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Research Paper

Surgical Options for Aggressive Vertebral Hemangiomas : A case series, literature review and treatment recommendations

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HIGHLIGHTS

- The surgical strategy for AVHs remains controversial due to its rarity
- We retrospectively study twenty-nine AVH surgical cases with neurological deficits to describe an optimal surgical strategy for AVHs in a single institute.
- The results indicate that decompression Plus VP achieves good tumor control and post-radiation helps avoid tumor recurrence.

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ABSTRACT

Purpose: We retrospectively study twenty-nine surgical cases of aggressive vertebral hemangiomas (AVHs) with neurological deficits and extradural compression to determine the optimal surgical treatment strategy for AVHs at a single institution.

Methods: Patients with AVHs with neurological deficits who underwent partial tumor resection plus decompression with or without vertebroplasty (VP), and radiotherapy between 2010 and 2021 were included in this study. Clinical characteristics, surgical outcomes, and follow-up data of the patients were reviewed retrospectively.

Results: Twenty-nine AVH cases with neurological deficits and spinal instability were included in this study and treated surgically. The mean operation time of patients with decompression surgery plus VP (Groupe A) was 215.9 (120-265 min), shorter than that of decompression surgery without VP (Group B) 240.2 (120-320 min). Intraoperative blood loss was 273.3 (100–550 mL) in group A and 635.3 (200–1600 mL) in group B. In addition, a significant reduction in blood loss was observed in group A compared to the group B (p=0.0001). All patients experienced immediate pain relief and improvement in their neurological symptoms. Neurological function was assessed by the Frankel score, ASIA score, and the visual analogue scale (VAS) pain score decreased from 7.4 (4-9) to 1.3 (0-3). Of twenty-nine patients in this study, only 7% (2/29 patients) showed signs of recurrence.

Conclusion: Decompression plus VP achieve good tumor control and decrease surgical complication. Preoperative vascular embolization and VP can reduce intraoperative bleeding in the treatment of AVH surgery. Moreover, postoperative radiotherapy seems to be a good technique to prevent tumor recurrence.

1. Introduction

Vertebral hemangioma (VH) is one of the most common slow-growth benign tumors of the spine, accounting for 2–3 % of all spinal tumors. VH is typically asymptomatic and is often discovered incidentally on imaging examination [1–3]. However, about 0.9 %–1.2 % of VH can be symptomatic [6–8,15,16,25] (aggressive vertebral hemangioma) with spinal cord distension, pain and compression [4,5]. AVH can occur in cervical, thoracic, and lumbar regions and cause the enlargement of the posterior cortex of the vertebral body, destruction of the vertebral

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pedicle, and spinal canal stenosis, resulting in bleeding, pathological compression fractures, and neurological deficits. There are several options to treat patients with AVHs, although the surgical treatment strategies for AVHs are still controversial due to their rarity [6–8,16,28].

In recent years, various treatments have been studied alone or in combination for AVHs, including subtotal or total tumor resection, vertebroplasty, decompressive surgery without tumor resection, arterial embolization, and radiotherapy. Radical surgery resection is recommended for hemangiomas with an extraosseous extension that cause neurological symptoms [6]. Excellent rates local control and long-term survival can achieved with aggressive resection, but total en bloc spondylectomy resulted in significant bleeding and intraoperative morbidity [7]. Subtotal tumor resection has been widespread for several years, but is associated with an increased recurrence and adjuvant radiation therapy makes a second operation more difficult [8]. Decompression has also been used to circumvent neurological symptoms, but palliative debulking surgery increases the rate of early or delayed recurrence [9]. Vertebroplasty has been used in the treatment of AVHs to reduce the risk of recurrence and to relieve local pain. However, it can lead to cement leakage and disease of the adjacent segment. These surgical strategies have shown therapeutic effects in AVHs, but carry risks unique to each therapy. Furthermore, due to the rarity of tumors and the lack of research, there was no gold standard for surgical treatment or principle of combination therapy for AVHs [8,10–13].

This study investigates whether coupling decompression, intraoperative vertebroplasty (VP), and postoperative radiotherapy can effectively treat AVHs by retrospectively studying twenty-nine surgical AVH cases with neurological deficits in extradural compression at our institute.

2. Materials and methods

2.1. Patients

We retrospectively collected and analyzed data from patients with AVHs in the cervical, thoracic, and lumbar spine in our department between 2010 and 2021. We reviewed the record and radiological studies to identify the patients with AVHs; Each patient underwent posteroanterior and lateral spine radiographs, computed tomography (CT), and magnetic resonance imaging (MRI). Inclusion criteria were AVHs with neurological deficits and extradural compression. We excluded seven patients from the study because they had received other treatments for their hemangioma and ten patients because a lack of follow-up results. We used data such as deterioration symptoms, tumor location, multiple surgical treatments, operation time, estimated blood loss (EBL), and a visual analogue scale (VAS) to clinically evaluate pain levels. We performed neurological assessments to assess the patients' sensory symptoms, motor deficits, Frankel grade, ASIA score, post-radiation therapy, recurrence, and follow-up (FU).

2.2. Criteria for these surgical options

There is no consensus on the best treatment for symptomatic spinal hemangioma, so treatment strategies should be based on the patient's symptoms and imaging characteristics. Surgery involves removing tumors, decompress nerves, and stabilizing the spine. Our surgical approach was chosen according to tumor size and location, including decompression with VP (Fig. 1) and decompression laminectomy (Fig. 2). We proposed the following treatment algorithm (Fig. 3). (1) Decompression + VP (Group A): used when the anterior vertebral body is destroyed, and vertebral instability due to vertebral body collapse leads to nerve compression. (2) Decompression (Group B): Laminectomy was chosen for patients whose tumor only invades the posterior structure of the spine (less than 1/3 vertebral body is destroyed) without soft tissue bulging. (3) The criteria of radiation: 1) decompression surgery with conventional radiation; 2) The VP may not completely fill the lesion.

