

Intercalary reconstruction with successful joint preservation by uncemented 3D-printed endoprosthesis following tumor resection in distal radius: A case report

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Abstract. The distal radius is an extremely rare site for epithelioid hemangioendothelioma. Joint preservation to maintain a good wrist joint function is rarely reported. The present study described a case of joint preservation surgery in the distal radius with an uncemented 3D-printed endoprosthesis and evaluated the endoprosthesis design and short-term outcomes. A 14-year-old boy was diagnosed with epithelioid hemangioendothelioma in radius. Due to the extensive defect and the excessively short length of the residual distal radius after resection, a custom-made 3D-printed custom-made endoprosthesis was designed and fabricated to reconstruct the defect, with the preservation of the wrist joint. The patient had a favorable wrist function and no endoprosthesis-related complications were observed. The present study presented a case of en bloc tumor resection with joint preservation of the wrist and reconstruction using a 3D-printed endoprosthesis. Satisfactory postoperative function and low complication rates were found.

Introduction

Epithelioid hemangioendothelioma (EHE), also known as low-grade anaplastic angiosarcoma, cellular hemangioma,

histiocytoid hemangioma and angioendothelioma, is a rare, well-differentiated but locally aggressive endothelial tumor (1). EHE occurs commonly in the calvarium, spine, femur, tibia and feet, but is rare in the distal radius (1,2). The therapeutic options vary owing to the wide spectrum of the tumor behavior (1-3). For benign-appearing lesions, curettage or marginal resection can be sufficient, while for more aggressive tumors, wide resection or even amputation is necessary.

With the progress in imaging and multimodality therapy, en bloc resection has been widely accepted in treating intermediate and malignant tumors involving the distal radius. In the literature, the distal radius was totally sacrificed and the patient's upper limb function was greatly impaired even if properly reconstructed (4). Resection with joint preservation can salvage the majority of wrist function but, as a challenging procedure, it can also lead to an ultra-critical sized bone defect which is technically demanding to reconstruct (5,6). Previous reports regarding wrist joint sparing surgery are rare (7,8). The present study reported a case of wrist preservation surgery using a 3D-printed custom-made porous endoprosthesis after en bloc resection of the EHE in the distal radius. 3D-printed custom-made porous endoprosthesis is a novel uncemented implant that has demonstrated great clinical success in the reconstruction of extensive bone defects (9-11). Due to its optimized-fit shape and porous surface, it was considered a feasible alternative for the massive defect of the distal radius to preserve the intact wrist.

Case presentation

A 14-year-old boy was referred to the Department of Orthopedics, West China Hospital, Sichuan University in January 2019, complaining of dull pain accompanied with a mass in the right forearm for a month. The radiographs showed expansive, osteolytic and poorly demarcated lesions in the middle and distal part of the right radius (Fig. 1A). Magnetic resonance imaging (MRI) showed that the tumor did not involve the growth plate (Fig. 1B and C). Tc-99 bone scans revealed increased uptake in the forearm (Fig. 1D). An open biopsy was performed. Immunohistochemistry was carried out using the EnVision two-step method. Pathological tissues were fixed using 4% paraformaldehyde solution for 8 h at room

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Abbreviations: EHE, epithelioid hemangioendothelioma; MRI, magnetic resonance imaging; CT, computerized tomography; ROM, range of motion; MSTTS-93, the 1993 version of the Musculoskeletal Tumor Society; DASH, the disabilities of the arm, shoulder and hand

Key words: distal radius, joint preservation, 3D-printed endoprosthesis, case report

