

A Review of the Surgical Treatment Methods of Kashin-Beck Disease, an Endemic Osteoarthritis in China

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Abstract

In China, with the decrease of the prevalence of Kashin-Beck disease (KBD) in children, the treatment methods for adult KBD patients have attracted more attention. Mostly, to alleviate the pains and improve the function of joints of moderate and severe KBD patients, surgical treatment is a better way. The methods of surgical treatment for different involved joints are usually extremely different. Arthroscopic debridement and total knee replacement are applicable for most KBD knee joints. Artificial ankle replacement and ankle arthrodesis are mainly suitable for serious KBD ankle joints. Total hip arthroplasty is currently recognized as an effective way to improve hip joint function. For other joints, including shoulder, elbow, wrist, metacarpophalangeal and so on, an appropriate surgical method has often been selected according to the pathological features of KBD in different stages, such as debridement, osteotomy and periosteum transplantation arthroplasty. This paper aims to summarize the related research progresses of KBD surgical treatments, and provides practical references for clinical KBD treatment. It is quite important to perform different surgical treatments based on different damage locations and levels in KBD patients. Making efforts to benefit every patient, also regarded as personalized medicine, may be the development trend of surgical treatments for KBD.

Keywords: Kashin-Beck disease; Joint; Clinical effect

Introduction

Kashin-Beck disease (KBD), an endemic and deformed osteoarthritis with unclear etiology, is multiple in children and adolescents. The labor capacity of the mild KBD patients, who always suffer from joints' thickening deformation, muscle atrophy and pain all year round, was affected to some extent. The severe KBD patients, who are suffering from the developmental disorders and phocomelia have lost their labor abilities and have difficulties in taking care of themselves. KBD brought heavy burdens to the family and society, leading to the general ward poverty. This is one of the reasons for block in the economic development in the endemic areas [1-3]. Most KBD patients, living in mountainous, are labor-intensive. The main feature of KBD is multiple hyaline cartilages of endochondral bone and focal cartilage necrosis during the process of adolescent development. The knee, ankle and other joints of lower limbs are often involved in. It was reported that all adult KBD patients often had two or more large joints involved, and the elbow and knee joints were the most common ones involved [4]. A research on adult KBD patients from

Rangtang county Aba Sichuan province showed that 9-12 large joints have been implicated in Grade II adult KBD patients [4]. The treatments for adult KBD are faced with great clinical challenges. So far, the treatments for KBD, such as physical therapy, oral administration or injection of non-steroidal anti-inflammatory drugs (NSAIDs) or hyaluronan acid [5] can help to some extent, however, they are still palliative. It was shown that physical therapy could increase mobility and function, and relieves pain. While for KBD treatments, it needs a long-term treatment to get effective outcomes. Drug therapy can slow down the progression course of KBD and reduces pain as well, but some side effects bring unspeakable suffering to the patients' physiology and psychology, like drug addiction, withdrawal syndrome, etc. Therefore, it is necessary for the doctors and patients to find treatments with reliable curative effects, short course and fewer complications. Surgical treatments attract doctors' and patients' attention, as they can alleviate the joints inflammation and improve the joints function rapidly by clearing the articular loose bodies, or replacing the diseased joints fundamentally [6]. It is reported that surgical treatment is mainly applied in the advanced KBD cases, which are diagnosed as Grade II and III according to the diagnostic criteria of KBD [6]. However, studies showed that if mild KBD patients (Grade I) could get timely surgical treatments, they will achieve a relatively satisfactory curative effect after surgery [7]. There are numerous reports of surgical treatment methods, such as percutaneous bone puncture decompression, joint debridement, arthroplasty, arthroscopic surgery and acupotomy [7-8]. As known, in adult KBD patients, many joints are always affected, which means it is difficult to perform surgical operations for them when multiple joints involved under different damage conditions. As there are no relevant reports showing how to select the surgical methods according to different involved joints. In this paper, the surgical treatments of different types of involved joints, including big joints such as knee, ankle, hip and other small joints in KBD patients were systematically reviewed (Table 1 and Figure 1).

