Letter to the Editor

Successful surgical treatment of a sacral hemangioma causing cauda equina dysfunction: a case description

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Introduction

Vertebral hemangioma (VH) is relatively common, occurring in 10% to 12% of the general population (1), and although benign, it is actually a vascular malformation (2). Most VHs are latent [Enneking stage 1 (st.1)] and do not require specific treatment, with only 1% of VHs become active and symptomatic, approximately 45% of which become aggressive and extend into the spinal canal and/or paravertebral space, leading to neurological dysfunction (Enneking st.3) (3). Due to the rarity of aggressive VH cases, the diagnosis and treatment options for these cases remain controversial and problematic. In 2010, Acosta *et al.* (4) reported that 10 patients with VHs underwent surgical treatment. However, surgical treatment of invasive sacral hemangioma causing cauda equina syndrome with good results has not been reported.

Case presentation

For patients with invasive spinal hemangioma accompanied by neurological dysfunction, our center has a set of diagnostic and treatment procedures, as shown in *Figure 1*.

Clinical history

The patient was a 61-year-old man who had lumbosacral pain with weakness in both lower extremities for 2 months, numbness in the back of both lower extremities, and difficulty defecating and urinating. No treatment was provided in other medical facilities. The patient had no family history of genetic or similar diseases.

The physical examination showed lumbosacral tenderness, hypoesthesia in the saddle area, hypoesthesia in the posterior lower limbs and dorsolateral feet, hypoesthesia in the hind heels, bilateral weak plantar flexion (grade 3), no ankle reflex, rectal prolapse, anal sphincter muscle strength weakness, and no anal reflexes. All procedures performed in this study were in accordance with the ethical standards of the Peking University First Hospital research committee and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this article and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Imaging

On magnetic resonance imaging (MRI), irregular epidural signals with low T1-weighted imaging (T1WI), high T2-weighted imaging (T2WI), and high fat-suppression T2-weighted imaging (fsT2WI) could be seen in the second sacral vertebra and the first to third sacral vertebrae of the spinal canal. The size of lesion was approximately 5.7 cm × 3.4 cm × 6.2 cm, and there was obvious enhancement on the enhanced scan. A lesion was observed growing along the sacral foramen in a slit-like manner. The bones of the posterior margin of the sacral vertebra 1 were compressed and thin, and only the normal bone of the anterior margin

