Surgical treatment of aggressive vertebral hemangiomas

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OBJECTIVE Vertebral hemangiomas are common tumors that are benign and generally asymptomatic. Occasionally these lesions can exhibit aggressive features such as bony expansion and erosion into the epidural space resulting in neurological symptoms. Surgery is often recommended in these cases, especially if symptoms are severe or rapidly progressive. Some surgeons perform decompression alone, others perform gross-total resection, while others perform en bloc resection. Radiation, embolization, vertebroplasty, and ethanol injection have also been used in combination with surgery. Despite the variety of available treatment options, the optimal management strategy is unclear because aggressive vertebral hemangiomas are uncommon lesions, making it difficult to perform large trials. For this reason, the authors chose instead to report their institutional experience along with a comprehensive review of the literature.

METHODS A departmental database was searched for patients with a pathological diagnosis of “hemangioma” between 2008 and 2015. Medical records were reviewed to identify patients with aggressive vertebral hemangiomas, and these cases were reviewed in detail.

RESULTS Five patients were identified who underwent surgery for treatment of aggressive vertebral hemangiomas during the specified time period. There were 2 lumbar and 3 thoracic lesions. One patient underwent en bloc spondylectomy, 2 patients had piecemeal gross-total resection, and the remaining 2 had subtotal tumor resection. Intraoperative vertebroplasty was used in 3 cases to augment the anterior column or to obliterate residual tumor. Adjuvant radiation was used in 1 case where there was residual tumor as well. The patient who underwent en bloc spondylectomy experienced several postoperative complications requiring additional medical care and reoperation. At an average follow-up of 31 months (range 3–65 months), no patient had any recurrence of disease and all were clinically asymptomatic, except the patient who underwent en bloc resection who continued to have back pain.

CONCLUSIONS Gross-total resection or subtotal resection in combination with vertebroplasty or adjuvant radiation therapy to treat residual tumor seems sufficient in the treatment of aggressive vertebral hemangiomas. En bloc resection appears to provide a similar oncological benefit, but it carries higher morbidity to the patient.

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VERTEBRAL hemangiomas are benign vasoformative neoplasms of endothelial cells that grow within marrow spaces in the bone and encase bony trabeculae. They are generally considered neoplasms, but due to the lack of aggressive histopathological features, some authors have referred to them as hamartomas or vascular malformations.^{32,37,45,72} There is at least one associated genetic rearrangement that has been described resulting in a new fusion gene, *EWSRI-NFATC1*, which suggests that vertebral hemangiomas may in fact be neoplasms, but this remains controversial.⁵ In the majority of patients, these lesions remain asymptomatic and do not require any treatment or surveillance. In 0.9%–1.2% of patients, however,

the lesions can cause symptoms due to neural compression as a result of bone expansion, erosion through cortex, fracture, or hematoma.^{8,19,24,27,52–54} These Enneking Stage 3 lesions—which are termed “aggressive vertebral hemangiomas”—are more likely than their asymptomatic counterparts to involve the entire vertebral body, extend into the posterior elements, and have an irregular honeycomb pattern with lytic areas on radiological evaluation.^{21,29,50} The presenting symptoms are dependent on the location of the tumor and the degree of spinal cord or nerve root compression.¹⁰ Female patients may be more prone to become symptomatic during the last trimester of pregnancy, presumably due to the hemodynamic effects of a gravid

ABBREVIATIONS EBL = estimated blood loss; NBCA = *N*-butyl cyanoacrylate.

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uterus.^{12,48,51,59,61,64,67} Aggressive vertebral hemangiomas are also more common in adults but have been reported in children as well.^{11,18,44,54}

It is generally agreed that surgical intervention is warranted when hemangiomas cause pain or neurological symptoms, however the optimal treatment strategy is controversial.^{1,31} Some surgeons have used less-invasive strategies, such as vertebroplasty, endovascular or percutaneous embolization, ethanol injection, and radiation therapy. Others recommend surgery in the form of decompression, gross-total resection, or even en bloc resection. Because of the relative infrequency of these lesions and sparse reporting in the literature, there are few studies with long-term follow-up that directly compare treatment modalities. Also, some authors reported on a heterogeneous group of patients who had local pain referable to intracompartmental vertebral hemangiomas without neural compression or aggressive features.⁵⁰ For this reason, we feel it is important to publish our own institutional experience along with a thorough review of the literature.

