Management of Aggressive Vertebral Hemangiomas Presenting with Spinal Cord Compression

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Background: *Vertebral hemangioma (VH) is the most common benign vertebral tumor. Most patients are asymptomatic. Neurological deficit due to spinal cord compression is a rare presentation of symptomatic VH.*
Objective: *This study aims to describe patients who developed compressive myelopathy owing to aggressive VH and were treated by surgical decompression.*
Material and Method: *Retrospective chart reviews of patients with symptomatic VH manifesting by compressive myelopathy were conducted. Spinal radiograph, magnetic resonance imaging (MRI) and angiography were collected.*
Results: *Three cases with spinal cord compression caused by VH were included in the study. VH was suspected by preoperative spinal MRI in only one case. Preoperative embolization was performed for reducing intraoperative blood loss. The remaining two did not undergo embolization before surgery. Significant blood loss during the operation made the surgeon terminate the procedure. The second-stage procedure was performed following embolization. The intraoperative blood loss was minimized.*
Conclusion: *VH is a differential diagnosis in patients with spinal tumor presenting with pain and myelopathy. Massive bleeding from the tumor during surgical decompression can be reduced by transarterial embolization. Multidisciplinary treatment is the mainstay in management of VH.*

Keywords: *Vertebral hemangioma, Spinal cord compression, Surgical decompression*

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Vertebral hemangioma (VH) is the most common, benign, primary tumor of the spine[1]. The incidence is about 10 to 12%, based on large autopsy series and reviews of plain radiograph of the spine[2-4]. VH is most frequently located in the thoracic spine[5], followed by the lumbar region[6,7]. Multiple level involvements were reported up to 30% of cases with VH[8]. Females are affected more often than males and young adults have a higher incidence of developing symptoms, compared with the elderly group[9]. Only 0.9 to 1.2% of the cases are symptomatic. The most frequent clinical presentation is pain, which is related to an osseous expansion or a pathologic fracture. Further tumor progression can lead to instability or neurologic symptoms, caused by compression to neural structures, such as nerve roots or spinal cord. These rare circumstances are called aggressive VH[2,9-16].

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Management of VH generally requires multidisciplinary treatment[17-23]. Radiation therapy is recommended for isolated pain symptom or after subtotal tumor removal[24-29]. Transarterial embolization to decrease intraoperative blood loss is recommended before surgical procedure[30-32]. Vertebroplasty and kyphoplasty are also optional for pain treatment.

With the limited report of this condition[33,34], it is difficult to recognize this rare condition. We analyzed retrospectively our experience in diagnosis and surgical treatment of aggressive VH.

Material and Method

Data were collected from chart reviews of neurosurgical patients who were diagnosed with hemangioma of the vertebral column, and whose presenting signs and symptoms consistent with spinal cord compression over the last 7 years.

Results

From March 2007 to December 2013, a total of 3 patients with myelopathy caused by spinal cord compression from VH were enrolled in the study. In all

patients (1 male, 2 females), the tumors were located in the thoracic spine, and involved both anterior and posterior elements. Age of the patients at the time of surgery ranged from 14 to 37 years (mean 27 years). All of them underwent transarterial embolization and surgical decompression. The diagnosis of VH by pathological examination was confirmed in all patients after the surgery. Neurological evaluation was routinely performed before, and after the operation, during follow-up period at the out-patient department by using Nurick score. None of patients could ambulate before the operation.

All patients had plain radiographs and magnetic resonance imaging (MRI), but VH was suspected in only one of them before the surgery. In this patient, pre-operative spinal angiography with transarterial embolization was carried out when the above studies suggested the presence of large feeding or draining vessels. In the remaining 2 without suspicion of VH before surgery, the first operation was terminated because intraoperative massive bleeding occurred. Subsequently, spinal angiography with embolization was performed in both patients. Minimal intraoperative blood loss was noted in the second operation of each patient. Total excision was performed in two; both anterior and posterior approaches were used for complete tumor removal in separated sessions of the operation.