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Table 1: Some representative studies of KBD operation methods

Studies	Operation Methods	Age of Patients (years)	Course of Disease (years)	Number of Joints	Index of Evaluation	Follow-up Period
Huang XH et al. 2012 [10]	Knee joint debridement	19-64	NM	24	Self-evaluation on clinical symptoms and pain index	8 years
Tang X et al. 2014 [15]	Total knee arthroplasty	55-80	May-40	18	VAS, HSS, FSAT-KBD	(23-62) months
Liu FD et al. 1998 [17]	Knee joint osteotomy	Dec-41	NM	247	Functional handicap and limitation of working ability	(1.5-12) years
Dai JN et al. 2015 [20]	Ankle arthrodesis	51-62	15-Oct	18	AOFAS	(8-14) months
Zhao W et al. 2005 [24]	Ankle joint replacement	54-65	10-Mar	5	McGuire score	(0.5-1.5) years
Li HL et al. 2015 [28]	Ankle joint debridement	23-57	NM	48	VAS, AOFAS, ROM	(3-24) months
Lv YY 2014 [29]	Total hip arthroplasty	45-55	NM	15	Joint pain, ROM	(1-14) years
Wang JC et al. 2006 [33]	Metacarpal periosteum transplantation arthroplasty	55-60	10-Apr	15	Pain, ROM, joint stability and gripping force.	(1-5) years

NM: Not Mention; **UC:** Unclear; **VAS:** Visual Analogue Scale; **HSS:** Hospital for Special Surgery; **FSAT-KBD:** Functional Score for Adult Tibetans with KBD; it was developed by orthopedic experts for evaluating changes in KBD symptoms and disability in Tibet according to the specific lifestyle of Tibetans; **AOFAS:** American Orthopedic Foot and Ankle Society; **ROM:** Range of Motion

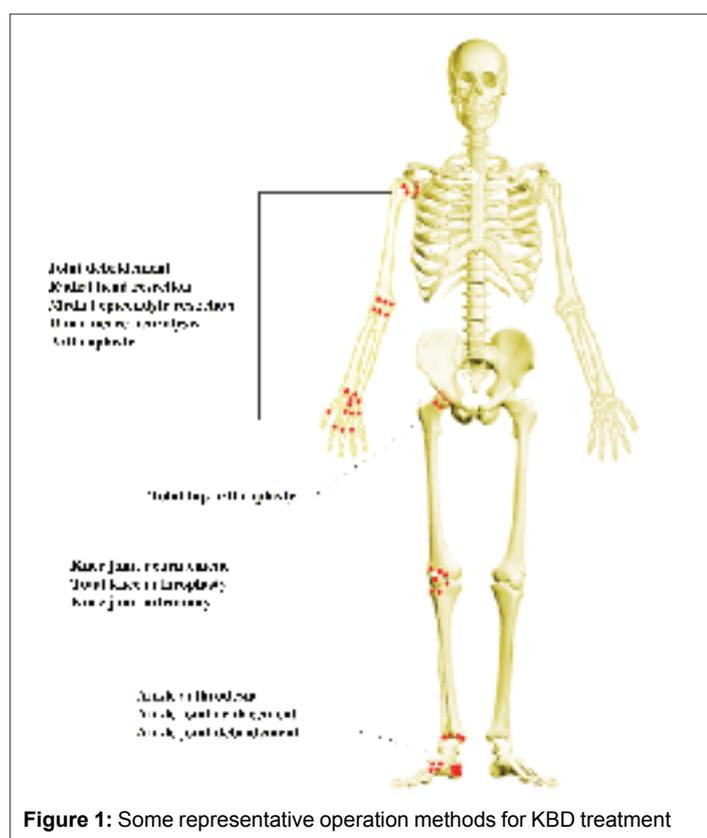


Figure 1: Some representative operation methods for KBD treatment

Review

Surgical treatments of knee joints in KBD patients

Knee joints are the most common joints observed with damages in KBD. Current surgical treatment methods of KBD knee lesion mainly include joint debridement (open debridement and arthroscopic debridement), arthroplasty and osteotomy. Joint loose bodies are cartilage debris or calcified cartilage freely in KBD patients' articular cavity, which could be formed mainly due to malnutrition, developmental disorders, cartilage atrophy, degeneration, necrosis and shedding [9]. They can cause joint pain, effusion, locking phenomenon, and directly affect the life and work ability of adults. Joint debridement, which was also called free bodies removal surgery, was used to clean up the excessive joint loose bodies so that it can help anti-inflammatory pain and improve

