Radiology Case Reports 19 (2024) 988–993

Available online at www.sciencedirect.com

ScienceDirect

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

Case Report

Clinical and radiographic response of a paravertebral hemangioma to radiotherapy ☆

Laura Burgess, MSc, MD[a,b], *Julia Malone, BSc*[a,b], *Marlise P. dos Santos, MSc,MPH, MD*[c,d,e], *John Sinclair, MD*[f], *Osbert Zalay, MD, PhD*[a,b], *Shawn Malone, MD*[a,b,*]

[a] *Department of Radiology, Division of Radiation Oncology, University of Ottawa, Ottawa, Ontario, Canada*
[b] *Department of Radiology, Division of Radiation Oncology, The Ottawa Hospital Research Institute, Ottawa, Ontario, Canada*
[c] *Department of Radiology, Radiation Oncology and Medical Physics, Section of Neuroradiology, University of Ottawa, Ottawa, Ontario, Canada*
[d] *Department of Surgery Divisions of Neurosurgery and Interventional Neuroradiology, The Ottawa Hospital Research Institute, Ottawa, Ontario, Canada*
[e] *Brain and Mind Research Institute, University of Ottawa, Ottawa, Ontario, Canada*
[f] *Department of Surgery, Division of Neurosurgery, University of Ottawa, Ottawa, Ontario, Canada*

ARTICLE INFO

Article history:
Received 24 May 2023
Revised 7 November 2023
Accepted 11 November 2023

Keywords:
Hemangioma
Stereotactic body radiotherapy
Image-guided radiotherapy

ABSTRACT

Hemangiomas can arise anywhere in the body. While vertebral hemangiomas are common, atypical hemangiomas with paraspinal and epidural extension are rare. We present a case of a patient who presented with persistent cough and anorexia from a paravertebral hemangioma that invaded the adjacent vertebrae and neural foramen causing moderate spinal canal stenosis. She was treated with stereotactic body radiotherapy to prevent the development of symptomatic spinal cord compression. The hemangioma underwent significant shrinkage and her cough resolved. This case demonstrates impressive and sustained clinical and radiographic response of a paraspinal hemangioma to stereotactic body radiotherapy.

Introduction

Hemangiomas are benign vascular tumors that can arise anywhere in the body. Vertebral hemangiomas occur in the spine and were first described by Virchow [1] and are common, with an incidence of 10%-12% in the population based on large autopsy studies [2,3]. However, it is very rare to see hemangiomas with epidural and paraspinal extension and, in fact, they are rarely mentioned in literature [4–6]. The more common vertebral tumors are typically asymptomatic with only 1% developing clinical symptoms [7], most commonly pain, likely

☆ Competing Interests: Shawn Malone has honoraria from the Astellas, Janssen, Abbvie, AstraZeneca, Amgen, Knight Therapeutics, and Bayer. Shawn Malone has travel grants from the following: Sanofi, TerSera. Marlise dos Santos has honoraria and travel grants from Stryker.

* Corresponding author.
E-mail address: smalone@toh.ca (S. Malone).

https://doi.org/10.1016/j.radcr.2023.11.032

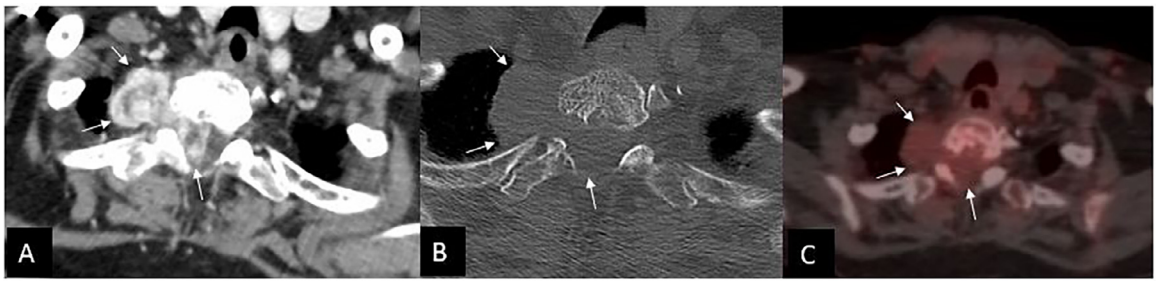

Fig. 1 – Initial imaging investigation. (A) Axial CT with contrast revealing a dumbbell-shaped heterogeneously enhancing epidural mass with right paraspinal extension, (B) Axial unenhanced CT bone window showing prominent trabeculae in the vertebral body and lamina of T1, and (C) FDG-PET revealing mild avidity within the lesion. The extent of the mass is marked with arrows.

