



ORIGINAL ARTICLE

One-step reconstruction with a 3D-printed, custom-made prosthesis after total en bloc sacrectomy: a technical note

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Abstract

Background Surgeries for primary malignancies involving upper sacrum require total en bloc sacrectomy followed by complex mechanical reconstruction, which might be simplified by application of the three-dimensional (3D) printing technique.

Purposes To describe the design of a 3D-printed custom-made prosthesis for reconstruction after total en bloc sacrectomy, the surgical technique, and the clinical and functional outcome of a patient.

Methods A 62-year-old patient with recurrent sacral chordoma was admitted in our center. One-stage total en bloc sacrectomy through posterior approach was planned, and a 3D-printed sacral prosthesis was prepared for reconstruction according to the anticipated osteotomic planes.

Results The patient received one-stage total en bloc sacrectomy through posterior approach followed by reconstruction with the 3D-printed sacral prosthesis. The whole procedure took 5 h, and intra-operative blood loss was 3400 ml. The patient recovered uneventfully and started ambulation at 3 weeks after surgery. An asymptomatic instrument failure was found radiographically at 8-month follow-up. At 1 year after surgery, the patient was disease free and could walk over short distance with crutches without pain or any mechanical instability.

Conclusions The advantages of our reconstruction method included: (1) the prosthesis provided an optimal

reconstruction of lumbosacral and pelvic ring by integrating spinal pelvic fixation, posterior pelvic ring fixation, and anterior spinal column fixation in one step and (2) its porous surface could induce bone ingrowth and might enhance stability. Although there was an instrumental failure, we considered that it could be one reconstructive option. More research is warranted focusing on the modification of locations, diameters, and quantity of screws and biomechanical characteristics. The long-term functional and bone in-growth outcome will be followed to validate the use of the prosthesis.

Keywords Total en bloc sacrectomy · Prosthesis · 3D printing · Reconstruction · Oncology · Chordoma

Introduction

Primary sacral tumor, especially the primary sacral malignancy, is rare [1]. Primary malignancies involving upper sacrum often require total en bloc sacrectomy in order to achieve adequate tumor-free surgical margin [2]. With the accumulation of clinical experiences and improvements of surgical techniques, one-stage total en bloc sacrectomy has become practical and feasible in selected cases [2, 5, 6]. The subsequent reconstruction of the spinopelvic continuity, however, remains challenging, and no standardized methods have been established despite decades of efforts [3–12].

Three-dimensional (3D) printing technique is capable of reconstructing bone defect precisely by tailored contour and inducing bone ingrowth by porously proofing treatment, which could help to reduce the long-term mechanical complications, such as loosening and fracture [13]. This novel technique has been successfully used in orthopedic

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surgery [14, 15]. To our knowledge, there were no reports on 3D-printed prosthesis for reconstruction after total en bloc sacrectomy.

We report a case of sacral chordoma treated by total en bloc sacrectomy followed by 3D-printed prosthesis reconstruction, aiming to describe the design of a 3D-printed custom-made prosthesis, the surgical technique, and clinical/functional outcome of the patient.

Materials and methods

Case report

A 62-year-old male patient came to our center complaining of increasingly severe sacrococcygeal pain and bowel/bladder dysfunction in 2015. He had undergone two operations in 2003 and 2013, respectively, because of sacral chordoma. After admission, imaging study showed local recurrence in the residual S1/2 with a huge mass protruding anteriorly and posteriorly without invading the ilia and L5 vertebrae (Fig. 1). Bone scan and chest CT showed no evidence of metastasis. Since the upper sacrum was involved, we planned to perform a one-stage total en bloc sacrectomy through posterior approach followed by reconstruction with a 3D-printed custom-made prosthesis.

Design and manufacture of prosthesis

Based on the pre-operative imaging studies, we determined the osteotomy planes and identified the shape of the bone defect. The design stemmed from the concept of a porous metal prosthesis that could connect lumbar spine and ilium, connect both sides of ilium, and rebuild the structure of loading transfer through anterior spinal column in one step while conducive to bone in-growth into the trabecular pores [10, 13]. The shape of the prosthesis was designed according to the bone defect by MIMICS 15.0 (Materialise NV, Leuven, Belgium). The prosthesis was produced from titanium alloy and manufactured by 3D printing technique. The bone-contacting surfaces were porous to facilitating the bone ingrowth. The diameter of pores and wires was 800 and 550 μm , respectively, with an average porosity of 50–80%. Electron beam melting (EBM) was used in fabrication by successive layering of melted titanium alloy according to a computer-aided design (CAD) model. The prosthesis was tested according to the National Standard of Implants for Surgery in China.