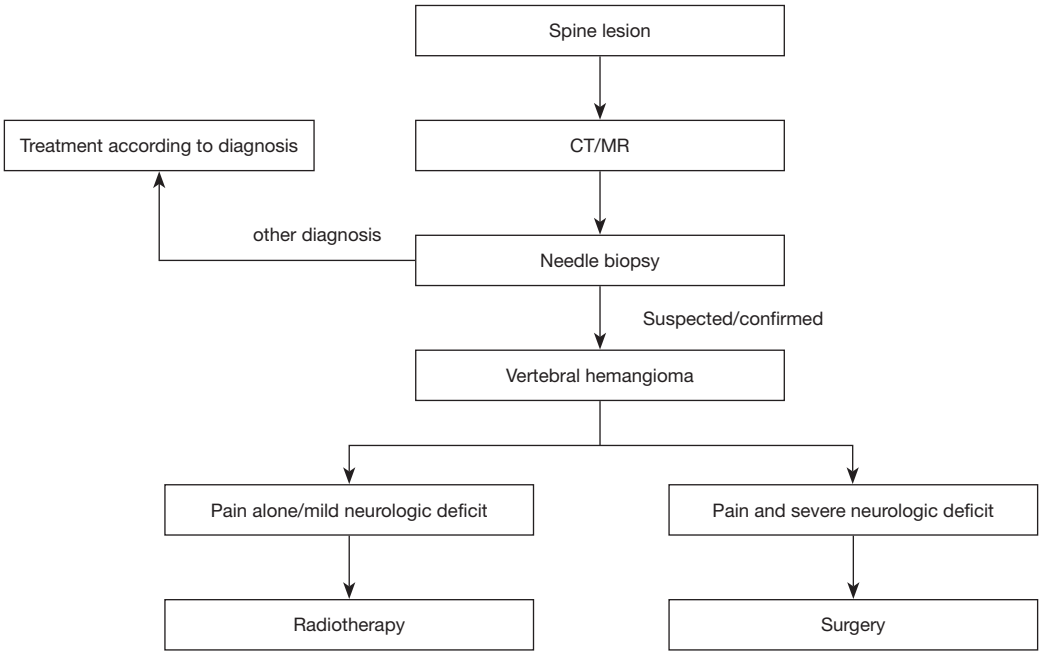

Figure 1 The flowchart for the treatment of invasive spinal hemangioma. CT, computed tomography; MR, magnetic resonance.

of the second sacral vertebra remained (*Figure 2*).

Fine needle aspiration pathological biopsy

The sacral mass was found composed of dead bone and bone marrow tissue with active local hyperplasia, vascular structures of different sizes, and uneven wall thickness and lumen size, thus suggesting vascular malformation.

Diagnosis

The ultimate diagnose was a sacral hemangioma.

Surgery

After discussion in the Department of Bone Oncology, Neurosurgery, and Interventional Vascular Surgery, it was decided to perform surgical treatment because the tumor had caused cauda equina nerve function loss but was otherwise benign and had not invaded the surrounding soft tissues. Because preoperative pathologic puncture indicated the possibility of a hemangioma, interventional embolization of local blood supply vessels of the tumor was performed before surgery to avoid massive bleeding during the operation. The local tumor blood supply before and after interventional surgery is shown in the *Figure 3*.

After general anesthesia, the lesion was located with the patient in the prone position, and the lumbosacral posterior median incision was approximately 10 cm long. The L5–S3 spinous process and laminae were exposed. A bone knife and lamina rongeur were used to remove part of L5 lamina and excise the S1–S3 laminae. Red soft solid masses were found in the vertebral canal, which surrounded the sacral nerve root and were attached to the nerve root. The tumor tissue was carefully separated, and the nerve root compression was relieved. The bone was deeply exposed and eroded, and the surface of part of the bone was rough. The bone surface was treated with a scraping spoon and bipolar electrocoagulation. Tumor tissue resection and nerve root lysis were satisfactory, the wound was rinsed, a hemostatic sponge was placed in the wound, a drainage tube was placed, and the incision was closed. The postoperative specimens are shown in *Figure 4*.

Pathology

Examination of the sacral lesions revealed that the fibrous adipocytes were lined with flat endothelial cells and contained a large number of red blood cells. The vascular tumors tended to be hemangiomas. An image of the postoperative hematoxylin and eosin (HE) staining of the tumor tissue under microscopy is shown in *Figure 5*.

