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# Multidisciplinary management of invasive vertebral hemangioma: the role of different neurointervention modalities—a retrospective study

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## Abstract

**Background** Catastrophic intraoperative bleeding is a major complication of surgical removal of invasive vertebral hemangioma. Interventional neuroradiology techniques could be more useful tools to manage such hemorrhagic lesions.

**Results** Retrospective analysis of cases of invasive vertebral hemangioma revealed 17 cases treated at the Alexandria University School of Medicine from 2006 to 2020. The study included 52.9% of males with a mean age of  $38.4 \pm 18.6$  years old. All patients reported local and sometimes radicular pain; 64.7% exhibited progressive neurological deficits. Imaging revealed thoracic spine affection in 11 cases, lumbar vertebral in four cases, and cervical vertebra in two cases. Vertebral involvement occurred in 10 cases and paravertebral extension in 13 cases. Neurointervention modalities included transarterial embolization followed by corpectomy and fixation (one case with C4 lesion), direct surgery with corpectomy and anterior fixation (one case with C7 lesion), vertebroplasty alone (four cases), vertebroplasty with fixation (seven cases), and direct transpedicular alcohol injection with immediate devascularization and necrosis of the vascular channels inside the lesions (six cases). The alcohol injection use ranged from 4 to 10 ml in each pedicle. All patients did well during the follow-period post-intervention. The neurological deficits improved over six months. All patients showed improved Nurick grade regardless of the intervention (preoperative mean  $2.7 \pm 1.9$  vs. postoperative mean  $1.1 \pm 1.3$ ,  $p$  value 0.0001). Two patients were completely paraplegic, but with intact deep sensation, they improved dramatically and can walk unsupported post-intervention.

**Conclusions** Vertebral hemangioma can present in an invasive manner that necessitates intervention. Preoperative embolization, alcohol injection, or vertebroplasty are helpful methods to decrease intraoperative catastrophic hemorrhage. Alcohol injection is cost-effective with immediate devascularization of the lesion. The extensive 360 surgery utilization can be decreased with the use of alcohol and vertebroplasty. More cases are needed to validate these conclusions.

**Keywords** Vertebral hemangioma, Aggressive, Invasive, Vertebroplasty, Absolute alcohol, Endovascular embolization, Percutaneous, Laminectomy, Fixation

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**Background**

Vertebral hemangioma (VH) has the highest incidence among vertebral column space-occupying lesions, and the estimated incidence is about 10–12% in population-based studies [1, 2]. In most cases, they are accidentally discovered without any clinical manifestations. The benign noninvasive lesions are confined to the vertebral body without posterior element involvement and represent 99% of cases. On the contrary, invasive vertebral hemangiomas (IVH) are rare representing only 1% of cases [3]. The invasive lesions extend beyond the vertebral body to posterior elements, spinal canal, and other paravertebral locations with progressive compression of the neural structures [4]. Pathological fractures are also a common sequela of IVH due to bone weakness. The clinical manifestation includes local pain, radicular pain, and weakness ranging from minimal weakness to complete paraplegia [1, 2].

Due to the extensive vascularity of invasive lesions, surgery is usually accompanied by massive bleeding if done without preoperative hemostatic techniques like embolization or vertebroplasty [1, 2]. Also, inadequate removal of invasive lesions is common leading to more neurological deterioration.

The rarity of IVH stands behind the lack of controlled studies which adds more challenges to optimizing the management of those difficult lesions [4]. The previously used treatment lines included direct tumor attack with decompression, transarterial embolization, percutaneous embolization with absolute alcohol or other liquid embolic agents, vertebroplasty, and radiotherapy. We aim to discuss the perioperative tools that allow easier management [3, 4].