from their extramedullary component [7,8]. Treatment options include surgical decompression, tumor resection, arterial embolization, percutaneous sclerotherapy, vertebroplasty, and radiotherapy [9–13]. Treatment decisions are individualized based on tumor and patient factors. Herein, we describe a case of an atypical hemangioma with epidural and paraspinal involvement and its clinical features and radiologic response to radiotherapy.

Case presentation

A 60-year-old female presented with persistent cough and anorexia for 6 months. She had no other associated respiratory complaints. She had a past medical history of mood disorder not otherwise specified and end-stage renal disease secondary to type 2 diabetes. Since starting dialysis, she has become fully dependent on her family for activities of daily living. Her nephrology team arranged for imaging investigations.

An computed tomography (CT) of the neck revealed an irregular soft tissue mass, isodense to muscle, centered in the right medial apical extra-pleural region with extension into the T1-2 neural foramen and paraspinal intrathoracic component (Fig. 1A). The T1 vertebral body, as well as its right-sided pedicle and transverse process, showed mildly expanded and thickened osseous marrow trabeculae typically described in vertebral hemangiomas (Fig. 1B). MRI showed a 34 × 23 × 30 mm (T × AP × CC) lobulated, dumbbell-shaped epidural soft tissue mass centered in the right T1-T2 neural foramen with ipsilateral paraspinal extension and hypointense T1 and hyperintense T2 -weighted signal relative to the spinal cord, and an avid enhancement following intravenous gadolinium administration (Fig. 2). Similar signal characteristics were noted involving the adjacent T1 vertebral body and its right-sided pedicle, transverse process and lamina and the adjacent right lateral aspect of T2 vertebral body and pedicle with a benign nonspecific appearance although atypical for a vertebral hemangioma. The thicker epidural component of the mass lesion measured approximately 13 × 8 × 30 mm (T × AP × CC) and was associated with a thin circumferential epidural enhancement (dural tail) extending from C7 to T4. The lesion caused moderate (50%-60%) spinal canal stenosis at the T1-T2 level without associated spinal cord edema or myelomalacia (Fig. 2I).

Positron emission tomography (PET) revealed homogeneous mild FDG uptake, SUV maximum 3.0 (Fig. 1C). CT-guided biopsy revealed small vascular spaces line with endothelial cells, without atypia, consistent with a hemangioma.

She was assessed by the spine surgical service and they did not recommend surgery. At the time of assessment, she did not have any focal neurological symptoms and, given her medical co-morbidities and poor performance status, their opinion was that surgery would pose a high risk with significant chance of negatively impacting her quality of life. In addition, due to the moderate cord compression (Bilsky score 2), imaging-guided percutaneous sclerotherapy or intra-arterial embolization was deemed too risky. Her case was reviewed at a thoracic multidisciplinary cancer conference. Pathology favored hemangioma and the recommendation was to proceed with definitive radiotherapy for management.

At the time of assessment by radiation oncology, she was unchanged from her baseline presentation. She did not have any chest pain or upper back pain, shortness of breath, hemoptysis, or upper GI symptoms. She was still having a persistent cough, especially after oral intake. She had no focal neurological deficits, no focal weakness or numbness, no urinary retention or fecal incontinence, and no saddle anesthesia.

She was consented to stereotactic body radiotherapy (SBRT) to the right paravertebral hemangioma and received 25 Gy in 5 fractions with a course of low-dose dexamethasone during treatment. Delivery of radiotherapy was by volumetric modulated arc therapy (VMAT) technique on an Elekta Linear Accelerator (Fig. 3). Daily cone beam imaging was used for image guidance. She tolerated treatment well without any side effects.

