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CASE REPORT

Three-dimensional printed talar prosthesis with biological function for giant cell tumor of the talus: A case report and review of the literature

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Author contributions: Tang KL and Tao X were responsible for the conception and design of the research and article validation; Mu MD and Yang QD were responsible for data collection and integration; Yang QD was the final author of the article.

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Abstract

BACKGROUND

Giant cell tumors (GCT) are most commonly seen in the distal femur. These tumors are uncommon in the small bones of the hand and feet, and a very few cases have been reported. A giant cell tumor of the talus is rarely seen clinically and could be a challenge to physicians.

CASE SUMMARY

We report a rare case of GCT of the talus in one patient who underwent a new reconstructive surgery technique using a three-dimensional (3D) printing talar prosthesis. The prosthesis shape was designed by tomographic image processing and segmentation using technology to match the intact side by mirror symmetry with 3D post-processing technologies. The patient recovered nearly full range of motion of the ankle after 6 mo. The visual analogue scale and American Orthopaedic Foot and Ankle Society scores were 1 and 89 points, respectively.

CONCLUSION

We demonstrated that 3D printing of a talar prosthesis is a beneficial option for GCT of the talus.

Key Words: Three-dimensional printing technology; Giant cell tumor; Talar prosthesis; Case report

Core Tip: Three-dimensional printing technology has been widely used in orthopedics.

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The purpose of this study is to evaluate the clinical results of the treatment of giant cell tumor of talus bone using a three-dimensional printing personalized talus prosthesis designed by our team. The casting process of this prosthesis is also discussed. Compared with other customized three-dimensional printing talus prostheses, our talus prosthesis is personalized and accurately constructed according to the anatomical data of the patient's normal foot.

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INTRODUCTION

Malignant tumors around the foot and ankle are rare, accounting for less than 1%[1] of all malignant tumors. However, 22%-39.2% of tumors occurring in these locations are malignant[2]. Tumors most commonly occur at the metatarsals and calcaneus followed by the phalanges and talus.

Giant cell tumors (GCTs), also called osteoclastomas, account for approximately 5% of bone tumors and 20% of benign bone tumors[3,4]. The highest incidence rate of GCTs occurs between the ages of 20 and 40 years with a peak at the age of 30. The incidence rate before the age of 10 is only approximately 1%[5]. These tumors often occur in individuals aged 30-40 years old and rarely occur in individuals under 20 years of age. For giant cell tumor of bone, it is important to completely remove tumor cells to prevent recurrence.

For bone tumors of the talus, currently available treatment methods include partial talus resection[6] and fusion surgery[7]. The more widely accepted surgical method is fusion surgery, including talocrural arthrodesis and shortening arthrodesis between the calcaneus and the tibia[8,9]. However, the loss of ankle joint function caused by fusion surgery is a problem that we cannot ignore. Fusion and other surgical methods seem to be difficult to deal with significantly large talar tumors. In particular, some studies have reported that complications, such as loss of motion of the ankle and adjacent joints and shortening of the affected limb after fusion surgery, are unacceptable to young patients[9].

With the development of three-dimensional (3D) printing technology, this methodology has been widely used in clinical practice and has yielded good results[10-12]; therefore, 3D printed, personalized, talus prostheses have been used in clinical surgery. The world's first replacement surgery with a 3D printed, personalized talus prosthesis with biological function was performed in our department in 2016. To the best of our knowledge, there are few related studies on the treatment of GCTs of the talus, especially with 3D printed, personalized talus prostheses. Thus, the purpose of this case report was to present the innovative treatment method and clinical outcomes. American Orthopaedic Foot and Ankle Society (AOFAS) and visual analogue scale (VAS) scores were used for clinical efficacy, and imaging measurement and evaluation were performed[13,14].

CASE PRESENTATION

Chief complaints

A 22-year-old patient visited our hospital because of pain in the left ankle for half a year.

History of present illness

The pain was persistent, became aggravated during walking and weight bearing, and was relieved during rest. The pain was associated with limited mobility of the ankle joint.

