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ORIGINAL PAPER

Computer-aided designed, three dimensional-printed hemipelvic prosthesis for peri-acetabular malignant bone tumour

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Abstract
Background Prosthetic reconstruction may be a promising treatment for peri-acetabular malignant bone tumour; however, it is associated with a high complication rate. Therefore, prosthetic design and approach of prosthetic reconstruction after tumour resection warrant study.
Methods We retrospectively analyzed 11 patients with peri-acetabular malignant bone tumours treated by personalized 3D-printed hemipelvic prostheses after *en bloc* resection between 2015 and 2016. Pre-operative and post-operative pain at rest was assessed according to a 10-cm VAS score. The results of functional improvement were evaluated using the MSTS-93 score at the final follow-up. We also analyzed tumour recurrence, metastases, and complications associated with the reconstruction procedure.
Results All patients were observed for six to 24 months with an average follow-up of 15.5 months. One patient had occasional pain of the involved hip at the final follow-up (VAS, pre vs. post 8 months: 3 vs. 2). The mean MSTS-93 score was 19.2 (range, 13–25). Hip dislocation was detected in two patients, while delayed wound healing occurred in one patient. One patient with mesenchymal chondrosarcoma had a left iliac bone metastasis. Local tumour recurrence was not observed.
Conclusions Reconstruction of bony defect after tumour resection using personalized 3D-printed hemipelvic prostheses can obtain acceptable functional results without severe complications. Based on previous reports and our results, we believe that reconstruction arthroplasty using 3D-printed hemipelvic prostheses will provide a promising alternative for those patients with peri-acetabular malignant bone tumours.
Level of Evidence: Level IV, therapeutic study.

Keywords Peri-acetabular tumour · Computer-aided design · 3D-printed hemipelvic prosthesis · Reconstruction arthroplasty · Complications

Introduction

The treatment of peri-acetabular malignant bone tumours is a great challenge to orthopaedic oncologists. Historically, the common method of managing these cases was hindquarter amputation or external hemipelvectomy [1]. Currently, with advances in radiotherapeutic, chemotherapeutic, and surgical techniques, limb-salvage surgery has become an accepted treatment [2–9]. Various methods, including arthrodesis, hip transposition, osteoarticular allograft/autograft and prosthetic implantation, have been used for hemipelvic reconstruction after peri-acetabular tumour resection [5–17]. However, the optimal reconstruction method remains controversial because of the relatively high rate of associated complications [5, 6, 18–23]. Arthrodesis using various bone grafts produces a good stability and entails no risk of long-term complications. However, it is difficult to achieve a painless rigid bone union because of the small contact area, and the hip function is sacrificed [10, 23]. Hip transposition retains the function of

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the hip joint, but it is associated with severe leg-length discrepancy [5]. Theoretically, reconstruction using a massive allograft/autograft can obtain biological reconstruction; however, the high deep infection rate (15–55%) and fracture risk (9–20%) greatly limit this technique [6, 14, 18, 24].

Prosthetic reconstruction may be a promising treatment because of acceptable cosmesis, immediate stability, and the possibility of rapid recovery, as well as early weight-bearing activity [7–9, 12, 13, 15–17]. At present, computer-aided design techniques have greatly improved the anatomic matching of prostheses [8, 13]. However, the patient-specific designed prostheses with complex shape are sometimes difficult to produce because of the limitations of the traditional manufacturing technology. More importantly, long-term non-integration between implant and host bone may result in inevitable reconstruction failure. 3D printing technology, also called additive manufacturing or rapid prototyping, may provide a solution. It can fabricate anatomy-conforming prostheses of any shape with a porous metal surface allowing osseointegration at the bone-implant junctions [25–28]. Recently, 3D-printed prostheses have been used in hemipelvic reconstruction and have obtained good short-term functional results [29–31]. In this study, to evaluate the feasibility of the 3D-printed hemipelvic prosthesis in pelvic reconstruction, 11 patients with peri-acetabular malignant bone tumours were treated by *en bloc* resection and reconstruction using newly computer-aided designed, 3D-printed hemipelvic prostheses. The purpose of this study was to evaluate the short-term functional results, complications and oncological outcomes in patients who underwent this procedure.