Three months after completing radiotherapy (Fig. 4), MRI revealed a slight enlargement of the paraspinal component of the lesion from 34 × 23 × 30 mm to 35 × 24 × 30 mm (T, AP, CC), due to postradiation intralesional necrosis, and a reduction in the size of its epidural component from 13 × 8 × 30

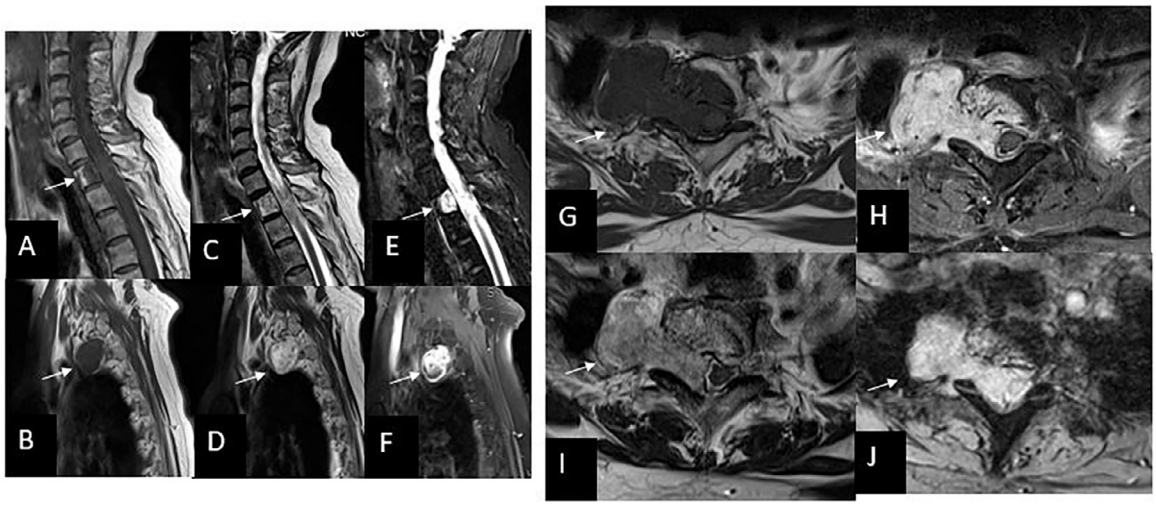

Fig. 2 – Baseline MRI showing the dumbbell-shaped epidural and paraspinal mass at the right T1-T2 level, with arrows highlighting the site of the disease. T1-TSE on (A) sagittal midline, (B) right parasagittal, and (G) axial planes; T2-TSE on (C) sagittal midline, (D) right parasagittal, and (I) axial planes; (E) T2-STIR at midline; T1-TSE fat-sat postgadolinium on (F) right parasagittal, and (H) axial planes; and (J) T2-MEDIC on axial plane.

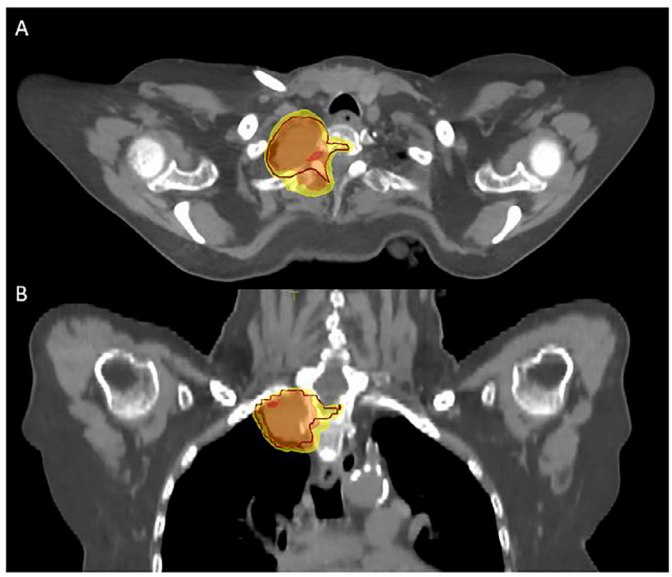

Fig. 3 – VMAT plan with PTV in red, 100% of the prescription dose in orange and 95% of prescription dose in yellow on both (A) Axial CT and (B) coronal CT images.

mm to 12 × 7 × 20 mm (T, AP, CC), with improvement of the mass effect on the spinal cord. She remained clinically asymptomatic without any upper back pain or neurologic dysfunction.

MRI spine at twenty-four months post-completion of radiotherapy (Fig. 5) revealed a further reduction in size of both the paraspinal (from 35 × 24 × 30 mm to 26 × 17 × 25 mm [T, AP, CC]) and the epidural components (from 12 × 7 × 20 mm to 12 × 7 × 19 mm [T, AP, CC]) with slightly less effacement of the spinal cord. She remained clinically well without any pain or neurologic dysfunction. Her cough had also completely resolved.

