



Review Article

Surgical management of symptomatic vertebral hemangiomas: A case report and literature review

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ABSTRACT

Background: Vertebral hemangiomas (VHs) are common benign tumors that only rarely become symptomatic. There is a paucity of data regarding their surgical management and outcomes. Here, we reported a case involving an aggressive cervical VH, discussed its surgical management and outcomes, and reviewed the literature.

Methods: We assessed the clinical, radiological, and surgical outcomes for a patient with an aggressive cervical VH. We also performed a systematic review of the literature according to Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines to describe surgical outcomes for symptomatic VH.

Results: A total of 154 studies including 535 patients with VH were included in the study. The majority of patients were female (62.8%), the average age was 43 years, and the thoracic spine was most commonly involved (80.6%). Utilizing Odom's criteria, outcomes were excellent in 81.7% (95% CI 73.2–90.2) of cases. For those presenting with myelopathy ($P = 0.045$) or focal neurological deficits ($P = 0.018$), outcomes were less likely to be excellent. Preoperative embolization was not associated with excellent outcome ($P = 0.328$).

Conclusion: Surgical outcomes for VH are predominantly favorable, but aggressive VHs have the potential to cause significant residual postoperative neurological morbidity.

Keywords: Preoperative embolization, Spine tumor, Surgery, Vascular tumor, Vertebral hemangioma

INTRODUCTION

Vertebral hemangiomas (VHs) are benign vascular tumors comprised capillaries and venous structures. They have a prevalence of 10–12%, are usually asymptomatic, and rarely require surgery.^[7] However, approximately 1% of VHs demonstrate aggressive features including damage to the surrounding bone and soft tissue with subsequent spinal cord and/or nerve root compression.^[5,6] Due to their rarity, the optimal surgical management of VH and predictors of postoperative outcomes are not well defined. Here, we reviewed an unusual case of a multilevel cervical VH and systematically reviewed the literature regarding their management and outcomes.

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Illustrative case

Presentation

A 14-year-old male presented with 12 weeks of mechanical neck pain, hand weakness, and distal upper extremity paresthesias. MRI revealed abnormal anterior/posterior bony element enhancement from C4–C6 with subtle epidural enhancement [Figure 1]. CT revealed a severe osteolytic fracture of the C5 body with retropulsion into the canal and osteolysis of the C4 and C6 endplates. After a fall, the patient became quadriplegic with 0/5 strength below C7 and a sensory level at the chest. The second CT demonstrated a Grade 4 retrolisthesis at C5–C6 with severe spinal canal stenosis [Figure 1c].

Operation

The patient underwent an emergent C3–C7 anterior corpectomy with fusion. The C4, C5, and C6 vertebral bodies were removed *en bloc* and an expandable cage was inserted into the corpectomy defect followed by a plate spanning C3–C7. The posterior longitudinal ligament was abnormally vascular. An additional C4–C6 laminectomy with posterior C2–T3 fusion was performed in the same setting.

Pathological findings

The histologic evaluation of the lesion [Figure 2] demonstrated marrow space replacement with thin-walled vessels, which surrounded bony trabeculae and focally eroded mature cortical bone, consistent with hemangioma. There was no evidence of malignancy.

Postoperative course

The patient did not neurologically improve and was discharged to an inpatient rehabilitation center. Postoperative CT showed adequate decompression of the spinal canal, reduction of the C5 angulation, and correction of the kyphosis [Figure 3]. The 5-month follow-up X-ray showed a stable construct [Figure 3b], but he failed to regain neurological function.

METHODS

A systematic review was performed utilizing the Preferred Reporting Items for Systematic Reviews and Meta-Analyses guidelines. MEDLINE, EMBASE, and Scopus were searched using the criteria “vertebral hemangioma” OR “spine hemangioma.” All case reports and series reporting outcomes after open surgery with or without instrumentation for symptomatic VH were included. Outcomes were categorized as excellent, good, fair, or poor according to Odom’s criteria. Random effects model was used to calculate the pooled rate of an excellent outcome. Weighted least squares regression was used to identify factors associated with an excellent outcome.