**Methods**

The data of patients with vertebral hemangioma admitted to the neurosurgery department at the Alexandria University School of Medicine from 2006 to 2020 were collected. We included all cases with symptomatic vertebral hemangioma either fresh or recurrent. Asymptomatic patients were excluded from this analysis and are not presented as a part of this case series. All patients

had CT scans, and MRIs to assess tumor extension and allow proper treatment options. The management option of choice was based on tumor extension and patient symptomatology. Minimally invasive percutaneous vertebroplasty was done in cases of painful lesions without deficits and no radiological evidence of compression. The presence of neurological deficits and neurological compression on imaging was an indication of a combined approach. Clinical and radiological follow-up was done from six months to three years. The preoperative and postoperative neurological status was assessed according to Nurick grades (Table 1) [2]. Statistical analysis was done using excel and STATA 17. Descriptive analyses were performed to characterize the distributions of variables. The analyses were conducted using frequencies for binary and categorical variables, mean for continuous variables, and student's t-test to compare pre- and post-operative neurological assessment.

**Results**

**Characteristics of the study population**

The patients included 9 females and 8 males with a mean age of  $38.35 \pm 18$  years old. All patients had local and sometimes radicular pain; progressive neurological deficit occurred in 11 cases. Imaging revealed thoracic spine affection in 11 cases, lumbar vertebral in four cases, and cervical vertebra in two cases. Vertebral body involvement occurred in nine cases, and paravertebral extension was encountered in 13 cases. Involvement of the posterior neural arch was observed in eight patients. The lesion was confined to the vertebral body in two cases. Ten patients had pathological fractures, and six patients had previous surgery. Four patients had a trial of excision and fixation, but surgery was omitted due to blood loss. (Table 2).

**Surgical approach**

Seven patients underwent transpedicular screw fixation and laminectomy. Two patients with thoracic spine hemangioma underwent anterior corpectomy and insertion of corpectomy cage followed by posterior laminectomy and fixation. Another patient with

**Table 1** Nurick grades

0	Signs or symptoms of root involvement but without evidence of spinal cord disease
1	Signs of spinal cord disease but no difficulty in walking
2	Slight difficulty in walking which did not prevent full-time employment
3	Difficulty in walking which prevented full-time employment or the ability to do all housework, but which was not so severe as to require someone else's help to walk
4	Able to walk only with someone else's help or with the aid of a frame
5	Chair-bound or bedridden

**Table 2** Demographic, clinical, and interventional details of the study cases

No	Age & sex	Level	Tumor extension	Path. fracture	pain	FND	Nurick grades preoperative	Previous surgery	Intervention	Follow-up in (month)	Improvement	Nurick grades postoperative
1	59 f	D10	WV, EC	Yes	Yes	Yes	4	Yes	A + V + F + L	6	Yes	2
2	67 F	D6	Vert body, pedicle, EC	No	Yes	Yes	3	No	A + V + F + L	24	Yes	1
3	30 m	D8	Vert body, pedicle, EC	Yes	Yes	Yes	5	No	A + V + F + L	8	No	5
4	49 F	D6	Vert body, pedicle	No	Yes	No	2	No	A + V	18	Yes	0
5	30 f	C4	WV, EC	Yes	Yes	No	1	Yes	E + C + F + RT	11	Yes	0
6	25 f	D9	WV, EC	Yes	Yes	Yes	5	Yes	E + C + F + L	36	Yes	2
7	22 m	D9	WV, EC	Yes	Yes	Yes	4	Yes	E + C + F + L	7	Yes	2
8	20 m	C7	Vert body, pedicle, EC	Yes	Yes	Yes	4	No	C + F + L	36	Yes	1
9	66 m	D10	WV, EC	YES	Yes	Yes	4	YES	A + V + F + L	13	Yes	2
10	53 m	D10	WV, EC	No	Yes	Yes	2	No	A + V + F + L	15	Yes	0
11	45 m	L3	WV, EC	No	Yes	yes	1	No	V + F + L	30	Yes	0
12	18 f	D6	WV, EC	Yes	Yes	YES	5	No	V + F + L	21	Yes	1
13	45 m	D4	WV, EC	Yes	Yes	Yes	2	Yes	A + V + L	11	Yes	1
14	65 m	L4	Vert body	No	Yes	No	0	No	V	6	Yes	0
15	17 f	L2	Vert body, pedicle	No	Yes	No	0	No	V	8	Yes	0
16	22 m	L3	Vert body	No	Yes	No	0	No	V	6	Yes	0
17	19 f	D11	WV, EC	Yes	Yes	No	5	No	V	6	Yes	1

