Aggressive Vertebral Hemangiomas Causing Compression Fracture with Neurological Deficits: A Retrospective Cohort Study

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Research article

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Abstract

Background: In rare instances, aggressive vertebral hemangiomas (VHs) can cause compression fracture, resulting in severe pain and neurological deficits. But the diagnosis and treatment of these aggressive lesions are challenging because of these lesions are rare and atypical. This study aimed to evaluate the safety and efficacy of surgical management for aggressive VHs with vertebral compression fracture by a modified multimodality surgery.

Methods: We retrospectively reviewed 6 cases suffering from aggressive VHs with compression fracture and neurological deficits in our department from July 2011 to April 2016. These patients were treated by the multimodality surgery, including preoperative embolization, intraoperative injection of gelfoam mixed with cement, and laminectomy decompression. The follow-up period was at least 3 years. Perioperative parameters, clinical outcomes, and radiographical data were collected and analyzed.

Results: The 6 patients involved 1 male and 5 females (mean age, 52.3 years). The levels involved were: in thoracic spine (5 cases) and lumbar spine (1 case). Preoperative CT-guided biopsy was conducted in all patients, with 5 patients had definitive pathologic diagnosis. All patients were treated successfully with the multimodal surgery, with no cement leakage and other severe complications. The mean operation time was 182.2 minutes, and the estimated blood loss was 908.3 ml. At an average follow-up of 49.8 months, clinical outcomes assessed by the visual analogue scale and Frankel grade were significantly improved. The vertebral body height and kyphosis angle of the fractured vertebra were also corrected postoperatively. No affected vertebra re-fracture and adjacent vertebral fracture were developed, and none of the patients experienced recurrence of tumor at final follow-up.

Conclusions: In cases of aggressive VHs causing compression fracture with neurological deficits, CT-guided biopsy is indicated for the diagnosis. The multimodality surgery (preoperative embolization, intraoperative injection of gelfoam mixed with bone cement combined laminectomy decompression) is effective and safe, and can be considered as an acceptable surgical choice.

Background

Vertebral hemangiomas (VHs), one of the common benign spinal diseases, frequently affect the thoracic and lumbar spine [1, 2]. Most of these hemangiomas are incidentally discovered and have no obvious symptoms. For asymptomatic VHs, treatment is unnecessary. However, in rare instances, VHs can present as a locally aggressive behavior by enlarging and extending into the spinal canal and/or paravertebral space, leading to spinal cord compression [3-6]. Aggressive VHs often exhibit several symptoms, including local back pain, radicular pain, or even neurological deficits. It has been reported that there are 0.9 to 1.2% of patients with symptomatic or aggressive VHs, which require medical treatment [1, 7].

Usually, most aggressive VHs present typical radiographical findings, such as a vertical striations sign, honeycomb morphology, or polka-dot change [8, 9]. Nevertheless, some aggressive lesions can cause underlying bone resorption and even compression fracture [10, 11]. Wang et al. [12] recently demonstrated that compression fracture is one of the atypical features, which may result in misdiagnosis. As such, the diagnosis and treatment protocol for vertebral compression fracture by aggressive VHs deserves further investigation.

Although various non-surgical treatment modalities have been described for treating aggressive VHs [5, 13-16], the outcome is sometimes unsatisfied. Surgical therapy is still the most radical method of clinical treatment of aggressive VHs. The surgical procedures mainly involved laminectomy decompression, intralesional vertebrectomy, and en-bloc spondylectomy; but the formulation of ideal surgery for treating these lesions is still a matter of debate because of the rarity of aggressive VHs [4, 6, 7, 17]. In this study, we reported 6 patients with compression fracture caused by aggressive VHs. These patients were treated with our modified multimodality surgery (preoperative embolization, intraoperative injection of gelfoam mixed with bone cement combined laminectomy decompression). The objective of this multimodality surgical technique was to minimize invasive, decompress spinal cord, and reduce the complication rate.

Methods

Patient population

This study was approved by the First Affiliated Hospital of Fujian Medical University Ethics Committee and written informed consent was obtained from all patients in this study. From July 2011 to April 2016, 6 consecutive adult patients suffering from aggressive VHs with compression fracture and neurological deficits were retrospectively reviewed. These patients were treated with the multimodality surgery, which included preoperative embolization, intraoperative injection of gelfoam with vertebroplasty, and laminectomy. The follow-up period was at least 3 years. Patients with incomplete medical records or lost to follow-up were excluded. All patients received complete preoperative radiological examinations, including X-ray radiography, computed tomography (CT), and magnetic resonance imaging (MRI) of the spine. Because compression fracture of the aggressive VHs is a relatively atypical radiographical feature, CT-guided percutaneous biopsy was indicated and conducted in all patients.