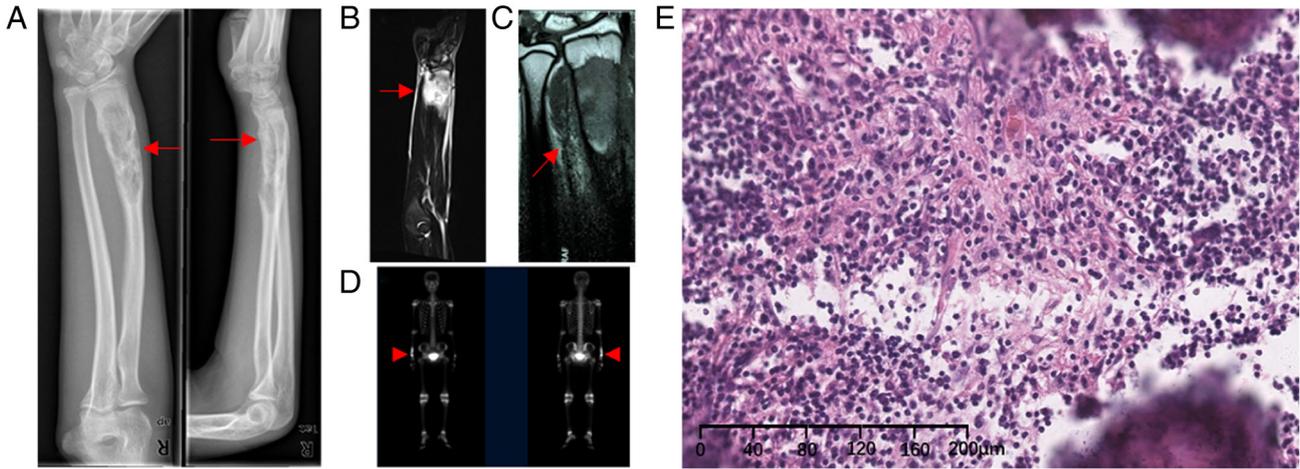


Figure 1. Preoperative imaging and postoperative pathology. (A) Radiography: Expandable osteolytic lesion was detected in the middle and distal part of the right radius where the arrow points. (B and C) MRI: A huge extraskelatal tumor surrounded the radius where the arrow points but growth plate was not affected. (D) Bone scan: Increased uptake was found in the forearm where the arrow points. (E) Cell morphology image of the tumor.

temperature. The tissue blocks were embedded in paraffin wax and the wax sections were 4 μm thick and blocked with PBS containing 5% goat serum for 30 min at room temperature. Primary antibodies: CD31 (cat. no. ZM-0044), CD34 (cat. no. ZM-0046), ERG (cat. no. ZM-0103), D2-40 (cat. no. ZM-0469), CAMTA1 (cat. no. ZA-0535), EMA (cat. no. ZM-0095) and WT-1 (cat. no. ZM-0269) were purchased from OriGene Technologies, Inc. CR (cat. no. MAB-0716) and S-100 (cat. no. RAB-0150) were purchased from Fuzhou Maixin Biotech Co., Ltd. The primary antibodies were diluted according to the instructions provided by the supplier and incubated overnight at 4°C. The polymorphic enzyme complexes (secondary antibody) were incubated for 30 min at room temperature. Under the microscope the bone tissue was infiltrated by small cells in the form of patches, some of which had nuclear sulci with scattered heterogeneous mononuclear and multinucleated giant cells with hemorrhagic necrosis. A number of hemangioma-like proliferations were observed. The tumor cells were epithelioid or long spindle-shaped, with obvious heterogeneity. The nuclei varied in size and morphology; some were densely stained and distorted and some were vesicular. Pathological results [immunohistochemistry: CD31(+); CD34(+); ERG(+); D2-40(+); CAMTA1(+); EMA(-); S100(-); WT-1(-); CR(-)] verified the diagnosis of EHE (Fig. 1E). Following multiple disciplinary team discussions and the wish of the patient, segmental resection with 3D-printed custom-made porous endoprosthesis reconstruction surgery was performed in February 2019.

Endoprosthesis design and fabrication. The clinical team designed the endoprosthesis which was fabricated by Beijing Chunlizhengda Medical Instruments Co., Ltd. The main components of the custom-made device were shaft and stem. For the first step, computerized tomography (CT) data was used to build virtual 3D radius models in Mimics V20.0 software (Materialise). The image fusion technique integrated MRI data to build a virtual tumor model (Fig. 2A). Thereafter, the osteotomy plane was modified in accordance with the surgical approach and tumor-free bone resection margin. Subsequently, a preliminary