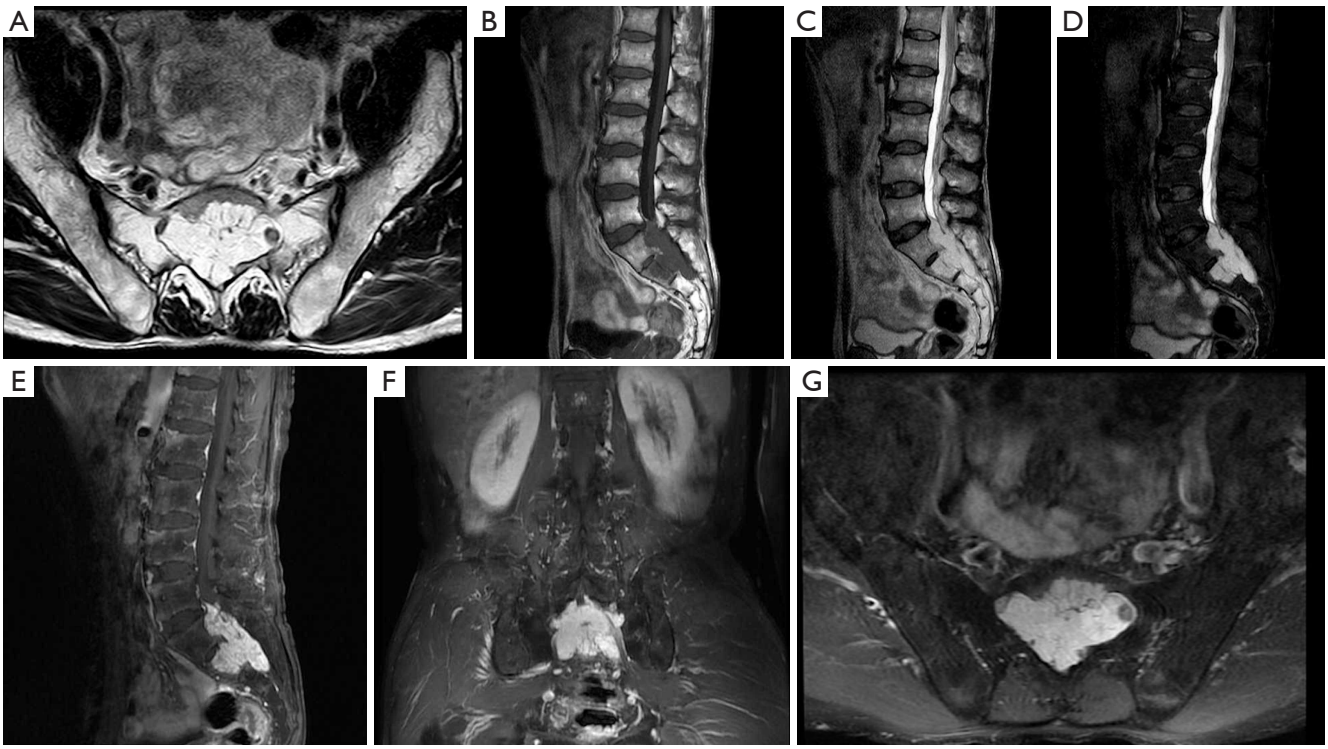

Figure 2 MRI of the sacral hemangioma. (A-D) MRI plain scan images. Irregular low T1WI, high T2WI, and high fat-saturated T2WI signals were seen in the second sacral vertebrae and the spinal canal of the first to third sacral vertebrae. The size of lesion was approximately 5.7 cm × 3.4 cm × 6.2 cm. (E-G) MRI enhancement images showing obvious and even enhancement. The lesion grew like a suture along the sacral foramina, the bones of the posterior margin of the sacral vertebral body were compressed and thin, and only the normal bone of the anterior edge remained in the sacral vertebral body of the sacral vertebral body. MRI, magnetic resonance imaging; T1WI, T1-weighted imaging; T2WI, T2-weighted imaging.

Results and follow-up

At 1 month after surgery, the symptoms of lumbosacral pain were significantly relieved, the urinary tube was pulled out, and urination function was gradually restored. Bowel function was slightly restored, but manual assistance was still needed. The sensation in the saddle area and both lower limbs gradually recovered. The plantar flexor strength of the foot was restored to level 4 (+). The ankle reflex was weak, the anal reflex was restored, and the sphincter muscle strength was enhanced. As of this writing, the wound is healing well.

Discussion

The most common tumors in the cauda equina are mucinous ependymoma (90%) and schwannoma, while other less common tumors include paraganglioma, intradural metastasis, hemangioblastoma, and ganglioglioma (5-11). Typically, hemangiomas are mainly located in the vertebrae, accounting for approximately 5% of all spinal vascular lesions, while only approximately 3% are found in the spinal canal with clear boundaries. However, invasive hemangiomas in the sacral vertebrae and that invade the cauda equina nerve root to cause cauda equina syndrome are very rare. In our case, intraoperative hemangioma was clearly found to be surrounding the adherent nerve roots, cause erosion of part of the bone, but the soft tissue around the sacrum was normal, as indicated on MRI. Recent studies have shown that typical cauda equina neurohemangioma is associated with severe neurological dysfunction, and the recovery rate of neurological function after surgical decompression is approximately 100% (3). This was also the approximate rate in our case. However, researchers have also reported cases of tumor recurrence after surgery for this type of aggressive hemangioma (12). In addition, there are some reports that surgical treatment

