, and resuming daily life at an early date. In patients with OcA, not all have only joint degeneration, and inflammatory indicators such as ESR and CRP can also be elevated[23]. A study found that the production of HGA products may lead to the activation of inflammatory pathways in patients and an increase in inflammatory factors above normal levels[24]. Compared to patients with osteoarthritis, if examination results in OcA patients show increased inflammatory factors, then the possibility of infection is greatly increased. Our patient with OcA had left knee joint infection, and combined with the theory of one-stage revision, total knee arthroplasty was performed in this patient. Postoperative inflammatory indicators such as white blood cell count, ESR, and CRP decreased gradually, and finally returned to normal. At the patient's review 4 mo after surgery, we observed that the left knee joint incision had healed well, local skin temperature was normal, and the range of motion of the left knee joint was 0°-110°. In addition, pain in the left knee joint had disappeared.

In this case, the short-term effect after operation was satisfactory. However, the follow-up period was short, and additional long-term follow-up is needed to clarify the long-term efficacy of this treatment.

CONCLUSION

We report a patient who developed OcA and left knee joint infection. This disease is rare, but should not be overlooked by clinicians. It is easily misdiagnosed as osteoarthritis, but it has clinical manifestations different from arthritis. Under the guidance of the one-stage revision theory, we treated a patient with OcA accompanied by knee infection with arthroplasty, and achieved satisfactory clinical results. This approach can be applied in patients with OcA and joint infection.

ACKNOWLEDGEMENTS

We would like to appreciate our patient for consenting to have his case presented and published.

FOOTNOTES

Author contributions: Wang XC, Zhang XM, Yan TS, and Cao XW designed the study; Wang XC, Zhang XM, Yan TS, Liu YH, He QL, and Cao XW performed the research; Wang XC, Cai WL, Li Z, and Ma C reviewed the literature and contributed to drafting the manuscript; all authors have read and approved the final manuscript.

Supported by Talent Training Project of Guangdong Provincial Bureau of Traditional Chinese Medicine, No. 0103030908; and Guangdong Provincial Hospital of Traditional Chinese Medicine and the School of Biomedicine,

Chinese University of Hong Kong School of Medicine, Basic Clinical Collaborative Innovation Project, No. YN2018HK04.

Informed consent statement: Informed written consent was obtained from the patient for publication of this report and any accompanying images.

Conflict-of-interest statement: The authors have no conflicts of interest to declare.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Open-Access: This article is an open-access article that was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution NonCommercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <https://creativecommons.org/licenses/by-nc/4.0/>

Country/Territory of origin: China

ORCID number: Xiao-Chao Wang 0000-0002-6992-8662; Xiao-Min Zhang 0000-0003-0104-1747; Wan-Ling Cai 0000-0003-4620-7341; Zhen Li 0000-0003-4961-0590; Chao Ma 0000-0002-6057-7413; Yi-Hai Liu 0000-0001-8461-0444; Qi-Lian He 0000-0003-1344-5933; Tian-Sheng Yan 0000-0003-0208-989X; Xue-Wei Cao 0000-0001-6805-5268.

