Customized "Whole-Cervical-Vertebral-Body" Reconstruction After Modified Subtotal Spondylectomy of C2-C7 Spinal Tumor Via Piezoelectric Surgery

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BACKGROUND: Radical resection is the first-line option in managing cervical primary chondrosarcoma. Favorable anterior reconstruction is challenging after multilevel total spondylectomy in the cervical spine.

OBJECTIVE: To illustrate the application of piezoelectric surgery and three-dimensional (3D) printing techniques in spine surgery.

METHODS: A 27-yr-old patient was referred to our center with complaints of nocturnal neck pain and right upper extremity weakness. A 2-stage radical tumor resection was conducted using piezoelectric surgery with pathologically tumor-free margins. A 3D-printed titanium microporous prosthesis (3D-PTMP) was designed to reconstruct the anterior column of the cervical spine between C1 and T1 for stability.

RESULTS: The whole intraoperative blood loss was 2300 mL over the 2 procedures. The patient had an uneventful recovery, regaining ambulatory status 3 wk after the 2 operations without ventilator support or other severe complications. By the final 14-mo follow-up, the patient had achieved marked pain relief and favorable neurological improvement; a postoperative computed tomography scan indicated a good position of the 3D-printed construct between the endplates with no sign of tumor recurrence or implant subsidence.

CONCLUSION: The applications of piezosurgery in total spondylectomy and in 3D-PTMP in reconstruction can be a favorable alternative for managing multilevel cervical spinal tumors. Further studies are warranted to validate this surgical strategy.

KEY WORDS: "Whole-cervical-vertebral-body" reconstruction, 3-dimensional printing, Chondrosarcoma, Subtotal spondylectomy, Piezoelectric surgery

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Chondrosarcoma (CHS) is the second most common primary tumor of bone, accounting for approximately 25% of all primary cases. CHS is a cartilaginous malignancy that can derive from normal bone or be secondary to benign neoplasms (eg, osteochondroma). CHS derived from the mobile spine is rare, accounting for approximately 6.5% to 10% of CHS.[1-3] The World Health Organization (WHO) grade, tumor size, and extent of disease are independent survival determinants for CHS patients.[1,4] Due to its relative resistance to radiotherapy and chemotherapy,[1,5-7] radical resection, another important factor affecting survival, remains the treatment of choice for localized spinal CHS. Because of complex cervical anatomy including the trachea, esophagus, and significant neurovascular tissues, total tumor removal in an "en bloc" fashion is technically challenging, especially for patients with multilevel large-size spinal tumors.[3,8-10]

ABBREVIATIONS: CT, computed tomography; **3D-PTMP,** three-dimensional-printed titanium microporous prosthesis; **CHS,** chondrosarcoma; **JOA,** Japanese Orthopaedic Association; **VA,** vertebral artery; **VAS,** Visual Analogue Scale; **WHO,** World Health Organization

The safety and effectiveness of piezoelectric surgery (based on the "piezoelectric effect" of microvibrations) has been reported in spine and hand surgeries.[11,12] This form of surgery protects the adjacent soft tissue by reducing blood loss, and the use of a piezoelectric ultrasonic osteotome makes it possible to efficiently obtain more regular margins, allowing better implantation. Three-dimensional (3D) printing techniques have been utilized for a number of years as a feasible method for reconstructing spine stability after large spinal tumor excisions. Due to their favorable mechanical strength and osteoinductivity, 3D-printed implants are highly useful in reconstruction after spinal tumor resection.[13-16]