Methods

A departmental database was searched for all patients who underwent surgery and had a pathological diagnosis of “hemangioma” between 2008 and 2015. Medical records and radiology studies were retrospectively reviewed to identify patients with aggressive vertebral hemangiomas. All patients underwent both CT and MRI routinely. In cases where the diagnosis was uncertain based on the imaging studies, a biopsy was performed to rule out other diagnoses that might require different treatment. All patients in this series underwent surgery for subtotal resection, gross-total resection, or en bloc resection, and preoperative embolization was performed when appropriate. Patients with mechanical instability of the spinal column following resection underwent concurrent instrumented fusion. In some cases in which a partial vertebrectomy was necessary for resection of the tumor, intraoperative vertebroplasty was used to augment the load-sharing capacity of the remaining vertebral body. In other cases in which a complete vertebrectomy was performed, a structural cage was placed. Patients with spinal tumors are managed by a multidisciplinary team of neurosurgeons, orthopedic surgeons, oncologists, and radiation oncologists at our institution. This team determined whether patients underwent adjuvant radiation therapy and at what dose. When reviewing patient records, information was collected regarding age, sex, location of the tumor, preoperative embolization, surgical technique, spinal reconstruction, estimated blood loss (EBL), complications, and recurrence. The patient’s clinical status at follow-up was also recorded.

Results

Five patients who underwent surgical treatment of aggressive vertebral hemangiomas were identified. In this series, 4 patients were female and 1 was male. The mean age at the time of surgery was 51.4 years (range 30–71 years). Four patients presented with neurological deficits that were referable to the hemangioma, and 1 patient presented with worsening back pain postpartum. Neurological defi-

cits included radiculopathy, myelopathy, and neurogenic claudication. Imaging in all patients demonstrated aggressive features such as involvement of the entire vertebral body, extension into the posterior elements, expansion of bone, and epidural extension. Two lesions were located in the lumbar spine and 3 were located in the thoracic spine.

Two patients underwent percutaneous biopsy prior to surgery. Four patients were treated with preoperative embolization to minimize bleeding during the operation. All surgeries were performed from a posterior-only approach. In 2 cases, subtotal resection was performed with a goal of decompressing the thecal sac; in 2 cases, piecemeal gross total resection was performed; and in 1 case, en bloc spondylectomy was performed. Both of the patients who underwent gross-total resection and one of the patients who underwent subtotal resection had intraoperative vertebroplasty (Fig. 1). Four patients required an instrumented fusion at the time of surgery to prevent the possibility of iatrogenic instability. The patient who underwent en bloc resection also required anterior column reconstruction with a titanium cage (Fig. 2). One patient (Case 5) who underwent piecemeal gross-total resection did not require instrumented fusion. Estimated blood loss was recorded in 4 of the 5 cases and ranged from 400 to 500 ml for patients who underwent subtotal or gross-total resection. The EBL for the patient (Case 1) who underwent en bloc spondylectomy was 1800 ml. This patient was also the only patient who experienced early or late complications as a result of surgery (Table 1).

One patient (Case 2), the patient who underwent subtotal resection but did not require vertebroplasty, received fractionated proton radiotherapy (45 Gy in 25 fractions) (Fig. 3). Patients were followed for an average of 31 months (range 3–65 months). No patient has experienced clinical or radiographic recurrence, and all of the patients’ symptoms have resolved, except for the patient who underwent en bloc resection, who continues to have back pain.

Illustrative Case

A 45-year-old female (Case 5) presented with right-sided flank pain consistent with a thoracic radiculopathy. She underwent a CT scan and subsequent MRI, which revealed an aggressive vertebral hemangioma centered within the T-8 vertebral body with bony and soft tissue extension into the spinal canal and right T8–9 neural foramen (Fig. 4).

Because her symptoms were due to a focal bony prominence displacing the right T-8 nerve root, it was felt that nonoperative treatment modalities, including radiation, embolization, vertebroplasty, or ethanol injection, would not be useful as stand-alone therapies. Surgery was therefore scheduled to resect the lesion with preoperative embolization under the same anesthesia to minimize the risk of blood loss.

On the day of surgery, an angiogram was performed for embolization. During the angiogram a hypervascular blush was observed at the T-8 vertebral body with supply from both the right and left T-8 segmental arteries. The embolization was completed with coils and *N*-butyl cyanoacrylate (NBCA), and following this the hypervascular blush was no longer seen.

FIG. 1. Images obtained in Case 3, which involved a 30-year-old woman who presented with progressive postpartum back pain. **A and B:** Axial (A) and sagittal (B) CT images demonstrating an extensive aggressive vertebral hemangioma involving the entire T-11 vertebra, including the transverse processes and spinous process. **C and D:** Axial (C) and sagittal (D) T2-weighted MR images demonstrating soft tissue extension of the tumor into the epidural space resulting in spinal cord compression. **E:** Postoperative lateral radiograph obtained after subtotal resection and vertebroplasty showing cement filling the residual vertebral body, which was involved in tumor. Coils from the preoperative embolization can also be seen here.