endoprosthesis was generated and modified to a more streamlined shape. To ensure satisfactory fitting with the radius, the shape of the endoprosthesis was optimized via computer simulation. Meanwhile, a longish length of the implant over defect was produced to create a strain. Next, the orientation of the screws was designed after considering the surgical approach and the 3D space anatomical distribution to obtain a convenient and durable fixation. Then four straight pores were designed for the fixation using nonabsorbable suture (Ethibond size 2; Johnson & Johnson, Ltd.) to the stump of the distal radius in case the primary stability provided by screws was unsatisfactory. The endoprosthesis was composed of an inside solid shaft to ensure mechanical strength and outside porous structures with a pore size of 600 μm and porosity of 70%. The medullary cavity for the press-fit of the stem was measured with a diameter of 6 mm in the distal and 4 mm in the proximal. The stem part was designed as a tapered shape. The diameter was 5.5 mm in the base and 3.5 mm in the tip.

The endoprosthesis was fabricated using the electron beam melting technique (Arcam Q10plus; GE Additive). Thereafter, the stem was coated with titanium and hydroxyapatite powder (Fig. 2B).

Surgical techniques. The senior surgeon (Chongqi Tu) performed the surgery. The patient was placed in the supine position and a tourniquet was placed in the right upper arm before the surgery. A longitudinal dorsal approach was used to access the lesion. During the procedure, the insertion of the pronator quadratus, the partial origin of flexor pollicis longus and the intraosseous membrane were released from the radius. According to the preoperative plan, proximal osteotomy was performed in priority and then distal osteotomy. The epiphysis was preserved successfully (Fig. 3A and B). Thereafter, the operation area was soaked in 10% povidone-iodine and pulsatile lavage was performed with isotonic sodium chloride solution to possibly reduce the likelihood of wound infection. Using bone-holding clamps to stabilize the proximal stump, the radial medulla was reamed to press-fit the endoprosthesis stem. While inserting the stem, the radial crest referred to avoid implant rotation. After the stem was fixed appropriately

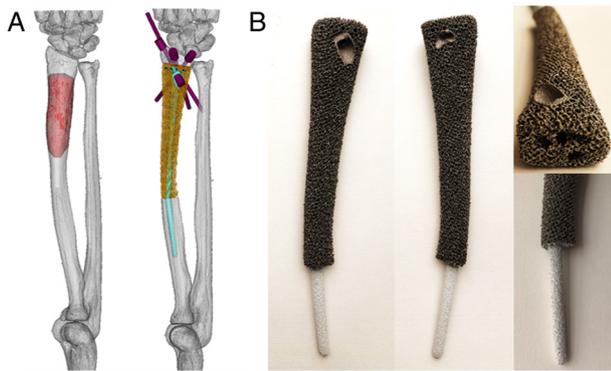


Figure 2. Design of 3D-printed prosthesis. (A) Reconstructed 3D models of the extent of the resection and the size of the distal radius defect. (B) Final product of 3D-printed prosthesis.

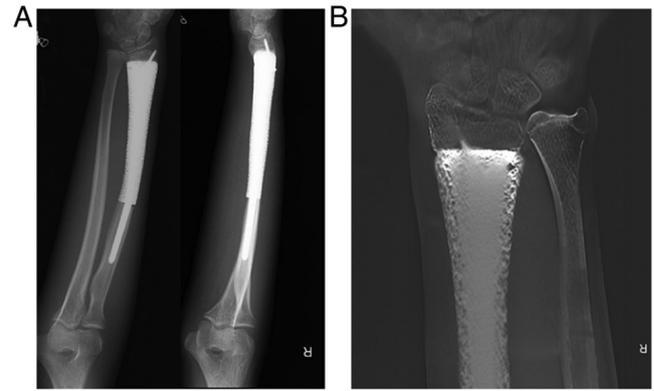


Figure 4. Postoperative imaging. (A) 24 month postsurgical anteroposterior and lateral radiographs. (B) Digital tomosynthesis showed the 3D prosthesis tightly integrated with the distal radius and partial bone substance could be observed to grow into the pores of the prosthesis.