Discussion

While hemangiomas can arise anywhere in the body, vertebral hemangiomas are common. They are usually detected as an incidental finding on imaging but can present with pain or neurologic dysfunction if associated with an extramedullary component causing stenosis [7,8]. Unlike typical vertebral hemangiomas, atypical hemangiomas with paraspinal and epidural extensions are rarely seen [4–6], with only a few case reports of these presentations in the literature. There are case reports of cervical paraspinal hemangiomas managed

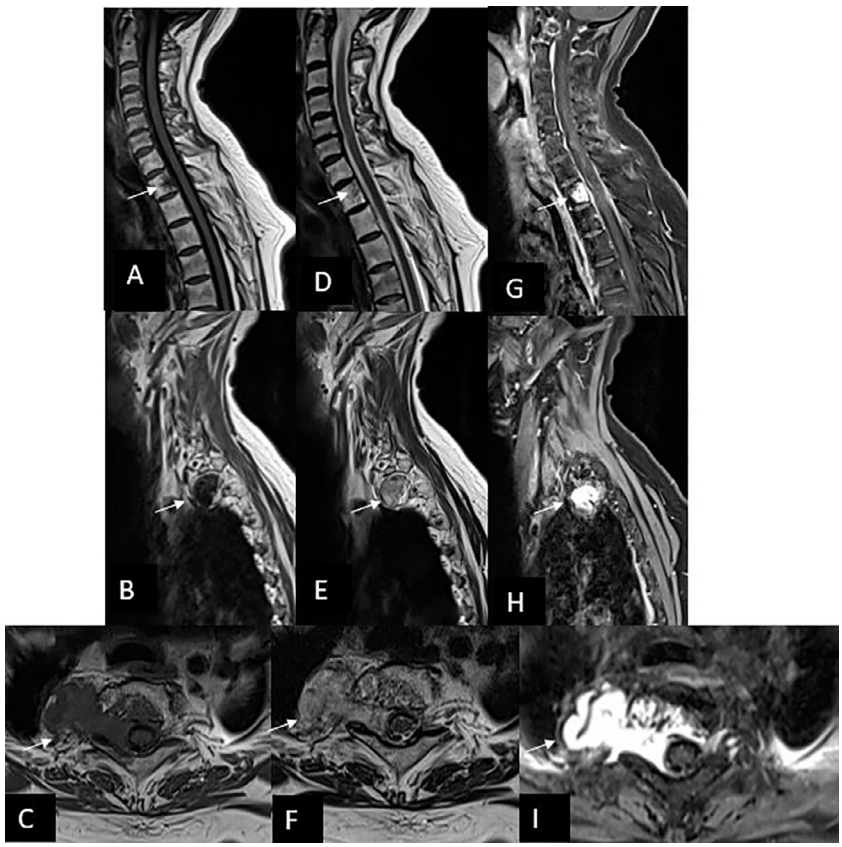

Fig. 4 – Three-month follow-up MRI showing reduction in size of the mass, with the mass arrowed, at the right T1-T2 level. T1-TSE on (A) sagittal midline, (B) right parasagittal, and (C) axial planes; T2-TSE on (D) sagittal midline, (E) right parasagittal, and (F) axial planes; T1-TSE fat-sat postgadolinium on (G) sagittal midline, (H) right parasagittal, and (I) axial planes.

surgically [4,5]. There are also reports of skin and subcutaneous hemangiomas associated with intraosseous hemangiomas, although these too are rare [6].

The patient in our report presented with a persistent cough and anorexia. Pulmonary symptoms may have resulted from pleural irritation by the tumor. The paraspinal hemangioma likely grew to invade the adjacent vertebrae and neural foramen causing severe neuroforaminal and moderate spinal canal stenoses. Interestingly, however, the patient did not complain of pain or focal neurological symptoms. Nevertheless, there was concern over the potential for development of symptomatic cord compression and consensus was that treatment was warranted in this case to prevent neurologic compromise.

Radiotherapy has long been used in the treatment of vertebral hemangiomas, either alone or in combination with another management strategy [14]. There is no standard approach for these patients, apart from those with frank spinal cord compression where decompressive surgery with or without postoperative radiotherapy is recommended [2,15]. Often, nonsurgical approaches are favored due to the highly vascular nature of the tumor, which could result in significant morbidity if intraoperative hemorrhage were to occur. As such, radiotherapy remains an attractive option for symptomatic lesions without spinal cord compression [15].