RESULTS

A total of 154 studies were included (1950–2020) comprising 535 patients [Figure 4 and Table 1]. The median (IQR) age was 43 years (27–57.5), and 37.0% were male versus 62.8% of females. The thoracic spine was involved in the majority of patients (80.6%), followed by the lumbar (16.4%), cervical (3%), and sacral (1.1%) levels. Hemangiomas typically

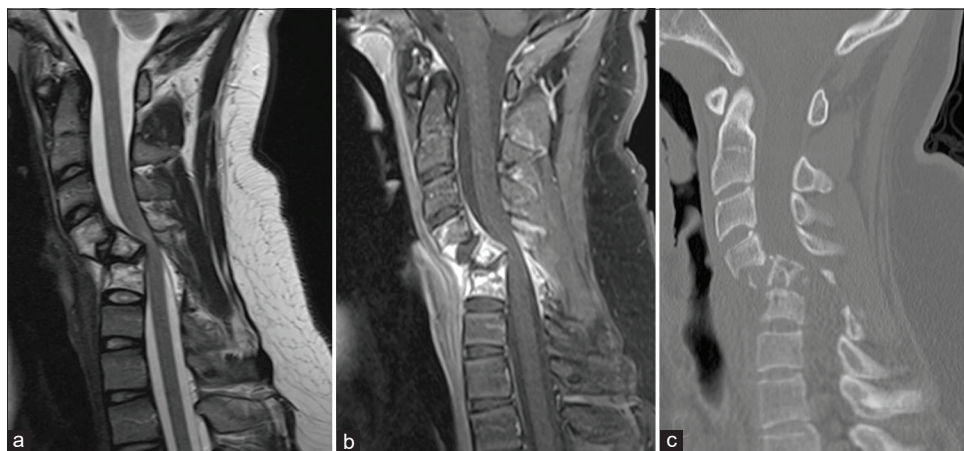


Figure 1: Preoperative sagittal T2-weighted (a) MRI demonstrates severe angulation and retropulsion of the C5 vertebral body causing marked spinal stenosis with hyperintensity within the spinal cord. Contrast-enhanced T1-weighted image (b) shows abnormal enhancement within the C4–C6 vertebral bodies, lamina, and spinous processes, as well as enhancement in the posterior epidural space with prominent flow voids. Plain sagittal CT obtained on presentation to the emergency department (c) demonstrates severe osteolysis at the levels of C4–C6 and increased retrolisthesis at C5.

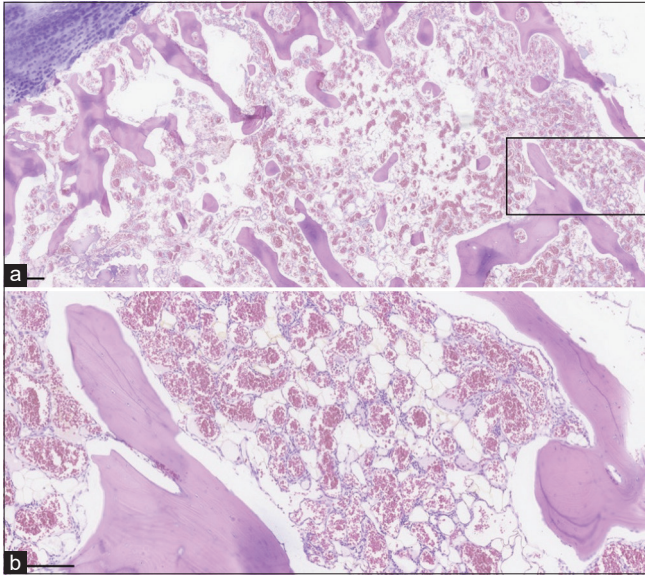


Figure 2: Microscopic sections demonstrate (a) mature cartilage and cortical bone with diffuse hemangioma replacement of bone marrow (total magnification = 20×, scale bar = 500 μm) and (b) higher power image demonstrates thin-walled vessels, focal adipose tissue, and minimal intervening stroma (total magnification = 100×, scale bar = 200 μm).