History of past illness

The patient had a free previous medical history.

Physical examination

The skin of the patient's foot was in good condition without swelling and ulceration. There was obvious tenderness in the ankle of the patient. Ankle range of motion was slightly limited

Laboratory examinations

The patient's preoperative laboratory examination was unremarkable, and a pathological biopsy revealed a giant cell tumor of the talus (Figure 1).

Imaging examinations

Computerized tomography (CT), magnetic resonance imaging (MRI), and whole-body bone scans were performed at our hospital, and the imaging findings suggested that the patient had a GCT of the talus bone, which was also demonstrated by pathological examination

FINAL DIAGNOSIS

Giant cell tumor of the left talus.

TREATMENT

Complete data of the affected area were acquired by CT image processing and segmentation using 3D CT postprocessing technology (Figure 2A), and the 3D raw data of the affected side were obtained by reconstruction and matching performed by mirror and data registration technology (Figure 2B). Severe defects in the data of the necrotic talus were repaired using reverse repair technology so that non-defective raw data could be used for talar reconstruction. Then, the data of the tibiotalar and subtalar articular facets were analyzed and processed (Figure 2C). After collecting complete patient talus data, an electron beam 3D printer ARCAM Q10 (Sweden, GE) with a maximum print scanning speed of 8000 m/s and a layer thickness of 0.05 mm was used to print the talus prosthesis. Finally, accurate 3D reconstruction of the talar prosthesis was completed (Figure 2D).

The column of the talar prosthesis at the calcaneal side and the position of the cannulated screws for fixation of the subtalar joints were determined. Then, the talar prosthesis was located after drilling was performed in accordance with the test model. The 3D-printed structure was made porous on the sides of the subtalar joints. Full-range 3D printing was completed using the Arcam EBM Q10 system (United States). The specific casting process included mirror polishing of the tibial articular surface, polishing and trimming of the talus matrix, ultrasonic cleaning, fine cleaning, and drying. Finally, the articular surfaces of the talus and matrix were assembled and reviewed in the purification workshop and were packaged after sterilization. Titanium alloy powder was used as the talar structure material, and cobalt-chromium-molybdenum alloy powder was used as the articular facet material. The high-precision dovetail slot design process and screw channel fixation of the prosthetic tibiotalar articular facet were completed after assembly. The articular facet was subjected to bright polishing. Co-Cr-Mo material, which is more resistant to the friction between the talus and tibia, was used on the tibia side (Figure 3A and B). The articular surface of that subtalar joint was printed with micropores to increase surface roughness and to take advantage of the property of TI6-Al-4V alloy to promote bone growth (Figure 3C).

The innovative surgical technique was performed with the patient in a supine position under combined spinal epidural analgesia. A long incision in the middle of the ankle was made, and subcutaneous tissue was cut layer by layer, exposing the neck of the talus bone. The denatured and necrotic talus was broken with a narrow osteotome, and the talus was completely removed. After the hyperplastic synovium and cystic degeneration tissue were cleared, a pathological culture was obtained. The denatured subtalar articular cartilage was completely excised with a wide osteotome and remained fresh until the subchondral bone tissue was exposed. The articular cavity and bone fragments were washed with physiological saline and a large amount of iodophor. Then, the ankle joint was flexed. The 3D printed talus prosthesis was