The prosthesis consisted of three bone-contacting surfaces: the proximal surface fit to the contour of inferior endplate of L5 vertebrae to reconstruct the lumbarsacral joint; the surfaces on both flanks were matched to bilateral iliac osteotomies to reconstruct both sacroiliac joints.

Screw holes were predrilled on every bone-contacting surface for fixation. Two screw heads were placed on the dorsal surface for connection to pedicle screws of lumbar spine with titanium rods ($\Phi = 6.0 \text{ mm}$) (Fig. 2). The prosthesis was prepared in three different sizes to fit the real size of intra-operative bone defect.

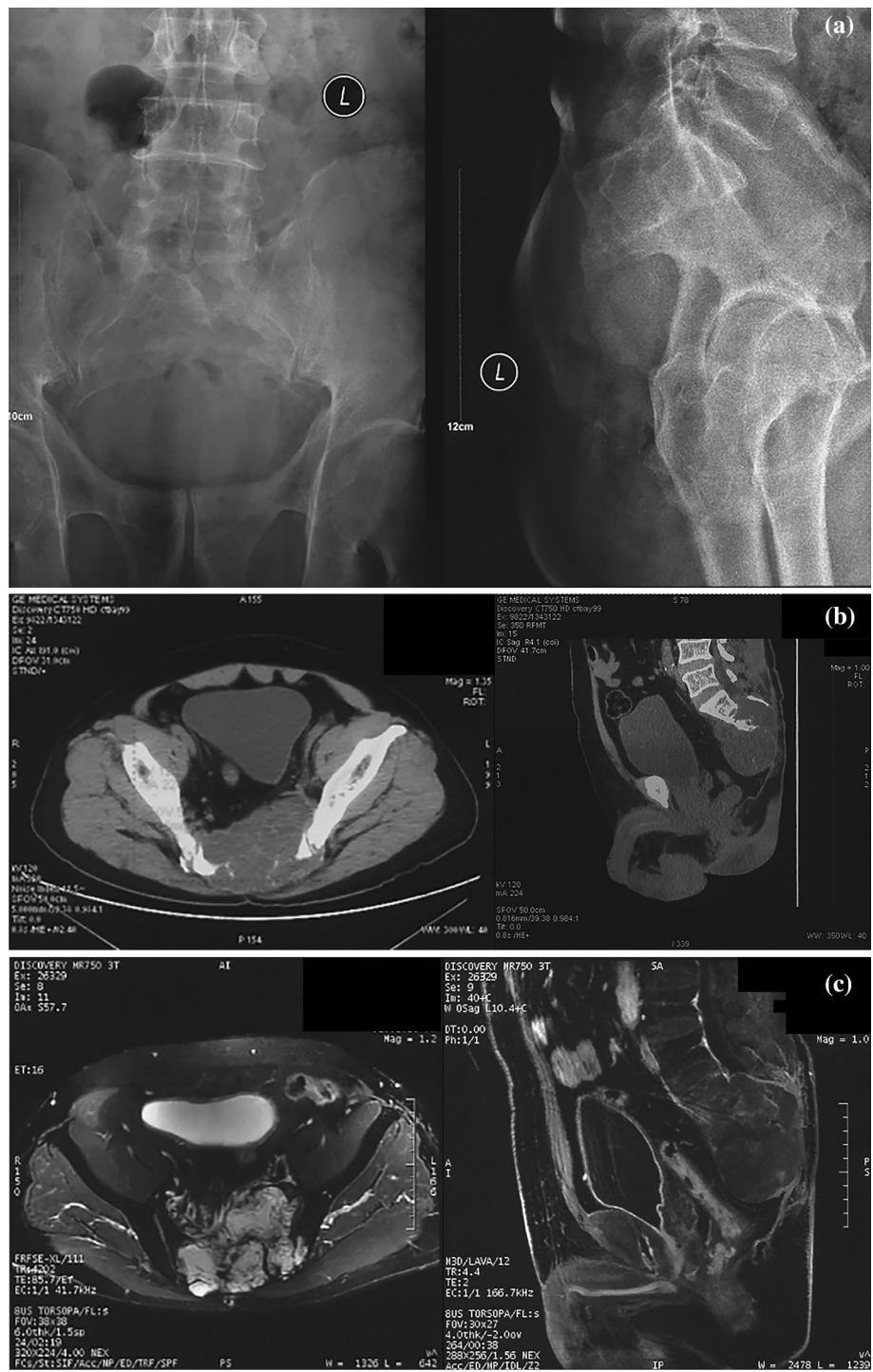
Results

Surgical procedure

Patient received selective arterial embolization before surgery and intra-operative aortic balloon occlusion to reduce surgical blood loss [16]. The operation was performed through a posterior approach as previous reported [5, 6]. In brief, patient was placed in prone position, and an inverted Y incision was made. The parasacral muscles, the gluteus maximus muscles, and the gluteus medius muscles were dissected to expose the L4–5 spinal process and lamina, posterior cortex of the sacrum, and back aspect of the bilateral ilium. The sacrum below S3 had been removed by previous surgeries. A huge lobulated mass was seen in the residual sacrum. The rectum was bluntly dissected from the tumor up to the S1 level with gauze packed into the pre-sacral space. The dura sac and bilateral L5 nerve roots were identified and preserved, while the dura sac below L5 was ligated and divided. The L5–S1 disc was removed. The bilateral transverse processes of L5 were removed to facilitate blunt finger dissection from the upper and lower edges of the sacroiliac joints. The lumbosacral trunk was identified, and gauze was packed into the space in front of the sacroiliac joints to protect the lumbosacral trunks