The patient was then taken to the operating room and turned prone on an open Jackson table. A semilunar incision was made, and a flap of skin, subcutaneous tissue, and fascia was rotated medially over the spinous processes. A right-sided periosteal exposure was then performed, revealing only the base of the spinous process, lamina, and facet joints from T-7 through T-9 while keeping the posterior longitudinal ligament and muscular attachments intact. A hemilaminectomy was then created from the inferior aspect of T-7 to the superior aspect of T-9. A medial facetectomy was performed at T7–8 and T8–9, and the medial portion of the T-8 and T-9 pedicles was removed with a high-speed drill. The T-8 nerve root was now exposed as it passed caudal to the pedicle, and it was ligated proximal to the dorsal root ganglion. At this point, the tumor was visible and was separated from the dura with a Penfield No. 4 dissector. Both a soft tissue and a bony component of the tumor were seen in the epidural space and tracking into the right T8–9 neural foramen. The soft tissue component was removed with a scalpel and a small pituitary rongeur. The bony component was subsequently removed with the drill. The tumor that remained in the foramen was removed with an angled curette. Using the drill followed by a series of straight and angled curettes the central portion of the hemangioma within the vertebral body was then removed. Because this required resection of a large portion of the vertebral body, intraoperative vertebroplasty was performed under fluoroscopic guidance to

restore the anterior load-sharing capacity at that segment. Instrumentation was not required due to the preservation of the midline tension band and the facets. The wound was closed in layers.

Following surgery, the patient recovered to her neurological baseline. At her first follow-up appointment her symptoms were completely resolved and she reported only a small area of numbness in the distribution of the right T-8 nerve root, which had been sacrificed. At her most recent follow-up visit, 8 months after surgery, she continued to do well with no clinical or radiographic signs of recurrence.

Discussion

Vertebral hemangiomas were first reported by Virchow in 1863 and were first associated with neurological symptoms by Gerhardt in 1895. Later, in 1927, Makrykostas described how “ballooning” of the vertebrae or epidural extension of a vertebral hemangioma could result in spinal canal stenosis and neurological symptoms.⁵² Despite our long-standing recognition of aggressive vertebral hemangiomas, there is still controversy regarding the optimal treatment strategy, and numerous therapeutic options have been described (Tables 2–4).

Radiation Therapy

Radiation therapy was recognized as an effective strat-

FIG. 2. Images obtained in Case 1, which involved a 48-year-old man who presented with back pain and radiculopathy. **A and B:** Axial (A) and sagittal (B) CT images demonstrating an aggressive vertebral hemangioma involving the entire vertebral body of L-1 and extending through both pedicles into the posterior elements. **C and D:** Axial (C) and sagittal (D) T2-weighted MR images showing additional epidural soft tissue extension within the spinal canal resulting in compression of the conus medullaris and cauda equina. The patient underwent en bloc resection requiring extensive spinal reconstruction. **E:** Lateral radiograph obtained 28 months after the initial surgery when the patient presented with severe back pain and was found to have a fractured rod.

egy for the treatment of aggressive vertebral hemangiomas by the 1930s. Since that time it has been recommended as a primary modality only for the treatment of slowly progressive lesions primarily because the therapeutic effects are delayed.^{19,32,45,52} In 1951, Manning⁵² described several cases in which radiation therapy resulted in complete resolution of symptoms and controlled local progression of disease. Glanzmann et al.³⁰ described a series of 62 patients treated with radiation therapy between 1939 and 1975 and reported that 60% experienced permanent improvement. In 1985, Faria et al.²⁴ described a series of 9 patients with symptomatic vertebral hemangiomas treated with radiation therapy and reported that 77% had complete or near-complete resolution of symptoms, including a woman who had presented with paraplegia during pregnancy. They also reported that no patient who initially responded to treatment experienced recurrence of disease. Yang et al.⁷⁰ reported on 23 patients with aggressive hemangiomas who underwent radiation therapy and found that over 80% experienced relief of pain and sensory symptoms and 5 of 7 patients who presented with paraplegia regained the ability to walk. In another study of 17 cases in which patients were treated with radiation, 87.5% had complete resolution of pain, 66.7% of patients had complete resolution of numbness or paresis, and 66.6% of patients with paraplegia recovered completely. In this study the patients who did not recover had long-standing paraplegia and presumably irreversible spinal cord injury.⁶ In 2014, Jiang et al.⁴⁵

published a retrospective series of 29 consecutive patients with aggressive vertebral hemangiomas. In this series 10 patients with slowly progressive neurological deficits underwent radiation therapy alone, but the treatment failed in 2 cases, and those 2 patients required surgery.