EC epidural component, vert vertebral, FND focal neurological deficit, A alcohol, V vertebroplasty, F fixation, L laminectomy, C corpectomy, E embolization, WBodyole vertebra (body, pedicle, &lamina), ALS (Aminoff and Logue disability scale)

C7 hemangioma underwent direct surgery with a corpectomy and 360-degree fixation. Operative blood loss reached 2000 cc. The patient with a C4 lesion underwent anterior corpectomy and anterior fixation after the transarterial embolization.

### Vertebroplasty

Vertebroplasty was used in 13 cases to augment the anterior column. Fixation with vertebroplasty was done in seven cases. Vertebroplasty alone was done in four cases. Vertebroplasty with alcohol was used in two cases (Fig. 1).

### Embolization and alcohol injection

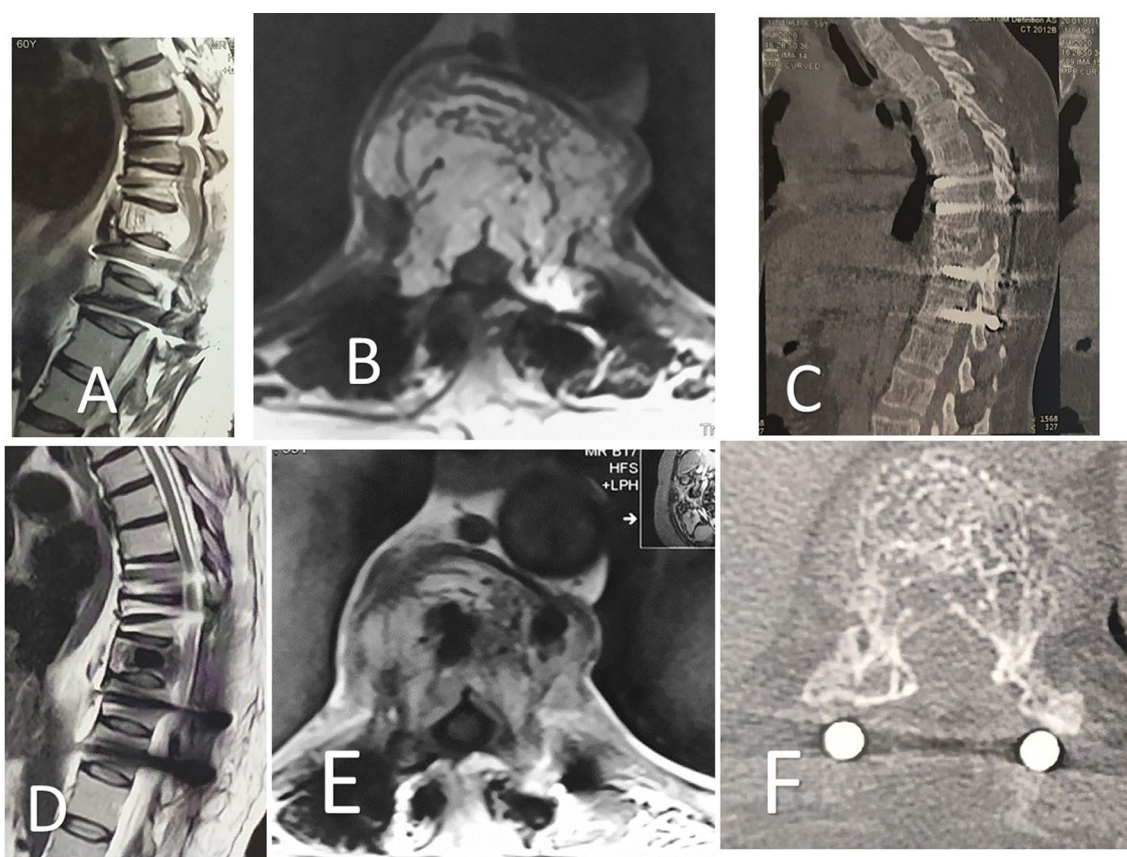
Alcohol injection was used in seven cases; 4–10 ml were injected in each pedicle. Before deciding on alcohol infusion, contrast should not be washed rapidly on fluoroscopy. Transarterial microcatheter embolization with Onyx was done in one case with a C4 lesion through the