Intraoperative blood loss ranged from 1,000 to 6,700 ml (mean 4,257 ml). Operating time ranged from 345 to 415 minutes (mean 388 minutes). There was no perioperative complication in all patients. None of them received postoperative radiation therapy. The mean postoperative follow-up time was 22.3 months (range 10 to 43 months). Neurological status of the patients was gradually improved, they could ambulate by themselves at the last follow-up and no one had recurrent tumor.

Illustration cases

Case 1

A 14-year-old boy had symptom of paraparesis (Nurick score of 5 or chair-bound) after minor trauma that progressed in few months. Spinal MRI revealed VH with extraosseous extension at the T6 vertebral level. The tumor showed vivid enhancement on T1-weighted image (T1WI) with gadolinium. It severely compressed the spinal cord from the epidural extension posteriorly (Fig. 1). Surgical decompression of the spinal cord was planned. After preoperative transarterial embolization (Fig. 2), piecemeal resection of the tumor at the posterior elements and removal of epidural part were performed. His paraparesis was alleviated after

Fig. 1 Contrast-enhanced T1WI of the thoracic spine showing vivid enhancement of the vertebral body (asterisk), posterior element (arrow) and epidural extension of the tumor (arrowhead); VH with spinal cord compression is suspected.

Fig. 2 Spinal angiography of the segmental arteries showing stagnation of contrast media in VH.

the surgery. At 2 years after the surgery, the patient was completely asymptomatic (Nurick score of 1 or normal) and had no progression of the tumor (Fig. 3).

Case 2

A 51-year-old woman presented with a thyroid mass and mild paraparesis (Nurick score of 2 or gait difficulty but fully employed). Spinal MRI showed a mass at the T1 vertebral body; the provisional diagnosis was thyroid cancer with vertebral metastasis. We intended to perform thyroidectomy with corpectomy of the T1 vertebra, but the procedure was terminated due to intraoperative massive bleeding in the corpectomy. Spinal angiography with transarterial embolization was performed (Fig. 4). The second-stage of T1 corpectomy was done in the same admission with minimal intraoperative blood loss. The third operation (posterior decompression by laminectomy) without preoperative transarterial embolization was done at six months after the second operation. Significant intraoperative blood loss was found, but it could be surgically controllable. At 6 months after the last operation, the patient had no neurological deficit (Nurick score of 1 or normal).

Discussion

VH is the most common primary vertebral tumor. Most of VH are asymptomatic lesions discovered incidentally on screening radiographs. Spinal cord compression is a rare clinical manifestation of this type of tumor. Progressive neurologic deterioration with radiographic findings may masquerade malignant diseases. Primary concern of this disease can prevent intraoperative disaster event and help in management of the tumor.

Radiographic findings are helpful in diagnosis of VH. Classically, plain radiographs show coarse vertical striations or honeycombing of the vertebral body caused by the presence of thickened trabeculae. Computed tomography (CT) is the best diagnostic tool for defining the bony architecture and giving diagnosis of VH by "polka dots" appearance[35].

Spinal MRI is the investigation of choice when neurological deficit develops. The spinal cord and neighboring structures can be visualized very well. VH typically shows isointense signal on T1WI, which represents stable fat stroma and has high signal intensity on T2-weighted images (T2WI) associated with hypervascularized soft tissue involvement[35-38].

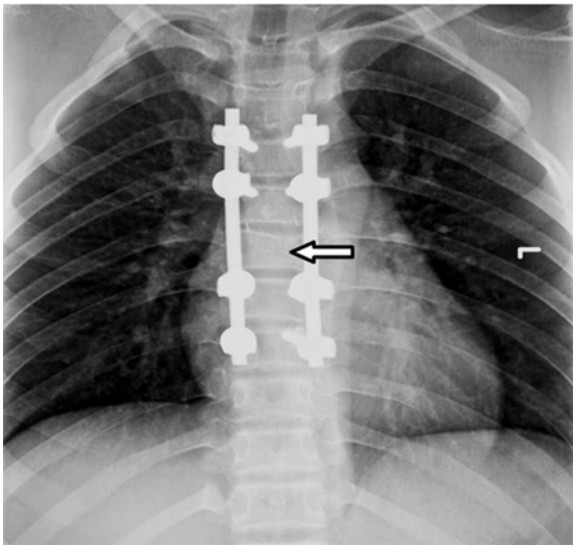

Fig. 3 Spinal instrumentation following surgical decompression of the affected vertebra (arrow); the patient had no neurological deficit and no progression of the tumor.