2.3. Follow-up

During follow-up, we evaluated the neurological status using the American Spinal Injury Association (ASIA) impairment scale. We used the VAS score to record back pain preoperatively and postoperatively at the last follow-up visit at 44.9 (12–138 months). In addition, MRI was used every three months for the first two years to evaluate AVH recurrence.

3. Results

3.1. Clinical characteristic of patients

Twenty-nine AVH cases were included in this study and treated surgically. As shown in Table 1, eighteen females and eleven males with a mean age of 55.76 years (19–84 years) were enrolled and the mean follow-up time was 44.9 (12–138 months). Fourteen patients had myelopathy (Frankel grade A, B, C, and D in three, seven, three, and one patients, respectively); Eleven patients presented with pain and numbness (Frankel grade A, B, and C in one, eight, and two patients, respectively) and four had radiculopathy (Frankel grade A, B, C, and D in one, one, one and one patient, respectively) for only several months (Table 2). AVH lesions were located in the cervical spine (between C2 and C7), thoracic spine (between T2 and T11), and lumbar spine (between L1 and L5) in 3, fourteen, and twelve patients, respectively (Table 1). Three surgeons from our department performed the procedures. Among them, the patients received different treatment strategies. In this study, twenty-nine patients underwent decompression surgery with or without VP. In Group A, we treated eleven patients with preoperative transarterial embolization (PTE) and one without PTE. Besides, in Group B, eight patients underwent decompression alone with embolization and nine patients without embolization (Table 1). In addition, five patients in group A received post-radiation therapy. Nine patients received post-radiation therapy for group B (Table 2).

3.2. Surgery and outcomes

We have summarized our surgical information and clinical results in Tables 2 and 3. The mean operation time of patients with decompression surgery plus VP (Group A) was 215.9 (120-265 min), shorter than that of decompression surgery without VP (Group B) 240.2 (120-320 min). Intraoperative blood loss was 273.3 (100–550 mL) in group A and 635.3 (200–1600 mL) in group B. In addition, a significant reduction in blood loss was observed in group A compared to group B (p=0.0001) (Table 3).

In addition, all patients experienced immediate pain relief and resolution of their neurological symptoms; the VAS pain score decreased from 7.4 (4–9) to 1.3 (0–3), and statistical analysis showed a significant difference between these groups in preoperative or postoperative VAS scores ($P < 0.0001$). Of all twenty-nine patients, twenty-four achieved Frankel grade E, 5 Frankel D, twenty-five achieved Asian grade E, and 4 Asian grade D; at the final follow-up 44.9 (12–138 months) (Table 2 and 3).

3.3. Complication and recurrence

There were three surgical complications, including 1 case of pleural effusion, which recovered with closed thoracic drainage. In another case, pneumonia was resolved with antibiotics treatment. The third patient who underwent decompression developed a deep infection at the surgical site and was managed with irrigation and debridement without revision of the instruments. Among twenty-nine patients, only 7% (2/29 patients) who underwent decompression therapy developed a recurrence. One patient died of unrelated cause (lung cancer) 22 months postoperatively.