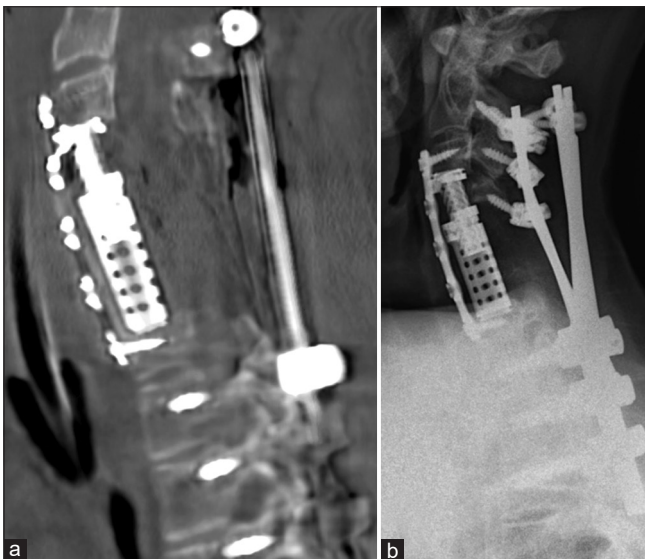


Figure 3: Immediate postoperative sagittal CT (a) demonstrates corpectomy defect from C4–C6 with placement of cage and anterior plate. Lateral X-ray (b) shows the remainder of the construct at 5-month follow-up.

involved one level (89.3%). The most common surgical intervention was decompression without fusion (48.4%) followed by instrumented fusion (41.5%) and corpectomy (10.1%). Excellent outcomes were observed in 81.7% (95% CI 73.2–90.2) of patients. The Q-statistic was 31.26 on 153 df

Table 1: Presenting symptoms and outcomes of 535 patients.

Symptom	n*	%
Axial pain	277	51.8
Myelopathy	275	51.4
Radiculopathy	28	5.2
Neurologic deficit	207	38.7
Cauda equina syndrome	4	0.7
Conus medullaris syndrome	1	0.2
Outcome		
Excellent	437	81.7
Good	68	12.7
Fair	11	2.1
Poor	19	3.5

*Several patients presented with more than 1 symptom

Table 2: Results from the weighted least squares regression identifying factors associated with an excellent outcome by Odom’s criteria. Statistically significant variables are denoted in bold type.

Variable*	B	95% CI	P
Age	0.002	-0.001–0.01	0.125
Female sex	0.03	-0.13–0.18	0.745
Presenting symptom			
Pain	-0.01	-0.12–0.11	0.926
Myelopathy	-0.16	-0.31–-0.004	0.045
Radiculopathy	-0.07	-0.37–0.23	0.652
Neurologic deficit	-0.18	-0.33–-0.03	0.018
Mean no. of levels involved	-0.03	-0.14–0.09	0.622
Surgery			
Fusion	0.07	-0.03–0.17	0.149
Corpectomy	0.07	-0.12–0.25	0.474
Preoperative tumor embolization	0.05	-0.05–0.15	0.328
Spinal segments effected			
Cervical	-0.29	-0.96–0.39	0.406
Thoracic	-0.22	-0.83–0.40	0.485
Lumbar	-0.32	-0.90–0.27	0.288
Sacral	0.13	-0.86–1.12	0.800

*Surgery with decompression alone did not contribute to the model

($P = 1.00$). Patients who presented with myelopathy ($\beta = -0.16$, $P = 0.045$) or a neurologic deficit ($\beta = -0.18$, $P = 0.018$) were less likely to have excellent results [Table 2]. A total of 198 (37%) patients underwent preoperative embolization but this was not associated with excellent outcomes.

DISCUSSION

Our case is unique for several reasons. Less than 6% of the cases in our review involved three or more levels as ours did. Only 3% involved the cervical spine. Presentation in a pediatric patient is also rare, with <15 cases reported