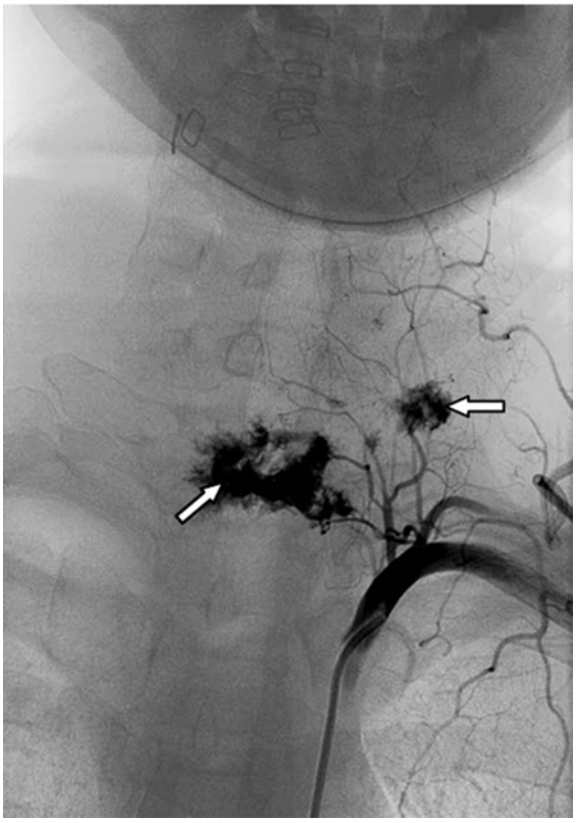

Fig. 4 Spinal angiography of the left subclavian artery showing tumor stain (arrow) and multiple arterial branches supplying VH

Spinal angiography with embolization is an optional modality and can be used as a preoperative adjunctive therapy to decrease intraoperative blood loss. Transarterial embolization can also relieve spinal cord compression caused by VH, reduce tumor size, provide pain relief, and relieve blockage of the subarachnoid space[17,24,39,40]. We suggest early surgical procedure following transarterial embolization because this tumor may resume vascular supply lately after the embolization.

Significant or progressive neurological deficit is an indication for surgical decompression[2,15,41,42]. Option of operative procedure depends on location and extent of the tumor[2,15,41-44]. Laminectomy is appropriate for posterior spinal cord compression. On the other hand, corpectomy with strut graft reconstruction is suitable for the tumor producing anterior compression of the spinal cord[41,42,45]. The mortality rate of surgical procedure is 6%. All deaths are due to exsanguination while attempting to remove large tumors in the ventral part of the vertebral body[46].

Even though neurological recovery can be seen following external radiation alone, radiation therapy is still controversial in management of VH[32]. However, in patients undergoing subtotal tumor removal for spinal cord decompression, postoperative irradiation is the traditional treatment protocol for these lesions[2,17].

Conclusion

VH should be one of the differential diagnoses in patients who manifest by spinal pain and myelopathy. Minimizing delays in diagnosis through increased clinician awareness and use of advanced imaging modalities may limit or prevent the complications associated with this disease. Multidisciplinary management can help in management of the tumor.

What is already known from this topic?

Patients with aggressive VH may present with pain or progressive myelopathy caused by spinal nerve root or spinal cord compression. Pre-operative transarterial embolization is useful for reducing intraoperative blood loss in surgical resection of aggressive VH.

What this study adds?

Aggressive VH is an important differential diagnosis in patients presenting with spinal cord compression by hypervascularized lesion of the vertebral body on contrast-enhanced spinal MRI. In cases with massive intraoperative bleeding, the operation should be terminated after proper hemostasis. Re-evaluation of the vascular supply of the lesion by spinal angiography and transarterial embolization are mandatory before retrying surgical resection of VH for decompression of the spinal cord.

Potential conflicts of interest

None.