Patients and methods

Clinical data

We performed this retrospective study after obtaining approval from the ethical committee. Eleven patients with peri-acetabular malignant bone tumours underwent *en bloc* resection of the tumours and reconstruction arthroplasty using personalized 3D-printed hemipelvic prostheses between 2015 and 2016. Our study enrolled six females and five males, with a mean overall age of 47 years (range, 21–63 years). The most common presenting complaints were pain in the inguinal area and a painful fixed local mass. All patients underwent pre-operative plain radiography (PR) of the pelvis, computerized tomography (CT) of the pelvis and lung, magnetic resonance imaging (MRI) of the pelvis and electrical capacitance tomography (ECT) bone scan.

This study included only patients with local sarcoma or solitary bone metastasis/plasmacytoma involving the acetabulum. The pathological diagnoses of ten patients were obtained by pre-operative needle biopsy, while that of another patient was obtained by incisional biopsy (at another institution). Seven patients were diagnosed with chondrosarcoma, two with osteosarcoma, one with solitary plasmacytoma, and one with metastatic bone tumour (breast cancer). To obtain a clear surgical margin and long-term survival prognosis, the patients with osteosarcoma and metastatic bone tumour received pre-operative adjuvant chemotherapy. According to the surgical staging system described by Enneking et al. [32], ten patients with chondrosarcoma, osteosarcoma and solitary plasmacytoma were stage IIB. The patient with a solitary bone metastasis from breast cancer also had an extraskeletal component.

En bloc resection was achieved in all 11 patients, with nine wide excisions and two marginal excisions. Of the two marginal excision patients, one had an osteosarcoma with the tumour invading the external iliac vessels, but he had a good pre-operative response to chemotherapy; the other patient had a chondrosarcoma with a large soft-tissue mass and underwent an inappropriate biopsy. The mean operation time was 271 minutes (range, 210–360 min). The mean intra-operative blood loss was 3236 mL (range, 1600–6500 mL), and the mean post-operative drainage volume was 1209 mL (range, 800–1800 mL) (Tables 1 and 2).

Prosthesis design and manufacture

CT and MRI were performed for the whole pelvis, including the sacrum, ilium, pubis, and ischium. The prostheses were designed to individual specifications based on these imaging data (Fig. 1), and then manufactured by 3D printing technology (Fig. 2). In contrast to traditional prostheses, 3D-printed titanium alloy prostheses have a porous metal interface allowing bone in-growth (Fig. 2). In order to increase the stability of the reconstruction, the screw holes on the iliac aspect of the prostheses include locking screw holes and universal pores.

It took approximately two weeks for the design and manufacture of each personalized 3D-printed hemipelvic prosthesis.

Surgical techniques

Pre-operative embolization can provide effective tumour devascularization, which significantly reduces the chance of intra-operative bleeding [33]. To assess tumour blood supply and reduce intra-operative blood loss, pre-operative arterio-angiography or embolization was performed in all patients. Surgery was performed one to two days after this procedure. To form a detailed pre-operative surgical plan and ensure an accurate osteotomy, 3D-printed pelvic tumour models and osteotomy guide plates were created according to pre-operative CT and MRI imaging data (Fig. 3).