A retrospective analysis of pooled data suggests that there is a dose-effect relationship for the treatment of symptomatic vertebral hemangiomas with radiation alone, and the authors recommended a total dose of 40 Gy.⁵⁸ In 2010, Aich et al.³ treated 7 consecutive patients with aggressive vertebral hemangiomas with 40 Gy of external beam radiation therapy over 4 weeks and noted that all of the patients tolerated treatment well and demonstrated improvement in motor strength. At last follow up, 6 of 7 had either no weakness or mild weakness only. With the development of intensity-modulated radiation therapy, it may be possible to safely deliver higher doses of radiation while avoiding complications such as radiation myelopathy and pulmonary radionecrosis, but this has not been shown in patients with vertebral hemangiomas.^{3,16,62}

It should be noted that the radiographic appearance of aggressive vertebral hemangiomas has not been reported to change after a number of years, even with successful treatment.^{24,52} This likely indicates that radiation is effective at controlling pathological vascular tissue but does not have a radiographically demonstrable effect on the surrounding bony tissue. For this reason authors have suggested that radiation alone may be less effective for

TABLE 1. Summary of patient demographic and clinical characteristics

Case No.	Age (yrs)* Sex	Presentation	Site	Bone Involvement	Percut Biopsy	Preop Embol	Surgical Approach	Method of Spinal Fixation					Complications	FU (mos)	Recurrence	Clinical Status
								EOR	Ant Column Reconstr	EBL (ml)	Adjuvant Radiation					
1	48, M	BP & radiculopathy	L-1	VB, both pedicles, pst elements	No	Yes	Pst	En bloc spondylectomy	Pst	Titanium cage	1800	No	CSF leak, wound infection requiring washout, DVT, hardware failure requiring revision, wound infection requiring washout	43	None	Continues to have BP
2	63, F	Myelopathy	T-4	VB, both pedicles, pst elements	No	Yes	Pst	STR	Pst	None	500	Yes	None	65	None	Sx resolved
3	30, F	BP	T-11	VB, both pedicles, pst elements	No	Yes	Pst	STR	Pst	VP	500	No	None	36	None	Sx resolved
4	71, F	BP & neurogenic claudication	L-4	VB	Yes	No	Pst	GTR	Pst	VP	NR	No	None	3†	None	Sx resolved
5	45, F	BP & radiculopathy	T-8	VB, base of rt pedicle	Yes	Yes	Pst	GTR	Pst	VP	400	No	None	8	None	Sx resolved

Ant = anterior; BP = back pain; DVT = deep venous thrombosis; Embol = embolization; EOR = extent of resection; FU = follow-up; GTR = gross-total resection; NR = not reported; pst = posterior; Percut = percutaneous; Reconstr = reconstruction; STR = subtotal resection; Sx = symptoms; VB = vertebral body; VP = vertebroplasty.

* Age at surgery.

† Lost to follow-up.

FIG. 3. Images obtained in Case 2, which involved a 63-year-old woman who presented with myelopathy. **A:** Axial CT image demonstrating an aggressive vertebral hemangioma at T-4 with a characteristic honeycomb pattern that expands the cortex and involves the entire vertebral body and both pedicles and extends into the posterior elements. **B:** Sagittal CT image showing a corduroy pattern characteristic of vertebral hemangiomas. **C and D:** Axial (C) and sagittal (D) T2-weighted MR images showing epidural spread of disease. This patient underwent a decompressive laminectomy and instrumented fusion for subtotal resection of the tumor followed by a course of radiation therapy. **E:** Postoperative anteroposterior radiograph.

patients who have neurological compression due to focal bone hypertrophy.¹⁶

Vertebroplasty

Vertebroplasty with methyl methacrylate cement has been described in the treatment of vertebral hemangiomas since the late 1980s.⁷⁴ This technique not only provides hemostatic embolization, but also improves the load-bearing capacity of the anterior column. Feydy et al.²⁵ reported 2 cases of painful cervical vertebral hemangiomas that were treated with vertebroplasty, resulting in immediate resolu-

tion of symptoms, but neither of these lesions had cortical expansion or epidural extension characteristic of aggressive vertebral hemangiomas. Some authors have recommended against the use of vertebroplasty alone in the treatment of aggressive hemangiomas as the cement may form a dense cast including the epidural portion of the cavity.^{1,32} Others have used vertebroplasty in these cases with success and have reported shrinkage of the epidural component due to decreased vascularity.^{34,45} One case in which vertebroplasty was used to treat an aggressive hemangioma with epidural disease was complicated by leaking of cement into the spinal canal.²³ Guarnieri et al.³⁴ published a series of 24 cases of vertebral hemangiomas including 6 with aggressive features and noted that none of the patients had recurrent symptoms at 4-year follow-up. Balloon kyphoplasty has been reported to decrease the risk of cement leakage.^{46,74} Either kyphoplasty or vertebroplasty may also be used intraoperatively in conjunction with decompressive surgery as we have done with several of our patients in the present series.⁴¹