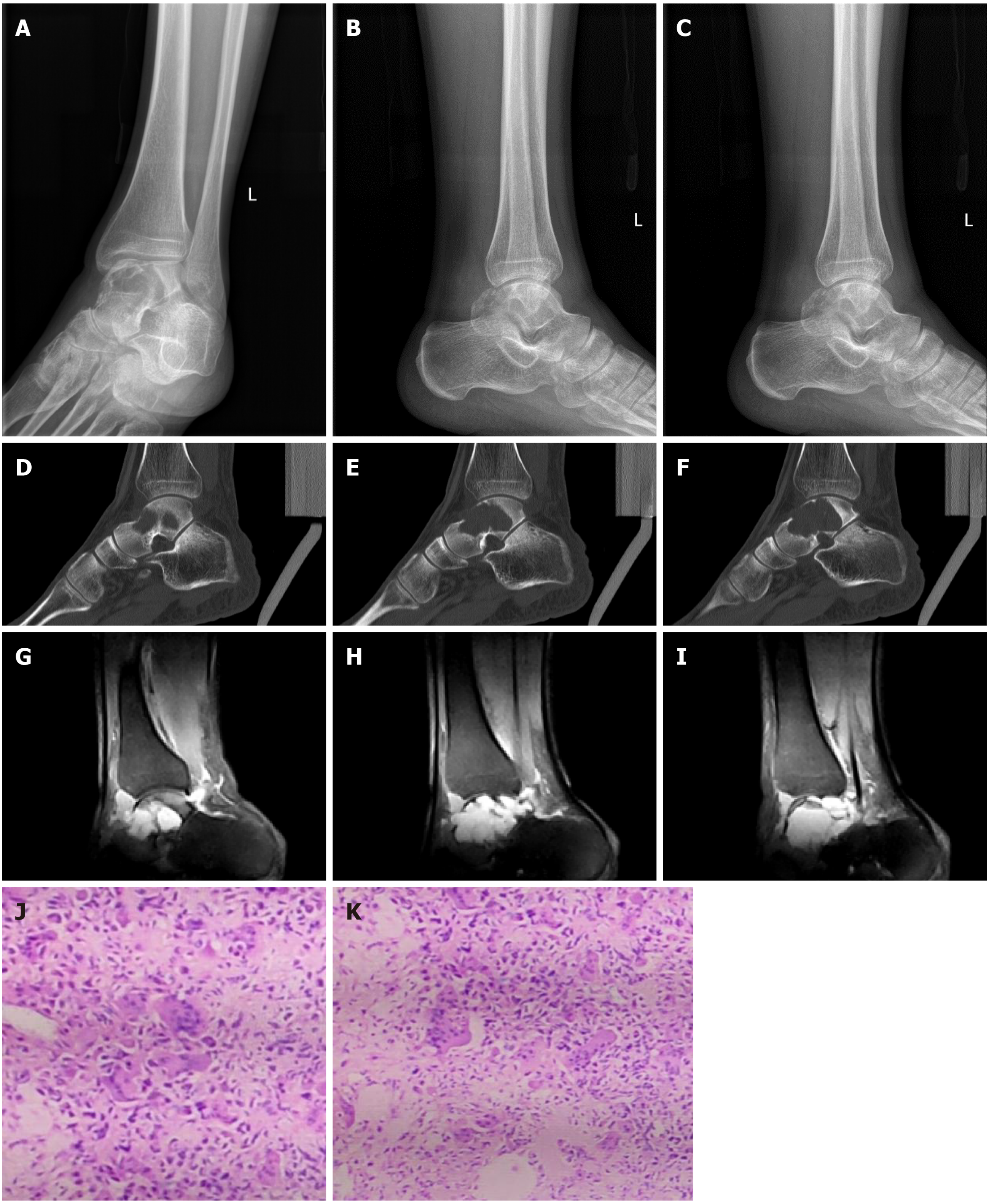

Figure 1 Preoperative imaging results. A-C: Preoperative radiographs; D-F: Preoperative computerized tomography images; G-I: Preoperative magnetic resonance imaging suggested a giant cell tumor of the talus; J and K: Pathological images.

implanted into the articular cavity until it completely fit the subtalar joint, and the ankle joint was moved again so that it adapted completely to the ankle joint. The prosthesis was fixed to the calcaneus with two titanium screws. Under fluoroscopy, the prosthesis was observed to be in a good position, and the ankle joint was able to move. A drainage tube was inserted. Then, the skin and subcutaneous tissue were sutured layer by layer (Figure 4). Imaging data on the day after operation are presented in Figure 5.