After general anaesthesia, the patient was placed in the lateral decubitus position with the ipsilateral hip up. A “T” incision (an extended ilioinguinal incision and a vertical incision toward the

Table 1 Summary data for all patients

Case number	Age (years)	Sex	Pathological diagnoses	Pre-op therapy	Enneking staging	Post-op therapy
1	21	Male	Osteosarcoma	CH	IIB	CH
2	58	Female	Chondrosarcoma	No	IIB	No
3	56	Female	Bone metastasis (Breast cancer)	CH	#	CH
4	45	Male	Chondrosarcoma	No	IIB	No
5	55	Female	Chondrosarcoma	No	IIB	No
6	60	Female	Solitary plasmacytoma	No	IIB	No
7	37	Female	Osteosarcoma	CH	IIB	CH
8	63	Male	Chondrosarcoma	No	IIB	No
9	38	Female	Chondrosarcoma	No	IIB	No
10	41	Male	Chondrosarcoma	No	IIB	No
11	43	Male	Chondrosarcoma	No	IIB	No

CH chemotherapy

[#] The patient with a solitary bone metastasis had an extraskeletal component

greater trochanter) was used for tumour resection. Soft tissue dissection was dependent on the presence or absence of tumour compromise. After the femoral neck was osteotomized, the femoral head was removed to expose the acetabular cavity. The bone surface near the planned resection sites was exposed. A 3D-printed osteotomy guide plate was firmly seated on the bone surface and then stabilized with 2.0-mm Kirschner wires. With the aid of a guide plate, the resections of the ilium, pubis, and/or ischium were performed. After tumour removal, a personalized 3D-printed hemipelvic prosthesis which precisely matched the bony defect was implanted. The iliac aspect of the prosthesis was fixed to the residual part of the iliac crest, sacroiliac joint, or sacrum using cancellous screws and locking screws. The pubic aspect of the prosthesis was fixed to the residual part of the pubis or contralateral pubic branch. A cemented elevated-rim acetabular liner with 42-mm outer diameter and 32-mm inner diameter was chosen for fixation. The hip joint was restored as in a conventional total hip arthroplasty. In order to reduce the risk of post-operative wound infection, the wound was flushed repeatedly with a pulsing squirt gun during operation, and sufficiently drained after surgery.

Among the 11 patients, nine with local sarcoma and solitary bone metastasis/plasmacytoma were treated with wide *en bloc* resection, while two patients with osteosarcoma and chondrosarcoma were treated with marginal *en bloc* resection.

Table 2 Data relating to the operation and follow-up of patients undergoing resection and reconstruction for peri-acetabular malignant bone tumours

Variables	Value
Operational data	
Operation time, mean (SD) mins	271 (45.5)
Intra-operative blood loss, mean (SD) ml	3236 (1665)
Post-operative drainage volume, mean (SD) ml	1209 (311)
Surgical margins (n, %)	
Wide	9 (81.8%)
Marginal	2 (18.2%)
Follow-up data	
Duration, mean (SD) mo	15.5 (5.7)
Survival status (*n*, %)	
DFS	9 (81.8%)
AWD	1 (9.1%)
DOD	1 (9.1%)
VAS (pre vs. post)	4.5 ± 2.5 vs. 1.8 ± 1.0[a]
MSTS-93 score, mean (%)	19.2 (64%)

SD standard deviation, *DFS* disease-free survival, *AWD* alive with disease, *DOD* died of disease

[a] All patients experienced some alleviation of pain two weeks after surgery ($P < 0.01$)

Post-operative assessments

All patients were evaluated every month during the first three months of follow-up and every three months thereafter with a physical examination, PR of the pelvis, PR or CT of the chest, and ECT bone scans (every three months) (Figs. 4 and 5). Pre-operative and post-operative pain at rest (two weeks after surgery) was assessed according to a 10-cm visual analogue scale (VAS) score. The clinical results of functional improvement were evaluated by the Musculoskeletal Tumour Society 93 (MSTS-93) score at the final follow-up [34].