Vascular endothelial cells are considered radiosensitive, but the exact mechanism of effect with radiotherapy remains controversial [16]. The two commonly hypothesized radiobiological mechanisms are vascular fibrosis obliterating abnormal blood vessels [17] and anti-inflammatory effects from doses of 20 Gy or higher [18].

A retrospective review of 7 German institutions reviewed patients treated for symptomatic vertebral hemangiomas from 1969 to 2008 [14]. They found 84 patients with 96 symptomatic lesions who were treated with radiotherapy to a median total dose of 34 Gy, and a median daily dose of 2 Gy. The series demonstrated an overall pain response rate of 90.5%. Multivariate logistic regression demonstrated that total doses of at least 34 Gy or higher led to significantly greater symptomatic relief [14]. Given the performance status of the patient in this case and tumor characteristics, neither standard fractionation nor a total dose of 34 Gy was deemed suitable. Instead, a shorter hypofractionated course using 5 Gy per fraction was delivered to a total dose of 25 Gy, which is a higher biologically effective dose (BED) than 34 Gy at 2 Gy per fraction (BED of 87.5 Gy_2 vs 68.0 Gy_2). The treatment regimen was deemed to be safe at the biologically effective dose.

Radiotherapy has been demonstrated to be effective in the treatment of other vascular pathologies affecting the central nervous system. For example, stereotactic radiosurgery (SRS)