CLINICAL PRESENTATION

A 27-yr-old patient weighing 135 kg presented to our center with primary complaints of right upper extremity weakness and repeated nighttime pain involving the posterior neck (8 to 9 out of 10 on the Visual Analogue Scale [VAS]) over the past year. These symptoms did not worry the patient until she developed astasia and motor dysfunction of the right upper limb. Physical examination revealed obvious cervical tenderness with a muscle strength of grade 1, high muscular tension of the right upper arm, hyperactive patellar and Achilles tendon reflexes and positivity of the right Hoffmann sign. Radiographic images and magnetic resonance images (MRI) indicated a very large tumor mass with ossification involving C2-C7, leading to an obvious compression of the right cervical nerve roots and epidural spinal cord (Figure 1). Vertebral artery (VA) angiography revealed abundant blood supply to the tumor mass. The right VA was infiltrated by the tumor and was much narrower than the left VA (dominant side; Figure 2). A balloon-occlusion test was performed to evaluate the feasibility of 1-sided VA sacrifice. Based on the patient's tolerance and collateral blood flow, the imaging results indicated that the right VA could be ligated if necessary. No metastatic lesion was detected via positron emission tomography-computed tomography. The Weinstein-Boriani-Biagini classification[17] of the most severe segment (C5) indicated the involvement of sector 1 to 3 and 7 to 12, layer A-D. The patient received a score of 14 on the Spinal Instability Neoplastic Score[18] and a score of 7 on the Japanese Orthopaedic Association (JOA) scale.[19] A 2-cm incision open biopsy was performed, yielding enough neoplastic tissue to immediately confirm its pathology, which indicated CHS with a WHO grade of II. The lesion was classified as IIB according to the Enneking Staging system. The tumor-infiltrating area was measured with the help of a radiologist based on the MRI, CT reconstruction, and CT angiography data. The 3D-printed construct was designed to be proximally fan-shaped to connect with the anterior arch of the atlas. Three duplications of slightly different sizes were prepared in advance to guarantee optimal placement.

Stage I: Posterior Approach Total Laminectomy with Screw-Rod Fixation

To maximize patient safety and therapeutic outcome, a 2-stage surgical procedure was identified to manage this case. Occiput-cervical fusion was selected to maximally reconstruct

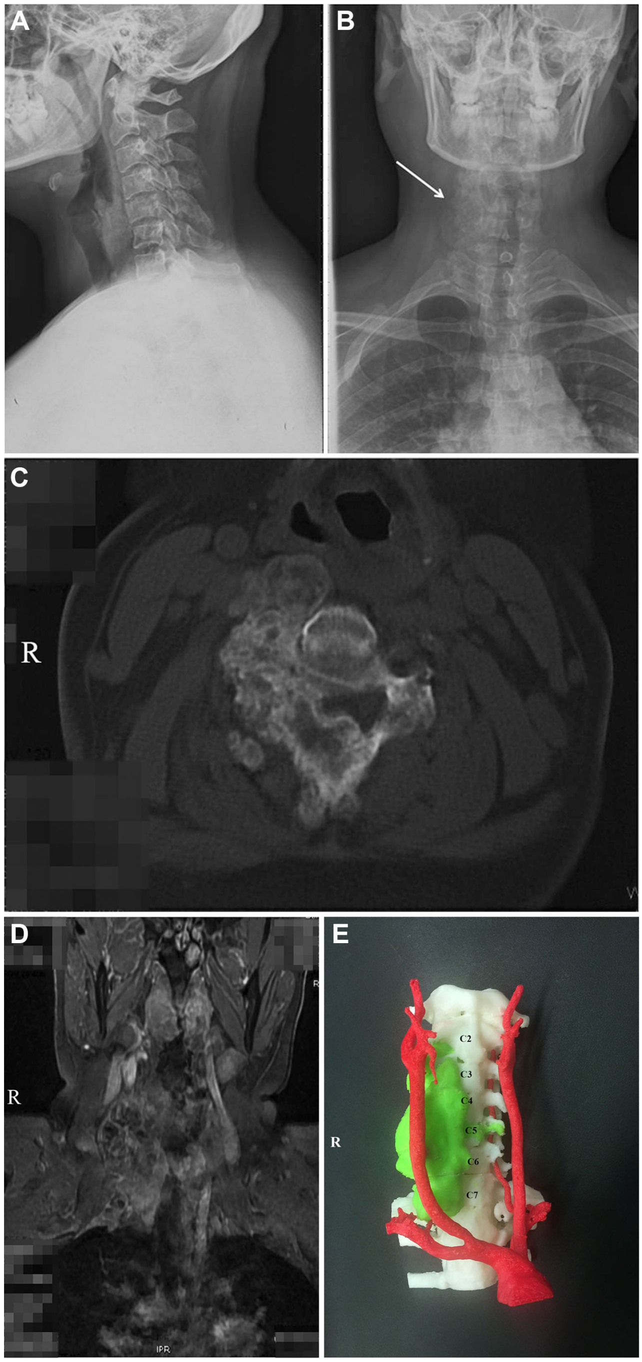

FIGURE 1. *Preoperative images of substantial cervical tumors involving C2-C7.* **A** *and* **B**, *White arrow on the X-ray indicates a substantial tumor mass.* **C**, *Obvious ossification was found intruding into the canal on the right side of the CT transverse section.* **D**, *No clear boundary was detected between tumor tissue and normal tissue; adjacent organs and tissue were compressed by a cancerous mass.* **E**, *3D-printed model (ratio 1:1) according to CT reconstruction and CTA data.*