Ethanol Ablation

Percutaneous ethanol ablation of aggressive vertebral hemangiomas was first described in 1994 by Heiss, Doppman, and Oldfield.³⁸ A series of 14 patients with aggressive vertebral hemangiomas treated with percutaneous ethanol ablation showed that 93% of patients had an “excellent” or “good” clinical result within 14 months of

FIG. 4. Images obtained in Case 5, which involved a 45-year-old woman who presented with right flank pain. Axial (left) and sagittal (right) CT images demonstrating irregular vertical trabeculae in the T-8 vertebral body as well as cortical expansion involving the posterior cortex and pedicle.

TABLE 2. Selective review of key literature on radiation therapy for aggressive vertebral hemangiomas

Authors & Year	Study Design	No. of Pts	Results	Conclusions
Manning, 1951	Retrospective	3	Clinical improvement in all 3 pts following RT w/ no recurrence at 4–10 yrs FU	RT is acceptable as a primary Tx for symptomatic vertebral hemangiomas w/o sudden cord compression
Glanzmann et al., 1977	Retrospective	62	Permanent clinical improvement seen in ~60% of pts following RT	Radiation may be used to treat symptomatic hemangiomas. In pts w/ severe cord compression, decompressive surgery should precede RT
Faria et al., 1985	Retrospective	9	77% had complete/near-complete resolution of Sx following RT w/ 6–44 mos FU & no complications	RT is safe & effective for Tx of symptomatic vertebral hemangiomas
Yang et al., 1985	Retrospective	23	88% experienced relief of BP, 80% experienced relief of numbness, 5 of 7 paraplegic pts regained ability to ambulate	Even w/ severe spinal cord compression, radiation can be used as primary Tx
Asthana et al., 1990	Retrospective	17	87.5% experienced complete relief of pain/tenderness, 66.7% experienced complete relief of numbness, 6 of 9 paraplegic pts recovered completely, & 2 had no response	Symptomatic vertebral hemangiomas can be treated w/ radiation. Radiation may be used as primary Tx
Aich et al., 2010	Retrospective	7	100% showed improvement in strength, pain relief, & increased quality of life	RT is dose dependent
Jiang et al., 2014	Retrospective	29	In 2 of 10 pts who underwent RT alone, Tx failed & surgery was required. No recurrences were seen following RT, average FU 51.1 mos	RT may be useful as a stand-alone Tx in pts w/ mild or slowly progressive neurological deficits

Pts = patients; RT = radiation therapy; Tx = treatment.

the procedure.³² Another series of 11 patients with aggressive vertebral hemangiomas treated with ethanol injection showed complete obliteration of the lesions on post-procedure angiography and no recurrence at follow-up of 15–76 months.¹⁷ Although this treatment strategy has been effective in ameliorating neurological symptoms, there is little long-term data on the rate of recurrence and numerous complications have been reported, including osteonecrosis, vertebral collapse, transient neurological deterioration, spinal cord injury, hemodynamic instability, and asystole.^{17,32,38,55,63,69} To avoid the risk of vertebral collapse, some surgeons may inject lower doses of ethanol or combine ethanol injection with percutaneous vertebroplasty.^{13,17} A case report by Chen et al.¹⁰ also describes the successful treatment of an aggressive vertebral hemangioma using ethanol injection in combination with en-

dovascular embolization with no clinical or radiographic recurrence at 21-month follow-up. Nevertheless, because there are other treatment options with comparable clinical outcomes and fewer complications, alcohol ablation has fallen out of favor.

Endovascular Embolization

Vertebral hemangiomas are highly vascular lesions and one of the greatest risks of surgical intervention is uncontrollable hemorrhage.^{4,6,19} In 1951, without the benefit of modern surgical technology, Manning estimated the operative mortality rate associated with resection of a vertebral hemangioma to be 20%–25% due to uncontrollable bleeding.⁵² In the 1970s it was suggested that angiography could be used not only to confirm the diagnosis but also to

TABLE 3. Selective review of key literature on embolization for aggressive vertebral hemangiomas

