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Case Report

Treatment of Talus Osteosarcoma with 3D-Printed Talar Prosthesis: A Case Report

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Abstract

Case: A 31-year-old male was diagnosed with a osteoblastic osteosarcoma of talus. Limb-salvage surgery for the talar osteosarcoma by replacing the intact talus with a 3D-printed talar prosthesis made from medical-grade titanium was planned. The prosthesis had three tunnels to simulate the ligaments around the talus. At the last follow-up, The functional and clinical outcomes were excellent. Our patient restored 93% functional score of MSTS and 93 points of TESS score, and there was no local recurrence or distant metastasis.

Conclusion: A 3D-printed talar prosthesis showed excellent functional and clinical outcomes for a osteosarcoma of talus. A 3-D printed implant is a feasible option for patients with osteosarcoma of the foot.

Keywords: 3D printing; Salvage; Talar prosthesis; Osteosarcoma; Talus

Introduction

Osteosarcoma, a malignant bone tumor, often affects the metaphyses of long bones [1,2]. Approximately 0.5% to 1.3% of all osteosarcomas occur in the bones of the foot, and 25% of them arise from the talus [1-4]. Of 20 talar osteosarcoma patients reported in the literature, 15 were managed with below-the-knee amputation5, and 4 were treated with limb-sparing surgery [2,5-7]. The limb-sparing surgery with traditional reconstruction techniques such as arthrodesis, allografting, or endoprosthetic arthroplasty needs to remove most of surrounding soft tissues as such little soft tissues could be used to cover the bone, and the recovery requires a complete bone union [2,8]. In this report, we present an alternative limb-sparing surgery for the talar osteosarcoma by replacing the intact talus with a 3D-printed talar prosthesis.

Case Report

A 31-year-old male complained of two-year history of pain and presented a swelled left ankle after falling. Physical examination revealed that the left ankle was slightly swelled with pain when moving and weight-bearing. All results of laboratory examinations were normal. The plain radiographs of the left ankle demonstrated a collapsed left talus with patchy high intensity shadow and narrowing of the left tibial astragaloid joint space (Figures 1A& B). Computed Tomography (CT) scanning of the left ankle showed a collapsed left talus with discontinuous cortex, a large patch of low intensity region with patchy high intensity shadow, and a narrow space of the left tibial astragaloid joint as shown in Figure 1C. Magnetic Resonance Imaging (MRI) revealed a wide destruction of the left talus with collapse, and soft-tissue in the left talus, which grew toward the anterior aspect of the left ankle. The signal intensity was low on T1-weighted images, high on T2-weighted images with a patch of higher signal intensity, and high on fat-suppressed images with moderate contrast enhancement (Figures 1D&E). Chest Computed Tomography (CT) revealed no definite focal lesions. The patient preferred a limb-salvage procedure to below-the-knee amputation. Based on his willing, we decided to perform a limb-salvage surgery by replacing a 3D-printed talar prosthesis.

Because of the collapse of the left talus, it was impossible to get intact images of the left ankle. Thus, CT images of the right ankle was taken and used to make a simulated left ankle *via* the mirror technique. The talus model was exported in Stereolithography (STL) format from the software (MIMICS, version 15, Belgium) and sent to AK Medical (Beijing) for processing and implant creation. The prosthesis was designed to have three tunnels to simulate the ligaments around the talus (Figure 2). The 3D talar prosthesis was printed by a powder-

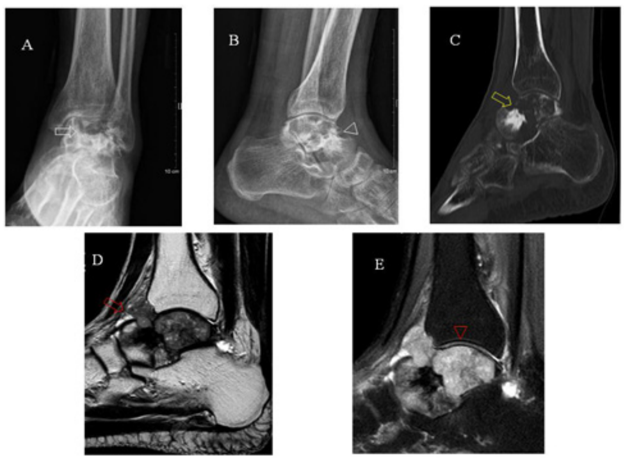

Figure 1: Anteroposterior (A) and lateral (B) radiographs of the left ankle demonstrated collapse of the left talus with patchy high intensity shadow and narrowing of the left tibial astragaloid joint space (white arrow and arrowhead). Computed Tomography (CT) of the left ankle showed collapse of the left talus with discontinuous cortex, large patch of low intensity region with patchy high intensity shadow, and narrowing of the left tibial astragaloid joint space (C, yellow arrow). Magnetic Resonance Imaging (MRI) revealed wide destruction of the left talus with collapse, soft-tissue in the left talus, which grew toward the anterior aspect of the left ankle. The signal intensity was low on T1-weighted images (D, red arrow), high on fat-suppressed images with moderate contrast enhancement (E, red arrowhead).