function. Joint debridement is a simple operation method, characterized by fewer traumas, low cost, good recovery of joint function. Therefore, the government has adopted a policy of free joint debridement surgery in hospital for severe KBD patients for some years. The surgery can relieve the long-term pain, prevent inflammation and restore the ability of life and work of the patients. Joint debridement has been welcomed by patients in the endemic regions. However, the operation effects of KBD knee joint debridement decreased with longer follow-up time gradually. One study on arthroscopic debridement with 31 KBD patients has been followed up for 2, 6 and 8 years after operation [10-11]. The pain index significantly decreased in 2, 6 and 8 years post operation compared with the preoperative. However, pain index was significant higher in 8 years than that in 2 years after operation [10]. The efficiencies of patients' self-evaluation on clinical symptoms improvement were 95.83%, 70.83%, and 87.50%, respectively in 2, 6 and 8 years after operation [10-11]. Total knee arthroplasty (TKA) had achieved good effects in the treatments of severe osteoarthritis patients. It not only improved the physical function of the knees of osteoarthritis patients, but also relieved their pain [12-14]. One study of effects of TKA in the treatments of severe KBD patients' knee has been performed with 18 knees in 15 elder KBD patients. As a result, all the patients underwent TKA achieved good clinical results, with a significant reduction in knee pain measured by visual analogue scale (VAS), a significant functional improvement measured by hospital for special surgery (HSS) and by functional score for adult Tibetans with KBD (FSAT-KBD) in a short-term follow-up after TKA [15]. Furthermore, no radiographic evidence of loosening, deformation, and fracture in component positioning was found at the final follow-up [15]. Additionally, Yang LG et al summarized the postoperative effects of the TKA in treatments of adult KBD knee with Genu Valgum mixed deformities within 2-72 months [16]. During the follow-up period, there were no cases of periprosthetic zona pellucida appeared, prosthesis subsidence or loosening, bone graft displacement, fracture, bone resorption, bone nonunion and tibial plateau collapse. At the end of the follow-up, the HSS score was 63.7 to 89.4, with an average of 82.6 points, and the results were excellent (66.67%), good (25.93%) and general (7.40%) in all knees. A wide range of mixed deformity angle has been corrected. Osteotomy is a valuable treatment to improve the knee deformities for the severe KBD cases, which is suitable for young and middle-aged KBD patients. It improves the ability of patients' work and life mainly by improving serious knee deformity of KBD patients. Liu et al and his colleagues showed that osteotomy with impaction and without internal or external fixation was an effective method in treating serious knee deformities in KBD patients (n = 195, age of 12-41 years old). In addition, the rate of normal work ability has increased to 96.4% from 59.5% after osteotomy [17].

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Surgical treatments of ankle joints in KBD patients

The ankle joint discomforts of KBD patients are mainly caused by talus osteonecrosis. The clinical symptoms are similar to arthritis, such as pain, swelling and deformity. Surgical treatments of talus necrosis include ankle fusion, vascularized fibular graft and ankle arthroplasty [18]. For severe talus necrosis, traditional ankle arthrodesis is applicable for its mature technology and certainly curative effect. Ankle arthrodesis can provide firm fixation for KBD ankle joints, and correct the ankle varus deformity and valgus deformity with satisfied effects. For ankle arthrodesis, the key factors to its success are strong internal fixation and precision attachment of resected surface of tibial and astragalus [19]. The materials for fixation are often divided into following classes: autologous bone, artificial bone, artificial bone and autologous bone mixed bone grafting. The autologous bone fixation shows advantages including high fusion rate, less complications, low medical cost, and satisfactory post-operational clinical effect rate; but it causes large trauma [20]. Complete artificial bone is difficult to heal. But artificial - autologous mixed bone grafting cannot only reduce the defects of complete artificial bone, but also make up the shortcomings of insufficient bone mass [21]. It was shown that 20 cases of talus necrosis in 20 KBD patients (average age: 55.4 years old) were cured by ankle fusion with the artificial bone and autologous bone mixed bone grafting. All of the patients were followed up for 15.8 months on average. There was no obvious rejection and nonspecific inflammatory reaction found in those patients, and X-ray showed that the bone healing time was from 4 to 6 months (mean, 5.2 months) [21]. Besides, one study showed that 18 cases of talus necrosis in 18 KBD patients had been cured by ankle arthrodesis with fibula fixation [20]. There was a mean follow-up period of 12.8 months after operation. X-ray suggested that ankle joints achieved bony fusion on average 14.4 weeks after operation. There were only 1 case of fat liquefaction and 1 case of incision edge black necrosis after symptomatic treatments; the two cases had been healed [20]. It was reported that arthroscopic ankle arthrodesis had been used to treat right ankle of a KBD patient, who suffered from bilateral pain. One year after the operation, the ankle had reached to adequate bone union according to the radiographs, and the patient had no feeling of pain at this ankle [22]. Ankle arthrodesis can effectively relieve the pain; but also can cause permanent loss of ankle function. Therefore, the ankle arthrodesis can be considered in extremely serious conditions, for example when the pain is intense, the patient's life has been seriously affected or the ankle function has been serious loss. Due to the design defect, early ankle joint replacement cannot completely relieve the pain; promote joint activities and long-term reliability. Therefore, many scholars do not support for ankle joint replacement surgery. Even though some researchers still support this operation scheme. Yin JH et al had carried out 13 cases of total ankle replacement with Scandinavian total ankle replacement (STAR) system (Waldemar, Hamburg, Germany) from October 1999 to February 2003. These patients had an average age of 63.4 years. According to the Kofoed ankle scoring system, the preoperative and postoperative pain degree, ankle joint function and ankle mobility were evaluated. Eight cases were followed up; the average follow-up time was 16 months. The 8 patients were with an average of 29 points for preoperative ankle score and an average of 82 points for postoperative ankle score. The average scores of ankle pain were 7 points and 48 points separately in before operation and after operation. All the patients were followed up by X-ray and no loosening or sinking of the prosthesis has been found [23]. Zhao et al followed up five ankle operation in three patients with STAR ankle joints prosthesis for 1.5 years. According to McGuire score, three ankle joints were excellent, two ankle joints were good, and no prosthesis loosening or sink were found [24]. Another study with 8 cases in total of artificial ankle replacement in 8 KBD patients has been performed from July 2004 to May 2007 [25]. All the patients were followed up for two years averagely with an average age of 62.3 years. The Kofoed ankle score was significantly

