Case Reports

Three-dimensional printing-assisted anterior and posterior combined surgery for treating a giant aggressive vertebral hemangioma

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Abstract
Aggressive vertebral hemangioma (AVH) is a type of non-neoplastic and congenital developmental abnormality of spinal cord blood vessels. We report the innovative application of three-dimensional (3D) printing-assisted anterior and posterior combined surgery for treating a giant AVH. This could be a novel treatment in the future. A 44-year-old man suffered from persistent neck pain and limited limb mobility for approximately 2 weeks. An imaging examination showed the destruction of C2–4 vertebral bodies, and a giant lesion invaded the spinal cord. He underwent 3D printing-assisted anterior and posterior combined surgery. Postoperatively, his symptoms of persistent neck pain and limited limb mobility were alleviated. An imaging examination showed that internal fixation and the prosthesis were fixed in place, and the spinal canal was unobstructed. Treating a giant AVH by 3D printing-assisted anterior and posterior combined surgery is feasible and effective.

Keywords
Three-dimensional printing, anterior and posterior combined surgery, spinal cord, vertebral hemangioma, giant lesion, neck pain

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**Introduction**

Aggressive vertebral hemangioma (AVH) is a non-neoplastic and congenital developmental abnormality of spinal cord blood vessels, and it is more common in intramedullary cervical and thoracic segments. Aggressive hemangioma is also known as Gorham–Stout syndrome, but its etiology is still unclear. Some scholars have suggested that aggressive hemangioma may be related to bone resorption caused by the presence of lymphoma or hemangioma of the bone. An AVH is often accompanied by severe neurological dysfunction, and emergency surgery should be performed to remove the lesions. Currently, total laminectomy is the most commonly applied surgical method for an AVH in the clinic. This method requires the stripping of bilateral paravertebral muscles and extensive removal of the posterior lamina and ligaments. Although the lesion can be fully exposed and removed, it has a certain effect on postoperative spinal stability. We report here innovative application of three-dimensional (3D) printing-assisted anterior and posterior combined surgery for treating a giant AVH. This could be a novel treatment in the future.

**Case presentation**

The reporting of this study conforms to the CARE guidelines. A 44-year-old man initially presented with persistent neck pain and limited limb mobility for approximately 2 weeks. A physical examination on admission showed that the left neck of the patient was swollen, and a soft fixed mass was faintly palpable, accompanied by tenderness. Additionally, the cervical function of flexion, extension, and rotation was limited. The Frankel classification was evaluated as E-class. An imaging examination showed the destruction of C2–4 vertebral bodies, and a giant lesion invaded the cervical spinal cord (Figure 1a–e). A biopsy of lesion tissue under ultrasound guidance showed a vertebral hemangioma (Figure 1f). On the basis of these findings, the patient was diagnosed with an AVH. Moreover, paralysis of the right limb occurred on the second day after the patient was admitted to our department, and active surgical intervention was urgent.

Effectively treating this case by a conventional surgical method was difficult because the AVH was extremely large and surrounded the vertebral artery. To maximize the removal of the lesion and maintain the stability of the whole cervical spine, the patient was scheduled to undergo 3D printing-assisted anterior and posterior combined surgery. In the first stage of surgery, we completed partial resection of the AVH via the posterior approach and performed subsequent decompression fixation using screw-rods at C1–6 (Figure 2a–d) to save nerve function and stabilize the cervical spine. One month after the first stage of surgery, the neurological function of the patient was good. Before the second stage of surgery, we used computed tomography (CT) scan data to reconstruct the overall 3D model of the patient’s diseased vertebral bodies and designed a personalized prosthesis through Mimics19.0 software (Materialise, Leuven, Belgium). Specifically, we conducted a virtual reset to restore the physiological curvature and the height of the diseased vertebral bodies. A personalized prosthesis was then manufactured by a 3D printer (Dongwang Technology, Inc., Xi’an, Shaanxi, China) and further simulated for implantation (Figure 2e–i). Subsequently, in the second stage of surgery, the patient underwent the operation of anterior cervical corpectomy decompression and fusion at C2–3 and the operation of anterior cervical discectomy and fusion at C4–5. The personalized prosthesis was further implanted after the autogenous iliac bone was filled (Figure 2j). Ultimately, the internal fixation
Position and spinal sequence were confirmed by C-arm fluoroscopy.