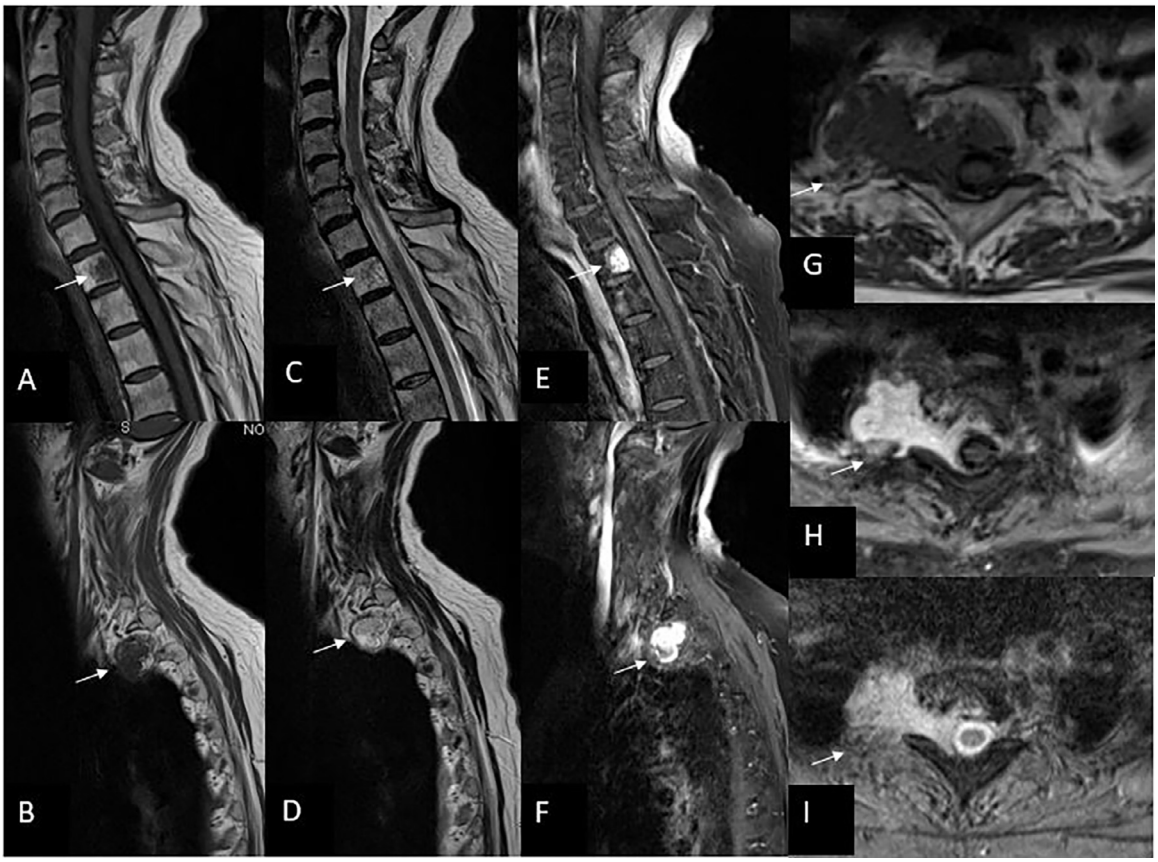

Fig. 5 – A 24-month follow-up MRI showing further reduction in the size of the mass, arrowed, at the right T1-T2 vertebral level. T1-TSE on (A) sagittal midline, (B) right parasagittal, and (G) axial planes; T2-TSE on (C) sagittal midline, and (D) right parasagittal planes; T1-TSE fat sat postgadolinium on (E) sagittal midline, (F) right parasagittal, and (H) axial planes; and (I) T2-MEDIC on axial plane.

for arteriovenous malformations (AVMs) with volumes of 3 mL or less has documented obliteration rates ranging from 72% to 96% [19,20]. Radiotherapy has also proven effective in larger AVMs with strategies including staged volume SRS [21] and hypofractionated radiotherapy [22,23]. Similarly, intramedullary spinal AVMs have been effectively treated with SRS and hypofractionated radiotherapy [24,25] using a BED of at least 60 Gy_2, although most receive a much higher effective dose than that.

Conclusion

In this report we present a rare case of paraspinal hemangioma, with only a handful of reports existing in the literature. The patient's symptoms at presentation were also atypical. She had a 6-month history of anorexia and a cough that resolved completely following treatment, despite imaging findings of epidural tumor invading the neural foramina and causing spinal canal stenosis, features that typically result in pain or neurologic dysfunction. Because of her medical comorbidities and poor performance status, surgery was deemed too high risk, and the location of the tumor pressing on the cord precluded embolization or percutaneous ablation. She was successfully treated with hypofractionated stereotactic body radiotherapy and has demonstrated an excellent and durable response both radiologically and clinically.

Patient consent

Consent was obtained from the power of attorney/substitute decision maker on behalf of the patient. We requested consent for publication of a case report regarding her paravertebral hemangioma and treatment. The nature of the case report was explained; that details of her clinical presentation and care would be included in a fully anonymized fashion. Consent was provided and this is documented in her chart.

REFERENCES

[1] Virchow R. Die Krankhaften Geschwulste. Hirschwald; 1863.
[2] Acosta FL, Dowd CF, Chin C, Tihan T, Ames CP, Weinstein PR. Current treatment strategies and outcomes in the management of symptomatic vertebral hemangiomas.