Authors & Year	Study Design	No. of Pts	Results	Conclusions
Gross et al., 1976	Retrospective	1	Neurological improvement w/in 48 hrs following endovascular embol w/ Gelfoam	Therapeutic embol should be considered in pts w/ aggressive hemangiomas
Hekster & Endtz, 1987	Retrospective	1	Paraplegic pt treated w/ embol followed by RT was in excellent clinical condition 7 mos later; pt remained in excellent condition at 15-yr FU	Endovascular embol may result in good long-term control when used in combination w/ RT
Raco et al., 1990	Retrospective	5	2 of 5 pts treated w/ embol experienced clinical improvement & did not require further Tx	Embolization alone is adequate for treatment of some symptomatic vertebral hemangiomas
Smith et al., 1993	Retrospective	8	No pt improved w/ embol alone	Embolization may not be useful as a stand-alone Tx
Kiroglu et al., 2009	Retrospective	1	Pt w/ pain & paraplegia underwent endovascular embol & 2 yrs later had recurrent spinal cord compression & required surgical Tx	Aggressive vertebral hemangiomas may recur after embol alone

TABLE 4. Selective review of key literature on surgery for aggressive vertebral hemangiomas

Authors & Year	Study Design	No. of Pts	Results	Conclusions
Djindjian et al., 1992	Retrospective	1	Pt w/ multiple aggressive vertebral hemangiomas treated w/ embol, laminectomy, RT & was subsequently asymptomatic for 6 yrs	Embolization, laminectomy, and RT may be used in cases in which total resection is not possible
Fox & Onofrio, 1993	Retrospective	11	Series included 11 pts w/ aggressive vertebral hemangiomas who underwent surgery. Of 5 pts who underwent STR w/o adjuvant RT, 2 developed recurrence. Of 5 pts who underwent STR w/ RT, 1 developed recurrence, but that pt may have received an ineffective dose of radiation	Patients w/ progressive neurological deficit should undergo preop embol & surgery. When STR is performed, adjuvant RT should be used
Bremnes et al., 1996	Retrospective	1	Pt who presented w/ paraparesis was treated w/ laminectomy, subtotal decompression, & RT and was neurologically normal 2 yrs after surgery	There is a beneficial role for RT in combination w/ surgical decompression
Jayakumar et al., 1997	Retrospective	12	Pts were treated w/ embol, laminectomy, & RT; 11 of 12 pts were clinically improved but 1 pt was Frankel Grade A & remained unchanged	Preop embol should be routine, & when it is used in combination w/ laminectomy & radiation, most pts have favorable outcomes
Inoue et al., 2007	Retrospective	1	Case report of hemangioma removed en bloc. The pt was asymptomatic w/o evidence of recurrence at 4-yr FU	En bloc resection is a treatment option for aggressive vertebral hemangiomas
Kato et al., 2010	Retrospective	5	2 pts underwent en bloc resection; 2, intralesional resection; 1, piecemeal resection. The pt who underwent piecemeal resection had the highest operative blood loss	En bloc resection may be associated w/ less blood loss than piecemeal resection
Acosta et al., 2011	Retrospective	10	Pts treated w/ preop embol followed by intralesional spondylectomy had improvement in BP VAS score. No pt had recurrence or required further therapy	GTR results in clinical improvement & good long-term control
Urrutia et al., 2011	Retrospective	4	Pts treated w/ preop embol & resection. At FU (mean 53 mos) only 1 pt had recurrence	Total resection or STR w/ RT may be used in treatment of aggressive vertebral hemangiomas
Jiang et al., 2014	Retrospective	29	21 pts underwent surgery for aggressive vertebral hemangiomas; 6 pts did not receive RT, & of those, 3 developed recurrence. There were no recurrences in pts who received RT or who had GTR	RT should be used after subtotal resection, except in cases where the tumor is obliterated by vertebroplasty cement
Goldstein et al., 2015	Prospective/retrospective	68	In 68 pts w/ symptomatic vertebral hemangiomas, recurrence rate was 3%	Surgery results in excellent local control of symptomatic vertebral hemangiomas. En bloc resection is not required for good clinical outcomes

VAS = visual analog scale.

perform endovascular embolization of the feeding arteries in order to reduce vascularity of the tumor and decrease blood loss during surgery.^{8,19,22,37,50,53,59,73} The benefits of embolization were confirmed in a systematic review that included 51 patients with aggressive vertebral hemangiomas. This study found that in the group that received preoperative embolization, blood loss was significantly less than in the group that did not (980 vs 1629 ml, respectively).⁶⁰ Cotten et al.¹⁵ reported that preoperative embolization can be used in combination with vertebroplasty to even further reduce blood loss during surgery.

Endovascular embolization has also been proposed as a definitive treatment for aggressive vertebral hemangiomas without surgery.^{7,33,39,57} Gross et al.³³ describe a case involving a patient who had a high-grade spinal block due to a vertebral hemangioma that was relieved following endovascular embolization with Gelfoam. Hekster and Endtz³⁹ treated a patient with an aggressive vertebral hemangioma causing paraplegia with embolization followed by radiotherapy and found that the patient had an excellent clinical outcome with no recurrence at 15 years. Raco et al.⁵⁷ reported on 2

patients who underwent embolization alone, both of whom had clinical improvement and remained free from recurrence at 18 and 36 months following the procedure.