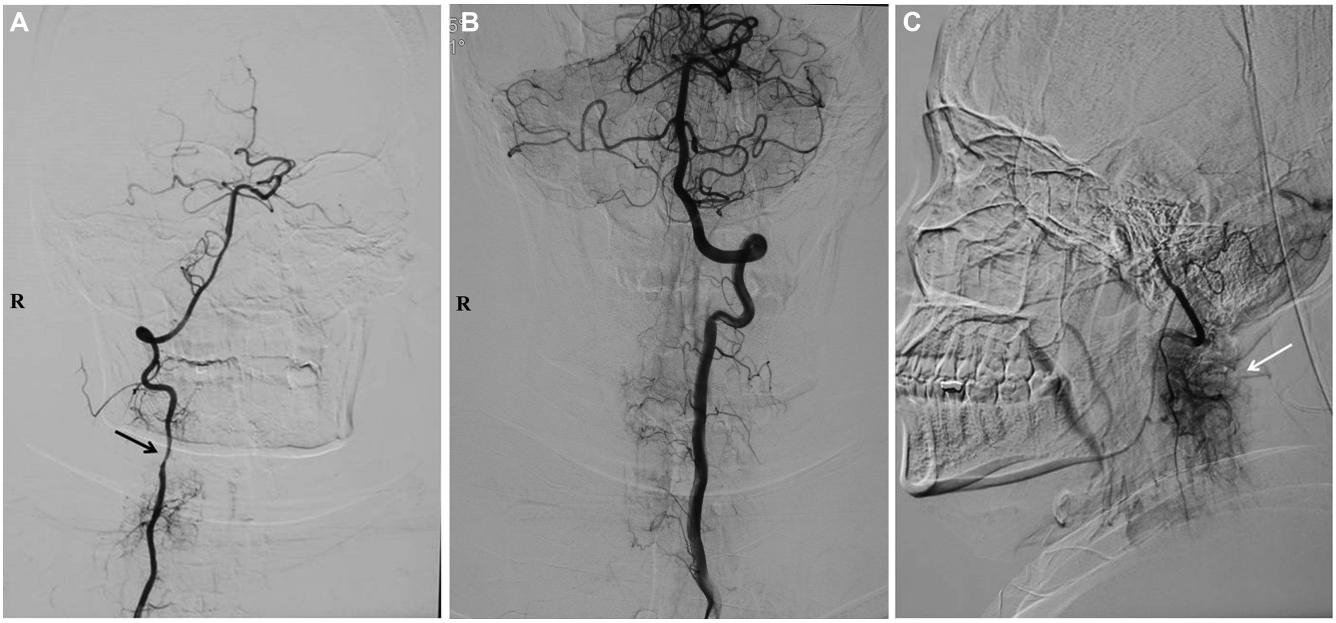

FIGURE 2. *Bilateral VA angiography.* **A** *and* **B**, *the right VA is narrower than the left VA (dominant side); the extreme narrow point is labeled by black arrow on Level C3/4, representing the tumor invasion.* **C**, *white arrow indicates an abundant blood supply of the tumor mass.*

