

A novel technology for 3D-printing artificial vertebral bodies for treating cervical spine cavernous hemangioma and cavernous lymphangioma mixed tumors: A case report

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Abstract

Background: Cavernous hemangioma and cavernous lymphangioma mixed tumor is a rare, complex tumor with minimal spine involvement.

Case description: A 34-year-old female patient presented with neck pain 6 years ago without obvious cause, combined with radiating pain in both shoulders and then developed obvious numbness and weakness in both upper limbs and a feeling of stepping on cotton in the lower limbs. MRI showed C7 vertebral hemangioma, which was not relieved well after conservative drug treatment, and the patient felt that neck and shoulder pain seriously affected his life recently. The anterior route sub-completely removed the C7 vertebral body, part of the vertebral body was taken for pathological biopsy, the appropriate 3D printed porous titanium alloy vertebral body was selected and filled with allogeneic bone and then placed into the excised vertebral space, and after the filling was satisfactory, the 6-hole steel plate was selected and placed in front of the neck 6-thoracic 1, and fixed with 4 screws. The postoperative results were satisfactory and there were no perioperative complications.

Conclusion: Cavernous hemangioma and cavernous lymphangioma mixed tumors involving the cervical spine are rare, and difficult to treat, and the symptoms caused need to be differentiated from cervical spondylosis, which can be differentiated by imaging at an early stage. After the preoperative surgical evaluation, the total tumor and subtotal vertebral body resection, and artificial vertebral implantation were performed during the operation, and the biomechanical stability of the spine was reconstructed through the internal fixation system, and satisfactory results were obtained.

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Keywords: Cavernous hemangioma; Spongy lymphangioma; Mixed tumors; 3D-printed artificial vertebral body; Subtotal vertebral body resection.

Introduction

Cavernous hemangioma and cavernous lymphangioma mixed tumor is a rare benign tumor, and cavernous hemangioma and cavernous lymphangioma mixed tumor is caused by abnormal production of blood vessels and lymphatic vessels during the developmental stage [1]. From a pathological point of view, it contains both dilated lymphatic vessels and blood vessels, filled with lymphatic fluid and blood, and separated by fibrous tissue into cavities of various shapes and sizes. Cavernous vascular lymphangiomas tend to occur in soft tissues such as the head and neck, limbs, chest wall, and abdominal wall, but also in the pubic area, vulvogenitals, and anorectum, and cases that occur in the vertebrae of the spine are very rare [2]. In this case report, we describe a patient with cavernous hemangioma and cavernous lymphangioma mixed tumors in the 7th cervical vertebrae and review the literature to discuss their clinical features and treatment measures.

Case description

A 34-year-old female patient developed neck pain 6 years ago with no obvious cause, accompanied by radiating pain and discomfort in both shoulders, followed by numbness and weakness in both upper limbs, and a feeling of stepping on cotton in the lower limbs in the morning. One year ago, the patient felt that the neck and shoulder pain was significantly aggravated, and the left shoulder was significantly limited in movement due to pain, accompanied by dizziness, nausea, numbness, and weakness in the left lower limb, and walking in the morning was like stepping on cotton. Then he was treated in multiple hospitals, and an MRI showed "cervical 7 vertebral hemangioma", and after conservative treatment with drugs, the numbness and discomfort of both upper limbs and the feeling of stepping on cotton in the lower limbs were significantly alleviated. Recently, the patient felt that neck and shoulder pain seriously affected his life and came to our hospital for further treatment. The patient has an unsteady walking gait, tenderness in the spinous process, paraspinous process, and intervertebral space in the lower cervical spine, limited movement of cervical spine forward flexion, posterior extension, and rotation, radiating pain in the left upper limb induced during activity, and normal muscle tone in the limbs. The results of the muscle strength comparison examination are as follows (left/right): deltoid (3/4), biceps brachii (4/5), triceps brachii (3/4), extens carpi (4/5), flexor carpi (4/5), extensor digitorum (3/4), flexor digitoris (3/4), and lower extremity muscles (4/4). All reflexes were normal and negative for Hoffmann's, Babinski's, and Kernig's syndromes. Squamous Cell Carcinoma antigen (SCC) was elevated (2.10 ng/ml) on the full tumor panel. X-ray of the cervical spine showed no abnormalities. CT examination of the cervical spine showed the irregular hypodense shadow of the C7 vertebral body, and the strip-like hyperdensity showed a "fence sign". MRI of the cervical spine showed an abnormal signal of the C7 vertebral body, and enhancement MRI showed an abnormal enhancing shadow of the C7 vertebral body (Figures 1 and 2).