Other authors have reported more negative experiences, however. Kiroglu et al.⁴⁸ published a case report describing a woman who developed symptoms from an aggressive vertebral hemangioma during pregnancy and was initially treated with embolization but developed recurrent spinal cord compression 2 years later. Likewise, in a retrospective review of 8 patients, Smith et al.⁶⁵ did not find that any of their patients experienced improvement with embolization alone. Embolization is also generally contraindicated when feeding vessels supply the artery of Adamkiewicz as well as the hemangioma; in addition, surgeons must be cautious of reflux of the embolic material into the intercostal or lumbar arteries.^{16,32} As an alternative, transpedicular embolization with NBCA has been described, and this may be useful when transarterial embolization is unsafe.^{28,71}

Surgery

Surgical decompression and gross-total resection are

commonly performed in the treatment of aggressive vertebral hemangiomas. Acosta et al.² performed a retrospective review of 10 cases involving patients who underwent gross-total resection of aggressive vertebral hemangiomas with preoperative embolization followed by intralesional spondylectomy. They reported no recurrence at an average follow-up of 2.42 years and concluded that gross-total resection decreased the likelihood of recurrence without the need for radiation therapy. Another retrospective review described 4 patients who were treated with preoperative embolization followed by either decompressive laminectomy and fusion or an anterior corpectomy for gross-total resection. In this study, 1 patient had a subtotal resection without adjuvant radiation therapy and later developed recurrence at 15 months. The other 3 patients, however, had no recurrent disease at 45–72 months.⁶⁸ Goldstein et al.³¹ recently described a large multicenter cohort of 68 patients with symptomatic vertebral hemangiomas treated with surgery and reported a local recurrence rate of 3%. In this series, 7 patients with aggressive vertebral hemangiomas underwent en bloc resection with no recurrence of tumor. Because of conflicting data in the article it is not clear how many patients with aggressive vertebral hemangiomas underwent intralesional resection—which in this study could mean either subtotal or gross-total resection—but there was at least 1 patient who had an intralesional resection without radiation therapy and developed a recurrence (at 5.3 years after surgery).

Many authors have recommended surgery followed by adjuvant radiation therapy. Jayakumar et al.⁴³ published a case series involving 12 patients with spinal cord compression from aggressive vertebral hemangiomas who were treated with preoperative embolization, decompressive laminectomy for subtotal resection, and adjuvant radiation therapy. Eleven of the 12 patients had favorable clinical outcomes, but long-term follow-up data on recurrence were not provided. Djindjian et al.¹⁶ presented a similar case involving a man with an aggressive vertebral hemangioma treated with embolization, decompressive surgery, and postoperative radiation who had complete resolution of his symptoms and no recurrence within 6 years. Bremnes et al.⁹ described yet another case involving a patient with an aggressive vertebral hemangioma who did well with decompressive laminectomy and subtotal tumor excision followed by radiation. In 1993, Fox and Onofrio²⁶ published a study including 10 patients with aggressive vertebral hemangiomas resulting in spinal cord compression who underwent subtotal resection and 1 patient who underwent gross-total resection. Two of 5 patients who underwent subtotal resection without adjuvant radiation developed recurrence at 5–6 years postoperatively. One of 5 patients who underwent subtotal resection with adjuvant radiation developed a recurrence 17 years postoperatively, but this patient only received 10 Gy radiation, whereas the other 4 patients without recurrence received between 26 and 45 Gy. Several studies discussed above suggest that 10 Gy is an ineffective dose, and this may be the reason for the late recurrence.^{3,40,58} The remaining patient had gross-total resection without adjuvant radiation therapy and did not have recurrence of disease.²⁶ In the retrospective series by Jiang et al.,⁴⁵ 21 cases involving patients with severe or quickly evolving neurological deficits underwent surgery for aggressive vertebral hemangiomas, including 2 patients

who had surgery after failure of radiation therapy. Most patients underwent preoperative embolization followed by decompressive surgery or subtotal resection, sometimes in combination with vertebroplasty. The authors found that 3 of the 6 patients who underwent decompression without radiation therapy developed recurrence 12–108 months postoperatively. None of the patients who had a total resection or were treated with a combination of radiation and surgery had any recurrence at 24–133 months' follow-up.

Within the last 10 years there have been occasional reports of en bloc resection of aggressive vertebral hemangiomas in the literature. En bloc tumor removal is technically challenging and is associated with a high rate of morbidity. One systematic review reported a 36.3% complication rate for en bloc resection spinal tumors.¹⁴ Another study reported higher rates of revision for patients who had en bloc resection when compared with those who had piecemeal resection of spinal tumors.²⁰ As a result, this operation is generally reserved for patients with a pathological diagnosis such as chordoma or chondrosarcoma for which there is a proven survival benefit.