cervical stability and maintain proper lordosis. Transpedicular or lateral mass screw fixation (DePuy Synthes Company, Raynham, Massachusetts) was performed through a posterior midline approach (covering the biopsy incision). Then, total laminectomy was performed using piezoelectric surgery (XD860A ultrasonic osteotome system, SMTP Technology Company, Jiangsu, China) to maximally remove the tumor mass involving the posterior column structures, which provided enough space to detach the spinal cord from the posterior longitudinal ligament. The right-side posterior elements invaded by the tumor were removed in an en bloc fashion. Moreover, right-side C3-C7 nerve roots were exposed and identified. Since the right VA was infiltrated by tumor tissue at Level C3/4 (which was shown to be relatively narrow in preoperative VA angiography), ligation of the right VA using titanium clips (Johnson & Johnson Company) was conducted at Level C3/4 to prevent extensive bleeding in the subsequent procedure (Figure 3A). The ligation of the right VA using a nonabsorbable suture (Johnson & Johnson Company) was performed later through an anterior approach to remove the tumor mass and involved VA as an integrity (Figure 3B). Cis-platinum (Haosen Pharmacy, Jiangsu, China) was administered for routine local infiltrating chemotherapy. Finally, sufficient compression was exerted on the screw-rod system to biomechanically maintain cervical lordosis. Total blood loss during the stage I operation was approximately 1900 mL without blood transfusion. The tracheal tube was not removed until 24 h after the first-stage operation in the event of asphyxia by laryngeal edema. Liquid food with high protein was allowed 36 h after surgery, followed by a gradual transition to solid food in 48 h. Cefuroxime (Glaxosmithkline manufacturing, Hounslow, United Kingdom) was used for routine infection prevention.

Stage II: Anterior-Submandibular Approach Total Tumor Excision with "Whole-Cervical-Vertebral-Body" 3D-printed microporous titanium prosthesis Reconstruction

After uneventful recovery for a week, the patient underwent a secondary surgery through a high anterior retropharyngeal approach. The incision was parallel with (2 cm below) the mandible, and the curve was made down across the submandibular and carotid triangle. Muscular tissue and complicated neurovascular structures were gently isolated and retracted, and the right VA (V1 section) was identified and ligated by nonabsorbable suture. After accurate vertebral positioning, piezoelectric surgery was utilized to remove tumor tissue in an en bloc fashion along the left medial border of C2-C7, leaving a margin around the tumor mass of at least 0.5 cm (Figures 3C, 3D, and 4A). The T1 superior endplate was decorticated using curette, and the C1 inferior articular surface was exposed completely for further fixation. A high-speed drill (Stryker) and curette were further used to reach normal osseous structure (Figure 4B). Gelfoam (Jinlin Pharmacy, Jiangsu, China) and a bipolar coagulator (Vedeng Company, Jiangsu, China) were used for bleeding control due to the defect by the resected tumor and epidural venous plexus. Then, a customized 3D-printed microporous titanium prosthesis