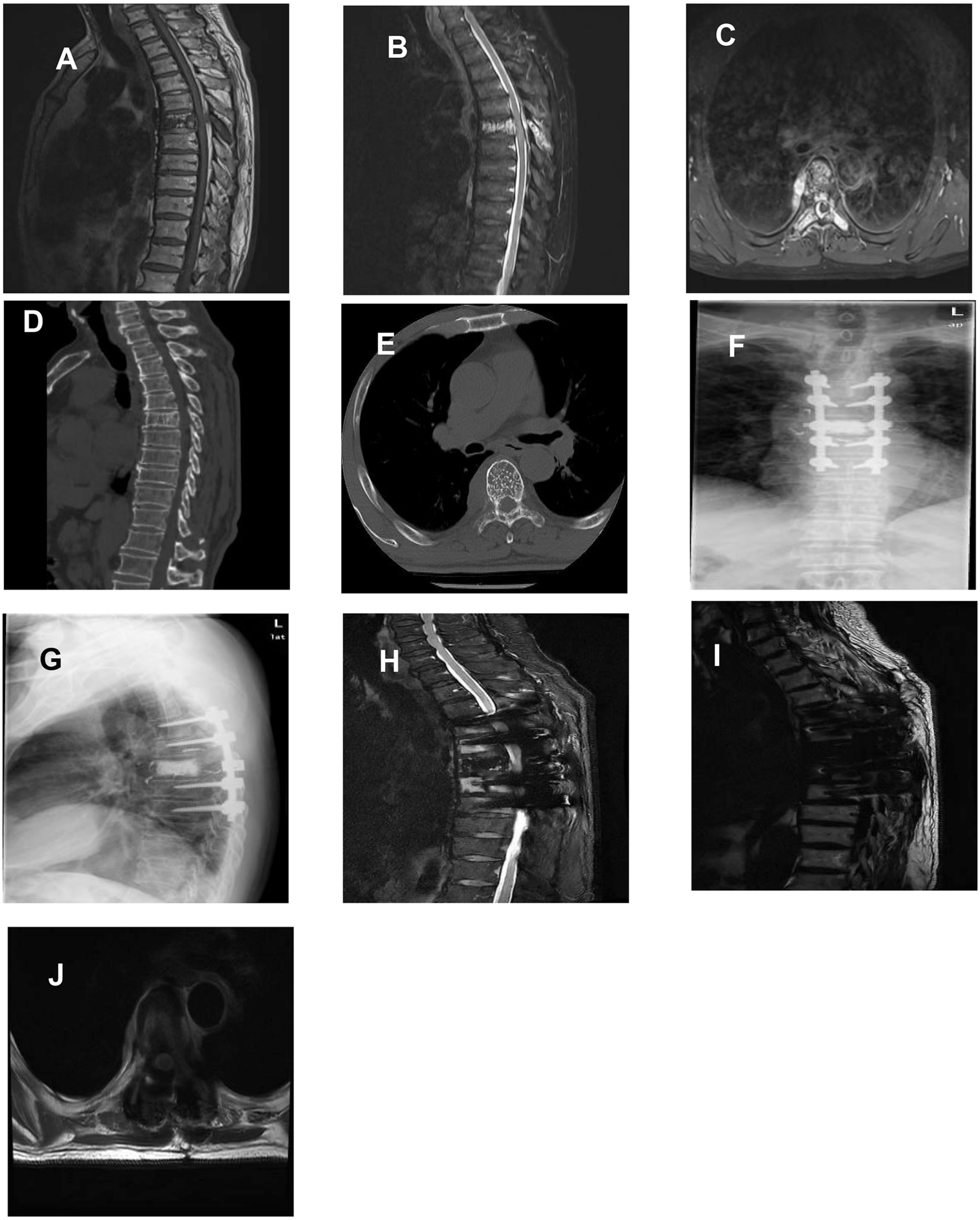

Fig. 1. Illustrated Case 1: We presented a 64-year-old male with a T5 spinal hemangioma who presented to the clinic with back pain with bilateral lower limb numbness of 3 months duration. MRI and CT scan obtained in an outpatient setting demonstrated an appearance of hemangioma of the T5 vertebra with epidural extension and cord compression (Fig. 1). She was sent to our department for further examinations. She had neither history of smoking nor any risk factors for malignancy. Se had no unintended weight loss, no limb weakness, and fever or cold. The patient underwent preoperative embolization, decompression plus VP, and posterior spinal fusion of T3-T4 to T6-T7 level using transpedicular screws and rods, T5 vertebroplasty iliac crest bone graft. At the latest follow-up, the patient's symptoms had resolved with no evidence of recurrent. Sagittal MR images showed the vertebral lesion to be hypointense on T1 (A) and hyperintense on T2 (B) and (C), showing intense enhancement of the vertebral lesion and epidural component. Sagittal (D) and axial (E) CT scan with the characteristic appearance of hemangioma of the T5 vertebra. F and G: Postoperative lateral radiograph obtained after the tumor resection. Coils from the preoperative embolization can also be seen. Postoperative T2W and T1W sagittal MRI showed a reduction in cord compression (H) and (I). MRI identified no recurrence or spinal cord compression at two years follow-ups (J).