References

1. Carnasale PG. Benign tumors of bone. In: Campbell WC, Canale ST, Beaty JH, editors. Campbell's operative orthopaedics. 10th ed. Philadelphia: Mosby/Elsevier; 2003: 793-812.
2. Fox MW, Onofrio BM. The natural history and management of symptomatic and asymptomatic vertebral hemangiomas. J Neurosurg 1993; 78: 36-45.
3. Lang EF Jr, Peserico L. Neurologic and surgical aspects of vertebral hemangiomas. Surg Clin North Am 1960; 40: 817-23.
4. Laredo JD, Reizine D, Bard M, Merland JJ. Vertebral hemangiomas: radiologic evaluation. Radiology 1986; 161: 183-9.
5. Rudnick J, Stern M. Symptomatic thoracic vertebral hemangioma: a case report and literature review. Arch Phys Med Rehabil 2004; 85: 1544-7.
6. Hemmy DC, McGee DM, Armbrust FH, Larson SJ. Resection of a vertebral hemangioma after preoperative embolization. Case report. J Neurosurg 1977; 47: 282-5.
7. Hemmy DC. Vertebral hemangiomas. In: Wilkins RH, Rengachary SS, editors. Neurosurgery. New York: McGraw-Hill; 1996: 1827-9.
8. Djindjian M, Nguyen JP, Gaston A, Pavlovitch JM, Poirier J, Awad IA. Multiple vertebral hemangiomas with neurological signs. Case report. J Neurosurg 1992; 76: 1025-8.
9. Camins MB, Oppenheim JS, Perrin RG. Tumors of the vertebral axis: benign, primary malignant and metastatic tumors. In: Youmans JR, editor. Neurological surgery. Philadelphia: W.B. Saunders; 1996: 3135-67.
10. Acosta FL Jr, Sanai N, Chi JH, Dowd CF, Chin C, Tihan T, et al. Comprehensive management of symptomatic and aggressive vertebral hemangiomas. Neurosurg Clin N Am 2008; 19: 17-29.
11. Aksu G, Fayda M, Saynak M, Karadeniz A. Spinal