S-Editor: Yan JP

L-Editor: Filipodia

P-Editor: Yan JP

REFERENCES

- 1 **Ranganath LR**, Cox TF. Natural history of alkaptonuria revisited: analyses based on scoring systems. *J Inherit Metab Dis* 2011; **34**: 1141-1151 [PMID: 21748407 DOI: 10.1007/s10545-011-9374-9]
- 2 **Braconi D**, Bernardini G, Bianchini C, Laschi M, Millucci L, Amato L, Tinti L, Serchi T, Chellini F, Spreafico A, Santucci A. Biochemical and proteomic characterization of alkaptonuric chondrocytes. *J Cell Physiol* 2012; **227**: 3333-3343 [PMID: 22213341 DOI: 10.1002/jcp.24033]
- 3 **Zhao BH**, Chen BC, Shao de C, Zhang Q. Osteoarthritis? *Knee Surg Sports Traumatol Arthrosc* 2009; **17**: 778-781 [PMID: 19381613 DOI: 10.1007/s00167-009-0778-0]
- 4 **Gómez-Lechón Quirós L**, Hidalgo Calleja C, Acosta de la Vega ME, Compán Fernández O, Pastor Navarro S, Montilla Morales C. Family history of ochronotic arthropathy. *Rheumatol Int* 2021; **41**: 1869-1874 [PMID: 32642830 DOI: 10.1007/s00296-020-04640-2]
- 5 **Gowda N**, Kumar MJ, Kumar AK. Black hip: a rare case treated by total hip replacement. *Ann Saudi Med* 2013; **33**: 368-371 [PMID: 24060716 DOI: 10.5144/0256-4947.2013.368]
- 6 **Ozmanevra R**, Güran O, Karatosun V, Günel I. Total knee arthroplasty in ochronosis: a case report and critical review of the literature. *Ekleml Hastalik Cerrahisi* 2013; **24**: 169-172 [PMID: 24191883 DOI: 10.5606/ehc.2013.36]
- 7 **Kapadia BH**, Berg RA, Daley JA, Fritz J, Bhawe A, Mont MA. Periprosthetic joint infection. *Lancet* 2016; **387**: 386-394 [PMID: 26135702 DOI: 10.1016/S0140-6736(14)61798-0]
- 8 **Oduwale KO**, Molony DC, Walls RJ, Bashir SP, Mulhall KJ. Increasing financial burden of revision total knee arthroplasty. *Knee Surg Sports Traumatol Arthrosc* 2010; **18**: 945-948 [PMID: 20148322 DOI: 10.1007/s00167-010-1074-8]
- 9 **Hernández-Vaquero D**, Fernández-Fairen M, Torres A, Menzie AM, Fernández-Carreira JM, Murcia-Mazon A, Gueraldo E, Merzthal L. Treatment of periprosthetic infections: an economic analysis. *ScientificWorldJournal* 2013; **2013**: 821650 [PMID: 23781163 DOI: 10.1155/2013/821650]
- 10 **Haddad FS**, Sukeik M, Alazzawi S. Is single-stage revision according to a strict protocol effective in treatment of chronic knee arthroplasty infections? *Clin Orthop Relat Res* 2015; **473**: 8-14 [PMID: 24923669 DOI: 10.1007/s11999-014-3721-8]
- 11 **Nagra NS**, Hamilton TW, Ganatra S, Murray DW, Pandit H. One-stage versus two-stage exchange arthroplasty for infected total knee arthroplasty: a systematic review. *Knee Surg Sports Traumatol Arthrosc* 2016; **24**: 3106-3114 [PMID: 26392344 DOI: 10.1007/s00167-015-3780-8]
- 12 **Zahar A**, Webb J, Gehrke T, Kendoff D. One-stage exchange for prosthetic joint infection of the hip. *Hip Int* 2015; **25**: 301-307 [PMID: 26109160 DOI: 10.5301/hipint.5000264]
- 13 **Phornphutkul C**, Introne WJ, Perry MB, Bernardini I, Murphey MD, Fitzpatrick DL, Anderson PD, Huizing M, Anikster Y, Gerber LH, Gahl WA. Natural history of alkaptonuria. *N Engl J Med* 2002; **347**: 2111-2121 [PMID: 12501223 DOI: 10.1056/NEJMoa021736]
- 14 **Mistry JB**, Bukhari M, Taylor AM. Alkaptonuria. *Rare Dis* 2013; **1**: e27475 [PMID: 25003018 DOI: 10.4161/rdis.27475]
- 15 **Demir S**. Alkaptonuric ochronosis: a case with multiple joint replacement arthroplasties. *Clin Rheumatol* 2003; **22**: 437-439 [PMID: 14677022 DOI: 10.1007/s10067-003-0760-z]
- 16 **Sharma L**. Osteoarthritis of the Knee. *N Engl J Med* 2021; **384**: 51-59 [PMID: 33406330 DOI: 10.1056/NEJMcp1903768]

- 17 **Sparks JA.** Rheumatoid Arthritis. *Ann Intern Med* 2019; **170**: ITC1-ITC16 [PMID: [30596879](#) DOI: [10.7326/AITC201901010](#)]
- 18 **Karaoglu S,** Karaaslan F, Mermerkaya MU. Long-term result of arthroplasty in the treatment of a case of ochronotic arthropathy. *Acta Orthop Traumatol Turc* 2016; **50**: 584-586 [PMID: [27817975](#) DOI: [10.1016/j.aott.2016.08.018](#)]
- 19 **Yabuuchi E,** Oyama A. *Achromobacter xylosoxidans* n. sp. from human ear discharge. *Jpn J Microbiol* 1971; **15**: 477-481 [PMID: [5316576](#) DOI: [10.1111/j.1348-0421.1971.tb00607.x](#)]
- 20 **Kengni Tameze J,** Korpak K, Compagnie M, Levie H, Cherifi S, Lali SE. Mitral endocarditis caused by *Achromobacter xylosoxidans* in an older patient: Case report and literature review. *IDCases* 2022; **27**: e01421 [PMID: [35198382](#) DOI: [10.1016/j.idcr.2022.e01421](#)]
- 21 **Marion-Sanchez K,** Lion F, Olive C, Cailleaux G, Roques F. Mediastinitis superinfected by *Achromobacter xylosoxidans*. A case report. *J Infect Chemother* 2018; **24**: 987-989 [PMID: [29895453](#) DOI: [10.1016/j.jiac.2018.05.005](#)]
- 22 **Nakamoto S,** Sakamoto M, Sugimura K, Honmura Y, Yamamoto Y, Goda N, Tamaki H, Burioka N. Environmental Distribution and Drug Susceptibility of *Achromobacter Xylosoxidans* Isolated from Outdoor and Indoor Environments. *Yonago Acta Med* 2017; **60**: 67-70 [PMID: [28331426](#)]
- 23 **Mannoni A,** Selvi E, Lorenzini S, Giorgi M, Airó P, Cammelli D, Andreotti L, Marcolongo R, Porfirio B. Alkaptonuria, ochronosis, and ochronotic arthropathy. *Semin Arthritis Rheum* 2004; **33**: 239-248 [PMID: [14978662](#) DOI: [10.1053/s0049-0172\(03\)00080-5](#)]
- 24 **Yuce Inel T,** Kisa PT, Balci A, Uslu S, Arslan Z, Hismi BO, Ucar U, Arslan N, Onen F, Sari I. Inflammatory rheumatic diseases in patients with ochronotic arthropathy. *Mod Rheumatol* 2021; **31**: 1031-1037 [PMID: [33427541](#) DOI: [10.1080/14397595.2020.1868121](#)]



Published by **Baishideng Publishing Group Inc**
7041 Koll Center Parkway, Suite 160, Pleasanton, CA 94566, USA

Telephone: +1-925-3991568

E-mail: bpgoffice@wjgnet.com

Help Desk: <https://www.f6publishing.com/helpdesk>

<https://www.wjgnet.com>