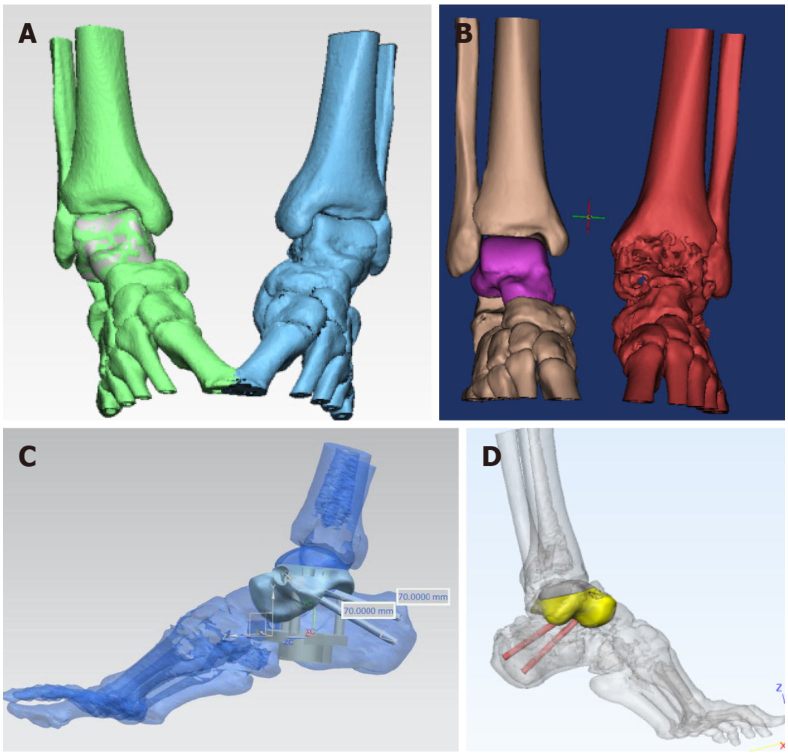

Figure 2 Casting process for the three-dimensional printed personalized prosthesis. Through three-dimensional (3D) computerized tomography and magnetic resonance imaging post-processing technology, the tomographic image processing and segmentation were carried out to obtain the complete data of the patient area, and the image technology and data registration technology were used to reconstruct and match the intact three-dimensional original data of the patient side. The serious data defects of necrotic talus on the affected side were repaired by reverse repair technology, the original data of defect-free talus reconstruction were obtained, and the tibial distance, distance boat, and subtalar joint surface were analyzed and smoothed to complete the personalized three-dimensional reconstruction of talus prosthesis. Reconstruction and manufacture of complex anatomical structure of soft and hard tissues were performed by three-dimensional printing technology. A: Complete data of the affected area were acquired by computerized tomography image processing and segmentation using 3D computerized tomography postprocessing technology; B: The 3D raw data of the affected side were obtained by reconstruction and matching performed with mirror and data registration technology; C: The data of the tibiotalar and subtalar articular facets were analyzed and processed; D: Accurate 3D reconstruction of the talar prosthesis was completed.

Figure 3 The three-dimensional printed personalized prosthesis with biological function A: Whole view of three dimensional (3D) print personalized talus prostheses; B: Articular side of 3D print personalized talus prosthesis tibialis; C: Subtalar joint side of 3D print personalized talus prosthesis. On the tibia side, cocrmo material was used, the subtalar joint and subtalar joint surface were treated with micropores, and hydroxyapatite coating and nano-treatment were used to promote bone growth.

OUTCOME AND FOLLOW-UP

The patient was able to move his ankle with mild pain during sport activities 6 mo postoperatively and nearly full range of motion and grasp force of the ankle was achieved in 12 mo (Table 1). Weakness and numbness of the ankle were not observed (Figures 6 and 7).

Degenerative arthritis and prosthetic dislocation were not detected on plain radiographs. The 3D printing talus prosthesis was placed in the original anatomic position. The VAS scores and AOFAS scores were 1 point and 89 points, respectively.