cord compression due to vertebral hemangioma. Orthopedics 2008; 31: 169.
12. Eisenstein S, Spiro F, Browde S, Allen CM, Grobler L. The treatment of a symptomatic vertebral hemangioma by radiotherapy. A case report. Spine (Phila Pa 1976) 1986; 11: 640-2.
13. Goyal M, Mishra NK, Sharma A, Gaikwad SB, Mohanty BK, Sharma S. Alcohol ablation of symptomatic vertebral hemangiomas. Am J Neuroradiol 1999; 20: 1091-6.
14. Healy M, Herz DA, Pearl L. Spinal hemangiomas. Neurosurgery 1983; 13: 689-91.
15. Nguyen JP, Djindjian M, Gaston A, Gherardi R, Benhaiem N, Caron JP, et al. Vertebral hemangiomas presenting with neurologic symptoms. Surg Neurol 1987; 27: 391-7.
16. Reeves DL. Vertebral hemangioma with compression of the spinal cord. J Neurosurg 1964; 21: 710-2.
17. Jayakumar PN, Vasudev MK, Srikanth SG. Symptomatic vertebral haemangioma: endovascular treatment of 12 patients. Spinal Cord 1997; 35: 624-8.
18. Cotten A, Deramond H, Cortet B, Lejeune JP, Leclerc X, Chastanet P, et al. Preoperative percutaneous injection of methyl methacrylate and N-butyl cyanoacrylate in vertebral hemangiomas. Am J Neuroradiol 1996; 17: 137-42.
19. Gross CE, Hodge CH Jr, Binet EF, Kricheff II. Relief of spinal block during embolization of a vertebral body hemangioma. Case report. J Neurosurg 1976; 45: 327-30.
20. Hamza S, Meddeb N, Elleuch M, Rajhi H, Chour E, Sahu H, et al. Vertebroplasty success in a case of aggressive hemangioma. Tunis Med 2003; 81: 824-7.
21. Hekster RE, Luyendijk W, Tan TI. Spinal-cord compression caused by vertebral haemangioma relieved by percutaneous catheter embolisation. Neuroradiology 1972; 3: 160-4.
22. Ide C, Gangi A, Rimmelin A, Beaujeux R, Maitrot D, Buchheit F, et al. Vertebral haemangiomas with spinal cord compression: the place of preoperative percutaneous vertebroplasty with methyl methacrylate. Neuroradiology 1996; 38: 585-9.
23. Jones JO, Bruel BM, Vattam SR. Management of painful vertebral hemangiomas with kyphoplasty: a report of two cases and a literature review. Pain Physician 2009; 12: E297-303.
24. Smith TP, Koci T, Mehringer CM, Tsai FY, Fraser KW, Dowd CF, et al. Transarterial embolization of vertebral hemangioma. J Vasc Interv Radiol 1993; 4: 681-5.
25. Picard L, Bracard S, Roland J, Moreno A, Per A. Embolization of vertebral hemangioma. Technic-indications-results. Neurochirurgie 1989; 35: 289-8.
26. Mirra JM. Vascular tumors. In: Mirra JM, editor. Bone tumors. Vol.2. Philadelphia: Lea and Febiger; 1989: 1338-69.
27. Graham JJ, Yang WC. Vertebral hemangioma with compression fracture and paraparesis treated with preoperative embolization and vertebral resection. Spine (Phila Pa 1976) 1984; 9: 97-101.
28. Guarnieri G, Ambrosanio G, Vassallo P, Pezzullo MG, Galasso R, Lavanga A, et al. Vertebroplasty as treatment of aggressive and symptomatic vertebral hemangiomas: up to 4 years of follow-up. Neuroradiology 2009; 51: 471-6.
29. Hernigou P, Djindjian M, Ricolfi F, Dahhan P. Neuro-aggressive dorsal vertebral hemangioma and vertebrectomy. Apropos of 2 cases. Review of the literature. Rev Chir Orthop Reparatrice Appar Mot 1994; 80: 542-50.
30. Faria SL, Schlupp WR, Chiminazzo H Jr. Radiotherapy in the treatment of vertebral hemangiomas. Int J Radiat Oncol Biol Phys 1985; 11: 387-90.
31. Pavlovitch JM, Nguyen JP, Djindjian M, Mazeron JJ, Piedbois P, Le Bourgeois JP. Radiotherapy of vertebral hemangioma with neurologic complications. Neurochirurgie 1989; 35: 296-8.
32. Yang ZY, Zhang LJ, Chen ZX, Hu HY. Hemangioma of the vertebral column. A report on twenty-three patients with special reference to functional recovery after radiation therapy. Acta Radiol Oncol 1985; 24: 129-32.
33. Poungvarin N, Bhoopat W. Symptomatic vertebral haemangiomas: report of two cases. J Med Assoc Thai 1991; 74: 363-8.
34. Sirikulchayanonta V, Jinawath A, Jaovisidha S. Epithelioid hemangioma involving three contiguous bones: a case report with a review of the literature. Korean J Radiol 2010; 11: 692-6.
35. Ross JS, Masaryk TJ, Modic MT, Carter JR, Mapstone T, Dengel FH. Vertebral hemangiomas: MR imaging. Radiology 1987; 165: 165-9.
36. Hiari A, Nawaiseh B, Jeber H. Magnatic resonance imaging in the diagnosis of vertebral hemangiomas. East Mediterr Health J 1998; 4: 149-55.
37. Laredo JD, Assouline E, Gelbert F, Wybier M, Merland JJ, Tubiana JM. Vertebral hemangiomas:

