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Technical Notes & Surgical Techniques

Effectiveness of postoperative radiation therapy for thoracic spine hemangioma recurrence

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ABSTRACT

Vertebral hemangiomas are the most common benign tumor of the spine. Although usually asymptomatic, there is a higher risk of severe neurological deficits when hemangiomas afflict the thoracic spine. The management of vertebral hemangioma patients with extraosseous extension causing neurological dysfunction has not yet been established. We described the effectiveness of surgical and radiation therapy in a case of recurrent hemangioma in the thoracic spine with extraosseous extension causing progressive paraparesis. We retrospectively reviewed a case of vertebral recurrent hemangioma treated by posterior spinal decompression and fusion and subsequent radiation therapy.

A 61-year-old woman suffered from neurological deficits caused by a hemangioma in the thoracic spine. Despite initial treatment by posterior decompression, subtotal transpedicular tumor resection, and posterior spinal fusion, her neurological symptoms due to tumor compression recurred 5 years later. Posterior-approach anterior spinal cord decompression by subtotal transpedicular tumor resection was attempted next but her paraparesis continued to worsen. Radiotherapy of a total of 32 Gy in 16 fractions restored mobility and lesion size was markedly reduced 7 months later. Even 14 months after surgery, she has been able to walk although she has slightly paralysis of the lower extremity remaining.

Subtotal transpedicular tumor resection, and especially additional radiation therapy, was effective in reducing tumor volume and improving neurological dysfunction caused by a thoracic spine recurrent hemangioma.

1. Introduction

Vertebral hemangiomas are the most common benign tumor of the spine [1], representing 2–3% of all spinal tumors and being found in approximately 11% of vertebral autopsies [2]. Only 0.9–1.2% of vertebral hemangiomas are symptomatic [3,4]. When hemangiomas occur in the thoracic spine, they are more likely to cause problems due to the narrow vertebral canal, which mandates more aggressive management to avoid severe neurological deficits [5,6]. To date, the treatment of vertebral hemangioma with extraosseous extension with accompanying neurological dysfunction remains to be established.

We herein report a case of recurrent hemangioma in the thoracic spine with extraosseous extension causing progressive paraparesis that was successfully treated by surgical and radiological therapy.

2. Case presentation

This study was approved by the ethics committee of our hospital. Informed consent was obtained from the patient. A 61-year-old woman presented with a history of lower extremity pain that had begun a month before admission in April 2011. Afterwards, she began to exhibit progressive weakness in the lower extremities and gait disturbance. Her medical history indicated no abnormalities. Physical examination on admission revealed slight bilateral lower extremity weakness that was most severe in the hip flexors and gait disturbance by spasticity. No sensory disturbance was noted and increased tone and hyper-reflexia in the lower extremities were observed. A T2-weighted sagittal magnetic resonance imaging (MRI) scan of the thoracic spine showed a high-signal mass in the region anterior to the 3rd thoracic vertebra with extraosseous extension and spinal cord compression (Fig. 1(a, b)).

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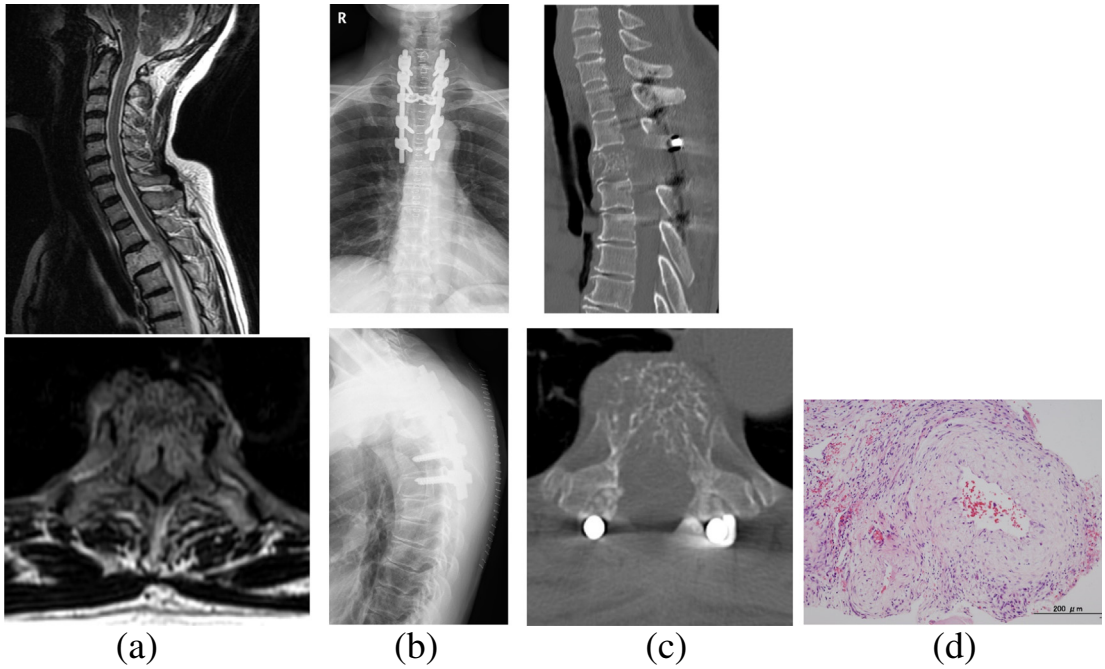