Table 1 Comparison of the wrist range of motion and grasp force

	The affected ankle	The intact ankle
Dorsal extension	10	18
Palmar flexion	15	22

Figure 4 Surgical technique. A: Preoperative anterior median approach marking; B: Resection of the talus and implantation of the prosthesis; C: Skin incision along anterior median approach.

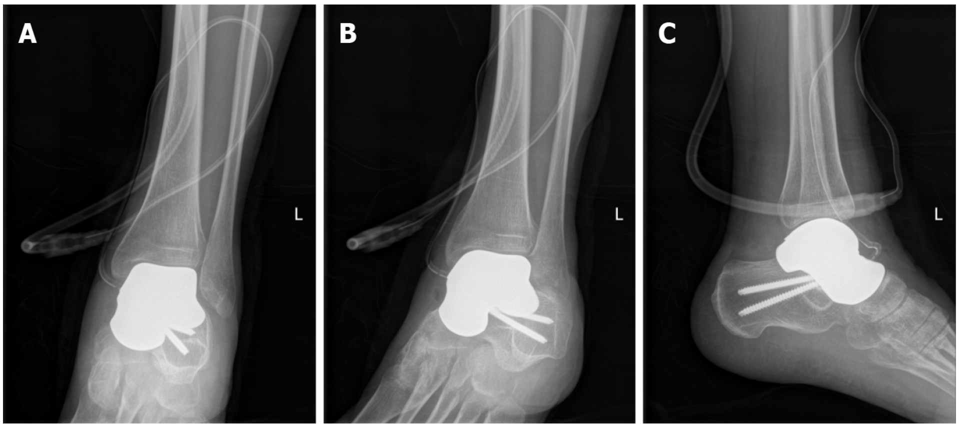

Figure 5 Radiographs on the day after surgery. A and B: X-ray films of the ankle joint immediately after operation (front); C: X-ray film of the ankle joint immediately after operation (flank).

In addition, we measured the talar arc length, talar height, talar width, tibial alignment angle, talar tilt angle, Bohler's angle, and Meary's angle preoperatively and postoperatively. We found that talar height and Meary's angle were significantly changed (Table 2).

DISCUSSION

GCTs are benign tumors that have a tendency to exhibit local aggressiveness and have a high risk of recurrence. The most common sites are the distal end of the femur, upper end of the tibia, and lower end of the radius[15]. GCTs of the talus are rare in clinical practice and are challenging to treat. In the surgical management of malignancy of the talus, it is difficult to achieve both adequate surgical margins and functional reconstruction.

One treatment option is partial talus resection. Since the talus is located at the ankle, most conventional surgical approaches cannot completely expose the talus, and there is an area occluded in the field of vision. Moreover, GCTs of the talus often invade the whole talus, so it is difficult to clear them completely. Therefore, curettage for GCTs of the talus is associated with a high recurrence rate. The local recurrence rate can be reduced by additionally performing procedures, such as carbolic acid smearing and bone cement filling. However, the recurrence rate is still as high as 30%[8].

Table 2 Imaging measurement data before and after operation

	Before surgery	At the last visit
Talar arc length (mm)	60	58
Talar height (mm)	30	36
Talar width (mm)	43	45
Tibial alignment angle (°)	84	86
Talar tilt angle (°)	2	3
Bohler's angle (°)	43	45
Meary's angle (°)	11	9