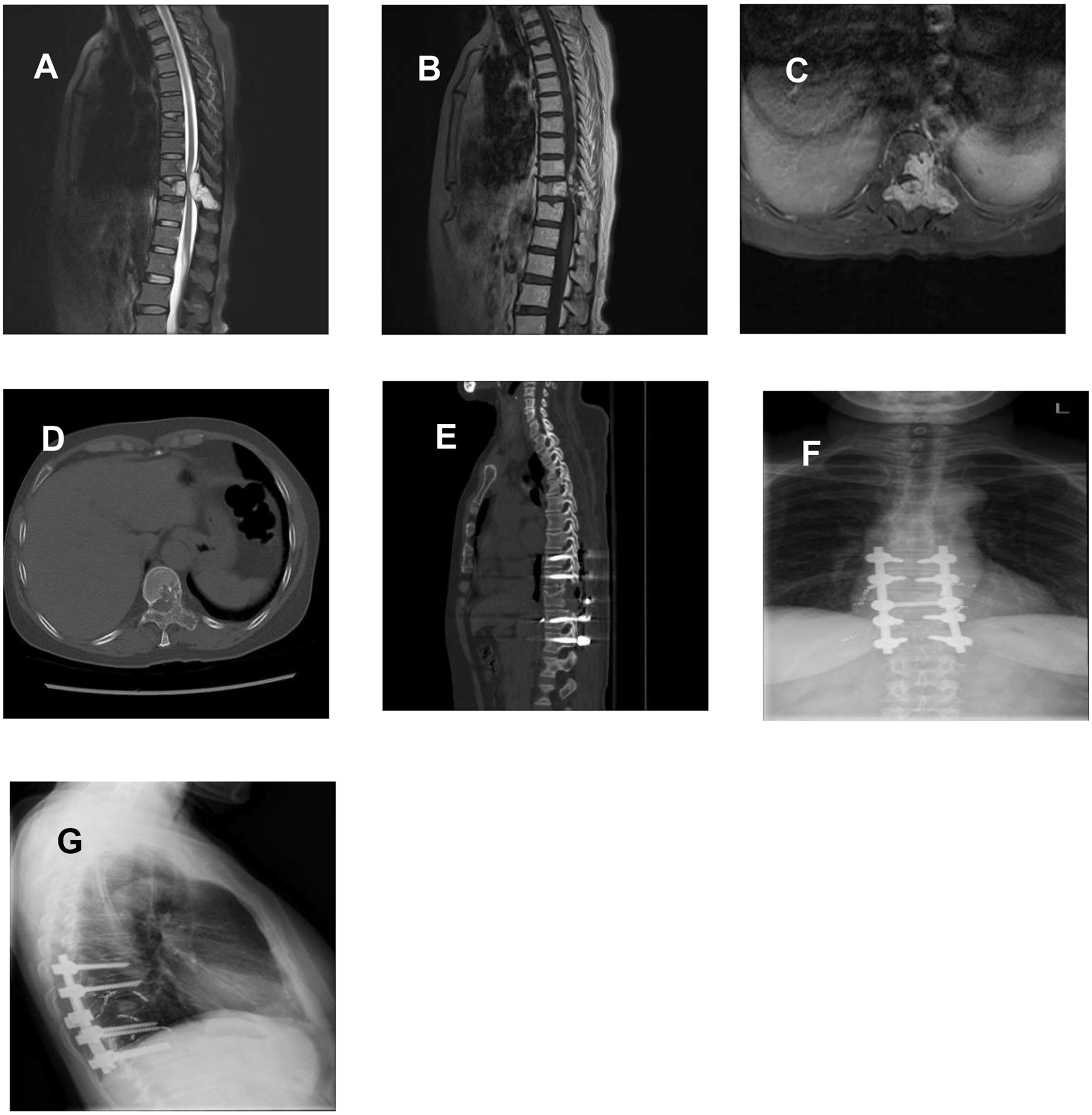

Fig. 2. Illustrated Case 2: Images were obtained in case 3, which involved a 58-year-old woman who presented with numbness and weakness in both lower limbs for two months. After vascular embolization, posterior laminectomy, decompression of T10 tumors, tumor curettage, and internal fixation were performed (Fig. 2). At the latest FU, the patient's symptoms had resolved with no evidence of recurrent. MRI dorsal spine Sagittal T2W (A), T1W (B), and Axial T2W (C) showing extraosseous intraspinal hyperintense lesion in T2W and hypointense lesion T1W with post-contrast enhancement at the level of T10 showing expansile mass with posterior element involvement causing cord compression and signs of extraosseous soft tissue expansion into the spinal canal. Axial (D) CT scan with the characteristic appearance of hemangioma of the T10 vertebra. Sagittal (E) shows good neural decompression with pedicle screws inserted two levels above and below the vertebra. Postoperative anterior and lateral X-rays (F) and (G) were obtained after the tumor resection, showing laminectomy at T10 and instrumented stabilization from T8-T12. Coils from the preoperative embolization were observed.

4. Discussion

Surgery is usually indicated in patients with severe neurological deficits or spinal instability [1], [13], [14]. VH was classified into four types: For type I VHs, only observation is needed [28]; for type II VHs, annual MRI follow-up is recommended, and treatment should only be considered if there is a risk of vertebral collapse [14]; as for type III VHs, Doppman et al. underlined the need of early detection and treatment to prevent neurological impairment. Type IV VHs are considerably more diverse; reported treatments include radiotherapy [32], vertebroplasty, direct alcohol injection [28], embolization of the feeding arteries, surgery, and a combination of these modalities. Various treatment options for AVH such as total En bloc spondylectomy, intralesional vertebrectomy, and decompression surgery, have been combined and shown satisfactory results [6,9,10,13–18]. However, the choice of surgical treatment of AVHs remains controversial [6–8,16,28]. We included patients with severe neurological deficits and extradural compression in this study. We present results of multi-modality surgical management of these aggressive lesions, including decompression with VP, and decompression alone.

Circumferential anterior vertebral body involvement and extraosseous epidural expansion cause in vertebral destruction and extradural compression, resulting in back pain and rapidly progressive neurological deficits. TES (total En bloc spondylectomy) has been reported to remove

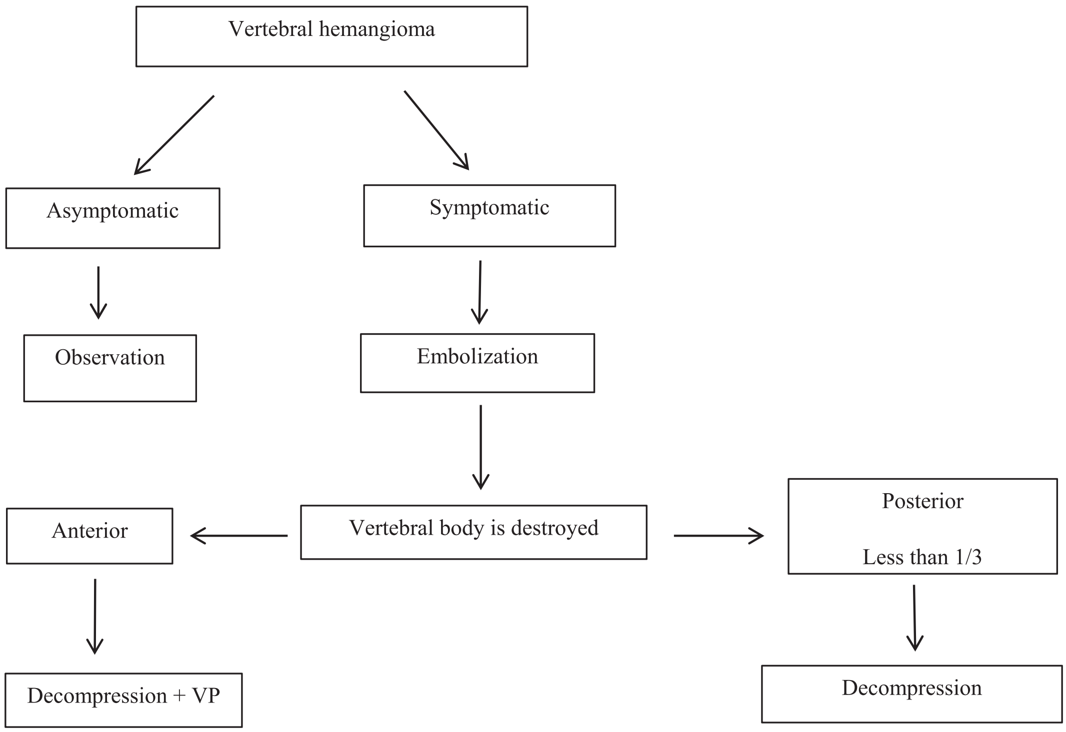

Fig. 3. Proposed treatment algorithm for aggressive vertebral haemangiomas.