Fig. 1. Findings of 1st surgery. (a) Preoperative MRI. T2-weighted sagittal magnetic resonance imaging scan of the thoracic spine at the first admission showed a high-signal mass in the region anterior to the 3rd thoracic vertebra with extraosseous extension and spinal cord compression. (b) Postoperative radiograph. We performed posterior decompression from the 2nd to 4th thoracic vertebra, subtotal tumor resection, and posterior spinal fusion from the 1st to 5th thoracic vertebra. (c) Postoperative CT. Postoperative CT showed posterior decompression from the 2nd to 4th thoracic vertebra. (d) Histological findings. Histological findings from tissue resected during the first surgery showed edematous stroma with increased capillaries that included giant osteoclasts and spindle-shaped cells. An aneurysmal bone cyst was suspected.

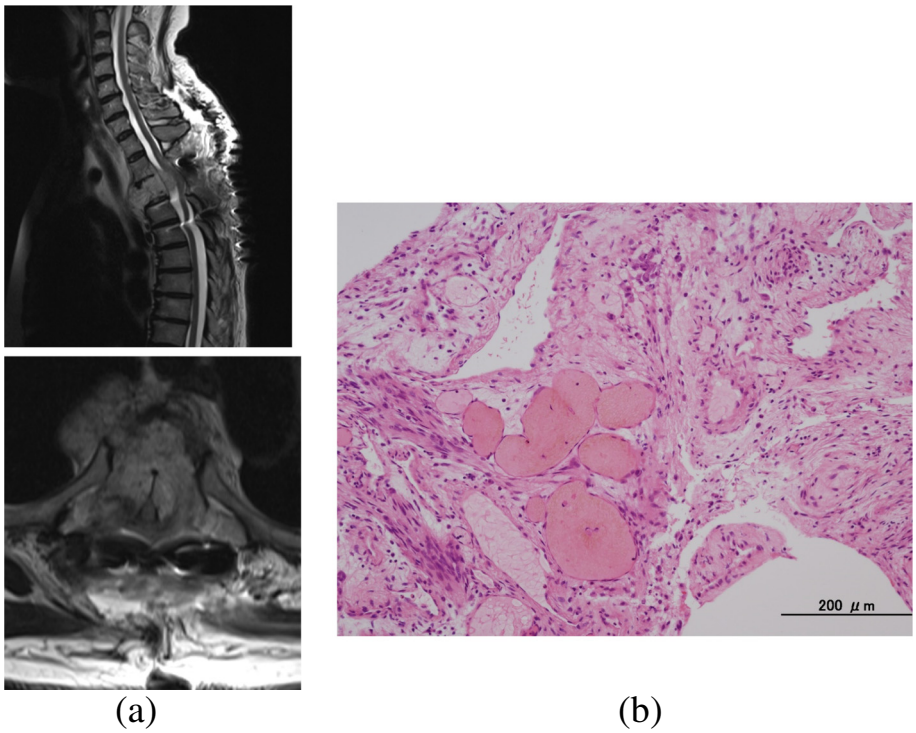

Fig. 2. The findings of 2nd surgery. (a) Preoperative MRI. Magnetic resonance imaging revealed an increase in lesion size and spinal cord compression 5 years after the initial surgery. (b) Histological findings. Histological findings from tissue resected during the second surgery revealed dilated blood vessels. A hemangioma was suspected.

Percutaneous vertebral biopsy was negative.