and internal iliac vessels during iliac osteotomy. Silicone tubes were introduced through the anterior space of the sacroiliac joints, so that a wire saw could be passed through to cut bilateral ilia at the anticipated osteotomic planes. After osteotomy of bilateral ilia, the sacrum was mobilized. The surrounding vessels were ligated and sacral nerve roots were resected. Finally, sacrum and bilateral partial ilia were removed with satisfactory margin (Fig. 3).

As for reconstruction, two transpedicle screws were placed into the L4 pedicles. The middle-size sterilized prosthesis was placed into the bone defect area and matched the osteotomic planes of L5 and ilia precisely. Screws were introduced through reserved holes into vertebrae of L5 and bilateral ilia. While inserting the second screw to the left ilium, we found that the residual ilium was too thin and osteoporotic to hold the screw. We gave up after several failed attempts. Two rods were used to connect the L4 pedicle screws and prosthesis (Fig. 4). The whole procedure took 5 h, and the intra-operative hemorrhage was 3400 ml.

Fig. 1 Pre-operative imaging study. **a** X-ray. **b** Computed tomography (CT). **c** Magnetic resonance imaging (MRI)



Histological findings and post-operative management

Microscopically, the tumor cells were vacuolar or acidophilic and were rich of cytoplasm which was lightly stained. The nucleoli were prominent and mild atypia. The pathological diagnosis was chordoma, and the margin was clear.

In the post-operative period, the silicone drainage tubes were kept for about 19 days and intravenous antibiotic was administered until the drainage was removed. Subcutaneous anticoagulation and oral antiplatelet agents were administered after the drainage volume was stable. The wound healing was uneventful, and no peri-operative complications occurred. The mobilization started 3 weeks