The patient was only 34 years old, had symptoms of cervical spondylosis for 6 years, and the imaging examination showed cervical 7 vertebral hemangioma, so the preliminary diagnosis was C7 hemangioma, and the pathological examination of the specimen confirmed that it was a mixed tumor of cavernous hemangioma and cavernous lymphangioma. After a multidisciplinary consultation, including oncology, interventional, and spine surgery, the decision was made to

remove the tumor surgically. The anterolateral approach of the neck was selected, the diseased vertebral body was sub-completely resected, and a 3D-printed porous titanium alloy artificial vertebral body was implanted.

After successful general anesthesia, the patient was placed in a supine position with the neck extended backward. Disinfect and spread towels. A transverse incision was made on the right side of the vertebral body plane of the flat neck 7, which was about 5 cm long. Slice by layer to reveal vertebral bodies C6, C7, and T1. The intervertebral discs of C6-C7 and C7-T1 were resected, and the complete tumor was removed, that is, the C7 vertebral body was sub-totally resected, and part of the tissue was sent for pathological examination (Figure 3). After using fluid gelatin and hemostatic yarn to completely stop the bleeding, select a suitable 3D printed porous titanium alloy artificial vertebral body and fill it with allogeneic bone (Figure 3), fill the vertebral body gap, select a 6-hole steel plate and place it in front of C6-T1, and fix it with 4 screws and lock it. The C-arm fluoroscopy fixation implantation position is good, the wound is irrigated and a drainage tube is placed, and the wound is sutured layer by layer. The anterolateral x-ray of the cervical spine was re-examined 3 days after surgery, and the anterior internal fixation position of C6-T1 was good, the artificial vertebral body of C7 vertebral body was in good position, and the intervertebral space was filled satisfactorily (Figure 4).

After surgery, histopathological examination confirmed the diagnosis of cavernous angioma and cavernous lymphangioma mixture, and the tumor tissue was composed of dilated blood vessels and lymphatic vessels of different sizes (Figure 5). Vital signs such as blood pressure and heart rate were normal after surgery.

Discussion

Cavernous hemangiomas and cavernous lymphangiomas are both benign tumors or developmental malformations that arise from mesenchymal tissues [3,4]. Cavernous hemangiomas tend to occur in the skull and orbits, and rarely occur in the vertebral body [5]. Cavernous lymphangiomas occur in the face, neck, and abdominal cavity, and rarely in the vertebral body [3]. Their mixed tumors occur in the vertebral bodies of the spine even more rarely. The symptoms of this type of mixed tumor depend mainly on the location and size of the occurrence, etc., and the main manifestations are soft and elastic lumps, which will compress the adjacent tissues and produce corresponding symptoms, such as headache, dizziness and seizures that occur in the skull [6]. In this case, symptoms that resembled those of cervical myelopathy were present in the vertebral body. Due to the few reports of cavernous hemangioma and cavernous lymphangioma mixed tumors, the incidence and recurrence rates are still unclear. Long-term research is still needed.