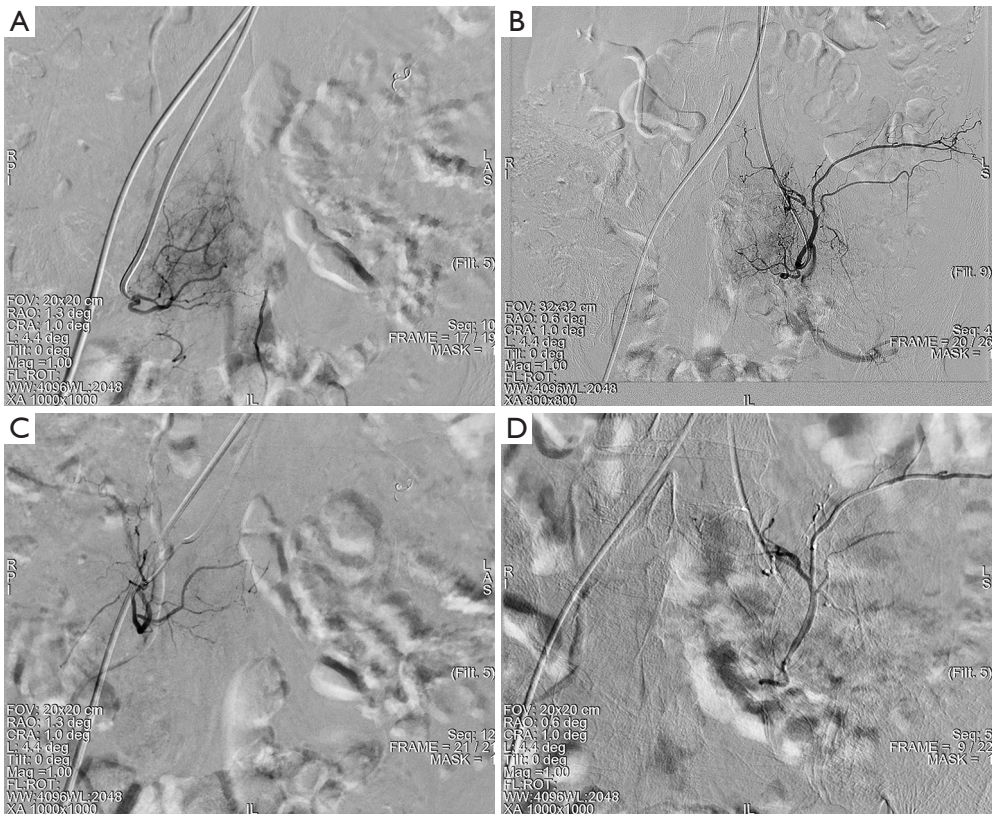

Figure 3 The right lateral sacral artery and left lateral sacral artery before and after interventional embolization. (A) A angiographic image of the right lateral sacral artery before interventional surgery. (B) The right lateral sacral artery after interventional surgery. (C) Angiographic image of the left lateral sacral artery before interventional surgery. (D) The left lateral sacral artery after interventional surgery. RPI, right posterior inferior; IL, inferior left; LAS, left anterior superior; FOV, field of view; RAO, right anterior oblique; CRA, cranial; L, lateral; FL, flipped; ROT, rotation; WW, window width; WL, window level; XA, X-ray angiography; Mag, magnification; RI, right inferior; LS, left superior.

