Radiology Case Reports 16 (2021) 938–941

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journal homepage: www.elsevier.com/locate/radcr

Case report

Thoracic spine hemangioma causing rapidly progressive myelopathy and mimicking a malignant tumor: A case report ☆,☆☆

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ARTICLE INFO

Article history:
Received 15 January 2021
Revised 29 January 2021
Accepted 31 January 2021

Keywords:
Hemangioma
Spine
Myelopathy
Vertebral
Tumor
Extraosseous

ABSTRACT

Vertebral hemangiomas are common benign tumors that are mostly asymptomatic and are discovered incidentally. Only 0.9–1.2% of all vertebral hemangiomas, termed aggressive vertebral hemangiomas, expand to cause pain and neural compression. We present an extremely rare case of a 49-year-old woman who had an aggressive vertebral hemangioma of the thoracic spine that caused rapidly progressive myelopathy with remarkable irregular extraosseous bone proliferation, which mimicked a malignant vertebral tumor. In this case, despite the lesion's hostile appearance during imaging, the pathological diagnosis was benign and symptom-based surgical treatment with posterior decompression and stabilization provided good clinical outcomes during the postoperative 18 months follow-up period. In this case, despite the use of standard imaging modalities (radiograph, CT, and MRI), making a preoperative imaging diagnosis of an aggressive vertebral hemangioma was difficult, and although aggressive vertebral hemangiomas with atypical radiological features are rare, they should be considered as a differential diagnosis.

Introduction

Vertebral hemangiomas (VHs) are benign vascular lesions of the spine with a prevalence of 10–12% in the general population, and accounts for approximately 2%–3% of all spinal

☆ Sources of funding: None.

☆☆ Competing Interest: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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https://doi.org/10.1016/j.radcr.2021.01.060

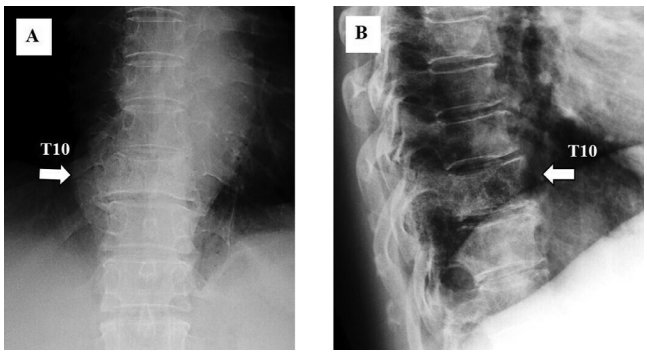

Fig. 1 – (A). Preoperative anterior-posterior radiograph shows a compressive deformity of the T10 vertebra (arrow) with obscurity of the shape of the right pedicle. (B). Preoperative lateral radiograph shows the T10 vertebral column (arrow) with heterogeneous sclerosis.

tumors. [1] The majority of VHs are asymptomatic and are discovered incidentally on computed tomography (CT) or magnetic resonance imaging (MRI) of the spine. [1] Histologically, VHs are benign vascular lesions of bones, composed of multiple thin-walled vessels surrounded by fat that infiltrate the medullary cavity between the bony trabeculae. [2] Only 0.9%–1.2% of all VHs expand and cause pain and neural compression—this clinical manifestation is known as aggressive VH [1,2]. Here, we present an extremely rare case of aggressive VH of the thoracic spine that caused rapidly progressive myelopathy with remarkable irregular extraosseous bone proliferation that mimicked a malignant vertebral tumor. In this case, we experienced difficulty in making a preoperative diagnosis of aggressive VH despite the use of standard imaging techniques, including radiography, CT, and MRI.

Case report

A 49-year-old woman without a significant medical or obstetric history presented with back pain that began 5 months prior and a rapid progression of numbness and paralysis of both lower limbs that persisted for 5 days. Neurological examination revealed marked paresthesia and weakness of the proximal and distal muscles groups in both lower extremities (manual muscle testing [MMT] grade: 3/5); therefore, she was promptly hospitalized. Radiographs of the spine revealed a compressive deformity and heterogenous sclerosis of the T10 vertebra (Fig. 1). A preoperative MRI revealed a T10 vertebral tumor that produced low signal intensity on T1-weighted image and a moderately high signal on T2-weighted image. The axial gadolinium-enhanced view revealed extraosseous tumoral extensions that caused spinal cord compression (Fig. 2 A–D), and were suggestive of metastatic deposit or a primary malignant vertebral tumor. Therefore, contrast-enhanced CT of the neck, chest, spine, abdomen, and pelvis was performed to further characterize the lesion and to identify the primary lesion. CT revealed heterogeneous sclerosis and remarkable irregular extraosseous bone proliferation of the T10 vertebral tumor (Figs. 2 E and F). No primary tumor was shown on neck, chest, abdomen, and pelvic CT, and no positive tumor markers were identified in blood test.