ascending cervical arteries (Fig. 2). Direct transpedicular onyx injection was used in two cases. The advantage of onyx use over alcohol relies on its radiopacity; however, onyx is much more expensive.

### Perioperative complications and follow-up:

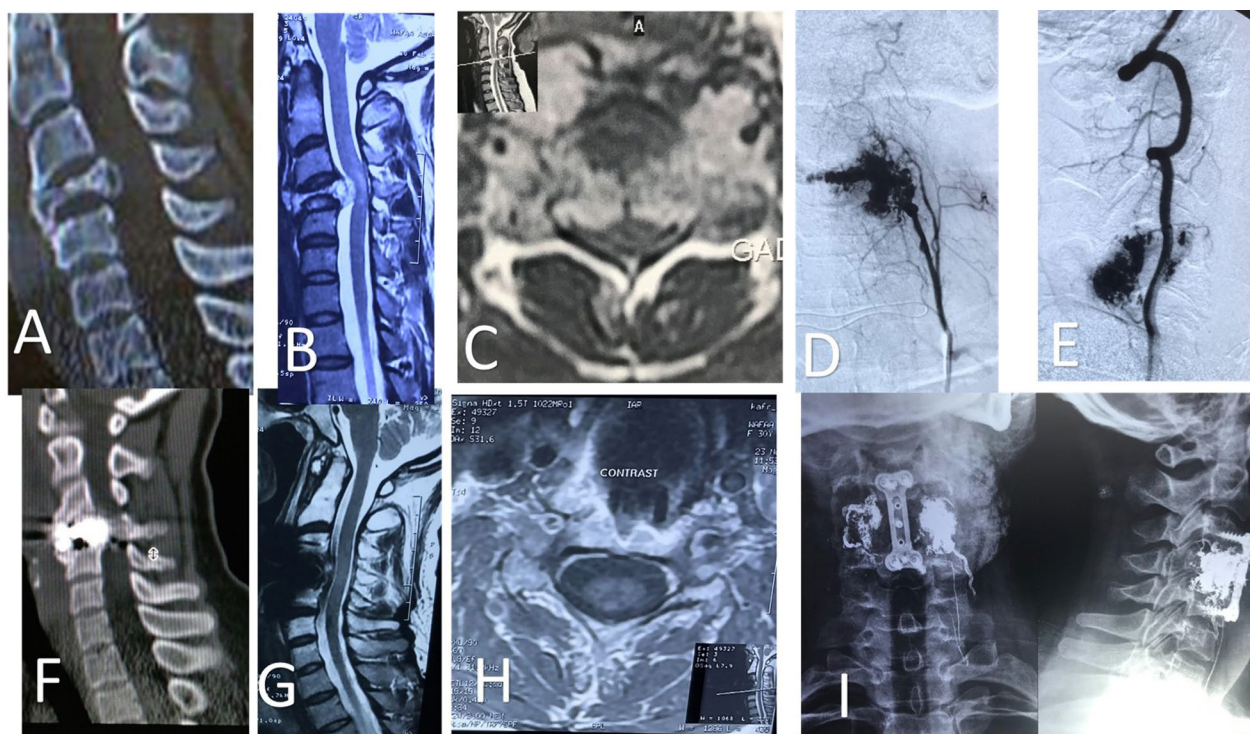
Follow-up (FU) of the patients ranged from 6 months to 3 years (mean FU was 15 months) (Table 2). No mortality was seen in this case series. A patient with D4 Lesion had grade 2 weakness that improved to grade 4 immediately after alcohol injection and posterior fixation. This patient experienced some deterioration on the 10th postoperative day. MRI showed epidural fluid collection. Evacuation resulted in improvement of the patient (Fig. 3).