In 2007, Inoue et al.⁴² reported performing an en bloc spondylectomy for removal of an aggressive vertebral hemangioma involving the vertebral body, pedicle, and lamina of L-2. When the decision to perform an en bloc resection was made, the authors had obtained a nondiagnostic biopsy and believed the tumor was a more ominous primary spinal column tumor, evidence that would suggest it should be removed en bloc. However the postoperative pathology revealed an aggressive vertebral hemangioma. The authors acknowledge that, had they known the correct pathological diagnosis, they would have performed only a percutaneous vertebroplasty instead. Although they reported that the patient did well following surgery, the operation itself required an anterior and posterior approach, took over 9 hours, and involved 4000 ml of blood loss even with preoperative embolization. Kato et al.⁴⁷ performed a retrospective review of 5 cases involving patients with aggressive vertebral hemangiomas treated with preoperative embolization followed by total excision. Two patients underwent en bloc excision, 2 patients underwent a combination of en bloc and piecemeal excision, and the remaining patient underwent piecemeal excision. Regardless of surgical approach, all patients remained free of recurrence at 92–163 months of follow-up. Despite the lack of oncological benefit, the authors recommended en bloc excision as the treatment of choice because the single patient who underwent piecemeal resection had increased blood loss when compared with the patients who underwent en bloc resection (3400 ml vs an average of 2300 ml). Other reports of en bloc resection of hemangiomas have been published in the literature within the last few years. Although they do not report any complications from surgery or recurrence of tumor, they do not demonstrate superiority of en bloc surgery to piecemeal gross-total resection.^{31,35,56,66}

Institutional Experience

In our series of 5 cases, regardless of the surgical strategy—subtotal resection, gross-total resection, or en bloc resection—none of the patients had developed recurrent disease at average follow-up of 31 months. Although the 2

patients who underwent subtotal resection both had definite tumor left behind at the time of surgery, they each received either vertebroplasty or adjuvant radiation therapy to obliterate the remaining tumor, so we considered these lesions to be fully treated. Our experience suggests that there is only a low likelihood of recurrence for aggressive vertebral hemangiomas, and this has been confirmed throughout the existing literature. The patient who had the worst outcome and the most difficult treatment course was the patient who underwent en bloc resection and had multiple complications requiring several further surgeries. Although this is only one case, it is known from other tumor pathologies that en bloc resection is a highly morbid operation and that gross-total resection or subtotal resection with vertebroplasty or adjuvant radiation might provide similar control of the tumor with much lower morbidity.

Conclusions

Treatment of aggressive vertebral hemangiomas has been controversial in neurosurgery for over a century. Multiple therapeutic options have been described with varying degrees of success, including radiation, vertebroplasty, ethanol injection, embolization, surgery, and any combination thereof. The technical approach to surgery has also been debated, with some authors recommending decompression alone, others supporting gross-total resection, and still others who have recommended en bloc resection. Because aggressive vertebral hemangiomas are uncommon lesions, there are no large studies that directly compare treatment modalities, and there is therefore no strong evidence to support one particular therapeutic algorithm.

Despite this, based on our own experience and a careful review of the literature we feel that certain principles should still be followed. For patients with an uncertain radiological diagnosis, percutaneous biopsy should be performed to rule out lesions that can mimic an aggressive hemangioma.^{29,36,45,49} For patients with aggressive vertebral hemangiomas who present with mild or slowly progressive neurological symptoms, it is reasonable to attempt nonoperative management with embolization, vertebroplasty, or radiation therapy provided the symptoms are not due to compression by a focal bony prominence, which is unlikely to resolve without surgery. For patients with severe or rapidly progressive symptoms, surgery should be considered, with preoperative embolization when possible. Piecemeal gross-total resection and en bloc resection both seem to have good clinical outcomes with a low likelihood of tumor recurrence, but piecemeal gross-total resection is preferable due to lower rates of complication and reoperation. If it is only possible to perform a decompression or subtotal resection, vertebroplasty or adjuvant radiation therapy should be considered to reduce the risk of recurrence. Aggressive vertebral hemangiomas are benign lesions that do not have metastatic potential and are not associated with mortality.³¹ So even if a tumor recurs after subtotal resection, a second decompressive surgery may still be performed with a low risk of morbidity.¹

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Conception and design: all authors. Acquisition of data: Vasudeva. Analysis and interpretation of data: Vasudeva. Drafting the article: Vasudeva. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Vasudeva.

Supplemental Information

Previous Presentations

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