Cavernous hemangioma and cavernous lymphangioma mixed tumors need to be distinguished from cavernous hemangiomas and cavernous lymphangiomas. Cavernous hemangiomas are mainly composed of enlarged vascular lumens, most of which are venous malformations with varying lumen sizes, flattened endothelial cells, and a small amount of connective tissue around them [7]. Clinically, it mostly appears as a mass of tissue, with a soft texture and unclear borders, which can be dark red or blue-purple [8]. Cavernous lymphangioma consists of many small atrial cavities lined with endothelial cells and surrounded by a thin layer of connective tissue containing lymphatic fluid and dominated by lymphatic vessels [9]. Clinically, it usually

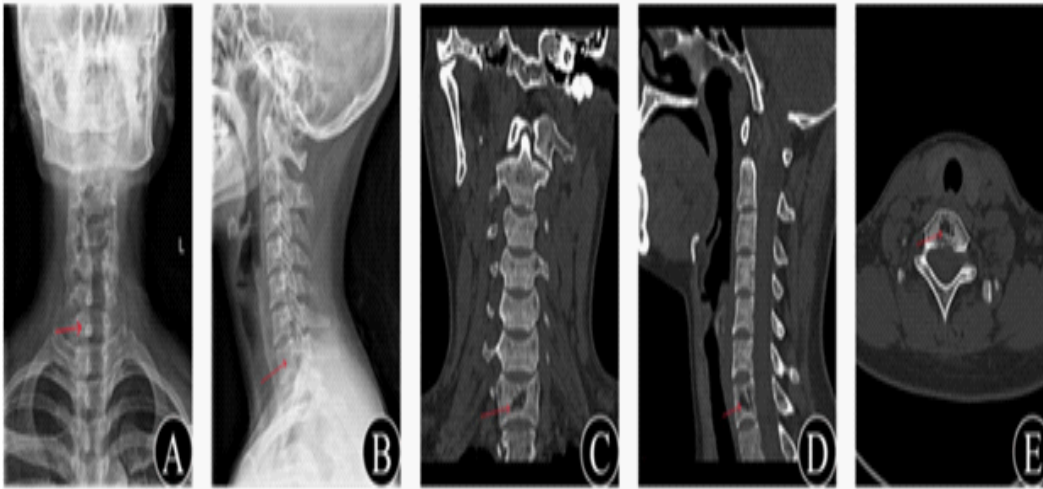


Figure 1: (A,B) X-ray of the cervical spine showed abnormal C7 transmittance in the anterolateral position. (C-E) CT of the cervical spine showed C7 irregular hypodense shadows, and strip-like hyperdensities were seen in them, showing a “fence sign”.

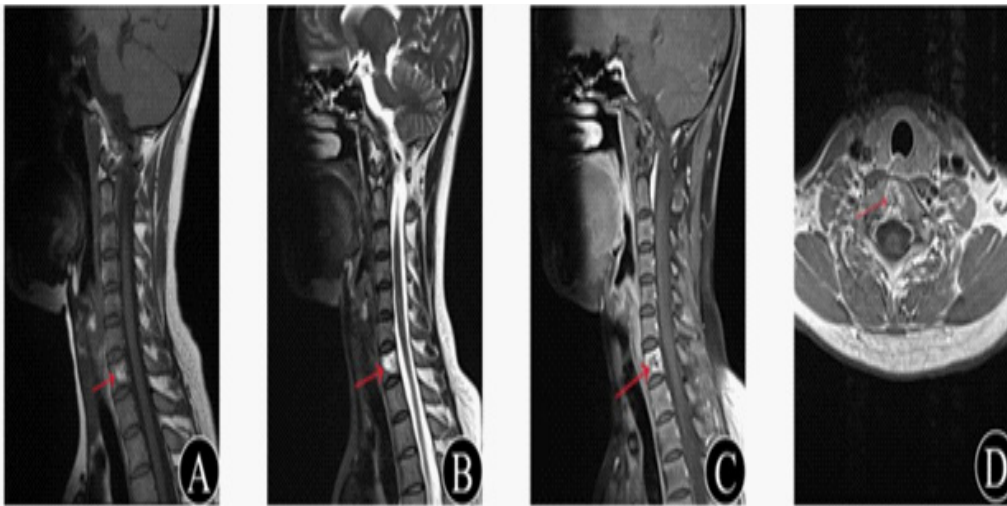


Figure 2: (A-D) MRI of the cervical spine showed abnormal C7 signal, patchy short T1 and long T2 signal shadows in the lesions, patchy long T2 signal shadows on contrast-enhanced MRI, and uneven enhancement on contrast-enhanced scanning.