Table 1
Patient characteristics.

No	Gender	Age	Site	Symptoms	VBC	PTE	Type of surgery	Comp	HD
1	F	59	L2	Numbness and pain	Yes	Yes	Dec + VP	None	7
2	F	84	L2	Myelopathy	Yes	Yes	Dec + VP	None	5
3	M	63	T9	Myelopathy	Yes	Yes	Dec + VP	None	5
4	F	44	L2	Radiculopathy	Yes	Yes	Dec + VP	None	4
5	F	19	T9	Myelopathy	Yes	Yes	Dec + VP	P	5
6	F	52	L1	Myelopathy	Yes	Yes	Dec + VP	None	7
7	M	68	T10	Radiculopathy	Yes	No	Dec + VP	None	6
8	M	64	L5	Numbness and pain	Yes	Yes	Dec + VP	None	5
9	M	36	L5	Myelopathy	Yes	Yes	Dec + VP	None	4
10	F	55	T3	Myelopathy	Yes	Yes	Dec + VP	None	4
11	M	72	T8	Myelopathy	Yes	Yes	Dec + VP	None	5
12	M	54	L4	Numbness and pain	Yes	Yes	Dec + VP	None	4
13	F	72	T11	Myelopathy	Yes	Yes	Decompression	None	5
14	F	54	T2	Myelopathy	Yes	Yes	Decompression	None	5
15	F	58	C7	Numbness and pain	Yes	Yes	Decompression	None	6
16	F	54	L2	Numbness and pain	No	No	Decompression	None	7
17	M	53	T9	Myelopathy	No	No	Decompression	PE	5
18	F	54	L4	Numbness and pain	Yes	Yes	Decompression	None	4
19	M	27	C2	Numbness and pain	No	Yes	Decompression	None	7
20	F	81	L5	Numbness and pain	No	No	Decompression	None	6
21	F	40	T7	Myelopathy	No	No	Decompression	None	5
22	F	54	C7	Numbness and pain	No	No	Decompression	None	6
23	F	78	T9	Radiculopathy	No	Yes	Decompression	None	4
24	M	77	T5	Myelopathy	No	No	Decompression	WI	5
25	M	40	T7	Myelopathy	No	N0	Decompression	None	6
26	F	55	T10	Numbness and pain	No	Yes	Decompression	None	5
27	F	32	L5	Radiculopathy	No	Yes	Decompression	None	4
28	F	64	L2	Numbness and pain	No	No	Decompression	None	5
29	M	54	T2	Myelopathy	No	No	Decompression	None	4

No, number; F, female; M, male; PTE, preoperative transarterial embolization; VBC, vertebral body collapse; HD, hospitalization durations; Comp, complications; Dec, decompression; VP, vertebroplasty; W I, wound infection; PE, pleural effusion; P, pneumonia.

circumferential compression of the spinal cord and increase blood flow and functional recovery of the spinal cord [11]. Although TES in AVHs was correlated with low recurrence rates, significant bleeding increased intraoperative/postoperative morbidity [9,15,16,20], thus prolonged recovery time. Tomita et al. [21] and Ogawa et al. [22] described radical TES in AVHs as time-consuming, hemorrhagic, and technically challenging. Goldstein et al. [7] demonstrated that the rate of AVH recurrence after intralesional vertebrectomy was 5.4 % (2/37), with no recurrence after other procedures including decompression (n = 17) and En bloc resection (n = 7). Moreover, they concluded that formal TES is

Table 2
Surgery and Outcomes.

No	Frankel Score		ASIA Score		VAS Score		EBL (ml)	OT (min)	Post RT	Recurrence	FU (month)
	Pre-O	Po-O	Pre-O	Po-O	Pre-O	Po-O					
1	B	E	B	E	7	1	300	240	No	None	48
2	A	D	B	D	9	1	215	240	No	None	30
3	B	E	C	E	6	1	100	120	No	None	42
4	C	E	B	E	7	1	265	250	No	None	37
5	B	E	C	E	8	1	300	140	Yes	None	55
6	A	E	B	E	7	2	200	265	Yes	None	78
7	B	E	C	E	7	1	200	140	Yes	None	42
8	B	E	C	E	8	0	300	215	Yes	None	12
9	C	E	C	E	6	1	300	205	No	None	12
10	B	E	B	E	8	1	250	245	Yes	None	13
11	C	D	B	D	7	2	300	265	No	None	13
12	B	E	C	E	6	1	550	190	No	None	14
13	B	E	C	E	8	1	600	185	No	None	26
14	A	E	B	E	9	2	500	262	No	None	53
15	B	E	C	E	9	2	500	285	Yes	None	77
16	C	E	B	E	8	2	800	255	Yes	None	54
17	B	E	B	E	7	1	1600	260	Yes	None	32
18	C	E	C	E	6	2	500	265	Yes	None	88
19	B	E	B	E	8	1	200	120	Yes	None	89
20	A	D	B	D	9	3	1000	320	No	Yes	90
21	B	E	B	E	8	2	600	132	Yes	None	138
22	B	D	A	D	9	2	600	309	No	Yes	75
23	D	E	D	E	5	1	200	140	Yes	None	55
24	D	E	C	E	4	1	800	265	Yes	None	45
25	C	E	D	E	6	2	500	275	Yes	None	22
26	B	E	B	E	9	1	500	225	No	None	16
27	A	E	A	E	8	1	500	275	No	None	12
28	B	D	A	E	8	1	1000	255	No	None	16
29	B	E	A	E	8	1	400	255	No	None	17

No, number; ASIA, American Spinal Injury Association; VAS, Visual Analogue Score; Pre-O, preoperative; Po-O, postoperative; EBL, estimated blood loss; OT, operation time; RT, radiotherapy.