Posterior decompression from the 2nd to 4th thoracic vertebra, subtotal transpedicular tumor resection, and posterior fusion from the 1st to 5th thoracic vertebra was performed in response to worsening paraparesis (Fig. 1(c, d)). Surgical time was 222 min and blood loss volume was 800 g. Histological findings from resected tissue suggested an aneurysmal bone cyst (Fig. 1(d)). She could walk with a T-cane after the surgery.

The patient's gait gradually worsened over the subsequent 5 years. MRI disclosed an increase in lesion size and spinal cord compression (Fig. 2(a)). Two days following a transpedicular biopsy of the spine, her lower limb paralysis became markedly increased, necessitating

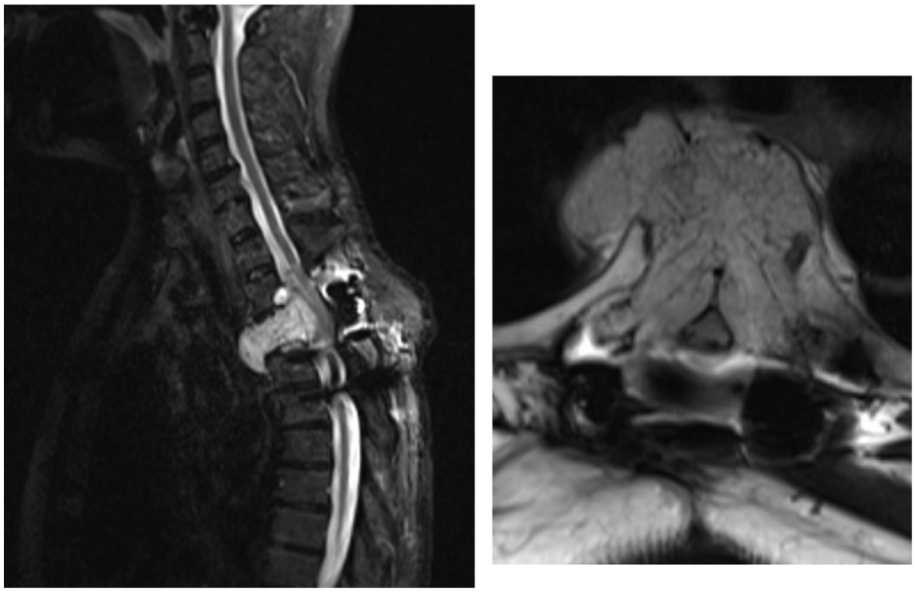

Fig. 3. Postoperative magnetic resonance imaging showed the lesion to be reduced in size with enlargement of spinal cord space 14 months after surgery.

posterior-approach anterior spinal cord decompression by subtotal transpedicular tumor resection. Surgical time was 200 min and blood loss volume was 1300 g. Histological findings from resected tissue indicated a hemangioma (Fig. 2(b)). Even after decompression surgery, however, her paraparesis progressed and she began suffering from urinary retention. Radiation therapy of a total of 32 Gy in 16 fractions soon enabled walking and self-urination. The lesion was reduced in size with enlargement of spinal cord space 14 months after the second surgery (Fig. 3). Since 21 months after surgery, she has been able to walk despite slight paralysis of the lower extremities.

3. Discussion

In the present case of recurrence vertebral hemangioma with severe spinal cord compression causing neurological deficits, posterior decompression and fusion surgery with additional radiation therapy were effective to reduce tumor volume and improve neurological dysfunction.

The treatment options for vertebral hemangioma include radiotherapy [5], arterial embolization [7], percutaneous sclerotherapy [8], vertebroplasty [9], surgical decompression [10], subtotal tumor resection [10], total tumor resection [11], or a combination of the above. However, a consensus on the management of vertebral hemangioma patients with extraosseous extension causing neurological impairment has not yet been reached.

In 5 earlier cases of total tumor resection for aggressive hemangioma, no tumor recurrence was reported, although mean intraoperative blood loss was 2424 mL [11]. Accordingly, total tumor resection has been associated with high intraoperative morbidity due to massive hemorrhage. We performed repeated subtotal transpedicular tumor resections with a mean blood loss volume of 1050 g in this case, but neurological deficits caused by the tumor recurred 5 years after the initial surgery. Thus, the rate of tumor recurrence might be higher for subtotal tumor resection, but it was considered safer due to lower blood loss.

Vertebral hemangiomas are responsive to radiotherapy, which has become the most common treatment for lesions causing pain [12]. However, radiation for patients with progressive neurological deficits is controversial. Surgical decompression is favored in most cases, with irradiation reserved as an adjuvant [12]. In our patient, radiotherapy markedly reduced lesion size after insufficient surgical decompression to improve gait and bladder disturbance, and thus appeared to be a good treatment option, although larger studies are needed to confirm this.