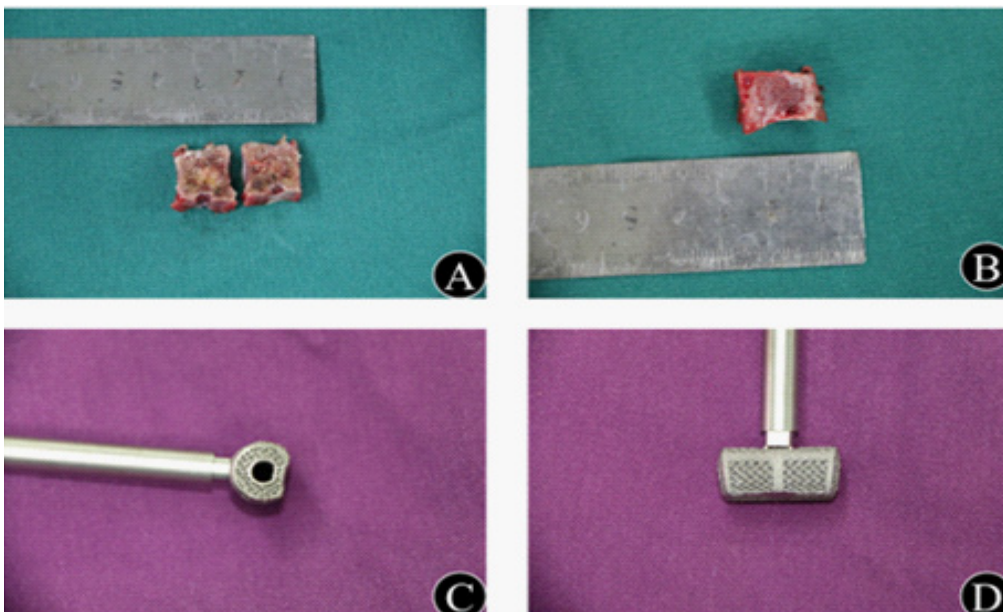


Figure 3: (A,B) The removed whole piece of bone tissue (volume: 3.5×1.8×1.5 cm) is detached from the middle, exposing the tumor tissue. (C,D) 3D printed titanium artificial vertebral body in anteroposterior and lateral positions