higher than that before operation, and the curative effect was excellent [25]. So the key for total ankle replacement was prosthetic materials and designs. The absolute contraindications for total ankle replacement were neurogenic arthropathy, active phase of infection, ischemic osteonecrosis (especially necrosis in more than half of the talus), abnormality of rebuild ankle function, and severe lesions of the tissue around ankle, high demand in postoperative exercise degree (strenuous exercises such as running, jumping, etc.) [23]. In addition, Kofoed [26] claimed that the ankle replacement is more excellent than ankle fusion in relieving pain, improving function, reducing the rate of infection without secondary subtalar joint osteoarthritis patients. Joint debridement is an important operation of KBD ankle surgery. It is mainly suitable for the mild KBD patients with fewer traumas. Its advantages are quick postoperative recovery, less pain and elimination of inflammation. But its disadvantage is easy to relapse. Gu et al treated 15 KBD patients with ankle osteoarthritis (mean age 49) by ankle arthroscopic debridement; after the follow-up of 22 months, the joint pain and interlocking symptoms remission rate was 93.3% [27]. Besides, there was another study to evaluate the effect of joint debridement by the American Orthopedic Foot and Ankle Society (AOFAS) score [28]. It was found that the scores of the patients in 3, 6, 12 and 24 month after operation were significantly higher than that before operation. The score was the highest in the 3 month of postoperative period, and then the score gradually decreased with the prolongation of the follow-up time after operation [28].

Surgical treatments of hip joint in KBD patients

Mostly, joints of the adult KBD patients, such as hand, wrist, ankle and knee, have different degrees of degeneration. Hip joint is also involved, however, when the lesion causes hip fibrous or bony ankyloses, the daily life of the patients has been seriously affected. Total hip arthroplasty (THA) is currently recognized as an effective way to improve the hip joint function. Total hip arthroplasty has been performed in 15 cases of femoral head necrosis in 14 KBD patients for analyzing the curative effect and indication evaluation [29]. KBD patients (mean age 50) were followed up for average 3 years after THA; the average excellent rate was 80%. Three years after THA, the patients with hip joint pain relief, function improvement, could participate in daily labor [29]. This study indicated that THA had good effect on Grade I (8 cases) and II (5 cases) KBD patients who had a vascular necrosis of the femoral head. Hip pain of the patients was significantly reduced and the function of the hip joint significantly was improved [29]. However, no efficacious effect has been found from Grade III (2 cases) KBD patients after THA [29]. Therefore, THA should be carefully considered for treating Grade III KBD patients with femoral head necrosis [29]. The effects of two materials of artificial hip joints (bone cement prosthesis and non-bone cement prosthesis) used for THA of KBD patients have been investigated. Both materials for THA can alleviate the joint pain and improve joint movable function. It showed that there was no statistically significant difference between both the two materials [30]. In order to achieve a good clinical result of THA for late stage of KBD patients, it is very important to establish a strict preoperative plan including selection of a suitable prosthesis and proper surgical treatment, as well as good preparation for all kinds of complications.