Statistical analysis

Survival was analyzed using Kaplan-Meier method. Pre-operative and post-operative data were compared using Wilcoxon signed-rank test (Statistical Package for the Social

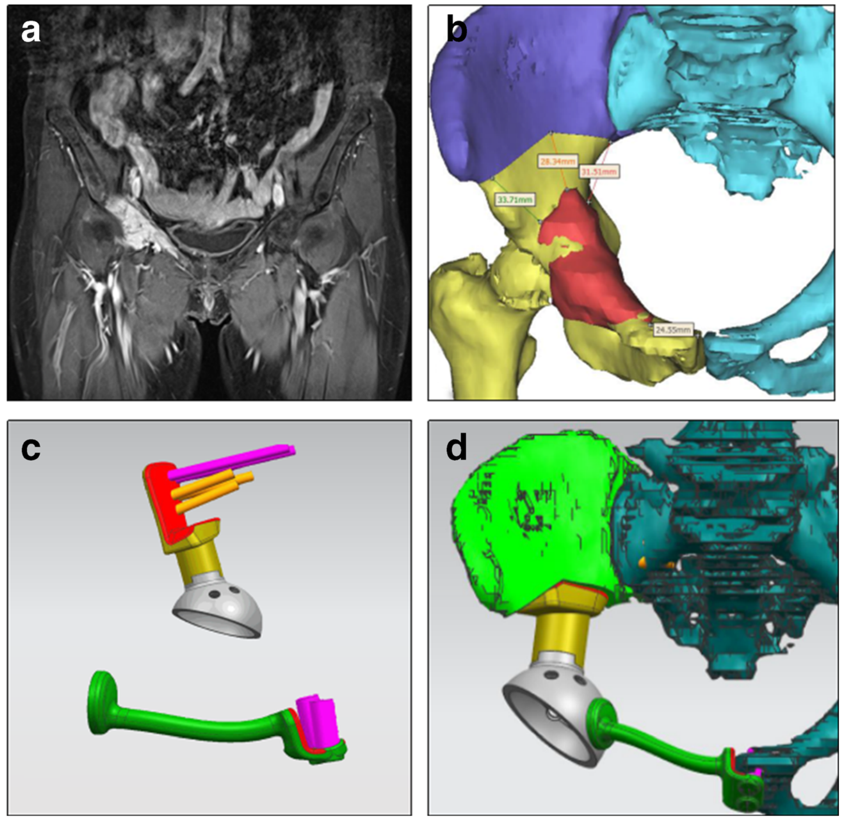

Fig. 1 (**a**) A coronal contrast-enhanced T2-weighted MR image in a patient with a solitary plasmacytoma involving the right acetabulum. (**b, c**) Design of a 3D-printed hemipelvic prosthesis based on pre-operative CT and MRI imaging data. (**b**) The extent of tumour (*red* in color) and osteotomy plane were outlined on a computerized 3D pelvic model. (**c**) Design of iliac and pubic aspects of the prosthesis. Screw positions and lengths (locking screws, *pink* in color; cancellous screws, *yellow* in color) were planned based on the quality of remaining bone after resection of the tumour. (**d**) Implant installation and screw fixation were simulated on a computerized 3D pelvic model

Sciences, Version 16.0; SPSS, Chicago, Illinois). A *P*-value of 0.05 was considered to be statistically significant.

Results

All patients were observed for six to 24 months with an average follow-up of 15.5 months. Nine patients were alive without evidence of disease, one patient with mesenchymal chondrosarcoma was alive with a left iliac bone metastasis and new-onset breast cancer (three months after surgery), and one patient with osteosarcoma had died of pulmonary metastasis 15 months after surgery (Fig. 6). No local tumour recurrence was observed in this series.

The functional results allowed patients good pain control and return to reasonable function. All patients experienced some alleviation of pain two weeks after surgery (VAS, pre vs. post; 4.5 ± 2.5 vs. 1.8 ± 1.0). Although one patient complained of occasional pain of the involved hip at the final follow-up (VAS, pre vs. post 8 months: 3 vs. 2), the pain could be relieved by nonsteroidal anti-inflammatory drugs (NSAIDs). The recovery of the affected limb function was assessed according to the MSTS-93 score at the final follow-up. The mean MSTS-93 lower extremity functional outcome score was 19.2 (range, 13–25).