Postoperatively, his symptoms of persistent neck pain and limited limb mobility were considerably alleviated. A postoperative imaging examination showed that the internal fixation and prosthesis were fixed in place, and the spinal canal was also unobstructed (Figure 2k, l). To date, the patient has been followed up for 15 months, and an imaging examination showed that the internal fixation was stable (Figure 2m, n). Additionally, there was no apparent limitation of the mobility of the cervical spine and limbs.

Discussion

Most AVHs originate from the vertebral body and extend to the epidural space, while vertebral hemangiomas occurring in the spinal canal are relatively rare, accounting for only 5% to 12% of AVHs. AVH is a type of neovascular tumor-like hyperplasia. AVHs are composed of thin, elastin-deficient or smooth muscle-deficient blood vessels. Moreover, AVHs can occur in the entire central nervous system, or in the spine, pelvis, scapula, liver, spleen, skin, or other tissues and organs alone or at the same time. Additionally, AVHs usually
manifests as progressive spinal cord dysfunction. At the beginning of the disease course, there may be local radicular pain, limb weakness, and sphincter dysfunction. Paroxysmal neurological dysfunction and different degrees of recovery during the disease is the typical course of an AVH.\(^6\)

A magnetic resonance imaging examination is important for the diagnosis of AVH. Observation by dynamic magnetic resonance imaging can show changes in the volume of the lesions. Because of acute or chronic dilation of blood vessels, when the volume reaches a certain degree, it might be complicated by bleeding due to rupture. This bleeding can occur repeatedly, and the neurological symptoms can last for several days. Therefore, distinguishing the clinical symptoms from extramedullary benign tumors is difficult.

AVHs without symptoms of spinal cord dysfunction usually do not require special treatment, and active surgical interventions should be performed in patients with symptoms, especially with neurological deterioration.\(^7\) Moreover, AVHs are mostly located in the lateral dorsal side of spinal dura mater, with a layer of the glial band as
the boundary with the normal spinal cord, and there is no obvious blood supply by an artery and a drainage vein. The capsule is complete, the boundary is clear, and there is no obvious adhesion with the dura mater. Therefore, an accurate intraoperative tumor location and proper application of bipolar electrocoagulation can often completely remove the lesion without injury to the spinal cord. Additionally, the local hematoma can be removed at the same time.

Our patient with a giant AVH had severe neurological symptoms, which required active and timely surgical intervention. The partial resection of the lesion via the posterior approach and corresponding decompression and fixation provided sufficient time to prepare for the 3D-printed prosthesis. A personalized 3D-printed prosthesis is a novel idea for reconstructing the stability of the cervical spine. In this process, with the assistance of 3D-printed guide plates, we successively implanted an anterior cervical pedicle screw (C3 right screw) and anterior atlantoaxial lateral mass screws in our patient, which provided a reliable fixation of the prosthesis. The correct placement of the prosthesis is important, and it determines whether the position of the anterior lateral mass screw is accurate. To achieve this placement, adequate surgical exposure and clearance of the lesion needed to be consistent with preoperative planning. We observed that the lesion tissue covered a large area and the bone destruction was severe. Posterior cervical C1–6 right lateral mass joint fusion and anterior artificial vertebral body fusion could only be used. To improve the fusion rate and increase the stability of the artificial vertebral body, anterior C4–5 interbody fusion and fixation were added. Ultimately, although this case of a giant AVH was not able to be resected in a single and en-bloc piece, after 3D printing-assisted anterior and posterior combined surgery, the prognosis of the patient was good.

**Conclusion**

Treating a giant AVH by 3D printing-assisted anterior and posterior combined surgery is feasible and effective. An operation of the posterior approach can remove part of the lesions. Additionally, an operation of the anterior approach is able to remove the remaining lesions and implant the prosthesis to maximize the stability of the cervical spine and improve the prognosis of the patient.

**Ethics statement**

This study was conducted in accordance with the declaration of Helsinki and was approved by the Ethics Committee of Jiangxi Provincial People’s Hospital Affiliated to Nanchang University (approval number: 8187306817). Written informed consent was obtained from the patient.

**Declaration of conflicting interest**

The authors declare that there is no conflict of interest.

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