Exacerbation of paralysis in both lower limbs (MMT grade of the proximal and distal muscles: 2/5) was observed 2 days after she was hospitalized. Given the progressive nature of neurologic deterioration, we proceeded with urgent surgical intervention. Preoperative needle biopsy was not performed as a pathological diagnosis required at least 7 days. We performed a preoperative embolization using fluoroscopically-controlled endovascular intervention 5 days after admission. Six days after admission, we performed T9-T10 posterior decompression along with tumoral resection of the posterior elements of the T10 vertebra, and T8–T12 posterior instrumented fusion (Fig. 3). Intraoperative findings revealed that the posterior elements of the T10 vertebra that were replaced by the bone tumor, were hemorrhagic. However, we were able to avoid excessive bleeding (estimated intraoperative blood loss: 575 mL) due to preoperative embolization.

Histopathological examination of the resected right lamina of the T10 vertebra demonstrated features of an underlying capillary VH and no evidence of a malignancy (Fig. 4). Immediately after the surgery, motor function in the lower extremities recovered. The patient was able to walk without aid 2 weeks postoperatively and maintained a good walking ability at the final follow-up visit 18 months postoperatively. CT performed 12 months postoperatively revealed that decompression of the spinal canal at the level of the T10 vertebra that had aggressive VH was sustained (Fig. 5).

Discussion

In typical cases, VHs usually show homogeneous enhancement on radiological images, and can be diagnosed using CT and MRI. On CT images, VHs are characterized by thickening of the vertically striated trabeculae, and are described as “polka-dot” and/or “corduroy cloth” in appearance. [3,4] In latent VHs, MRI reveals increased signal intensity on both T1- and T2-weighted images due to the fatty component and fewer vascular stroma, with alternating hypointense areas, leading to a “salt-and-pepper appearance.”[3] Meanwhile, aggressive VHs tend to show hypointensity on T1-weighted images associated with lesions that have a low fat content and distinct hypervascularity. [2,4] Although aggressive VHs are relatively rare, many reports have demonstrated the aforementioned radiological features of typical VHs that support preoperative diagnoses. [1,3–8] To the best of our knowledge, aggressive VHs with marked irregular extraosseous bone proliferation without typical radiological characteristics are extremely rare and only a few cases have been reported so far [9]. In this case, despite the use of standard imaging modalities (radiograph, CT, and MRI), a preoperative imaging diagnosis of aggressive VH was considered difficult. These atypical, aggressive VHs are rare; however, they should be considered as a differential diagnosis.

Regarding intraoperative bleeding, due to high vascularization of aggressive VHs, copious bleeding may complicate

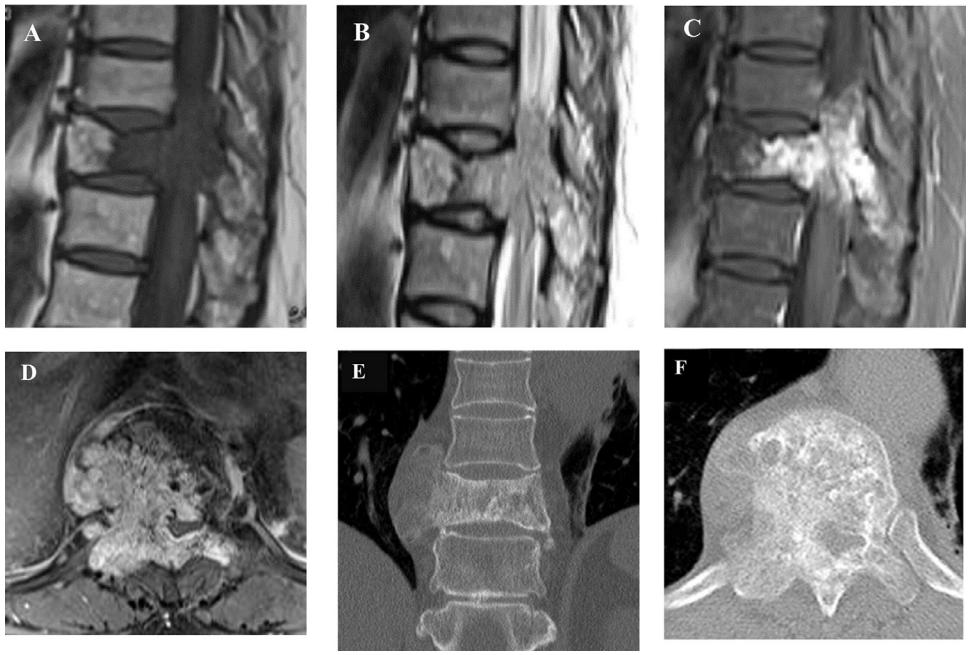

Fig. 2 – (A). Preoperative right-sagittal T1-weighted magnetic resonance (MR) image shows the T10 vertebral tumor with a low signal intensity. (B). Preoperative right-sagittal T2-weighted MR image shows the T10 vertebral tumor with a moderately heterogeneous high signal intensity. (C). Preoperative right-sagittal gadolinium-enhanced fat-suppressed MR image shows the T10 vertebral tumor with a moderately heterogeneous high signal intensity. (D). Preoperative axial gadolinium-enhanced fat-suppressed MR image shows extraosseous tumoral extensions causing spinal cord compression with enlargement of the right transverse process and pedicle. (E) Preoperative coronal computed tomography (CT) image of the T10 vertebra shows heterogeneous sclerosis and remarkable extraosseous bone proliferation on the right side. (F). Preoperative axial CT image of the T10 vertebra shows marked irregular extraosseous bone formations predominantly on the right side.