None of our patients experienced severe complications in this study. Delayed wound healing occurred in one patient. Although skin margin necrosis of the incision was observed, the patient received no special treatment, and there was a subcrustal healing 12 weeks after surgery. Hip dislocation occurred in two patients. After closed reduction of a posterior hip dislocation, the hip was maintained in neutral and abduction position with a hip fixation brace for eight weeks. Extensive hip joint exercise was delayed after reduction to allow healing of soft tissue. Recurrent hip dislocation was not observed in those two patients during the follow-up

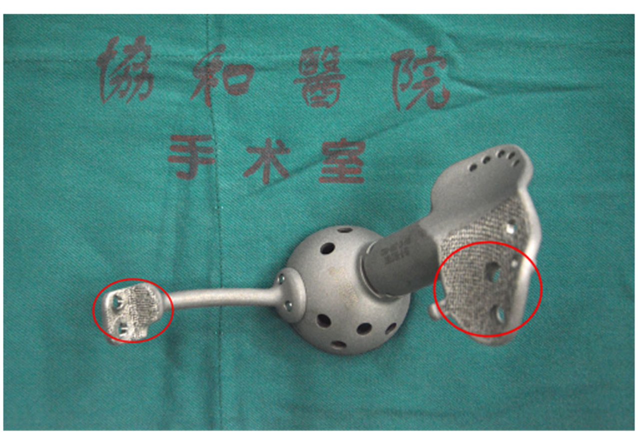

Fig. 2 A personalized 3D-printed hemipelvic prosthesis. The bone contacting surface of the prosthesis had the porous scaffold (*circle*) allowing bone in-growth. The scaffold has an interconnected network of pores with an average porosity of 60%

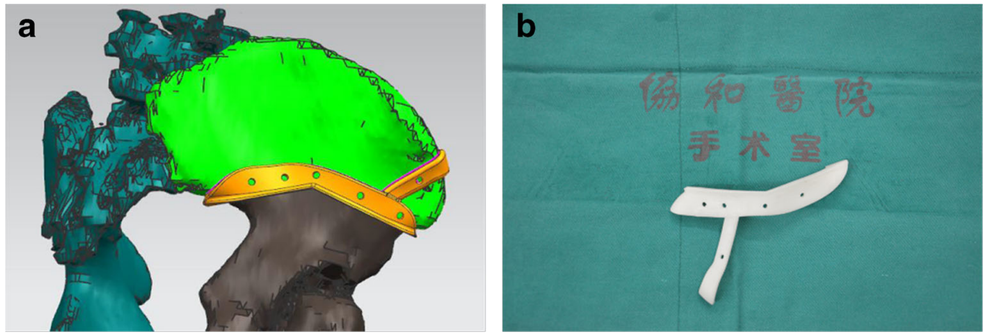

Fig. 3 (**a**) Design of a 3D-printed osteotomy guide plate based on pre-operative CT and MRI imaging data. The shape of the guide plate allowed a unique positioning on the surgical accessible bone surface determined by the surgeons creating the CAD software. (**b**) A 3D-printed osteotomy guide plate

period. No patient developed wound infection in such small series. Moreover, we could not find any evidence of aseptic loosening, bone resorption or periprosthetic fractures during the short-term follow-up.

Discussion

Peri-acetabular malignant bone tumours are rare, but when they occur, they can present formidable therapeutic challenges. In the past, hindquarter amputation was the most common treatment option because of the limitations of traditional techniques [1]. However, amputation surgery often leads to physical and psychological problems in the patient.

Currently, with the development of radiotherapeutic, chemotherapeutic and surgical techniques, most of these tumours can be limb-salvage resections [2–4, 6–8, 23, 35]. Wide *en bloc* resection is critical for primary peri-acetabular malignant bone tumours. During surgery, excision should be as wide as possible. For pelvic bone metastases, marginal resection may be enough [36]. However, wide *en bloc* resection was recommended for solitary bone metastases from renal or thyroid cancer without extraskeletal metastases [37]. Although there was an extraskeletal component, one of our patients, who had a solitary pelvic metastasis, underwent wide *en bloc* resection because of good pre-operative response to chemotherapy.