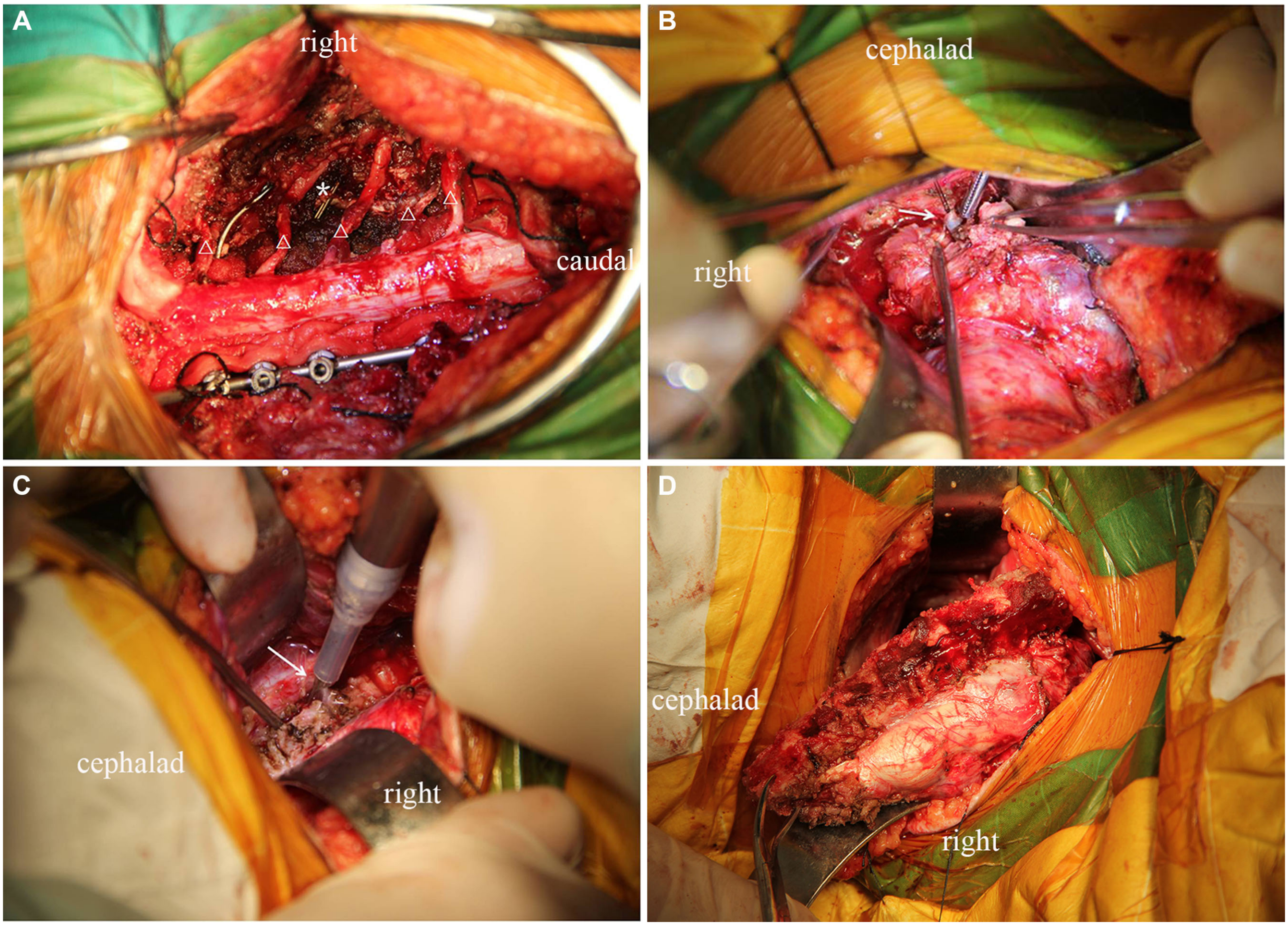

FIGURE 3. *Intraoperative images of a 2-stage operation.* **A**, *The cervical roots were exposed completely (Δ), and the right VA was ligated by titanium clips (*) due to tumor involvement.* **B**, *both ends of the right VA were ligated using a nonabsorbable suture (white arrow).* **C**, *Piezosurgery was utilized for better cancerous vertebral resection and soft tissue protection.* **D**, *The tumor mass was completely removed.*

(3D-PTMP) with an artificial spinal biomembrane (ZH-BIO Company, Shandong, China) was adopted to reconstruct spinal stability. The construct was modified with a slight lordosis to maintain biomechanical balance. Two short screws were fixed from the proximal wing into the C1 lateral mass through the inferior articular surface. The distal 3D-PTMP was fixed via conventional screws (DePuy Synthes). Notably, a special screw was fixed into the T1 vertebra at an oblique angle from anterior-superior to posterior-inferior (Figures 4C and 4D). This step was taken to prevent the dislocation of the interface by decreasing the probability of rotation. An absorbable suture was used to suture each individual layer. The whole blood loss during the second operation was approximately 400 mL, and 1000 mL of red blood cell suspension was recommended due to the large surgical field and the likelihood of oozing during the perioperative period. A nasointestinal tube was placed appropriately under fluoroscopic guidance to provide sufficient nutritional support and deliver Sulperazone (Pfizer Pharmacy) for infection prevention.

Outcomes

The entire 2-stage operation proceeded favorably despite the patient's 135 kg weight. No severe complications occurred during the perioperative period. The patient received oxygen support with 2 to 4 L per minute through a nasal tube in the 3 d (first day through tracheal tube) after the 2 surgeries. No artificial ventilator was utilized after assessing the patient's voluntary respiration and measuring blood oxygen saturation levels via pulse oximetry. Axillary temperature remained stable below 37°C; thus, the antibiotic was transitioned from sulperazone back to cefuroxime 5 d after the second operation. The nasointestinal tube was not withdrawn until the patient passed the peak period of esophageal swelling a week later. The 2 drainage tubes were removed when