Surgical techniques

Preoperative super-selective trans-arterial embolization was performed in all patients to reduce risk of uncontrollable intraoperative bleeding. The surgical method included following steps: (1) preinjection of gelfoam to further reduce blood loss; (2) vertebroplasty to obliterate the tumor and offer

stability of affected vertebra; (3) decompressive laminectomy to remove the extensive tumor and decompress the spinal cord. All operations were performed by the same experienced surgeon.

The surgery was performed with a prone position under general anesthesia. After the affected vertebra, adjacent laminae and facet joint were exposed, a puncture needle was placed into the center of affected vertebral body through the unilateral transpedicular approach under the C-arm fluoroscopic guidance. Two pieces of 6 cm × 2 cm × 0.5 cm of sterile absorbable gelfoam (Jinling Pharmaceutical Co., Ltd., Nanjing, China) were scraped into 1.5 mm × 1.5 mm × 1.5 mm pieces. We mixed these small pieces of gelfoam with 20 mL of contrast medium (iohexol) (Fig. 1), and injected into the vertebral body via the channel of the needle. Then, the bone cement was injected to fill the entire lesion as much as possible with C-arm fluoroscopy to prevent cement leakage. After injection, the puncture needle was extracted when the cement was solidified. Laminectomy decompression was then performed to excise the epidural lesion and remove the bone cement leaked into the spinal canal. Posterior pedicle screws and both pre-bent rods were installed finally.

Postoperative management and follow-up

Typically, we routinely used intravenous antibiotics and hemostatic drug within 24 hours after surgery to prevent deep infection and delayed hemorrhage. The negative drainage was taken out when drainage flow less than 50 ml/24 h. Patients were allowed to start exercising and walking gradually with their thoracic or lumbar brace one week postoperatively.

Immediately after surgery, lateral and anteroposterior X-rays were obtained routinely to evaluate the extent of decompression and position of cement and instrumentations. Then, the X-ray examinations were carried out at 3-, 6-, and 12-month follow-up and then once per year. The CT scan was taken on at the 3-month follow-up and examined annually after that. If the patient showed any symptoms and signs of recurrence, MRI and CT were performed immediately.

Outcome assessment

The perioperative parameters included operation time, blood loss, fused segments, complications, and pathologic diagnosis. Clinical assessment was evaluated using the visual analogue scale (VAS) and the modified Frankel grade. The VAS score was used to assess pain intensity. The Frankel grade was used to evaluate neurological status. The anterior vertebral body height (ABH), middle vertebral body height (MBH), and kyphosis angle were measured on lateral X-ray radiography. Kyphosis angle was defined as the angle formed by 2 lines parallel to the superior and inferior endplate of the fractured vertebrae, using the Cobb method[18].

Review of the literature

Aggressive VHs causing compression fracture are relatively rare. For further study, a brief literature review was conducted of the management of aggressive VHs with vertebral fracture in the PubMed, EBSCO, OVID, and Springer databases. The search criteria were publications related to aggressive VHs in recent years. Cases who had aggressive features (Enneking Stage 3, spinal cord compression, paravertebral extension or neurological deficits) with compression fracture were enrolled.

Statistical analyses

Statistical analyses were performed through SPSS version 22.0 (SPSS Inc., Chicago, IL, USA). Continuous data were presented as mean ± standard deviation. Comparisons between post- and preoperative parameters were conducted by using the ttest; a P-value < 0.05 was considered statistically significant.

Results

Table 1 shows the clinical features of the 6 patients. There were 1 male and 5 females. Their mean age was 52.3 ± 9.0 years (range from 41 to 63 years). Five patients had VH lesions in the thoracic spine, and 1 in the lumbar spine. In 5 cases, high-intensity signals on T2-weighted MRI were found, and the remaining was iso-intensity signal. Based on Tomita classification, 3 patients were type IV, and 3 were type V. The mean Spinal Instability Neoplastic Score (SINS) was 10.2 ± 1.6 (range from 8 to 12). All 6 patients presented with obviously neurological deficits preoperatively, including 1 patient with Frankel B grade, 1 with Frankel C grade, and 4 with Frankel D grade. These 6 patients had varying degrees of unsteady walking, limb numbness or weakness, radiculopathy and muscle atrophy. The mean duration of symptoms was 10.2 ± 7.9 months (range from 2 to 24 months). Physical examination found percussion pain in the paravertebral or spinous processes, decreased lower-limb sensation, weakened muscle strength and tender hyperreflexia. Neurological function examination showed positive Babinski's sign in 2 patients. Preoperative CT-guided percutaneous biopsy was conducted in all patients. Of them, 5 patients had definitive pathologic diagnosis. In the remaining patient, the samples consist of only osteocyte and blood cells, and this case was diagnosed by postoperative pathology examination.