Two patients were completely paraplegic, but with intact deep sensation, they improved dramatically and can walk unsupported. One patient with residual after the intervention was transferred to the oncology department for radiation. Another two patients underwent



**Fig. 1** 60-year-old female previously operated for laminectomy and two-level fixation of D10 vertebral. Her weakness improved for a few months, and then, progressive deterioration occurred again. Percutaneous alcohol injection followed by vertebroplasty was decided. The patient improved again within 3 months. **A** sagittal MRI showing D10 Hemangioma with spinal canal narrowing with the shadow of plate and screws. **B** Axial MRI with GAD showing epidural extension of the hemangioma with cord compression. **D, E** MRI one month after alcohol & vertebroplasty showing regression of the epidural component with the black signal of the vertebroplasty. **C, F** sagittal and axial CT showing D10 lesion before vertebroplasty with the plate and screws





**Fig. 2** 30-year-old female presented with neck pain with a previous history of surgical intervention of the lesion; fortunately, the patient had no neurological deficits: **A, B, C** are the preoperative CT and MRI showing pathological fracture of C4 with whole vertebral involvement, paravertebral expansion, and cervical spinal canal compromise. **D, E** angiographic images of the vertebral and ascending cervical arteries show the massive arterial blush of the hemangioma. **F** sagittal CT after onyx embolization. **G, H** late follow-up MRI with contrast done one year after corpectomy, fixation, and 6 months following radiotherapy showing lesion regression with good cord decompression. **I** postoperative plain X-ray anteroposterior and lateral views showing fixation plate and screws with residual onyx cast on both sides laterally

laminectomy and posterior fixation alone. The neurological symptoms improved for some time then deterioration occurred. The epidural component was compressing the cord (Fig. 1). Transpedicular alcohol and vertebroplasty were done. The patient experienced clinical improvement again. All patients showed improved Nurick grade regardless of the intervention except one (preoperative mean  $2.7 \pm 1.9$  vs. postoperative mean  $1.1 \pm 1.3$ ,  $p$  value 0.0001). A proposed algorithm for the treatment of those highly vascular lesions is added (Fig. 4).

### Discussion:

Invasive vertebral hemangioma represents a small percent of the hemangioma affecting the vertebral column. These invasive lesions extend outside the vertebral body in all directions. Neural compression occurs because of spinal canal compromise. Pathological fractures may occur in many cases, and spontaneous epidural hemorrhage was reported due to extensive vascularity [1–6].

Stand-alone surgical intervention is usually associated with massive blood loss and in most cases, and the procedure is aborted [1, 4]. Also, recurrence after initial improvement was reported in some studies. The

profuse bleeding during surgery makes the adequate decompression of the neural structures impossible. In this series, six cases (35.29%) had direct surgical intervention. The surgery was aborted in four cases due to blood loss and re-surgery was done after using hemostatic procedures including vertebroplasty and embolization. Two cases had passed surgical intervention with postoperative improvement despite the blood loss. This is similar to previously reported literature where Jiang et al. [3] reported two aborted surgeries (out of 29 case series) with blood loss ranging from 500 to 5500 ml. Chandra et al. [7] reported one case with an abandoned procedure and two with inadequate surgery requiring further intervention with the combined approach. Blecher et al. [8] reported massive bleeding after biopsy in one of their cases. Singh et al. [5] reported one case with recurrent symptoms after laminectomy and fixation.