Effective acetabular reconstruction after tumour resection is another key process. Various methods including arthrodesis, resection hip arthroplasty, osteoarticular allograft/ autograft reconstruction, and prosthetic replacement have been reported for hemipelvic reconstruction [4, 6, 8, 10–12, 23]. Of these methods, prosthetic reconstruction may be superior to other surgical options in restoring hip joint function and controlling associated complications. Arthrodesis between the ilium and femoral head is an excellent reconstruction because of low complication rate. However, the hip joint function is sacrificed [10, 23]. Allograft or autograft of the pelvis, which have been used for hip reconstruction, can obtain acceptable hip joint function, but this technique has been discontinued because of high deep infection rate and fracture risk [6, 14, 18, 24]. The prosthetic reconstruction is considered as the most effective method, as it can restore the anatomy of the acetabulum and preserve hip joint function, while avoiding complications seen with massive allograft or autograft [7–9, 12, 13, 15–17].

Several types of prostheses, such as the saddle prosthesis, modular prosthesis, and custom-made prosthesis, have been used for reconstructing bony defects after peri-acetabular tumour resections [7, 11, 12, 15, 17]. Since 1987, Jansen et al. used saddle prostheses to treat 17 patients with peri-acetabular tumours. After a mean of 12.1 years of long-term follow-up, they reported a mean MSTS-93 score of 47% and an 82% complication rate. Therefore, they deemed the saddle

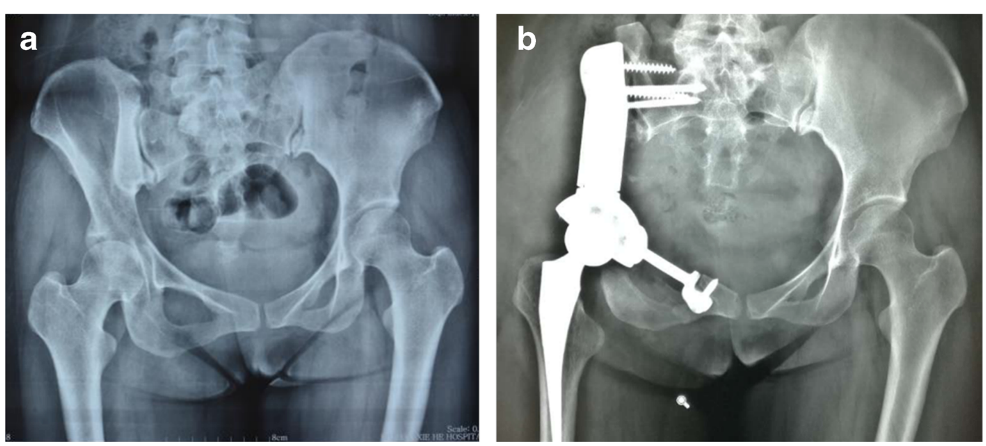

Fig. 4 Plain radiography obtained from a 38-year-old female patient with a chondrosarcoma involving the ilium and pubis. A personalized 3D-printed hemipelvic prosthesis was implanted after *en bloc* resection of the tumour. (**a**) Before surgery. (**b**) Eleven months after surgery