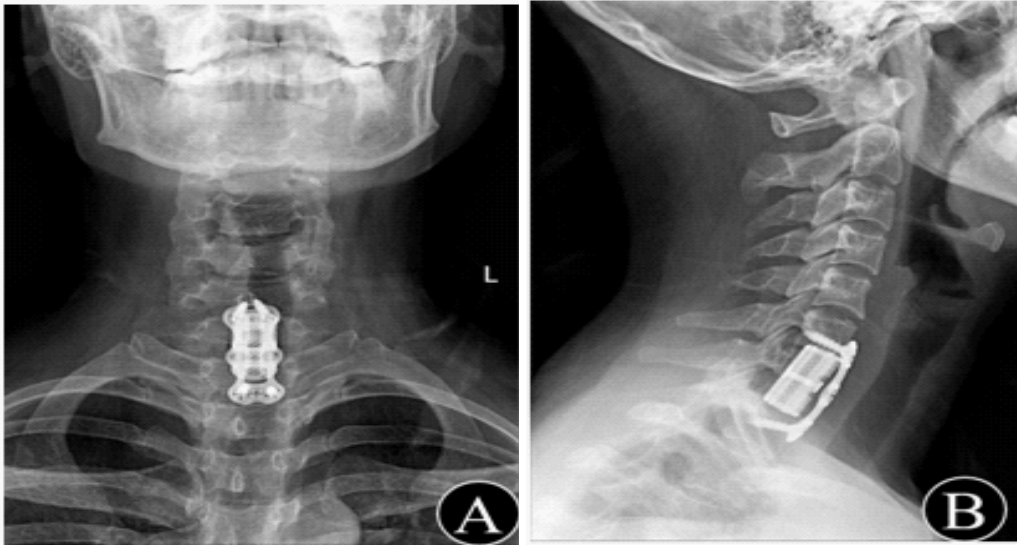


Figure 4: (A,B) The anterolateral X-ray of the cervical spine was re-examined 3 days after surgery, and the internal fixation position was good, and the intervertebral space was highly satisfactory.

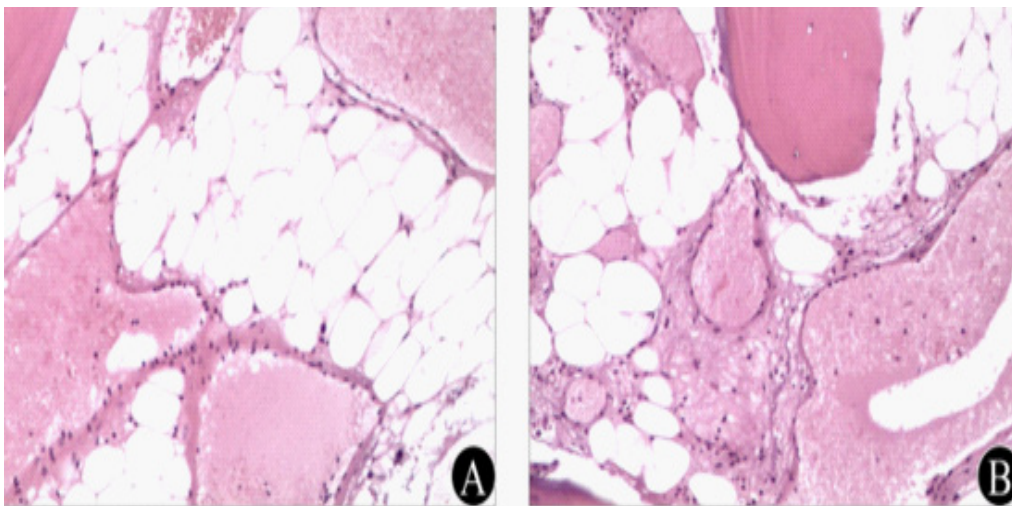


Figure 5: (A,B) Staining of pathological sections showed that the tumor was composed of dilated blood vessels and lymphatic vessels of various sizes.

manifests as a cystic mass of the skin or subcutaneous, pale yellow or light blue, with clear borders, soft texture, fluctuating feeling, and compressible, but does not change with body position [10].

Cavernous hemangiomas and cavernous lymphangioma mixed tumors are made up of dilated blood vessels and lymphatic vessels. These blood vessels and lymphatic vessels intertwine with each other to form a cavernous lacunar structure. Its cavities vary in size and are covered with flattened endothelial cells surrounded by fibrous connective tissue. Abnormal differentiation during development leads to this mixed malformation. The presence of red blood cells and lymph in the diseased tissue can be seen in the pathological section under the microscope, which is also one of the important bases for diagnosis.

Symptomatic treatment is the simplest measure to alleviate symptoms, and in this case, the patient was treated symptomatically with Non-Steroidal Anti-Inflammatory Drugs (NSAIDs) at the beginning of symptoms, and although the symptoms were relieved, the symptoms were masked and finally the symptoms became more severe until they seriously