Many options are available to facilitate intervention in those aggressive lesions. These include vertebroplasty, direct absolute alcohol infusion, transarterial or direct liquid embolic agent embolization (n-Butyl cyanoacrylate (NBCA) & Onyx), and radiotherapy. All these markedly



**Fig. 3** Forty-five-year-old male presented with progressive paraparesis with sensory level at lower chest level. **A, B** show the MRI of the dorsal spine with D4 invasive hemangioma involving the whole vertebral with spinal/canal narrowing from the epidural component. **C, D** shows the CT scan with characteristic Polka dot sign and striations of the hemangioma. **E & J**: plain X-ray of vertebroplasty procedure showing the needles. **F, G** Postoperative MRI showing regression of the hemangioma and disappearance of the epidural component. **H, I** Postoperative CT scan showing the hemangioma replaced with bone cement and laminectomy for decompression

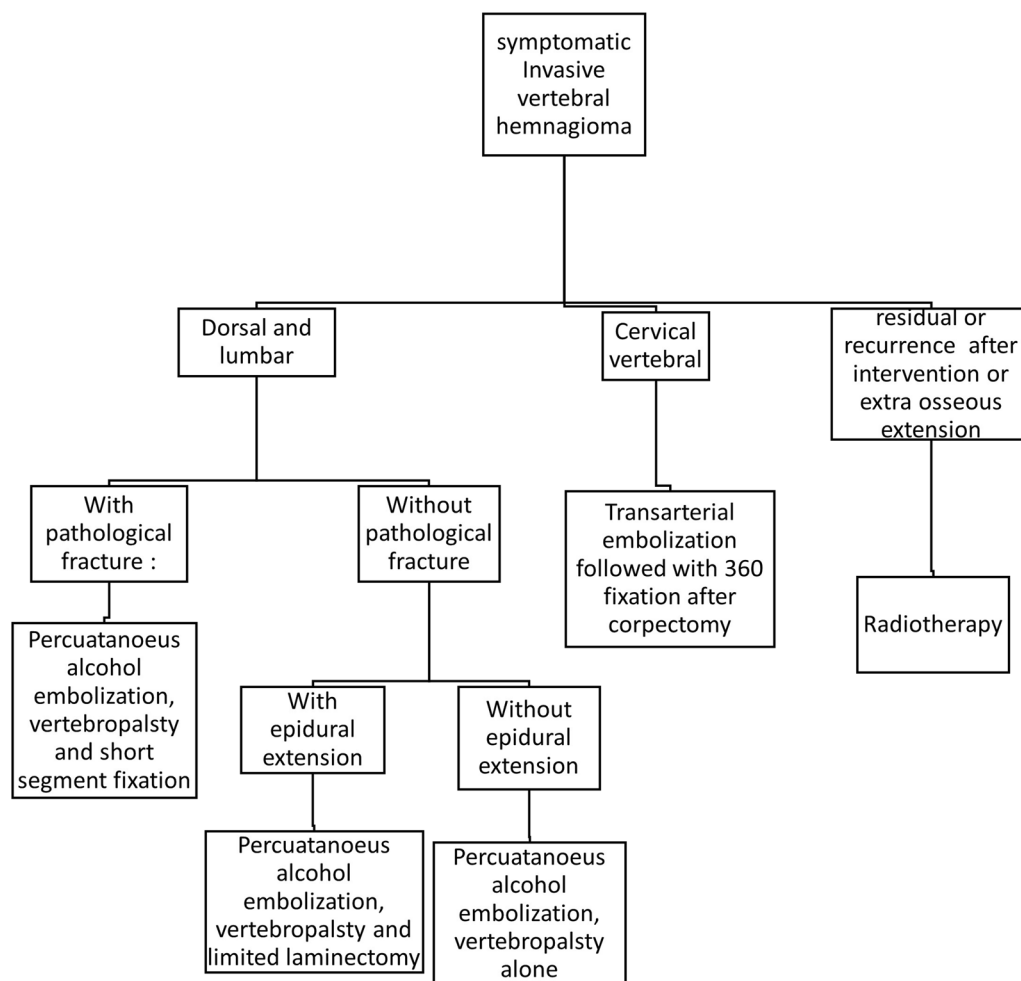
decrease the blood flow and allow safe surgical interventions. These can be used alone or in combination with surgical intervention [9–14].