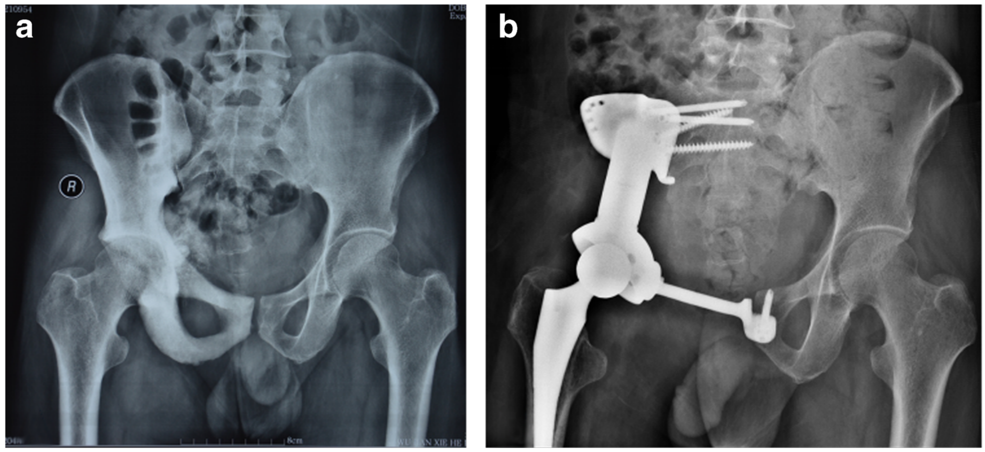

Fig. 5 Plain radiography obtained from a 41-year-old male patient with a chondrosarcoma involving the ilium, pubis and ischium. A patient-specific 3D-printed hemipelvic prosthesis was implanted after *en bloc* resection of the tumour. (**a**) Before surgery. (**b**) Twelve months after surgery

prostheses unsuitable for reconstruction following peri-acetabular tumour resection [11]. The modular prosthesis reconstruction is advantageous surgically due to the ease of working with flexible modular components, and can provide better functional results [7, 12, 15]. Guo et al. described the use of the modular prosthesis in 28 patients, which showed a mean MSTS score of 60% with an acceptable complication rate after a mean follow-up of 30 months [15]. The computer-aided custom-made prosthesis emerges as a step toward individually tailored anatomical designs and should, in theory, provide superior functional results [8, 13]. In addition to the reports of Ozaki et al. [16], reconstruction with computer-aided custom-made hemipelvic prostheses provided a more satisfactory function (MSTS score, approximately 70%) in most studies [8, 13]. The computer-aided designed prostheses are specifically modeled to perfectly match bony defects. However, the complex shapes, contours, and faces of custom-made prostheses are sometimes difficult or impossible to produce because of the limitations of the traditional manufacturing technology. More importantly, a revolution in hemipelvic prosthetic design seems inevitable because of the pervasive issue of long-term non-integration at the bone–implant interface. The recent development of 3D printing technology may provide an effective solution to these problems.