affected life. Sandmann et al. [11] studied 265 patients with cerebral cavernous hemangioma treated conservatively, and showed that most patients behaved normally under conservative treatment, and 14 patients received intervention such as surgery halfway. Two patients died directly from cerebral cavernous hemangiomas. Therefore, conservative treatment can be used as an early treatment measure, but not as a long-term measure. Cryotherapy is the use of cryogenic agents such as liquid nitrogen to cause frostbite and necrosis of tumor tissues. It is suitable for small tumors, but it may leave scars, etc., and it is easy to recur [12]. Kolarkodi et al. [13] reported a case of tongue hemangioma and treated it with cryotherapy and sclerotherapy with good results. Laser therapy is the use of the laser to necrosis tumor tissue, which is suitable for superficial tumors. Laser therapy is also an effective option when surgery is not suitable or when medications are contraindicated [14]. Gamma Knife radiation therapy is also an effective non-surgical treatment, Myeong et al. [15] included 79 patients with cerebral cavernous hemangiomas who underwent long-term radiation therapy with Gamma Knife for a mean follow-up of 14 years and found that Gamma Knife radiation therapy had a good effect on cerebral cavernous hemangiomas, especially for tumors

with a history of previous bleeding and brainstem location. Punukollu et al. [16] also demonstrated its effectiveness and low complications by studying Gamma Knife radiation therapy for orbital cavernous hemangioma. Sclerosing agent injection is the injection of sclerosing agent into the tumor, causing the blood vessels and lymphatic endothelial cells of the tumor to die, thereby causing the tumor to shrink. Commonly used hardeners are pingyangmycin, bleomycin, etc [17]. Injections require multiple treatments to achieve the desired therapeutic effect, and adverse reactions of local pain and discomfort may occur after injections. Bi et al. [18] reported a patient treated with hepatic cavernous hemangioma by pingyangmycin Drug-Eluting Beads Transarterial Embolization (DEB-TACE), who experienced adverse reactions such as abdominal pain after drug injection, and further reduced the tumor by secondary injection, but did not completely eradicate the tumor, and no recurrence was found in the follow-up examination 9 months after surgery, and it is still uncertain whether it will recur in the future. Doppman et al. [19] injected ethanol directly into vertebral hemangiomas, and 5 of the 6 paraplegic patients recovered. All patients had complete disappearance. Surgical treatment has a good effect on radical resection, which can completely remove the tumor and reduce the possibility of recurrence. It is suitable for cases where symptoms are obvious and conservative treatment is not effective. For example, the patient, in this case, caused symptoms similar to cervical spondylosis, which seriously affected his life, and after the tumor was removed by surgery, the 3D printed artificial vertebral body was implanted, which significantly alleviated the patient's symptoms. However, surgical treatment is risky, especially subtotal resection of the cervical spine, which has certain difficulties and risks. Kobayashi et al. [20] reported a case of intraosseous hemangioma at the base of the skull and surgically removed the tumor, and showed that subtotal resection is the only feasible radical procedure for skull base cavernous hemangioma. Kerolus et al. [21] reported the first case of intradural and extramedullary spongy lymphoma involving the thoracic spinal cord, which was treated with incomplete resection and led to recurrence three months later. Finally, the treatment effect is good through the shunt of cystic fluid to the peritoneum, which also provides a new idea for tumors that are difficult to remove by surgery. Wang et al. [22] followed 14 patients who underwent vertebral body resection and 3D printed artificial vertebral implantation for an average of 20 months, and found no cases of artificial vertebral body or screw fixation failure. Except for a few cases with prosthetic sinking, there were no other adverse events, and all of them achieved good treatment results. This further demonstrates the feasibility of vertebral body excision and 3D-printed artificial vertebral implantation.

Conclusion

Cervical vertebral cavernous hemangioma and cavernous lymphangioma mixed tumors are rare, and there is no uniform standard for treatment. Early diagnosis can be differentiated based on imaging, and indications for surgery should be evaluated preoperatively. During the operation, the cone subtotal resection and 3D printing artificial vertebral body were implanted and fixed by pedicle screws, and satisfactory results were obtained. After surgery, close care should be provided, multidisciplinary coordinated treatment and the patient's symptoms should be significantly improved and discharged from the hospital smoothly. Therefore, 3D printing artificial vertebral implantation is an effective treatment for

the treatment of spinal vertebral cavernous hemangioma and cavernous lymphangioma mixed tumors.

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