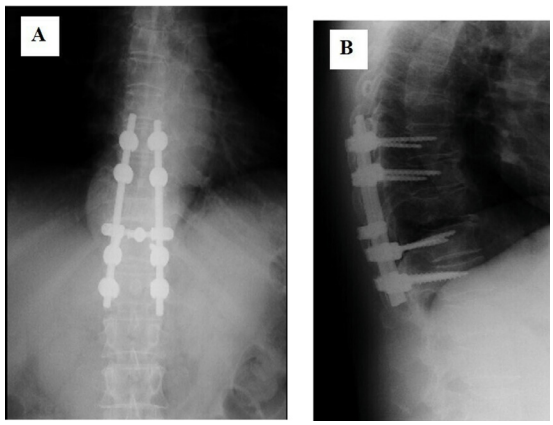

Fig. 3 – (A). Postoperative anterior-posterior radiograph of the spine. (B). Postoperative lateral radiograph of the spine.

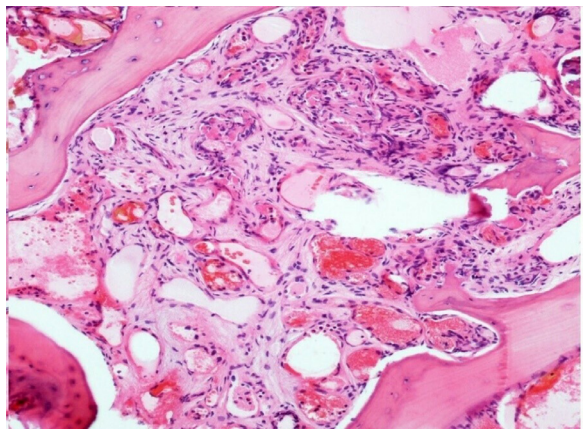

Fig. 4 – Histopathological study of the resected specimen from the right side of the T10 lamina shows congestion of capillary vessels without anastomosis, consistent with osseous capillary hemangioma (hematoxylin and eosin staining, x 100).

surgery and can even be life-threatening [10]. Previous papers have reported that embolization prior to surgery can significantly reduce bleeding in resection procedures for aggressive VHs. [10,11] In our case, although the preoperative diagnosis was not confirmed, we performed a preoperative embolization a day prior to the surgery and consequently, this endovascular intervention likely prevented excessive intraoperative blood loss.

Regarding the selection of a surgical treatment strategy, considering the possibility of a primary vertebral malignant

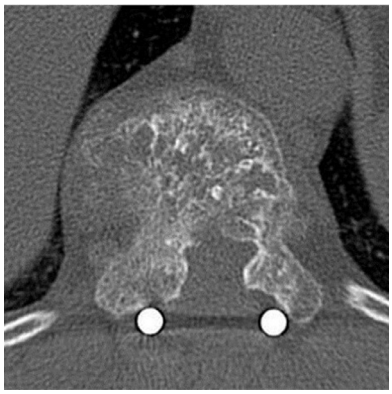

Fig. 5 – Axial computed tomography image of the T10 vertebra at 12 months postoperatively shows sustained decompression of the spinal canal.

tumor with marked osseous formation, such as osteosarcomas and chondrosarcomas, a two-staged surgical method may have been more ideal to reduce contamination due to the primary malignant tumor cells in the operative field: (1) performing decompression surgery for the compressed spinal cord and obtaining a tissue sample in an emergent setting; and (2) considering additional instrumented surgery when the pathological diagnosis is a benign tumor or metastasis. In our case, the pathological diagnosis was fortunately benign and we obtained a good clinical outcome following a single-staged procedure: posterior decompression and instrumented fusion.

In this case, despite the lesion's hostile appearance on imaging, histological examination revealed that it was benign and symptom-based surgical treatment (decompression and stabilization) has good outcomes during the follow-up period. However, other treatment modalities, such as radiation, vertebroplasty, and total en bloc spondylectomy may be considered in cases where tumor recurrence causes neurological decline. [11,12]

Patient consent statement

Written informed consent was obtained from the patient prior to submission of this case report.

Author contributions

Shunpei Iida wrote and prepared the manuscript. Satoshi Ogihara contributed to the conceptualization and design of the study, collected the patient's data, and edited the manuscript. Fumiaki Kobayashi, Ryutaro Kawano, and Kazuo Saita collected the patient's data. All authors have read, reviewed, and approved the article.

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