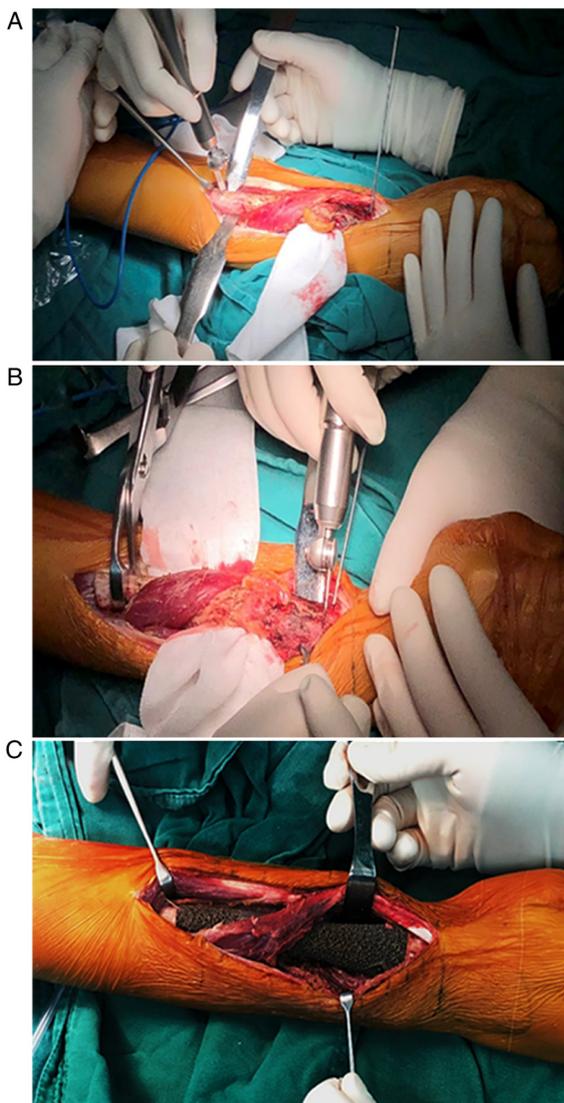


Figure 3. Intraoperative images indicating the resection of the middle and distal radius via a dorsal approach. (A) Osteotomy in the proximal side. (B) Osteotomy in the distal side. (C) Implanting the prosthesis was completed.

within the canal, the reduction of the distal end of the endoprosthesis proceeded. After confirming the right locations of

the proximal and distal parts of the endoprosthesis, we then inserted a 3.5-mm-diameter screw to enhance the primary stability (Fig. 3C). The released soft tissues were then reattached to the endoprosthesis. Pulsatile lavage with 10% povidone-iodine and another pulsatile lavage with isotonic sodium chloride solution was performed again, with a drain left in the right forearm thereafter. Resection margin was assessed and reported by the pathology department of our institution.

Postoperative management. The patient underwent plain radiography and digital tomosynthesis (Shimadzu metal artifact reduction technology) of the right forearm postoperatively (Fig. 4A and B). During the first two weeks postoperatively, the affected limb was immobilized in a volar resting brace. The grip exercise was carried out from the first day postoperatively and, at 2 weeks after the surgery, dorsiflexion and palmar flexion were encouraged without weight-bearing of the affected limb while immobilizing the wrist. At 4 weeks postoperatively, the brace was removed and the patient was allowed to rotate the forearm. The patient then gradually increased the intensity of training and initiate weight-bearing on the affected upper limb according to the patient's tolerance and recovery progress. The patient was followed up monthly in the first 3 months, then trimonthly thereafter. Functional outcome was assessed by range of motion (ROM), the 1993 version of the Musculoskeletal Tumor Society (MSTS-93) score (12) (range 0 to 30; a higher score is desirable), the disabilities of the arm, shoulder and hand (DASH) questionnaire (13) (range 0 to 100; a lower score is desirable) and Mayo wrist score (14) (range 0 to 100; a higher score is desirable). Osseointegration was evaluated by digital tomosynthesis.