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Figure 2: The prosthesis had three tunnels (red arrow, green arrow, blue arrow) to simulate the ligaments around the talus.

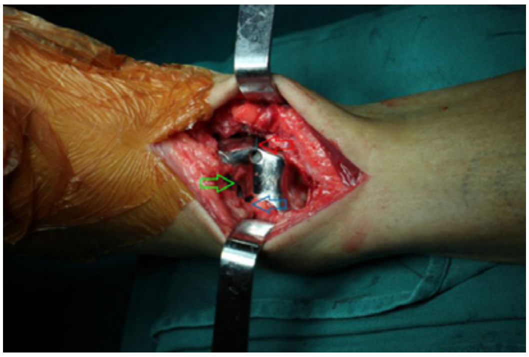

Figure 3: The 3D-Printed total talar prosthesis was inserted. The anterior tibiotalar part of deltoid ligament (red arrow), anterior talofibular ligament (green arrow) and talonavicular ligament (blue arrow) were sutured to the prosthesis by nonabsorbable Polyester sutures with proper tension.

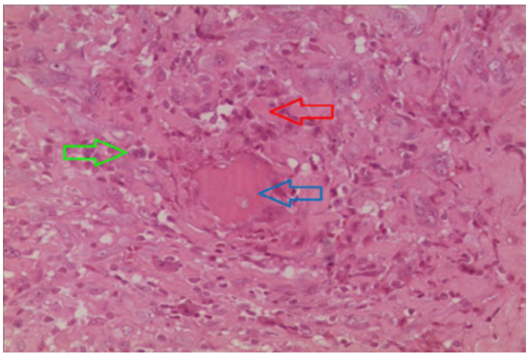

Figure 4: Histopathological examination revealed tumor-forming osteoid production (red arrow) and well-formed trabeculae of the woven bone (blue arrow) rimmed with osteoblasts associated with moderate atypia (green arrow). The host bone is embedded in an osteogenetic background cell proliferation. This infiltrative appearance confirms the osteoblastic osteosarcoma diagnosis (hematoxylin and eosin; original magnification, ×400).

based electron beam melting 3D printer (Arcam Q10plus; Arcam EBM, Sweden) with medical-grade titanium (Ti6Al4V ELI, extra low interstitial, ASTM F136).

The operation begun with a straight skin incision similar to the approach for a total ankle replacement over the anterior ankle [9]. The anterior capsule of the tibiotalar joint was incised, and the talus was exposed. The ligaments and neurovascular structures around the talus were preserved and the left talus was excised. Histopathological examination revealed tumor-forming osteoid production and well-formed trabeculae of the woven bone rimmed with osteoblasts associated with moderate atypia. The host bone was embedded in an osteogenetic background cell proliferation. This infiltrative appearance confirmed the osteoblastic osteosarcoma diagnosis (Figure 4). There were no pathological changes seen in the soft tissues sent for evaluation. Immunohistochemistry examination of the biopsy showed that SMA, Ki-67 and CD68 were positive, desmin and myogenin were negative (data not shown). According to the American Joint Committee on Cancer (AJCC) staging system, this case was considered as T1N0M0G3, stage IIA [10], and the surgical stage was G2T2M0, stage IIB, according to the Musculoskeletal Tumor Society (MSTS) Surgical staging system [11]. After removing the intact talus, the 3D-Printed total talar prosthesis was inserted as shown in Figure 3. The anterior tibiotalar part of deltoid ligament, anterior talofibular ligament and talonavicular ligament were sutured to the prosthesis by nonabsorbable Polyester sutures with proper tension. Finally, dorsiflexion and plantarflexion as well as inversion and eversion of the ankle were performed to assess articulations [9]. Multiple planes of movement of the ankle also revealed the flexibility at the midfoot [9]. The fluoroscopy before closure demonstrated that the implant was perfectly fitted with the adjacent joints (Figure 5A).