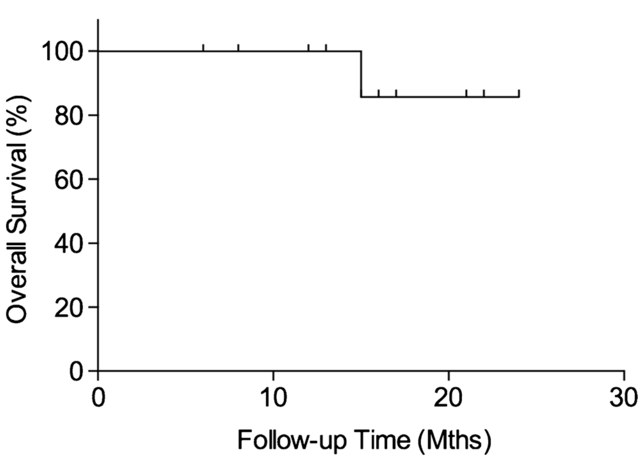

Fig. 6 Graph showing overall survival of the 11 patients

Compared to the traditional manufacturing technology, 3D printing technology has obvious advantages. First, it greatly expands our capability to produce guide plates or prostheses of complex shape and dimension. 3D-printed guide plates based on pre-operative CT and MRI images can create a rapid and accurate osteotomy without the aid of navigation, which facilitates precise matching of the implant and greatly shortens the operation time; 3D-printed prostheses can also perfectly match the bony defects left as a result of tumour resection, while avoiding repeated adjustments [28, 30, 31]. Second, 3D printing technology can fabricate a metal surface with porous scaffold. The porous scaffold allows the host bone to grow inside the construct to achieve a stable "mechanical" biological reconstruction [25–28]. Recently, 3D-printed prostheses have been used in hemipelvic reconstruction after peri-acetabular tumour resection [29–31]. Chen et al. reported the successful application of a patient-specific 3D-printed iliac prosthesis for the reconstruction of the hemipelvis in a 62-year-old patient with pelvic sarcoma [30]. However, pelvic reconstruction is more challenging if it involves the acetabulum. In this study, we introduced computer-aided designed, 3D-printed hemipelvic prostheses to reconstruct bony defects following peri-acetabular tumour resections, and evaluated our patient's short-term functional results, complications and oncological outcomes.

In the present series, the functional results allowed patients to return to reasonable function. The mean MSTS-93 lower extremity functional outcome score was 19.2 (range, 13–25). Recently, Liang et al. treated 32 patients with peri-acetabular tumours by *en bloc* approach and patient-specific 3D-printed hemipelvic prostheses (12 cases with a standard hemipelvic prosthesis, 20 cases with a screw-rod connected hemipelvic prosthesis), and the short-term function recovery was satisfactory (MSTS-93, range, standard: 15–26; screw-rod connected: 9–25) [29]. In contrast to the report by Liang et al. [29], all patients in this study underwent reconstruction of the pelvic ring which is critical for the stability of the pelvis. Although similar short-term functional results were observed in these two studies, we believe that the approach emphasizing pelvic ring reconstruction may obtain more satisfactory functional results in the long term.

Significant blood loss and a high complication rate often accompany resection and reconstruction of peri-acetabular tumours [13]. In our series, the mean intra-operative blood loss was 3236 mL, which is significantly lower than many previous reports [8, 13]. We attribute the effective bleeding control to pre-operative target arterial embolization and short operating times. Previous studies demonstrated a high rate (25%) of deep wound infection in patients who underwent hemipelvic prosthetic reconstruction [16, 22]. To reduce the incidence of infection, the wound was flushed repeatedly with a pulsing squirt gun during surgery, and drained sufficiently after surgery. Among our patients, none developed a wound infection during the follow-up period. The difficulty of wound healing is another common complication. Inconsistent with the findings of Dai et al. and Liang et al. [13, 29], only one patient suffered from delayed wound healing in this study. We identified multiple possible contributing factors, including different surgeons, incision design, soft tissue involvement and post-operative therapy. Although an elevated-rim acetabular liner was used to improve the stability of the total hip prosthesis, two patients still suffered from hip dislocation in such small series. This is in keeping with the findings of others who observed an association between reconstruction arthroplasty of the pelvis and a relatively high dislocation rate [13, 16, 29]. Moreover, we could not find any evidence of aseptic loosening, prosthetic breakage, or screw breakage, which could be due in part to the short follow-up period and novel “mechanical” biological reconstruction.

This study has some limitations. First, the short follow-up period of this study does not account for some mechanical complications that may arise as we follow these patients for a longer time. Second, the small sample size limits this study. However, although small, this study provides some interesting information for 3D-printed prosthetic reconstruction following peri-acetabular tumour resection. Larger scale studies will be necessary before this new generation of prostheses can be widely implemented. Finally, this study is a retrospective analysis. A long-term prospective study may be very difficult because of many uncertain factors such as tumour response to therapy, surgeon ability, and the quality of the 3D-printed prostheses.

Conclusions

Bony defect reconstruction following tumour resection using personalized 3D-printed hemipelvic prostheses can obtain acceptable functional results without severe complications. Although the follow-up period of this preliminary study is very short, based on previous reports and our results, we believe that reconstruction arthroplasty using 3D-printed hemipelvic prostheses will provide a promising alternative for those patients with peri-acetabular malignant bone tumours.

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