Results

Three months after the surgery, functional recovery of the wrist was favorable. The patient showed a significantly improved ROM. At the last follow-up at 27 months, wrist joint function of the affected side was almost normal. The patient achieved ROM of the affected wrist with active dorsiflexion to 90°, palmar flexion to 80°. At the forearm, the patient had supination of 90°

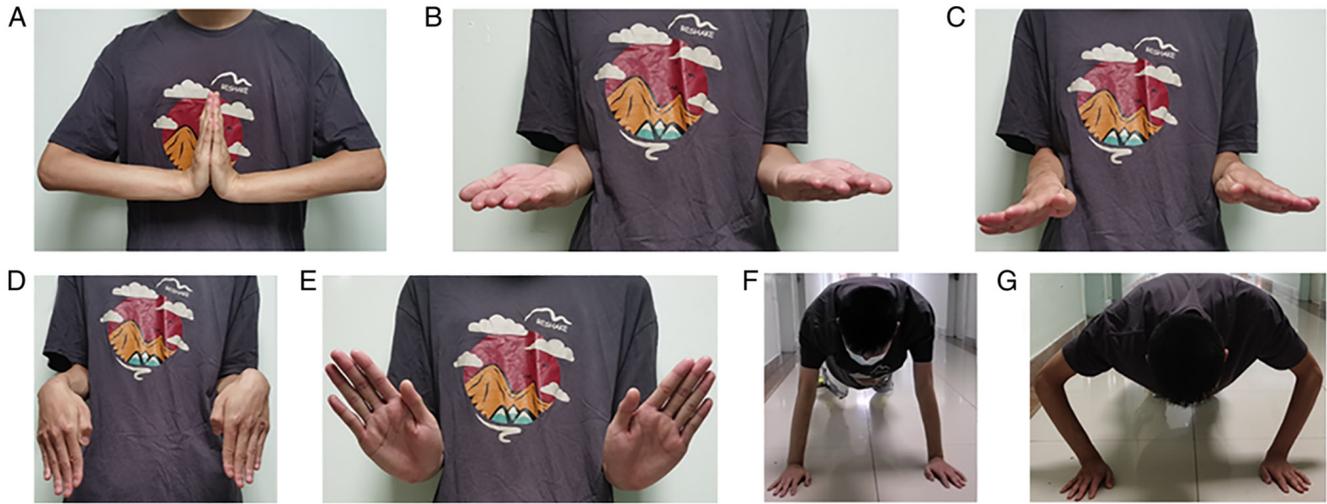


Figure 5. Functional outcomes of the affected arm 27 months after surgery. (A) Active dorsiflexion to 90°. (B) Supination to 90°. (C) Pronation to 85°. (D) Active palmar flexion to 80°. (E) Anterior view of active dorsiflexion. (F and G) The patient performing push-ups.

and pronation of 85° (Fig. 5A-E). The patient can engage in daily activities such as push-ups and arm wrestling with a healthy person (Fig. 5F and G). Functional scores were as follows: 95% in MSTS-93 score, 8 in DASH and 90 in Mayo wrist score. Digital tomosynthesis showed that the 3D-printed endoprosthesis was well integrated with both proximal and distal radius. During the follow-up, no complications were observed.

Discussion

Although the distal radius is not a rare site for benign bone tumors such as giant cell tumors of bone, it is an extremely uncommon skeletal site for EHE (1,15). En bloc resection with reconstruction is needed when a tumor does not appear benign (1). As a growing demand for limb salvage in favor of increasing postoperative function, a number of reconstructive techniques, including arthrodesis (16,17), autograft (18,19), allograft (20,21) and endoprosthesis (22,23), for massive defects following the en bloc resection of the tumor in the distal radius have been described.

Arthrodesis is still an option to salvage proper grip strength and reduce long-term complications. Total wrist arthrodesis has been demonstrated to restore 65% grip strength of the affected wrist comparing to the healthy side (24). However, ROM sacrifice of the wrist limits patients from performing daily activities and narrows its application. Therefore, partial wrist arthrodesis was introduced to result in less restriction of the ROM of the wrist, if the carpal bones can be retained (16). Nevertheless, the limited fusion contact area in partial wrist arthrodesis requires prolonged immobilization to achieve bone union. Generally, no matter total or partial joint arthrodesis, it is associated with a high incidence of complications, such as infection, fracture, delayed union and nonunion (17).