Surgeries for hemangioma depend on the extent of the lesion, degree of neural compression, spinal canal compromise, presence or absence of neurological deficit, and presence or absence of pathological fractures. Surgeries include anterior fixation, posterior fixation, and 360-degree fixation. Also, short, and long-segment struts were applied depending on the situation [2–5]. Long-segment fixation was done for 11 cases in this series. In previous studies, single- and two-level fixation were tried. Singh et al. [5] did single level fixation in their work in 10 patients. Chandra et al. [7] used short-segment fixation in their series. Shamhoo et al. [2] did only vertebroplasty and decompression without fixation, and the vertebral body height was not affected in their series. Acosta et al. [15] did total vertebrectomy in 9 patients with anterior column reconstruction after preoperative embolization long-segment fixation.

Absolute alcohol can be infused directly or percutaneously with C-arm guidance. Many previous studies showed the usefulness of absolute alcohol in devascularization, angioneurosis, and shrinkage of lesions [15–19]. Shrinkage and necrosis of the soft tissue component is immediate after alcohol injection which facilitates safe

and bloodless neural decompression [9–19]. We used alcohol in 6 cases with a volume from 4 to 10 ml in each pedicle. One of those patients had a progressive weakness after laminectomy and fixation with the presence of an epidural component. Percutaneous alcohol injection allowed shrinkage of the lesion inside the spinal canal and neural decompression with the improvement of the neurological status. Another patient had immediate postoperative improvement of his lower limb paraparesis. A stand-alone alcohol infusion may be associated with pathological fractures, especially with large volumes. The key factor for alcohol infusion is the ability of the lesion to retain contrast, rapid washout of the contrast is a contraindication for alcohol injection as it may lead to systemic toxicity. The amount of alcohol infusion is debatable in the literature. It is better to limit the amount of injection to avoid any morbidity [5]. The alcohol infusion is the cheapest method among all modalities and the epidural component can vanish immediately with alcohol [5]. In the study of Chandra et al [7], five patients were paraplegic and improved with alcohol embolization and short-segment fixation [5, 7, 9–12].

Vertebroplasty with bone cement is a well-known method for the treatment of vertebral hemangioma. Methyl methacrylate cement is characterized by its hemostatic effect, and its construct can support the



**Fig. 4** A proposed algorithm for the treatment of invasive vertebral hemangioma

anterior and middle columns of the vertebral body, especially in the presence of pathological fractures. The debate against its use is the presence of an epidural component. The epidural component may collapse after vertebroplasty due to a block of the blood supply. The addition of alcohol injection can help wipe out the epidural component and at the same time reconstruct the vertebral body thus preventing vertebral collapse. One case of this series with dorsal hemangioma had initial improvement followed by progressive weakness later due to progressive cord compression. Percutaneous alcohol injection was done which allowed regression of the intraspinal component. Vertebroplasty was added to support the vertebral body. In contained vertebral hemangioma without paravertebral extension, vertebroplasty can be used as a standalone intervention to ameliorate patient symptoms. Vertebroplasty was used with short-segment fixation in previous reports with

good results [2, 3, 13, 14]. Shamhoot et al. [2] used vertebroplasty alone for their cases. They mentioned that the vertebral height was maintained on follow-up.