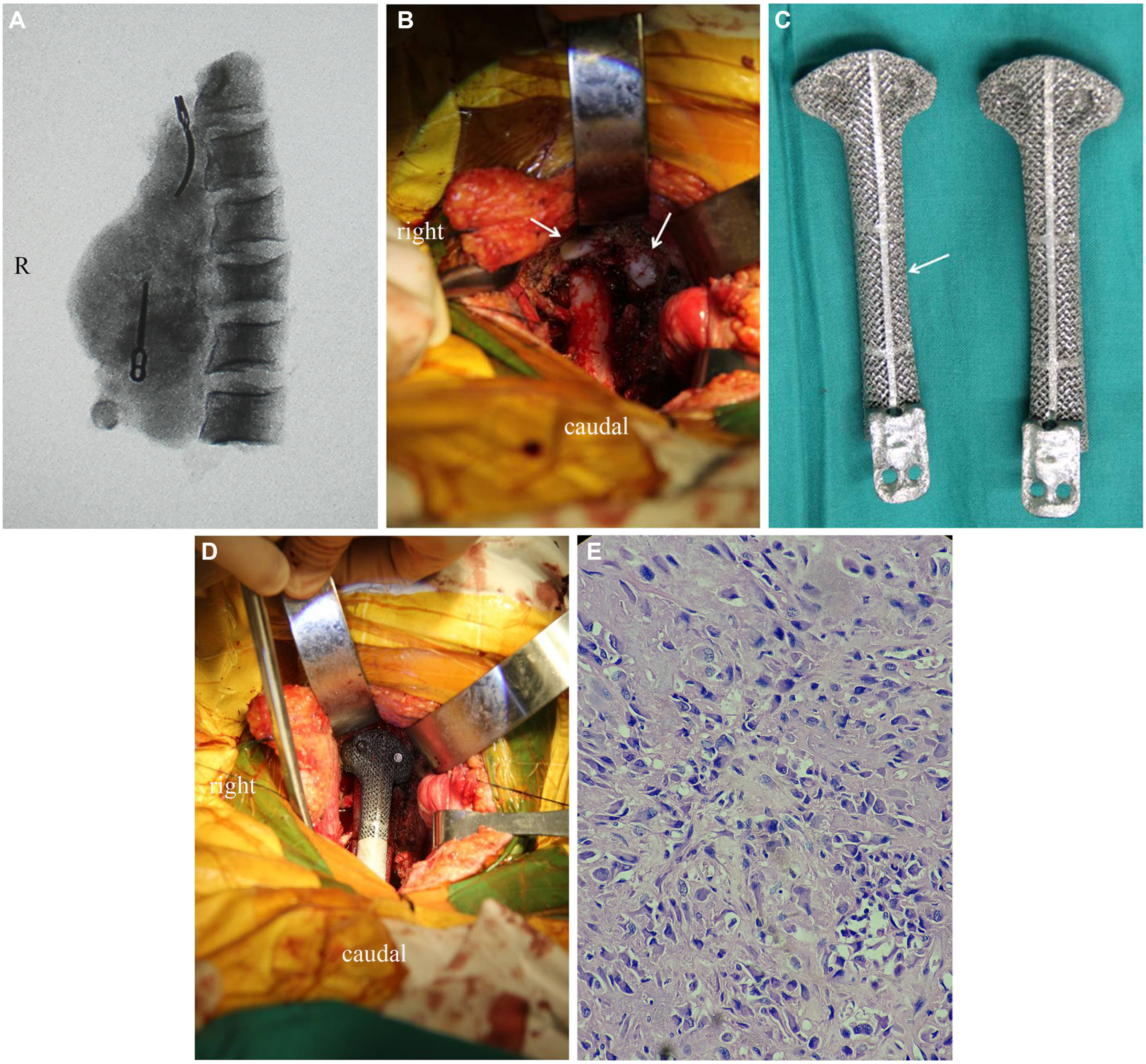

FIGURE 4. *Intraoperative images of a second-stage operation and pathological micrographs.* **A**, *Intraoperative X-ray of the tumor mass.* **B**, *Exposure of the C1 inferior articular surface (white arrow).* **C**, *Images of 3D-PTMP. i: amicroporous structure resembling natural trabecula (white arrow) and an oblique channel for third vertebral screw insertion; ii: bilateral access for C1 lateral mass screw insertion; iii: screws were inserted into the bilateral mass through the proximal wing of 3D-PTMP. iv: micrographs of hematoxylin-eosin-stained sections of CHS (100×).* **D**, *bilateral access for C1 lateral mass screw insertion through the proximal wing of 3D-PMTP.* **E**, *micrographs of hematoxylin-eosin-stained sections of CHS (40×).*