Figure 4 Postoperative specimen.

combined with postoperative radiotherapy can reduce the probability of recurrence (13). We believe that the choice of treatment should be based on the patient's tumor invasion, symptoms, and circumstances. Of course, there are many other treatment options for invasive hemangiomas of the spine that have shown good clinical results, such as radiotherapy alone, vertebroplasty, direct alcohol injection, and surgical decompression with or without radiotherapy (10). In our case, the patient already had symptoms of neurological impairment, so surgery was the preferred treatment; Preoperative MRI examination and further pathologic puncture indicated a hemangioma. Compared with MRI results, the results of pathological puncture are particularly important. The pathological results are the gold standard for our clinical diagnosis and may determine our surgical methods. In order to reduce

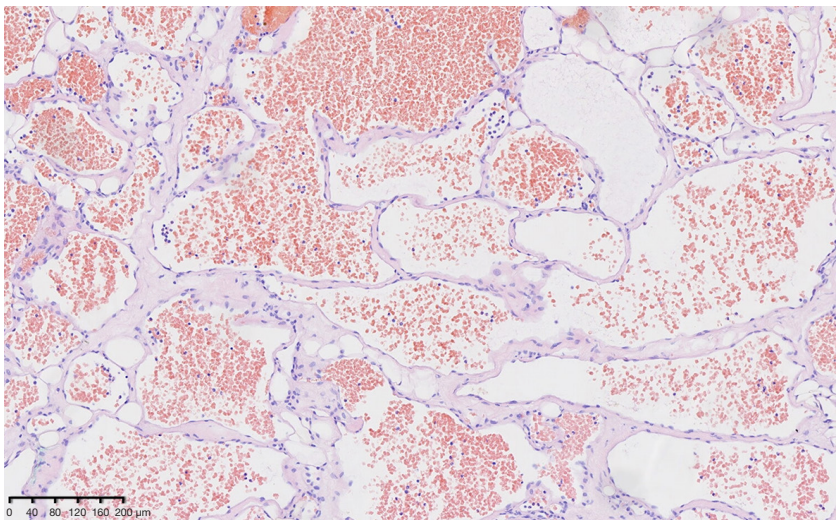

Figure 5 Image of postoperative hematoxylin and eosin staining of the tumor tissue under microscopy. Scale bar =200 μm.

bleeding, preoperative embolization of tumor blood supply vessels was performed. In addition, we found that the tumor had done little damage to the bone, and a hard bone cortex remained in some areas, so we did not consider vertebroplasty treatment. In addition, the tumor could be completely removed during the operation, so we did not consider complete vertebral resection or postoperative radiotherapy and chemotherapy before first completing a follow-up. If the preoperative pathologic puncture results had indicated that the tumor was malignant, we might have expanded the scope of surgical resection and needed to incorporate postoperative radiotherapy and chemotherapy, rather than simply exfoliating the tumor. In this case, we did not consider the use of alcohol injection. For one, most of the tumors were located in the posterior one-third of the vertebral body and in the spinal canal, and alcohol injection might have damaged the nerves in the spinal canal. For another, our experience with this technology had not yet matured to mastery. However, to prevent tumor recurrence, we burned the tumor and the vertebral interface with alcohol. As of this writing, the patient has shown good recovery after the operation, and there has been no recurrence. Surgery is a relatively effective treatment for invasive sacral hemangioma, but early diagnosis and treatment and postoperative rehabilitation are critical steps for comprehensive treatment, as they can ensure avoiding excessive damage to neurological function and facilitate the recovery of neurological function after surgery. In 2022, Brindisino *et al.* reported a case of invasive VH with spinal cord compression, which highlighted the importance of rapid clinical diagnosis and treatment as well as postoperative rehabilitation planning with rehabilitation physicians for postoperative neurological recovery (14).

Some limitations to our study should be mentioned. Although the short-term postoperative effect was obvious, long-term follow-up is still needed to observe whether the tumor has recurred and to determine the degree of improvement of nerve function. Generally, patients are highly satisfied with our medical services, as they typically experience a significantly improved quality of life.

Conclusions

Sacral hemangioma with cauda equina involvement is a rare disease that can lead to severe and permanent neurological dysfunction. In the absence of preoperative pathological examination, MRI may be the key examination method for the diagnosis of typical hemangiomas. This is because on MRI, hemangiomas demonstrate certain characteristic features that are conducive to diagnosis. However, in this case, histopathological diagnosis was more important than MRI and could guide further treatment. Even without pathological examination and accurate preoperative diagnosis, surgery may be the preferred treatment for patients with neurological dysfunction.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-23-1493/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional research committee and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this article and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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