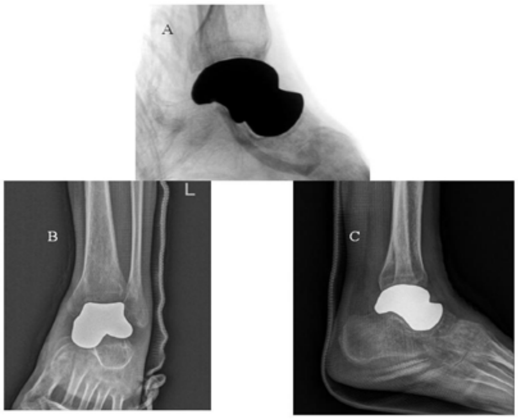

Figure 5: The fluoroscopy before closure demonstrated that the implant was perfectly fitted with the adjacent joints (A). Anteroposterior (B) and lateral (C) postoperative plain radiographs showed that the prosthesis was perfectly fitted with the adjacent joints.

Postoperative plain radiographs showed that the prosthesis was perfectly fitted with the adjacent joints (Figures 5B &C). After six courses of postoperative chemotherapy with cisplatin, doxorubicin, ifosfamade and high-dose methotrexate, there was no local recurrence or distant metastasis [2,7,12,13]. The result of the functional rating system of the Musculoskeletal Tumor Society (MSTS) [14] was 93%, and the score of the Toronto Extremity Salvage Score (TESS) [15] was 93. The patient continued to be disease-free Thirteen months after surgery and was able to walk normally without support. The patient consented to the publication of this case report.

Discussion

Patients with talar osteosarcoma were treated with either amputation [5] or limb-sparing surgery [2,5,7,16]. Below-the-knee amputation is usually considered as the standard treatment for osteosarcoma of foot because it is associated with a low rate of complications and good local tumor control [2], but the patients need artificial legs after surgery. An alternative of limb-sparing surgery for management of foot osteosarcomas is acceptable because of the better patient satisfaction and the excellent walking ability without an artificial leg [2]. However, the limb-sparing surgery with traditional reconstruction techniques such as arthrodesis, allografting, or endoprosthetic arthroplasty is difficult to achieve the oncologic margin of resection and needs to remove most of surrounding soft tissues as such little soft tissues could be used to cover the bone; the surgery causes a high rate of complications, and the recovery requires a complete bone union [2, 8]. Katagiri et al. treated the first case of talar osteosarcoma by arthrodesis with autogenous bone graft and intramedullary nailing, [2] and Wang CW et al. reported a case treated by limb-sparing surgery with allogenic bone graft, ankle arthrodesis, and distal tibiofibular syndesmosis fixation [7]. The functional TESS[14] scores of the two patients were 892 and 89.5 points, respectively, and the MSTS[13] scores were 90% and 86%, respectively. Both of the patients underwent complete preoperative chemotherapy and postoperative chemotherapy [2,7] and had no evidence of disease of lung metastatic and local relapse after surgery [2,7]. The mean TESS scores of the patients with osteosarcoma treated with amputation or limb-sparing surgery were found to be 75.6 and 85.7, respectively [17]. Here, we report a case of talar osteosarcoma treated with a 3-D printed with talar prosthesis, and demonstrate that prosthesis was perfectly fitted with the adjacent joints, and pre-operational designed holes were appropriate for suturing ligaments [18]. Our patient restored 93% functional score of MSTS and 93 points of TESS score, and the patient was very satisfied with the results of the surgery.

We found that the customized 3-D implant reconstructed the talus functionally and anatomically [9,18,19]. In addition, the talar prosthesis used in the surgery greatly reduced the surgical time, avoided intraoperative fluoroscopy, reconstructed ligaments around the talus, and resulted in an excellent walking ability without any support [9]. The technique does not have the issues such as little soft tissues coverage of the bone, a high rate of complication, or the requirement of complete bone union that the traditional reconstruction techniques has [2]. However, the limb-sparing surgery with a 3-D printed implant for osteosarcoma of foot also has limitations. First, it is difficult to achieve a sound oncologic margin of resection, so that it will not have good local tumor control as compared to below-the-knee amputation. Second, high cost of the prosthesis may limit the wide use [9], and third, the metal implant may hinder for identifying local relapse of the osteosarcoma by CT scanning or MRI [18,20-25].

Conclusion

We report an alternative limb-sparing surgery for management of osteosarcoma of talus by replacement of a 3-D printed talar prosthesis. The functional and clinical outcomes of 3-D printed implant were more favorable than those of autografts, allografts and amputation. The technique had less complications and could overcome some limitations of autografts, allografts and amputation. Our experience indicates that the 3-D printed implant is a feasible option for patients with osteosarcoma of the foot.

Acknowledgement

The 3D printed talar prosthesis was made in AK Medical Co., Ltd.

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