the amount of blood loss did not exceed 50 mL over 2 consecutive days. The suture in the back was removed pursuant to satisfactory wound healing 10 d after surgery. Pain relief was effective with VAS descending from 8-9 to 0-1. Muscle strength in the right arm was upgraded to grade 4 without any weakness in the forearm or hand at the final 6-mo follow-up. The JOA score was upgraded to 13, with a 60% rate of improvement ([postoperative score − preoperative score]/[17 − preoperative score] × 100%). The histological findings confirmed CHS (Figure 4E) 1 wk after the operation.

Notably, the patient was able to stand still and ambulate 3 wk postoperatively with the protection of the individualized

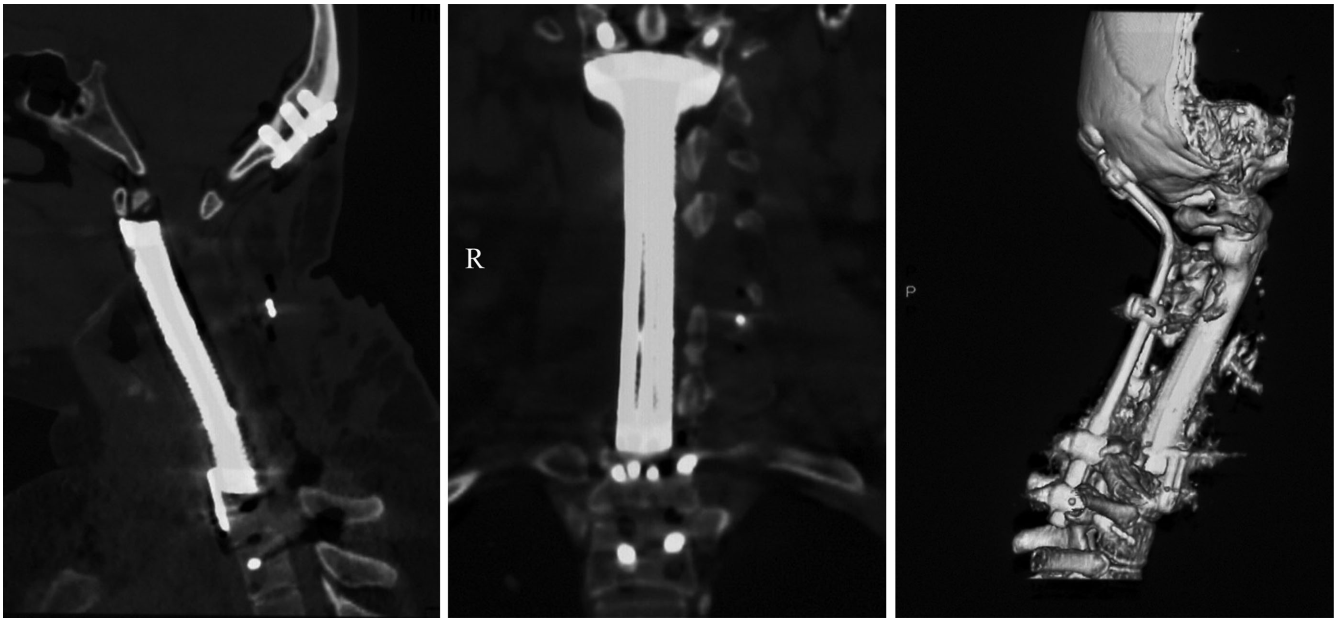

FIGURE 5. *Radiographic images at the 12-mo follow-up: the sagittal, coronary, and 3D CT images revealed a good position of the implant with no sign of subsidence.*

orthosis. The final 12-mo radiological follow-up revealed that the implant was in a good position without signs of hardware failure (Figure 5). Routine radiotherapy was scheduled. The patient was grateful to be able to lead an independent life and go back to work at full capacity by the final follow-up of 14 mo.

Ethical Approval

Informed consent was obtained from the patient included in the study. All procedures performed in studies involving human participants were in accordance with the ethical standards and approved by the hospital ethical committee.