Table 3
Comparison of the treatment information in the two groups.

Variable	Goup A (Dec + PVP) N = 12	Group B (Dec) N = 17	*p*
Operation time (min)	215.9 (120–265)	240.2 (120–320)	
Intraoperative EBL (mL)	273.3 (100–550)	635.3 (200–1600)	< 0.0001
Pre-O VAS score	7.2 (6–9)	7.6 (4–9)	
Po-O VAS score	1.1 (0–2)	1.5 (1–3)	< 0.0001
Pre-O ASIA Score			
A	0	4	
B	6	7	
C	6	4	
D	0	2	
Po-O ASIA Score			
D	2	2	
E	10	15	
Pre-O Frankel Score			
A	2	3	
B	7	9	
C	3	3	
D	0	2	
Po-O Frankel Score			
D	2	3	
E	10	14	
HD (days)	5.1 (4–7)	5.2 (4–7)	
Number of recurrence	0	2	

Dec, decompression; EBL, estimated blood loss; VAS, Visual Analogue Score; ASIA, American Spinal Injury Association; Pre-O, preoperative; Po-O, postoperative; HD, hospitalization durations.

not always mandatory; brilliant local control rates and long-term survival can be achieved with aggressive intralesional resection. In our series, no patient underwent En bloc resection.

Current literature reports that the coupling of partial tumor resection and decompression is also suitable for AVHs with progressive neurological deficits [5,8,10]. Previous studies have shown that surgical decompression is appropriate when there is a significant or progressive neurological deficit [6,16,19,22,24–26]. Cloran et al. [40] suggested an algorithm to treat AVH in their paper. However, the authors do not suggest which surgical technique, anterior or posterior, should be used if the patient has back pain and significant neurological deficit. Even in the presence of significant neurological deficits, the authors advocate minimally invasive treatments such as alcohol ablation and arterial embolization of the feeding artery if the patient is not a surgical candidate. Furthermore, the authors do not mention how to deal with recurrences after minimally invasive procedures. The choice of the surgical technique anterior or posterior surgical approach is determined by the location of the hemangioma and the degree of neurological deficit [5,13,24]. Rapid decompression by laminectomy must be performed in all cases of active and progressive neurological deterioration [25,26]. Previous studies have also shown recovery rates of 70 % to 80 % when laminectomy alone is used to treat lesions that only press on the posterior elements without soft tissue extension [13,25]. In our series, seventeen patients undergo circumferential decompressive laminectomy and resection of the bony tissue from the vertebrae for progressive pain, neurological deficit, and extradural compression. Hence, we have found that decompressive laminectomy helps in the treatment of progressive neurological deficits and extradural compression.

In this study, patients who underwent partial tumor resection and decompression experienced rapid relief from nerve compression, and postoperative follow-up data showed good patient neurological recovery. Only 7% (2/29 patients) in our study had recurrent lesions, including 0 patients in the decompression plus PV group, with no recurrence after prior resection and stabilization. In addition, our data demonstrated that decompression without VP takes longer operation time, leads to more bleeding and significant postoperative pain, and significantly prolongs the therapeutic of patients. These data indicate that brilliant local control rates and long-term survival can result from

Table 4
Review of the literature for treatment of vertebral haemangiomas.