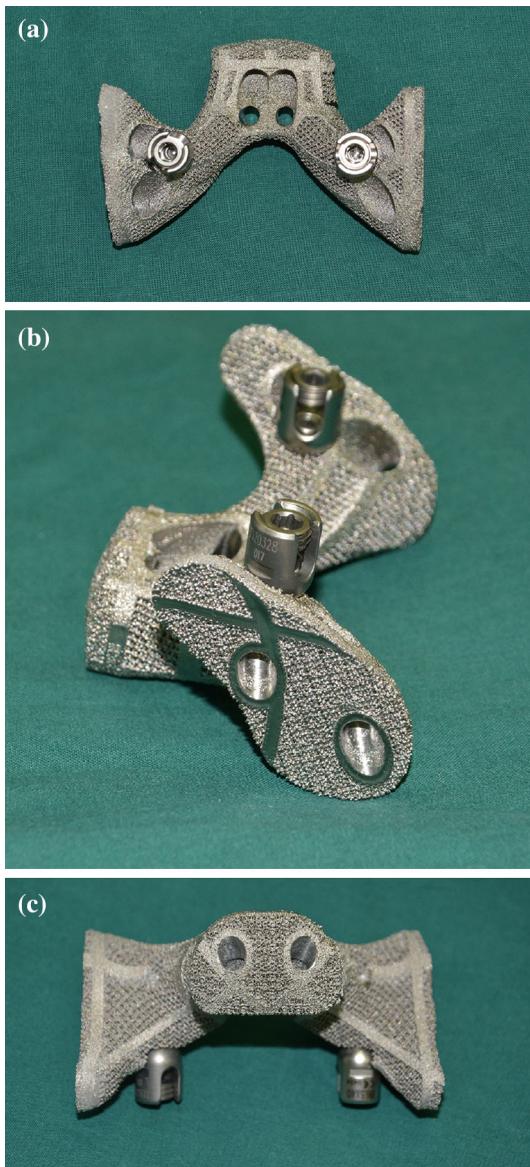
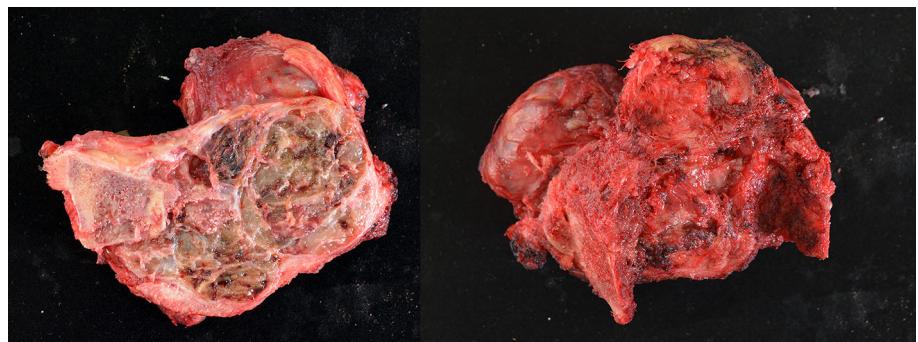


Fig. 2 3D-printed custom-made prosthesis for reconstruction after total en bloc sacrectomy. **a** Dorsal view. **b** Lateral view. **c** Superior view

Fig. 3 Resected tumor specimen. The *left image* showed that the bone destruction and mass involving S1 and S2. The *right image* showed the resection surfaces of both sides of ilium and L5/S1 intervertebral disc



post-operatively, and there was no pain or symptoms of instability.