Neurosurgery 2006;58:287–95. doi:10.1227/01.NEU.0000194846.55984.C8.
[3] Blankstein A, Spiegelmann R, Shacked I, Schinder E, Chechick A. Hemangioma of the thoracic spine involving multiple adjacent levels: case report. Paraplegia 1988;26:186–91. doi:10.1038/sc.1988.29.
[4] Yu D, Choi JH, Jeon I. Intramuscular hemangioma with hemorrhagic transformation arising from paraspinal muscles of posterior neck: a case report. Medicine (Baltimore) 2020;99:e21741. doi:10.1097/MD.0000000000021741.
[5] Makeieff M, Maurice N, Mondain M, Crampette L, Guerrier B. Intramuscular hemangioma of posterior neck muscles. Eur Arch Otorhinolaryngol 2001;258:28–30.
[6] Bansal H, Chaudhary A, Mahajan A. Large cervical paraspinal haemangioma in association with intraosseous haemangioma: a rare case report. J Clin Diagn Res 2015;9:PD01–2. doi:10.7860/JCDR/2015/13513.6292.
[7] Pastushyn AI, Ph D, Slin EI, Mirzoyeva GM. Management, natural history and clinicopathological correlates in 86 patients. Spinal Cord 1998;50:535–47.
[8] Harrison MJ, Eisenberg MB, Ullman JS, Oppenheim JS, Camins MB, Post KD. Symptomatic cavernous malformations affecting the spine and spinal cord. Neurosurgery 1995;37:195–204.
[9] Aich RK, Deb AR, Banerjee A, Karim R, Gupta P. Symptomatic vertebral hemangioma: treatment with radiotherapy. J Cancer Res Ther 2010;6:199–203. doi:10.4103/0973-1482.65248.
[10] Kato S, Kawahara N, Murakami H, Demura S, Yoshioka K, Okayama T, et al. Surgical management of aggressive vertebral hemangiomas causing spinal cord compression: long-term clinical follow-up of five cases. J Orthop Sci 2010;15:350–6. doi:10.1007/s00776-010-1483-z.
[11] Gupta AK, Phukan P, Bodhey N. Percutaneous vertebroplasty for the treatment of symptomatic vertebral hemangioma with long-term follow-up. Interdiscip Neurosurg Adv Tech Case Manag 2021;23:100968. doi:10.1016/j.inat.2020.100968.
[12] Gabal AM. Percutaneous technique for sclerotherapy of vertebral hemangioma compressing spinal cord. Cardiovasc Intervent Radiol 2002;25:494–500. doi:10.1007/s00270-002-1944-7.
[13] Robinson Y, Sheta R, Salci K, Willander J. Blood loss in surgery for aggressive vertebral haemangioma with and without embolization. Asian Spine J 2015;9:483–91. doi:10.4184/asj.2015.9.3.483.
[14] Heyd R, Seegenschmiedt MH, Rades D, Winkler C, Eich HT, Bruns F, et al. Radiotherapy for symptomatic vertebral hemangiomas: results of a multicenter study and literature review. Int J Radiat Oncol Biol Phys 2010;77:217–25. doi:10.1016/j.ijrobp.2009.04.055.
[15] Fox MF, Onofrio BM. The natural history and management of symptomatic and asymptomatic vertebral hemangiomas. J Neurosurg 1993;78:36–45.
[16] Heyd R, Seegenschmiedt MH. Vertebral hemangioma (SVH). Radiother. non-malignant disord 2008:425–40 Contemp. concepts Clin. results..
[17] Faria SL, Schlupp WR, Chiminazzo H. Radiotherapy in the treatment of vertebral hemangiomas. Int J Radiat Oncol Biol Phys 1985;11:387–90. doi:10.1016/0360-3016(85)90162-2.
[18] Miszczyk L, Ficek K, Trela K, Spindel J. The efficacy of radiotherapy for vertebral hemangiomas. Neoplasma 2001;48:82–4.
[19] Friedman WA, Bova FJ, Mendenhall WM. Linear accelerator radiosurgery for arteriovenous malformations: the relationship of size to outcome. J Neurosurg 1995;82:180–9. doi:10.3171/jns.1995.82.2.0180.
[20] Yamamoto Y, Coffey RJ, Nichols DA, Shaw EG. Interim report on the radiosurgical treatment of cerebral arteriovenous malformations. The influence of size, dose, time, and technical factors on obliteration rate. J Neurosurg 1995;83:832–7. doi:10.3171/jns.1995.83.5.0832.
[21] Pollock BE, Kline RW, Stafford SL, Foote RL, Schomberg PJ. The rationale and technique of staged-volume arteriovenous malformation radiosurgery. Int J Radiat Oncol Biol Phys 2000;48:817–24. doi:10.1016/S0360-3016(00)00696-9.
[22] Chang TC, Shirato H, Aoyama H, Ushikoshi S, Kato N, Kuroda S, et al. Stereotactic irradiation for intracranial arteriovenous malformation using stereotactic radiosurgery or hypofractionated stereotactic radiotherapy. Int J Radiat Oncol Biol Phys 2004;60:861–70. doi:10.1016/j.ijrobp.2004.04.041.
[23] Karlsson B, Lindqvist M, Blomgren H, Wan-Yeo G, Söderman M, Lax I, et al. Long-term results after fractionated radiation therapy for large brain arteriovenous malformations. Neurosurgery 2005;57:42–8. doi:10.1227/01.NEU.0000163095.56638.26.
[24] Sinclair J, Chang SD, Gibbs IC, Adler JR. Multisession CyberKnife radiosurgery for intramedullary spinal cord arteriovenous malformations. Neurosurgery 2006;58:1081–8. doi:10.1227/01.NEU.0000215891.25153.BA.
[25] Hida K, Shirato H, Isu T, Seki T, Onimaru R, Aoyama H, et al. Focal fractionated radiotherapy for intramedullary spinal arteriovenous malformations: 10-year experience. J Neurosurg 2003;99:34–8. doi:10.3171/spi.2003.99.1.0034.