Authors	Year	No of patients	Follow-up period	Treatments	Complications
Graham et al. [46]	1984	1	15 months	PE and Vertebral resection	No
Asthana et al. [61]	1990	17	–	Radiotherapy with 35–40 Gy	Not mentioned
Fox et al. [8]	1993	11	77.4 months	Decompression surgery + Arterial embolization. Decompression surgery + Radiotherapy. Subtotal tumor excision	Profuse intraoperative bleeding (n = 2) Arachnoiditis (n = 2) Epidural hemorrhage (n = 1)
Sakata et al. [62]	1997	14	–	Radiotherapy with 36 Gy	No
Schwartz et al. [47]	2000	1	–	Decompression (anterior approach) and RT	No
Doppman et al. [28]	2000	11	6–180 months	CT-guide injections of ethanol	Pathological fractures (n = 2)
Hadjipavlou et al. [48]	2007	1	36 months	Balloon kyphoplasty	No
Acosta et al. [13]	2008	22	2–240 months	16 surgeries: Preoperative embolization (n = 10) Decompression surgery (n = 7) Intralesional spondylectomy (n = 9) vertebroplasty or embolization (n = 6)	Excessive intraoperative bleeding (n = 1)
Kato et al. [6]	2010	5	92–163 months	Preoperative embolizations and total excisions (en bloc or piecemeal)	No
Vinay et al. [49]	2011	1	6 months	Decompression and Vertebroplasty	No
Singh et al. [58]	2011	10	12–26 months	Decompressions with intraoperative ethanol embolization	No
Acosta et al. [18]	2011	10	2.4 years (0.8–5.5yrs)	Preoperative embolizations and Intralesional spondylectomies without adjuvant radiotherapy.	No
Nair et al. [50]	2012	7	24	Anterior corpectomy + Posterior instrumentation	Not mentioned
Armaga nian et al. [51]	2013	1	12	Percutaneous osteosynthesis, embolization and kyphoplasty	No
Haque et al. [52]	2013	1	12	PE and vertebral corpectomy	No
Narayana et al. [63]	2014	14	36	Percutaneous vertebroplasty	No
Jiang et al. [16]	2015	20	4.3 years (2–11.1) years	Decompressive partial tumor excision radiotherapy (12), Decompressive partial tumor vertebroplasty radiotherapy (8)	No
Goldstein et al. [7]	2015	68	Mean 3.9 yrs	33 preoperative embolizations; 17 palliative decompressions, including 3 with adjuvant radiotherapy; 37 intralesional spondylectomies, including 2 with radiotherapy; 7 en bloc spondylectomies including 1 with radiotherapy; 7 surgeries without details	Not mentioned
Cloran et al. [40]	2015	16	20 months	CT guided alcohol ablation, Surgery	No
Li et al. [53]	2016	28	24 months	Decompressio (n:14) Decompression and VP (n:14)	Cement leakage occurred in 2 cases in Decompression and Vertebroplasty group
Zhang et al. [26]	2016	06	23 months	Decompression + Vertebroplasty Gel foam + Bone cement	No
Singh et al. [54]	2016	08	12 months	Alcohol ablation + posterior decompression and instrumentation.	No
Vasudeva et al. [59]	2016	05	8–43 months	4 preoperative embolizations; 1 en bloc spondylectomy; 2 piecemeal gross-total resections; 2 subtotal resections; 3 intraoperative VPs; and 1 adjuvant radiotherapy	CSF leak, wound infection DVT, and hardware failure in 1 case
Gajaseni et al. [55]	2017	01	12 months	PE and total T10 resection	No
Ben Wang et al. [45]	2018	20	20 months	Role of Radiotherapy and surgery	Not mentioned
Prabhuraj et al. [64]	2019	05	12 months	Surgery + perioperative glue embolization.	No
Ji et al. [56]	2019	07	Mean 51.4 months	Total en bloc spondylectomy	No
Wei hong Xu et al. [67]	2020	13	Average 62 = 19 months	Decompression + Vertebroplasty + Gelatin sponge + Cement (n = 6) Decompression + Intraoperative vertebroplasty (n = 7)	No
Yu et al. [57]	2020	06	25.8 months	Single-fraction SBRT Dose was 18 Gy (range, 13–20)	No
Makoto Handa et al. [60]	2020	15	128.4 = 88.6 months	Total en bloc spondylectomy (n = 10) Piecemeal total tumor excision(n = 1) Revision procedure 2 weeks after ineffective laminectomy (n = 4)	No
Xu D et al. [65]	2021	51	Average 5.3 = 2.1 years	16 Embolization + Decompression 19 Decompression + vertebroplasty 16 Embolisation + Decompression + Vertebroplasty	No
Guna et al. [66]	2023	21	Mean 55.78 = 25 months	3 Instrumented decompression 9 Instrumented decompression + vertebroplasty 9 Intralesional tumor resection + anterior reconstruction	Neurlogical worsening (n = 1) Wound dehiscence (n = 2)
Current study	2023	29	44.9 (12–138) months	Vertebroplasty + Decompression (n = 12) Decompression alone (n = 17)	Pneumonia (n = 1) Pleural effusion (n = 1) Wound infection (n = 1)

aggressive intralesional resection combined decompression for patients with a definite diagnosis.

Surgery is the only treatment option for AVH with back pain and neurological symptoms. The purpose of surgery is to remove or ablate the tumor that is compressing the neural components and to do bony decompression [41]. The conventional procedures are posterior decompressive laminectomy with vertebroplasty [45] or direct alcohol injection [40] and anterior corpectomy and reconstruction with previous feeding vessel embolization [18]. Pre-operation feeding vessel embolization is no longer indicated with these procedures. Post-operative radiation treatment may be advised if a residual lesion is seen in the follow-up contrast-enhanced CT [41].

Vertebroplasty is considered an independent treatment of aggressive vertebral hemangioma and offers advantages in addition to surgical resection and decompression [10,14,26,27]. Vertebroplasty is effective in reducing the risk of recurrence, reducing bleeding and relieving local pain [4,14,27,29]. In their study, Lie Dang et al. [41] proposed an algorithm based on the tumor's epidural and paravertebral extension. In patients with haemangioma with epidural or paravertebral extension, he recommends vertebroplasty, radiation therapy, or alcohol ablation, even if the patient is symptom- or pain-free. He promotes surgery in patients who have failed on the preceding lines of treatment, even if they are neurologically intact, and considers post-operative radiation therapy in all AVH patients who have developed significant or quickly progressing neurological deficits. After classifying the patient as having AVH based on radiological parameters, Subramaniam et al. [42] suggested an algorithm based on the patient's clinical presentation in the most recent published systematic review. Percutaneous vertebroplasty, guided alcohol ablation, or external beam radiation can be used to treat individuals with localized back pain who are not responding to medicinal treatment, as well as patients with back pain and radicular symptoms. Thus, Singh et al. [31] described that intraoperative alcohol injection into the vertebral body enabled optimal blood loss control. Previous studies have shown the benefits of embolization [10,30] for patients with AVH. Robinson Y et al. [30] reported that blood loss was significantly less in the group that received preoperative embolization than in the group that did not (980 vs. 1629 mL) [30]. As the blood loss was significantly less, we were able to quickly remove the intraspinal epidural component of the lesion without causing hypotension during surgery. Complete obliteration of the lesion with bone cement is indicated to prevent recurrence. Of the twelve patients treated with decompression plus vertebroplasty in our series, seven patients with a relatively higher rate of bone cement filling after surgery received no radiotherapy. None showed a recurrence. Therefore, effective decompression coupling VP can improve tumor control and symptom relief. Consistent with the previous result, our results showed a significant reduction in blood loss in the decompression plus VP group 273.3 (100–550 mL) compared to the decompression group 635.3 (200–1600 mL), indicating that VP also decreased the blood loss during decompression surgery. Our study uses embolization for patients with abundant arterial blood supply or large tumors on enhanced CT. In contrast, our department used embolization for small scale lesions or an upper CT artery with no apparent enhancement. Our study showed that preoperative embolization can effectively reduce blood loss and intraoperative complications during decompression surgery. Therefore, VP and pre-embolization are beneficial to reduce blood loss during AVH surgery.