Follow-up

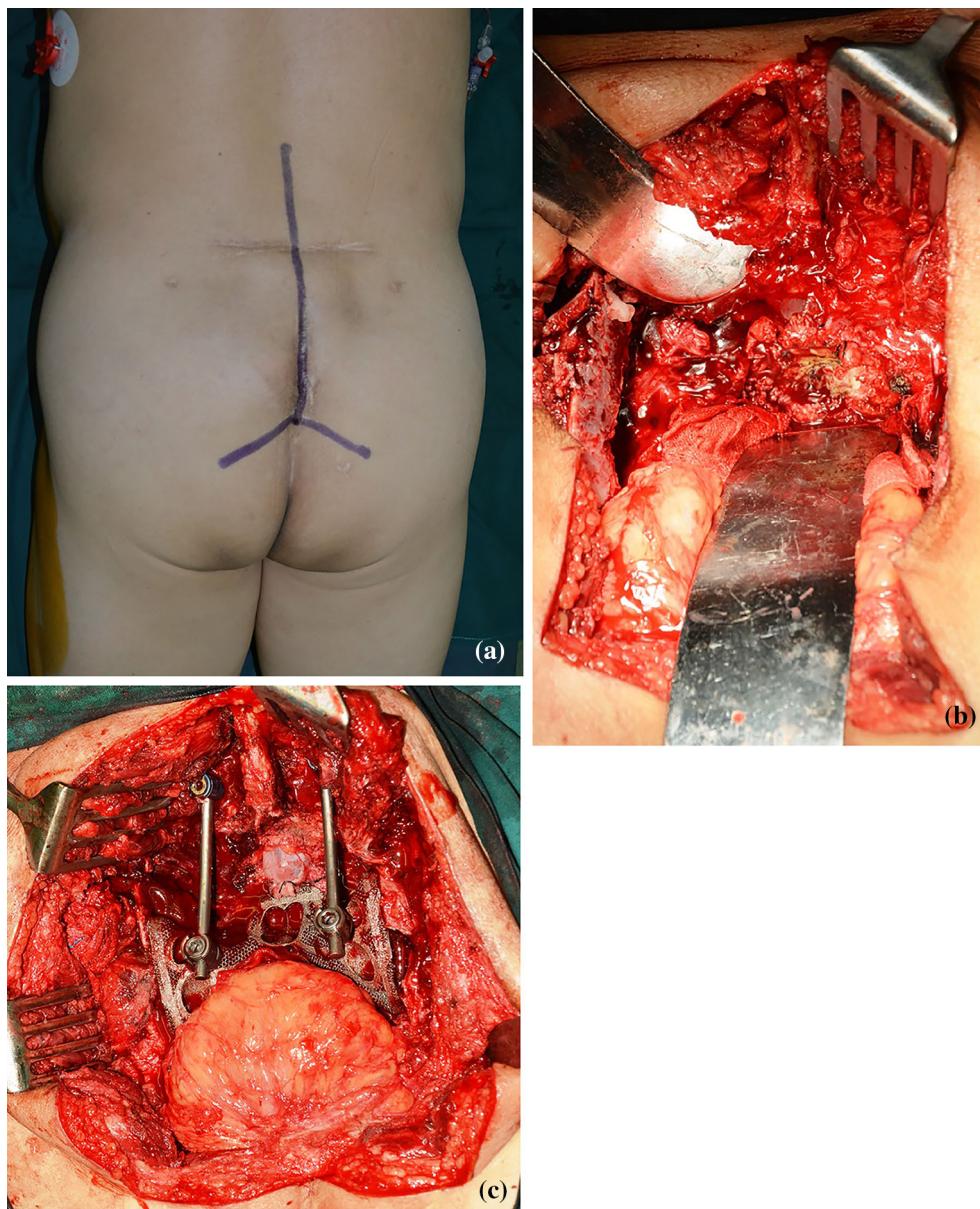
At 3-month follow-up, X-ray study showed no evidence of loosening or fracture (Fig. 5b) compared with that performed right after surgery (Fig. 5a). At 8 months after surgery, the X-ray showed two fractured screws in bilateral ilia, while the CT showed some new bone formation around the prosthesis–ilium interface (Fig. 5c). The patient remained asymptomatic. At 1-year follow-up, patient was disease free and could walk indoor and outdoor over short distances with crutches. He did not feel any pain or spinopelvic instability.

Discussion

Primary sacral tumors are rare and account for only 1–7% of spinal neoplasms [1]. The most common primary malignancies in sacrum include chordoma, chondrosarcoma, Ewing sarcoma, and osteosarcoma, of which surgical resection is the main treatment [2]. Nevertheless, adequate surgical margin had been difficult to obtain because of the complex anatomic structures and massive intra-operative bleeding. As for primary sacral malignancy involving the upper sacrum, total en bloc sacrectomy was encouraged, but was complicated by a high incidence of peri-operative complications [3]. With the advancement of surgical techniques in the past decade, total en bloc sacrectomy has become much more practical and feasible with shortened operation time and reduced incidence of complications [2, 5, 6].

The bone defect due to total en bloc sacrectomy leads to disconnection between pelvis and spine. Effective reconstruction of the spinopelvic continuity is of importance, as it could allow early ambulation and thus reduce post-operative complications caused by long-time bed-ridden

Fig. 4 Surgical procedure.
a Patient was placed in prone position and an inverted Y incision was made. **b** Image showed the bone defect between L5 and both sides of ilium after en bloc resection of tumor.
c Prosthesis was settled