DISCUSSION

CHS arising from the cervical spine is relatively rare. Effective surgical treatment is the optimal choice for such primary malignancies because of insensitivity to adjuvant palliative therapies.[1,3,5-7,20,21] The long-term prognosis of patients undergoing radical resection has been reported to be more favorable than that of patients who received subtotal removal, tumor debulking or resection with positive surgical margins.[2,3,22,23] However, en bloc resection of cervical spine tumors is quite challenging and is rarely feasible considering the unique anatomical complexities of the cervical spine. To date, only a few cases have been reported to receive en bloc resections with favorable outcomes, and the extent of CHS in those cases has been limited to posterior elements and/or less than 2-segment invasion.[2,3] Lee et al[24] successfully performed a subtotal en bloc spondylectomy of C6 CHS (Enneking Staging IIB). However, there have been no reports of en bloc resection of CHS involving more than 3 cervical vertebrae. Multilevel total spondylectomy is preferred due to the positive functional outcome, favorable survival rate, and promising local control despite its higher complication rate. Additionally, correction of cervical lordosis after tumor resection is important to maintain sagittal stability and biomechanical balance. The present case was selected for this report to illustrate the potential for the radical tumor resection of C2-7 CHS and to show that reconstruction after radical resection is feasible and effective using 3D printing techniques.[25]

The Application of Piezoelectric Surgery in Spinal Tumor Surgery

Piezoelectric surgery has been utilized frequently for osteotomy, as seen in Figure 3C. Widely applied in craniofacial and stomatological surgery, piezoelectric surgery has been reported to provide safe, precise and high-probability bone cutting with the promise of protecting adjacent soft tissue.[12,26] Compared with conventional rotary osteotomy, piezoelectric surgery provides the following advantages:

a. Piezoelectric surgery is capable of providing more regular margins for better implantation.[26,27]
b. The utilization of piezoelectric surgery saves time with less blood loss than rotary osteotomy. Piezoelectric surgery works on the basis of the "piezoelectric effect" caused by microvibrations.[28,29] Thus, it enables spine surgeons to attain the maximum preservation of soft tissue including spinal cord, thecal sac, epidural fat and ligaments.

c. The piezoelectric surgery device offers water cooling artificially during surgical procedures, while rotary osteotomy requires an additional assistant to provide water cooling.
d. Unlike conventional RB, the piezoelectric surgical device can function normally in the presence of the gelfoam and brain cotton that are commonly used in spine surgery.

To date, only a few studies have focused on the piezoelectric surgery methods used in spine and hand surgery.[11,12] The present case illustrates the feasibility and benefits of piezoelectric surgery usage in cervical spinal tumor surgery. However, piezoelectric surgery methods should be employed with care by an experienced surgeon because misuse and inexperience may cause severe complications, which may affect the prognosis.

Description of 3D-PTMP

The application of 3D-printed implants has previously been reported in reconstruction after cervical tumor resection.[13,14] Compared with artificial vertebrae and conventional 3D-printed implants, 3D-PTMP has some advantages as follows:

a. 3D-PTMP was designed using 3D computing to integrate conventional metal plates and artificial vertebral bodies based on the cervical physiological curve index. In addition, with respect to the cervical zero-profile screw system,[30] the notch was placed relatively low to prevent foreign body sensation or even dysphagia.
b. The distal fixation of 3D-PTMP was assigned as "3D screw insertion," which added an oblique channel for another screw fixation. This addition enhances the antirotation and anti-pull-out properties of the implant (Figure 5).
c. The "successive-layer" design provides multipoint support compared to titanium mesh, thus reducing the subsidence rate of prosthesis.[31]
d. The porous microstructure, originating from titanium alloy powder, resembles the natural trabecula to allow the adjacent osteoblast in-growth to achieve bony fusion ideally. This intrinsic osteoinductivity overcomes the problem of autogenous bone insufficiency and eliminates the need for unnecessary substantial allografts.[13,14]

With the progress in material technology and reductions in cost, 3D-PTMP has become a good option for spinal tumor surgeons, especially for achieving successful reconstruction after multisegmental tumor resection.

CONCLUSION

For patients with multilevel substantial cervical tumors, radical resection via piezoelectric surgery with the use of 3D-printed implants for reconstruction should be strongly considered by spine surgeons.

Disclosures

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