fat content as a sign of aggressiveness. Radiology 1990; 177: 467-72.
38. Fujita T, Kawahara K, Tomita K, Ueda Y. Analysis on the MRI and histologic findings of symptomatic vertebral hemangioma. J Cent Jpn Orthop Traum 1999; 42: 571-2.
39. Gaston A, Nguyen JP, Djindjian M, Le Bras F, Gherardi R, Benhaiem N, et al. Vertebral haemangioma: CT and arteriographic features in three cases. J Neuroradiol 1985; 12: 21-33.
40. Gross CE, Hodge CH Jr, Binet EF, Kricheff II. Relief of spinal block during embolization of a vertebral body hemangioma. Case report. J Neurosurg 1976; 45: 327-30.
41. McAllister VL, Kendall BE, Bull JW. Symptomatic vertebral haemangiomas. Brain 1975; 98: 71-80.
42. Mohan V, Gupta SK, Tuli SM, Sanyal B. Symptomatic vertebral haemangiomas. Clin Radiol 1980; 31: 575-9.
43. Sar C, Eralp L. Double thoracic vertebral hemangioma causing complete paraplegia. Am J Orthop (Belle Mead NJ) 2004; 33: 81-4, 57.
44. Shah KC, Chacko AG. Extensive vertebral haemangioma with cord compression in two patients: review of the literature. Br J Neurosurg 2004; 18: 250-2.
45. Nguyen JP, Djindjian M, Pavlovitch JM, Badiane S. Vertebral hemangioma with neurologic signs. Therapeutic results. Survey of the French Society of Neurosurgery. Neurochirurgie 1989; 35: 299-8.
46. Pastushyn AI, Slin'ko EI, Mirzoyeva GM. Vertebral hemangiomas: diagnosis, management, natural history and clinicopathological correlates in 86 patients. Surg Neurol 1998; 50: 535-47.

การรักษาเนื้องอกหลอดเลือดของกระดูกสันหลังที่มีการกดทับไขสันหลัง

ธนภณ งามมณี, อัคคพงษ์ นิติสิงห์, บรรพต สิทธินามสุวรรณ, หลักชัย พลวิจิตร

ภูมิหลัง: *เนื้องอกหลอดเลือดของกระดูกสันหลังเป็นเนื้องอกที่พบบ่อยที่สุดของกระดูกสันหลัง ผู้ป่วยส่วนใหญ่ไม่มีอาการ พบความผิดปกติของระบบประสาทจากการกดทับไขสันหลังโดยเนื้องอกชนิดนี้ได้น้อย*

วัตถุประสงค์: *รายงานผู้ป่วยที่ได้รับการผ่าตัดรักษาการกดทับไขสันหลังโดยเนื้องอกหลอดเลือดของกระดูกสันหลัง*

วัสดุและวิธีการ: *การศึกษานี้เป็นการศึกษาย้อนหลังในผู้ป่วยเนื้องอกหลอดเลือดของกระดูกสันหลังที่มีการกดทับไขสันหลัง มีการรวบรวมผลการตรวจทางรังสี ภาพแม่เหล็กไฟฟ้าและการตรวจหลอดเลือดของกระดูกสันหลัง*

ผลการศึกษา: *ผู้ป่วยสามรายที่มีการกดทับไขสันหลังโดยเนื้องอกเข้าร่วมการศึกษา มีเพียงแค่รายเดียวที่สงสัยเนื้องอกหลอดเลือดของกระดูกสันหลังจากภาพแม่เหล็กไฟฟ้าก่อนผ่าตัด ผู้ป่วยรายดังกล่าวได้รับการอุดหลอดเลือดที่ไปเลี้ยงเนื้องอกเพื่อลดการเสียเลือดในขณะผ่าตัด ในผู้ป่วยอีกสองรายไม่ได้สงสัยเนื้องอกชนิดนี้ก่อนผ่าตัดจึงได้รับการผ่าตัดโดยไม่ได้รับการอุดหลอดเลือดที่ไปเลี้ยงเนื้องอก พบว่าเสียเลือดมากในขณะผ่าตัดจึงจำเป็นต้องหยุดการผ่าตัด แล้วจึงได้รับการอุดหลอดเลือดที่ไปเลี้ยงเนื้องอกก่อนที่จะผ่าตัดเนื้องอกครั้งที่สอง ซึ่งการผ่าตัดครั้งหลังมีการเสียเลือดน้อยมาก*

สรุป: *เนื้องอกหลอดเลือดของกระดูกสันหลังเป็นหนึ่งในโรคที่ควรคิดถึงในผู้ป่วยเนื้องอกกระดูกสันหลังที่มีอาการปวดและการกดทับไขสันหลัง สามารถลดการเสียเลือดขณะผ่าตัดได้โดยการอุดหลอดเลือดที่ไปเลี้ยงเนื้องอกผ่านทางหลอดเลือดแดง การรักษาแบบสหสาขาวิชาชีพเป็นสิ่งสำคัญในการรักษาเนื้องอกชนิดนี้*