The extensive hemorrhage associated with surgery of those highly vascular lesions made the use of endovascular embolization (EVE) logic of high priority. EVE can be transarterial through the radicular spinal feeders using liquid embolic agents like Onyx and NBCA. The embolic agents obliterate the vascular channels inside the lesion allowing safe non-bloody resection of the epidural component. Those embolic agents can be injected through the pedicles of the affected vertebra. This is technically easier than transarterial embolization and avoids the navigation through the vessels supplying the cord, especially the artery of Adamkiewicz. Direct transpedicular EVE decreases the possibility of ischemic complications, decrease the cost of using catheters, and decreases radiation exposure. Unfortunately, EVE cannot be used as



a stand-alone treatment according to many previous studies as the recurrence and regrowth of the lesion can occur [4, 8, 21–25]. Acosta et al. [20] made preoperative embolization before vertebrectomy in 10 myelopathic cases with invasive hemangioma.

Radiation treatment for invasive IVH is well-known. It is used for patients without neurological deficits. Regression of the lesion can occur with radiotherapy. It can be also used for remnants after surgery. Previous reports showed clinical improvement and functional recovery from 60 to 100%. The effect of radiation is dose-dependent and ranged from 30 to 40 Gy in previous case series [3, 10, 26, 27]. One case of this series with C4 hemangioma was sent for radiation as she had a remnant after surgery. The remnant decreased on follow-up imaging. This is similar to the study of Acosta et al. [20] that send a patient with a residual lesion for radiation treatment [20]. Jiang et al. [3] reported the disappearance of extraosseous extension on follow-up after irradiation with recalcification in 2 cases. They did vertebroplasty, later on, to prevent pathological fracture in one case [3]. Despite the effectiveness of radiation, it carries early and late side effects. It is not recommended as routine treatment. Jiang et al. [3] reported incidence of pathological fractures after radiation therapy of vertebral hemangioma. [3]

Clinical improvement was observed in most previous series with percutaneous alcohol and vertebroplasty with or without fixation. All patients in this series showed improved Nurick grades regardless of the intervention except in one case. Singh et al. [2] reported clinical improvement in all cases after the intervention. Shanhoot et al. [5] had similar results. Jiang et al. [3] reported that their patients were symptomless on follow-up except for 5 patients with numbness in both lower limbs.

## Conclusions

Vertebral hemangioma can present in an invasive manner that necessitates intervention. Preoperative embolization, alcohol injection, or vertebroplasty are helpful methods to decrease intraoperative catastrophic hemorrhage. Alcohol injection is cost-effective with immediate devascularization of the lesion. The extensive 360 surgery utilization can be decreased with the use of alcohol and vertebroplasty.

## Abbreviations

IVH	Invasive vertebral hemangioma
NBCA	n-Butyl cyanoacrylate
EC	Epidural component,
FND	Focal neurological deficit
WV	Whole vertebra
EVE	Endovascular embolization

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## Author contributions

AES analyzed and interpreted the patients' data regarding the risk factors, operative details, and clinical outcomes. MA and AE performed statistical analysis and English editing. TH shared in the clinical assessments and operative interventions and also revised the manuscript. All authors performed clinical evaluations of patients, and surgical interventions, and helped in reviewing and editing the manuscript. All authors read and approved the final manuscript.

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## Availability of data and materials

The data sets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

## Declarations

### Ethics approval and consent to participate

The research protocol was approved by the ethical committee in the Faculty of Medicine at Alexandria University in its monthly session. Informed written consent was obtained from each patient. The reference number is: Member of ICLAS, <http://iclas.org/members/member-list>, <http://www.hhs.gov/ohrp/assurances/index.html>. IRB NO: 00012098(Expires 6–10-2022) -F WA NO: 00018699 (Expires April 2021), serial no.:0304846.

### Consent for publication

Not applicable.

### Competing interests

The authors declare that they have no competing interests.

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