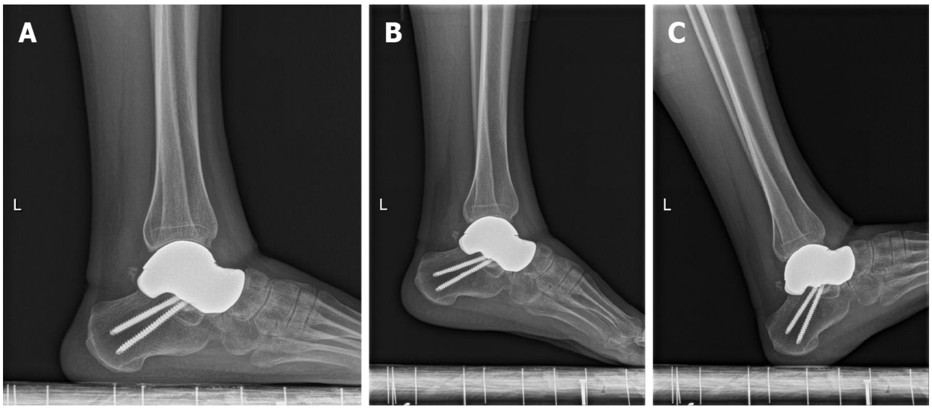

Figure 6 Postoperative radiographs (12 mo). A: Lateral X-ray film of the ankle joint at 12 mo after operation; B: Extreme plantar flexion position; C: Extreme dorsiflexion position. The talus prosthesis was in place without displacement or subsidence, the surrounding bone was in good condition, and there was no instability or fracture around the prosthesis.

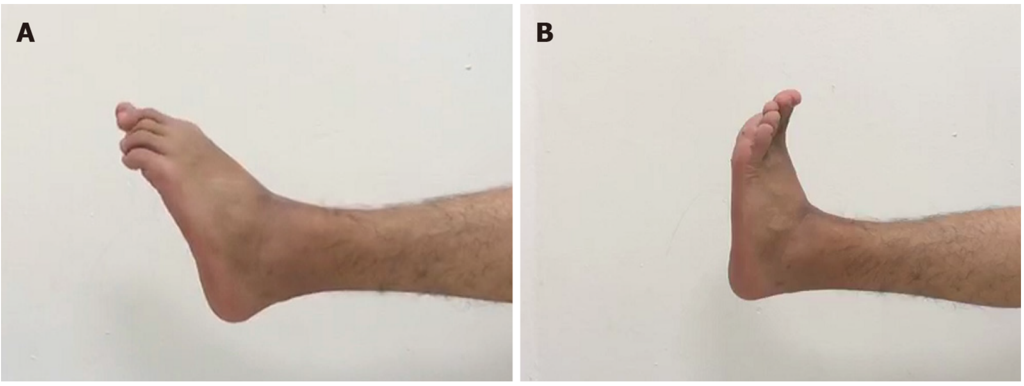

Figure 7 Postoperative range of motion (12 mo). A: Extreme plantar flexion position; B: Extreme dorsiflexion position

Another widely accepted option is arthrodesis. Dennison *et al*[9] reported a case of GCT of the talus treated by talus resection and tibial calcaneal fusion. There was no recurrence at 18 mo after the operation. However, the postoperative recovery process was long[16], and there was limited ankle function[17] due to arthrodesis.

Inspired by the recent success in treating stage IIIc Kienböck's disease with a 3D printed lunar prosthesis[18], our team successfully used 3D printing to generate a personalized talus prosthesis with biological function replacement technology to treat seven patients with severe talus collapse and necrosis. Moreover, the tumor in this case is too large to be treated using conventional treatment; therefore, we attempted to treat this case by 3D printing technology to completely solve the problem of easy recurrence of giant cell tumor of bone and restore the patient's walking function close to normal. In fact, most medical applications of 3D printing technology have been prostheses[19]. Three-dimensional printing technology serves as an innovative method of fabricating the complex shape and structure of the talus[20,21]. The talus imaging data were segmented and reconstructed in 3D based on CT and MRI data. Three-dimensional printing technology enables surgeons to design and manufacture anatomically matched implants for use in surgical operations[22]. In our study, the complete talus model was matched by comparing the healthy side and the affected side using mirror symmetry and multidimensional computer reconstruction techniques.