Wrist hemiarthroplasty can preserve improved postoperative wrist function by providing a more flexible wrist joint (25). Several materials can be used for wrist hemiarthroplasty, including allografts, autografts and endoprosthesis. The osteoarticular allograft offers reasonable wrist-specific matching and favorable functional outcomes without

donor-site morbidity (26). However, complications including fractures, nonunion and bony resorption, are not rare using this technique (26-28). The fibular head autografts have been demonstrated to have considerable anatomical similarity to the distal radius (18). However, such a demanding procedure is associated with wrist instability and inevitable drawbacks of autograft itself (19,22). In addition, the preservation of function is not as well as expected. Hemi-arthroplasty using endoprosthesis is a viable alternative to salvage proper wrist function (23,25). However, apart from conventional endoprosthesis-related complications including infection and aseptic loosening, subluxation is not uncommon with an incidence of 20% (22). The high subluxation rate of hemiarthroplasty can both impair upper limb function and cause a cosmetic problem; therefore, joint sparing surgeries without access to the articular cavity are considered to have the potential to avoid such disadvantages.

Resection with joint preservation is available when the growth plate is not involved (29). The joint-sparing resection in distal radius has only been reported in two case reports (7,8). Free fibular shaft and devitalized autograft were utilized to reconstruct the consequent intercalary bone defect. The postoperative function was deemed good with active dorsiflexion ranged from 85-90° and a palmar flexion ranged from 45-80° after a follow-up duration ranged 14-41 months (20). Higuchi *et al* (7), using devitalized autograft, present improved postoperative function with pronation and supination of 90°, an MSTS score of 100%, a DASH score of 12.5 and a Toronto Extremity Salvage Score (30) of 93.5. However, as aforementioned, free autograft has its limitations and devitalized autograft can fail or fracture if the tumors are osteolytic. Additionally, complete bone union time is reported to 6 and 9 months, prolonging the duration to achieve optimal function (14). In the present case report, a 3D-printed custom-made endoprosthesis was used to reconstruct the ultra-critical intercalary bone defect and, consequently, close to normal wrist function after a relatively short follow-up duration was observed.

Endoprosthetic reconstruction of ultra-critical bone defect is demanding and prone to mechanical complications (11,31,32).

Currently, no endoprosthesis has been applied to reconstruct an ultra-critical bone defect in radius. For the patient in the present study, durable structural reconstruction, optimized functional restoration and minimized complication incidence was considered during the endoprosthesis design. First, the distal interface was highly porous to promote the friction and prevent migration of the endoprosthesis; second, the increase in endoprosthesis length confirmed the soft tissue tension to stable the implant and reserved partial space for ulna growth; third, the screws went through the bone-implant interface of distal radius to augment the primary stability and interface compression for further osseointegration; fourth, the tapered stem with titanium and hydroxyapatite coating could be press-fitted into the proximal cavity to ensure fixation durability; finally, the inside solid structure contributed to the avoidance of endoprosthesis fracture. Apart from purposeful endoprosthesis design, an early and scheduled rehabilitation program is also important for the favorable functional recovery in this patient.

There were some limitations of the present study. First, the follow-up duration was only 27 months. Endoprosthetic-related complications might arise in a follow up for longer periods. Second, the one case involved was insufficient to verify the efficacy of this surgical techniques. More patients and a larger multi-institutional study are needed to compare this approach with other types of reconstruction. Third, finite element analysis should be performed to optimize endoprosthesis design.

The present study presented a case using 3D-printed endoprosthesis reconstruction for EHE of the distal radius. The patient achieved a favorable functional outcome and no complications were observed. The result suggested that the 3D-printed custom-made porous endoprosthesis may be a feasible option and benefit selected patients.

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Availability of data and materials

All data used or analyzed during this study are included in the published article.

Authors' contributions

SW, YL, YZ and CT were involved with the conception and design of the present study. SW, TG, YW and CZ were involved with the acquisition of subjects and data. LM, YL, YZ and CT were involved in the design of the endoprosthesis.

TG and CT were involved in the post-surgical evaluation of the patient. LM, YZ and CT were involved with the writing and revision of the manuscript. All authors have read and approved the manuscript. CT and YZ confirm the authenticity of all the raw data.

Ethics approval and consent to participate

The present study was approved by the West China Hospital's Ethics Committee, Sichuan University (Chengdu) and was permitted to be published. Written informed consent to have the case details and accompanying images published was obtained from the patient's parents.

Consent for publication

Written informed consent was obtained from the patient and his parents for publication of this case report and accompanying images.

Competing interests

The authors declare that they have no competing interests.

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