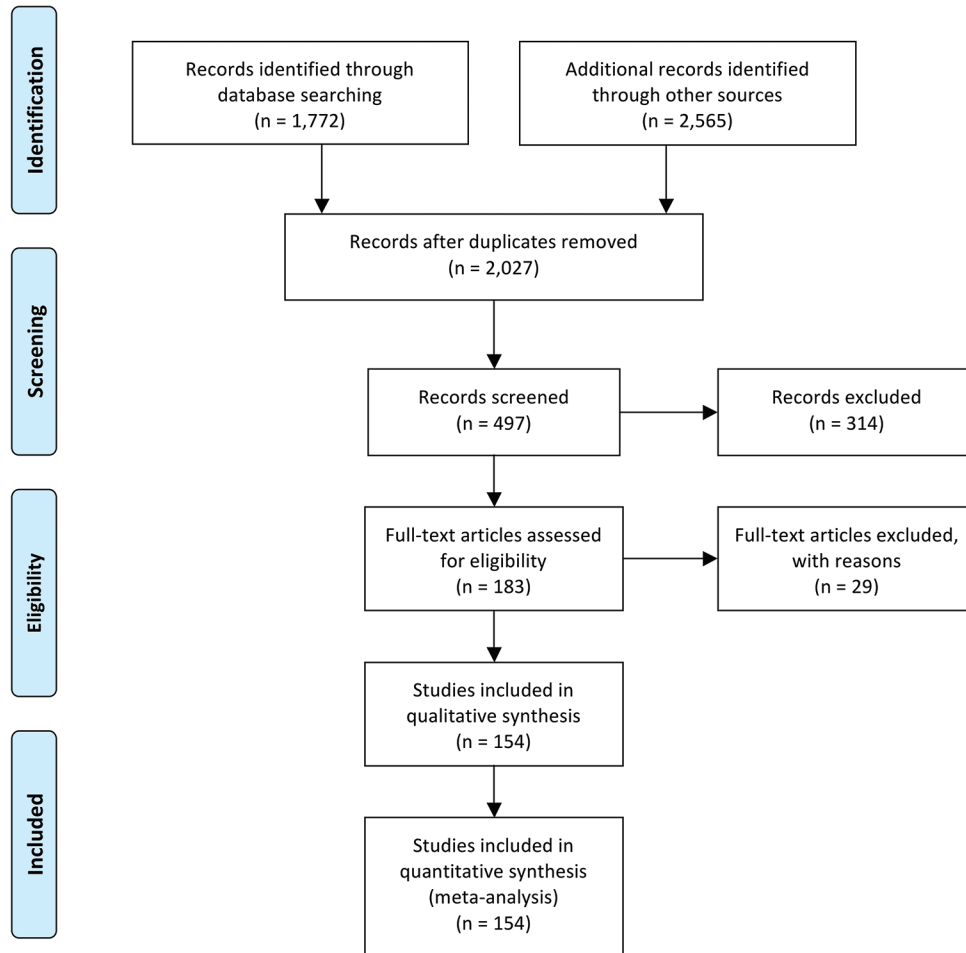


Figure 4: Preferred Reporting Items for Systematic Reviews and Meta-Analyses diagram depicting the results of the literature review.

worldwide. To the best of our knowledge, the only other report of a pediatric patient with an aggressive, symptomatic, multilevel VH involved the thoracic spine.^[3]

The 535 patients undergoing surgery in this review displayed similar characteristics as seen in other VH series. We identified a female predominance, which may be due to the growth-stimulating effect of progesterone on the hemangioma. The median age in our analysis was 43 years, which is similar to other cohorts. The most common surgical technique was decompression without fusion. This was likely due to VHs predilection for the thoracic spine, which permits more aggressive decompression without instrumentation as the ribs provide stability. Although not included in our review, percutaneous vertebroplasty or radiotherapy is reasonable treatment options for patients presenting with only axial pain.^[2,10]

Both myelopathy and neurologic deficit at presentation were negatively associated with excellent outcomes, suggesting that injury to neural elements is less likely to be reversed from surgery. Thus, early surgery before the development of

neurologic compromise may be indicated for symptomatic VHs, especially if they display an epidural component. Further, there are multiple reports describing rapid progression of neurologic deficits.^[1,4,8,9,11-13] Although percutaneous or endovascular embolization of symptomatic VH has been proposed as standalone treatments, we favor surgery to relieve mass effect and increase the chance of a favorable outcome.

CONCLUSION

VHs can cause significant neurologic morbidity and outcomes after surgical intervention are predominantly favorable. The presence of preoperative myelopathy and neurologic deficit was negatively associated with an excellent outcome, suggesting that a prophylactic approach to symptomatic VHs is warranted.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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