The operation was performed by an experienced foot and ankle surgeon who had performed more than 10000 operations. Our team also used 3D printed personalized prostheses with biological function for other cases who were suffering from irreversible ankle osteoarthritis. All of the patients exhibited good clinical effects as demonstrated by significant changes in AOFAS and VAS scores. The trend of imaging manifestation is similar to that noted in the study performed by Tracey *et al*[23] in 2019. Among these patients, the fixation methods adopted include subtalar joint and talonavicularis joint fixation and subtalar joint fixation only. In this case study, the fixation method used was subtalar joint fixation only. All prostheses are made of Ti-6Al-4V alloy. Given its superior biological properties (bone ingrowth characteristics), obvious bone ingrowth is observed in the postoperative imaging follow-up process both in the cases where the subtalar joint and the talonavicularis joint were previously fixed, and in this case where only the subtalar joint was fixed. Although the operation sacrificed the range of motion of the subtalar joint and the talonavicularis joint, the range of motion of the ankle joint was preserved to the greatest extent, and all patients were able to return to normal life.

Regarding talus prostheses for replacement operations, the first reported talus prosthesis replacement was completed and published by Harnroongroj and Vanadurongwan[24] in 1997. All of the eight patients whom they treated exhibited good prognoses within 11-15 years. However, in their subsequent studies, the long-term complication that occurred was loosening of the neck region of the talus prosthesis[25], which is related to the design of the first-generation prosthesis. The first research report on the second-generation talus prosthesis was completed by Taniguchi *et al*[26]. In total, 12 patients were treated with the second-generation prosthesis, and the results showed good clinical efficacy within 7 years[26]. To date, there have been some studies on the third generation of talus prostheses, and all of them have demonstrated good therapeutic effects[27-29] within a short-term follow-up period. Other case reports have also confirmed the clinical efficacy of the third-generation talus prostheses, and third-generation talus prostheses[30,31] are recommended.

Unlike other prostheses, this prosthesis has the following advantages: (1) All parts of the 3D printed structure are designed according to the functional anatomy of talus and Wolff's Law, which solves the problem regarding the anatomical and biomechanical applicability of prosthesis; (2) This prosthesis overcomes the technical difficulties in the composite technology of friction interface and surface coating; enhances the stiffness, toughness, and fatigue resistance; and realizes the required structural composition and high performance; (3) The surface of artificial tibia is made of cobalt-chromium-molybdenum alloy, which has effective rigidity similar to that of tibial plateau and shows enhanced performance for tibial prosthesis, including low friction and high wear and degradation resistance, which maximizes the recovery of joint motion function and the service life of the prosthesis. The lower surface material is composed of Ti6Al4V, which can promote bone growth and increase stability. Although Ti-6Al-4V has been confirmed to release toxic ions, such as aluminum and vanadium, it may lead to long-term health problems, such as Alzheimer's disease, neuropathy, and osteoporosis. However, considering the biocompatibility of the implant (excellent bone growth performance of Ti-6Al-4V) and minimal concentration of ions released in the foot, we still choose this alloy to replace pure titanium alloy[32]; and (4) Given the bone ingrowth characteristics of Ti6-Al4-V alloy, the subtalar joint surface of the prosthesis was subject to a micropore-generating treatment and hydroxyapatite surface coating (50 μm) to promote the rapid fusion and growth of bone at the interface of prosthesis. Therefore, we believe that 3D printed, personalized talar prostheses with biological function are an advanced and reliable treatment option for GCTs of the talus and other tumors of the talus.

Furthermore, this study has some limitations. First, the follow-up time was too short. The prognoses in terms of the prosthesis's clinical function should be assessed over a longer period. We will continue to follow-up this patient for a long-term follow-up study. Second, the 3D printed, personalized talus prosthesis replacement surgery was performed relatively late. To date, only seven patients in the world have undergone our surgery. The team is also performing relevant research on the biomechanical mechanism of prosthesis replacement. In summary, studies on many topics need to be conducted in the future.

CONCLUSION

We demonstrated that 3D printing of a talar prosthesis is a beneficial option for GCT of the talus.

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