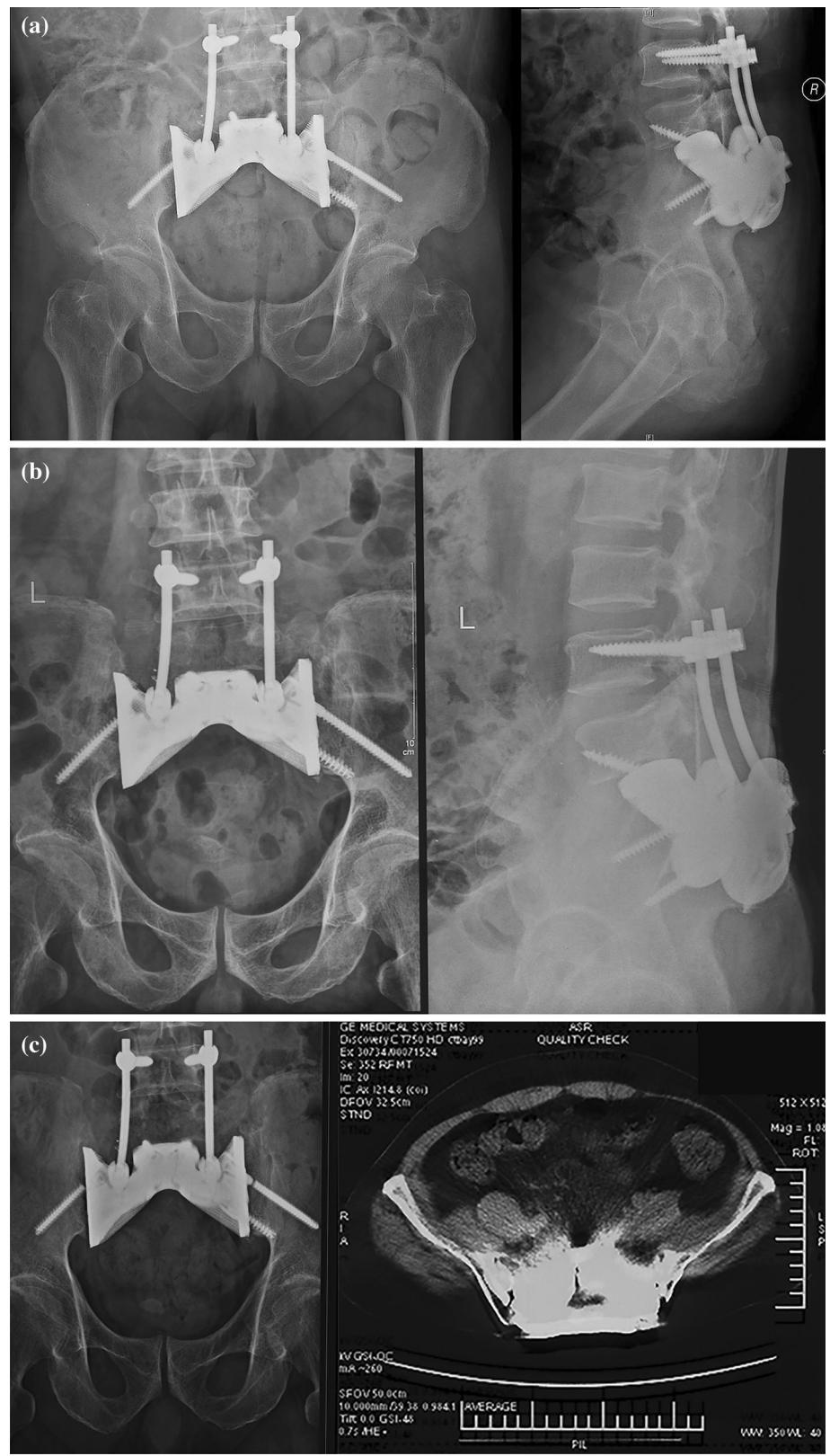


would be reduced [3]. However, unlike the maturation in sacrectomy techniques, the methods for reconstruction remained barely satisfactory and controversial despite decades of endeavor.