Radiotherapy has been an effective therapeutic for VHs since 1930 [28,32,33,38]. Radiotherapy destroys the veins and capillaries of the spinal hemangioma by causing endothelial damage, thereby reducing the size of the tumor. Moreover, studies suggest that VH radiotherapy depends on a dose–response relationship [39]. Some studies reported that external irradiation with a total dose of 36–40 Gy could achieve the best efficacy in AVHs [8,32–34]. Conti et al. [43] attempted to demonstrate the therapeutic role of stereotactic body radiation therapy, often known as radiosurgery, in managing AVH. In their series, the authors find an overall satisfactory local response (no disease progression) and acceptable pain alleviation after radiosurgery at 94.1% and 87.5%, respectively. Nevertheless, their study revealed these factors were statistically insignificant ($p = .7$, $p = .2$). Although radiosurgery is considered more precision-focused than external beam radiation, the authors' review indicates that 22.3% ($p = .02$) of surrounding tissues are damaged after radiosurgery. However, high radiation therapy doses have potential risks such as radionecrosis [23,35–37]. Although radiation cannot decrease tumor compression, it can significantly relieve pain [8,33], and radiotherapy has been reported to be effective in reversing neurological deficits of vertebral hemangiomas [37]. In the last century, external beam radiation therapy has been the treatment of choice [41]. It produces avascular necrosis in tumor cells and has a solid anti-inflammatory impact on them. The typical protocol was a 40 Gy radiation dosage delivered in 10-20 cycles over four weeks. Radiation-induced myelitis and subsequent malignancy are two of its most serious side effects. Mreatment of vertebral hemangiomas with absolute alcohsignificant side effects [41]. CT-guided alcohol ablation is a feasible treatment option for AVH. Alcohol injection into the vertebral body produces vascular thrombosis and endothelial damage, leading to lesion devascularisation and subsequent shrinking, culminating in cord and nerve root decompression. According to Goyal et al. [44], the major cause of recurrences is the non-uniform dispersion of alcohol into the haemangiomatous vertebral body following injection. This explains the research cohort's high recurrence rate of 66.6%. In this study, fourteen patients received 4500 cGy and 4000 cGy conventional radiation therapy. The results showed that AVH recurrence occurred in 2 patients who underwent decompression surgery without radiation therapy and no recurrence was observed in the post-radiotherapy group. This suggests that canal decompression does not always result in a permanent cure; radiation therapy can be used to prevent local recurrence after surgery.

5. Conclusion

This study presented different surgical treatments for aggressive vertebral hemangioma, including decompression with or without vertebroplasty and internal fixation. We have demonstrated that coupling decompression and VP can also achieve good tumor control and reduce surgical complications. Moreover, preoperative vascular embolization and VP can reduce intraoperative bleeding in AVH surgery. In addition, postoperative radiotherapy is a good technique to prevent tumor recurrence when VP does not fill the vertebral body and tumor.

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Author Contributions

All authors contributed to the study conception and design. Material preparation, data collection and analysis were performed by Mohamed Diaty Diarra, Zengjie Zhang, Zhan Wang, Eloy Yinwang, Hengyuan Li, Shengdong Wang, and Peng Lin. The first draft of the manuscript was written by Mohamed Diaty Diarra. Xin Huang and Zhaoming Ye revised the manuscript. All authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Ethics approval

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This study was approved by the Ethical Board of the Second Affiliated Hospital, Zhejiang University School of Medicine (No. 20220415).

Consent to participate

Informed consent was obtained from all individual participants included in the study.

Consent to publish

The authors affirm that human research participants provided informed consent for publication of the images in Figs. 1, 2.

Code availability

Not applicable

CRediT authorship contribution statement

Mohamed Diaty Diarra: Conceptualization, Data curation, Methodology, Supervision, Validation, Writing – original draft, Writing – review & editing, Visualization. **Zengjie Zhang:** Investigation, Visualization. **Zhan Wang:** Funding acquisition, Investigation, Visualization. **Eloy Yinwang:** Formal analysis, Software. **Hengyuan Li:** Investigation, Methodology, Supervision. **Shengdong Wang:** Investigation, Supervision, Visualization. **Peng Lin:** Investigation, Visualization. **Xin Huang:** Investigation, Validation, Writing – review & editing. **Zhaoming Ye:** Funding acquisition, Investigation, Project administration, Supervision, Validation, Visualization, Writing – review & editing.

Declaration of competing interest

The authors declare the following financial interests/personal relationships which may be considered as potential competing interests: Zhan Wang reports financial support was provided by The Second Affiliated Hospital Zhejiang University School of Medicine.

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