Surgical treatments of other joints in KBD patients

Although the KBD often damages the load-bearing joints (including hip, knee and ankle joints), the involvements of the shoulder, elbow, wrist, metacarpophalangeal joints and other small joints were also observed. Open debridement operations have been performed for 13 joints of elbow (9), wrist (2), and meta carpophalangeal (2). It showed an improvement of all operated joints [31]. One previous study also reported the excellent post operative effect of open debridement for three elbow joints [32]. The indications of the metacarpal periosteum transplantation

arthroplasty were mainly traumatic arthritis and degenerative arthritis, rheumatoid arthritis and congenital malformation. The therapeutic effects of metacarpal periosteum transplantation arthroplasty on the cartilage destruction in palm and finger of KBD patients have been studied [33]. 11 KBD patients with the average age of 57.5 years old participated in the study, 10 cases were followed up, and one case was lost. The follow-up time was 1-5 years (average 4 years). Metacarpophalangeal joints of 10 arthroplasty had no pain. Metacarpophalangeal joints active flexion extension range was improved from 35 degrees to 76 degrees (average 52.2 degrees). 6 cases of severe ulnar deformity were corrected. Usually, the indications of the periosteum transplantation arthroplasty were mainly pain. Stiff joints without pain were not suitable for this surgery because the increased range of postoperative joint motion is often unsatisfactory. The other research showed that the postoperative range averaged rose from 20 degrees (preoperative range) to 37 degrees, which indicated that the operation also had certain effects on stiff joint. Therefore, periosteum transplantation arthroplasty is a desirable method for repairing the cartilage destruction, relieving the pain and improving the dysfunction of articular cartilage in the palm and finger caused by KBD [33]. One study was carried out with different surgical methods on 287 cases of KBD patients. Different surgical methods have been selected for the patients according to the classification of X-ray results [34]. The surgical operations including joint debridement, incision debridement plus osteotomy, radial head resection, resection of the medial epicondyle, ulnar nerve neurolysis and subcutaneous epineurium incision, carpal tunnel incision of epineurium incision and decompression have been performed for 17 elbow joints and four wrist joints in this study [34]. The total postoperative excellent rate was 76.5% and 75% respectively, which indicated that appropriate selection of surgical methods according to the pathological features of KBD in different stages did not need long-term medication, so it could reduce the impact of drug dependence and side effects on the body. When compared with non-operative therapy, the effect of surgical methods was reliable for curative effect and easy to consolidate [34].

Suggestions for surgical treatment methods of systemic multiple KBD joints

Most adult KBD patients often had two or more large joints damage involved in disease progress [4]. Therefore performing different surgical treatment methods for different involved positions could relieve the pain and minimize the lesion of people suffering from KBD. Generally, the joints with obvious symptoms, severe deformity or poor function, should be treated in advance. Besides, the operation order should be decided based on the principle of joint position from top to bottom. For example, if the hip and knee joints were involved with similar degree of lesions at the same time, the operation of hip joint should be performed first, followed by the operation of knee joints, avoiding operation simultaneously in one anesthesia; the staging interval should be more than 3 months [34]. Moreover, the age of patients should also be considered, because the physical health statuses, activity intensity and expectation are disparate between the youth and the elder. Thus making optimal surgery plans for KBD patients according to the demands and conditions is highly recommended.

Conclusion

This paper reviewed some common surgical treatments on the joint damage of KBD patients. It mainly included joint debridement, arthroplasty, osteotomy and other surgeries. For early KBD joint damage, joint debridement is a preferred method because of small traumas, quickly recovering, the pain relief and the improvement of joint function. This method is also accepted for the majority of KBD patients. Osteotomy is commonly used in younger patients with KBD to improve joint deformity,

but there were a lot of complications of osteotomy, such as infection, insufficient correction, local paresthesia, secondary displacement and so on. This operation was not frequently used because of the decline of KBD incidence and less young KBD patients. Joint replacement is commonly used in older, heavier lesions of KBD patients, where hip joint replacement and knee joint replacement are relatively mature surgical treatments for KBD patients. However, the arthroplasty should be carefully chosen when the KBD patients have pathological changes of Grade III or higher. In terms of the current common clinical surgical treatments, Arthroscopic debridement and total knee replacement are applicable for most KBD knee joints. Artificial ankle replacement and ankle arthrodesis are mainly suitable for serious KBD ankle joints. Total hip arthroplasty is currently recognized as the effective way to improve hip joint function. For the other small joints, the appropriate surgical methods are often selected according to the pathological features of KBD in different stages; such as debridement, osteotomy and periosteum transplantation arthroplasty. In general, the developing trend of surgical treatment methods is individualization. It requests excellent and high quality standards in the field of surgical operation. In order to adapt to this tendency, it is necessary for the surgeon to get proficient in various operation methods and indications. Consequently, a prudent choice can be made for KBD patients.

Conflict of Interest

No conflict of interest.

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