The conventional reconstruction methods after total en bloc sacrectomy could be categorized into three types: spinal pelvic fixation (SPF), posterior pelvic ring fixation (PPRF), and anterior spinal column fixation (ASCF). SPF method refers to the surgical constructs that connect the lumbar spine to the ilium and is the basement of all reconstructive methods. It had developed from Galveston technique [17] to modified Galveston technique [18] which included single-rod dual-iliac-screw method, dual-rod dual-iliac-screw method, and U-shaped rod method

[5, 6, 19]. The advantages of SPF include fair mechanical stability, operational simplicity, and acceptable rate of complications. Several biomechanical studies found that dual-rod dual-iliac-screw method yielded the best stability [20, 21], while other studies also demonstrated concentrated stress at the titanium rods and rotational instability around the horizontal axis that might eventually result in fracture or loosening [22]. Moreover, the lack of transverse sacral bars in SPF could hardly prevent the ilia from spreading in a lateral anterior direction [12]. The stabilization of SPF would mostly depend on a biological sling formed by muscles and scar tissue between the pelvis and the spine, which indicates that the instability would occur at early stage.

Fig. 5 Radiography 3 weeks (a), 3 months (b), and 8 months (c) after surgery



PPRF method means connecting only the ilia together without fixation to the lumbar spine, mainly by bone graft to conduct bone fusion between two ilia [12] or interiliac

cross-connecting rods [11]. PPRF is frequently carried out with SPF to compensate for the shortages of SPF as mentioned above [7].

ASCF method aims to rebuild the structure of loading transfer through anterior spinal column. It is often carried out with SPF or with both SPF and PPRF [8, 23, 24]. Clark et al. compared the stability of SPF + PPRF and SPF + PPRF + ASCF in biomechanical study and showed that the latter reconstructive method performed better in both range of motion (ROM) test and fatigue test [25]. In a systemic review enrolling 43 patients in 23 studies of reconstruction after total en bloc sacrectomy, Bederman et al. found that the fixation failure rate was lower in cases with ASCF than those without, indicating that the reliable reconstruction method should include all SPF, PPRF, and ASCF [4].

Several unconventional reconstruction methods had also been reported, such as cannulated screws, inserted from the lateral of posterior superior iliac spine to the vertebrae of L5 in combination with SPF [26] and re-implantation of extra-corporeally irradiated sacrum [9], which, however, could hardly show advantages over another due to the limited number of cases.

Although custom-made prosthesis has been successfully used in orthopedic surgery, there were few reports about using prosthesis for reconstruction after total en bloc sacrectomy. Wuisman et al. reported one case using custom-made prosthesis for reconstruction of bone defect after total en bloc sacrectomy. There were no post-operative complications and no signs of prosthetic instability in 3 years after surgery [10].

The 3D printing technique has raised new conceptions in clinical practice especially in plastic and orthopedic surgery. It has been successfully applied in reconstruction for bone defect on upper cervical spine and pelvis [14, 15]. The contour of a 3D-printed prosthesis could perfectly fit to the bone defect, and the bone-contacting surfaces could be processed to be porous for the purpose of conducting bone in-growth and increasing long-term stability [13].

In our case, by 3D printing technique, a custom-made sacrum prosthesis was manufactured based on the shape of bone defect. During surgery, the prosthesis matched the osteotomic planes of L5 and bilateral ilia precisely. The advantages of our reconstruction method included: (1) the prosthesis provided an optimal reconstruction of lumbosacral and pelvic ring by integrating SPF, PPRF and ASCF in one step. As mentioned above, the most reliable reconstruction should include all SPF, PPRF, and ASCF. Our prosthesis was designed to integrate SPF, PPRF, and ASCF together and to provide a comprehensive and stable reconstruction. On the other hand, the prosthesis was connected to pelvis and lumbar by screws and rods through holes and screw heads reserved on the prosthesis, which is much more convenient and simpler than the reconstructive methods reported in literature (e.g., double titanium cages or double fibula autograft). (2) Its porous surface could induce bone ingrowth and might

enhance stability. We considered that it could be one options for reconstruction after total en bloc sacrectomy.

However, there was an instrumental failure occurred at 8 months after surgery without impairment to the quality of life and ambulation. The breakage of screws might attribute to the insufficient screw placement and/or short lumbar fixation. Patient showed no pain or spinopelvic instability, which might attribute to the bone in-growth on the prosthesis-bone interface and biological sling formed by muscles and scar tissue between the pelvis and the spine. More research is warranted focusing on the modification of locations, diameters, and quantity of screws and biomechanical characteristics. The long-term functional and bone in-growth outcome will be followed to validate the use of the prosthesis.

Our report is of only one case; however, we propose that it could be one of the options for the reconstruction after total en bloc sacrectomy.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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