Several reports have described the effectiveness of radiation therapy for aneurysmal bone cysts [13–15]. We did not perform radiotherapy before the second operation since emergency decompression had become necessary for progressive paralysis of the lower limbs 2 days after her diagnostic transpedicular biopsy. Another reason why the second surgical resection was ineffective might have been insufficient decompression due to reoperation on the weakened spinal cord, possibly resulting in paralysis exacerbated by the surgical technique.

Based on the above findings, subtotal hemangioma resection with radiation therapy appears to be a good treatment for vertebral hemangioma, and testing of larger cohorts is warranted.

4. Conclusion

Subtotal hemangioma resection along with radiation therapy reduced tumor volume and improved neurological deficits. The latter modality was especially effective in decreasing tumor size.

Disclosure

We received no specific funding for this study.

References

[1] J.O. Jones, B.M. Bruel, S.R. Vattam, Management of painful vertebral hemangiomas with kyphoplasty: a report of two cases and a literature review, Pain Physician 12 (4) (2009) E297–E303.
[2] H.C. Suparna, B.M. Vadhiraja, R.C. Apsani, T. Seetharamaiah, D.J. Fernandes, K. Rao, et al., Symptomatic vertebral hemangiomas – results of treatment with radiotherapy, Ind. J. Radiol. Imag. 16 (1) (2006) 37–40.
[3] M. Healy, D.A. Herz, L. Pearl, Spinal hemangiomas, Neurosurgery 13 (6) (1983) 689–691.
[4] J.P. Nguyen, M. Djindjian, A. Gaston, Vertebral hemangiomas presenting with neurologic symptoms, Surg. Neurol. 27 (4) (1987) 391–397.
[5] R.K. Aich, A.R. Deb, A. Banerjee, R. Karim, P. Gupta, Symptomatic vertebral hemangioma: treatment with radiotherapy, J. Cancer Res. Ther. 6 (2) (2010) 199–203.
[6] S.I. Nassar, F.S. Hanbali, M.C. Haddad, M.H. Fahl, Thoracic vertebral hemangioma with extradural extension and spinal cord compression. Case report, Clin. Imaging 22 (1) (1998) 65–68.
[7] Y. Robinson, R. Sheta, K. Salci, J. Willander, Blood loss in surgery for aggressive vertebral haemangioma with and without embolization, Asian Spine J. 9 (3) (2015) 483–491.
[8] Gabal Am, Percutaneous technique for sclerotherapy of vertebral hemangioma

compressing spinal cord, Cardiovasc. Intervent. Radiol. 25 (6) (2002) 494–500.
[9] J. Hao, Z. Hu, Percutaneous cement vertebroplasty in the treatment of symptomatic vertebral hemangiomas, Pain Physician 15 (1) (2012) 43–49.
[10] M. Shinozaki, A. Morita, K. Kamijo, A. Seichi, N. Saito, T. Kirino, Symptomatic T2 vertebral hemangioma in a pregnant woman treated by one stage combination surgery; posterior stabilization and anterior subtotal tumor resection. Case report, Neurol. Med. Chir. (Tokyo) 50 (8) (2010) 674–677.
[11] S. Kato, N. Kawahara, H. Murakami, S. Demura, K. Yoshioka, T. Okayama, et al., Surgical management of aggressive vertebral hemangiomas causing spinal cord compression: long-term clinical follow-up of five cases, J. Orthop. Sci. 15 (3) (2010) 350–356.
[12] M.W. Fox, B.M. Onofrio, The natural history and management of symptomatic and asymptomatic vertebral hemangiomas, J. Neurosurg. 78 (1) (1993) 36–45.
[13] B. Jereb, J. Smith, Giant aneurysmal bone cyst of the innominate bone treated with irradiation, Br. J. Radiol. 53 (629) (1980) 489–491.
[14] M. Maeda, H. Tateishi, H. Takaiwa, G. Kinoshita, N. Hatano, K. Nakano, High-energy, low-dose radiation therapy for aneurysmal bone cyst, Report of a case. Clin. Orthop. Relat. Res. (243) (1989) 200-3.
[15] K. Elsayad, J. Kriz, H. Seegenschmiedt, D. Imhoff, R. Heyd, H.T. Eich, et al., Radiotherapy for aneurysmal bone cyst: a rare indication, Strahlenther. Onkol. 193 (4) (2017) 332–340.