Table 1
1 Patient clinical features

No.	Gender	Age (years)	Site	Symptoms	Duration of symptoms (months)	T2WI	Tomita classification	SINS	Previous treatment
1	F	63	T8	Radiculopathy and pain	5	High	IV	8	NO
2	F	41	T12	Myelopathy and pain	12	High	V	12	NO
3	F	51	T3	Myelopathy and pain	2	High	V	12	NO
4	F	53	L2	Radiculopathy and pain	24	Iso	V	9	NO
5	M	62	T2	Myelopathy and pain	12	High	IV	10	NO
6	F	44	T8	Myelopathy and pain	6	High	IV	10	NO
<i>F</i> female, <i>M</i> male, <i>T2WI</i> T2-weighted MRI image, <i>High</i> high-intensity signal, <i>Iso</i> iso-intensity signal, <i>SINS</i> Spinal Instability Neoplastic Score									

All patients were treated successfully with the multimodality surgery, and the incision healed well. The average operation time was 182.2 ± 64.3 minutes (range from 120 to 300 minutes) (Table 2). The estimated intraoperative blood loss was 908.3 ± 349.9 ml (range from 500 to 1400 ml), and the average bone cement amount was 5.0 ± 1.7 ml (range from 3 to 7 ml). No bone cement leaked into the spinal canal was recorded. After surgery, the neurological symptoms were relieved for all patients, by varying degrees. The average follow-up period was 49.8 ± 12.6 months (range from 38 to 71 months). At final follow-up, the VAS score was significantly improved from 7.0 ± 1.5 to 1.5 ± 0.8 ($P < 0.01$), and Frankel E grade was achieved in 5 patients. Only one patient with preoperatively intramedullary T2-weighted MRI increased signal intensity achieved Frankel D grade at final follow-up, but this patient can sufficiently return to activities of daily living. Radiographical analyses showed the ABH and MBH significantly increased from 14.6 ± 2.2 mm to 16.6 ± 3.0 mm ($P = 0.021$) and from 12.1 ± 5.3 mm to 16.3 ± 4.2 mm at final follow-up ($P = 0.01$), respectively. The kyphosis angle was decreased from $12.2 \pm 4.3^\circ$ preoperatively to $6.4 \pm 1.6^\circ$ ($P = 0.01$).

Table 2
Patient surgical outcomes

No.	VAS score		Frankel grade		ORT (min)	EBL (ml)	Cement volume (ml)	ABH (mm)		MBH (mm)		Kyphosis angle ($^\circ$)		Complications	Follow-up (months)
	Pre	FFU	Pre	FFU				Pre	FFU	Pre	FFU	Pre	FFU		
1	9	3	D	E	120	700	6	14.4	15.9	13.5	16.7	7.4	5.0		43
2	5	1	D	E	153	1000	7	14.6	19.2	10.6	18.6	13.2	8.1		58
3	8	2	D	E	200	1400	5	13.9	14.4	9.1	14.7	9.9	5.9		40
4	6	1	B	D	300	1200	3	18.8	21.3	22.2	23.0	19.2	6.8	CSF leakage	38
5	8	1	D	E	140	650	6	12.7	13.9	8.1	13.2	14.4	8.3		71
6	6	1	C	E	180	500	3	13.2	14.7	8.8	11.4	9.0	4.4		49
<i>VAS</i> Visual Analogue Score, <i>Pre</i> preoperative, <i>FFU</i> final follow-up, <i>ORT</i> operation time, <i>EBL</i> estimated blood loss, <i>ABH</i> anterior vertebral body height, <i>MBH</i> middle vertebral body height, <i>CSF</i> cerebrospinal fluid															

There were no severe complications such as intraspinal hematomas, deep infection and thrombosis occurred in any patient. One patient had cerebrospinal fluid leakage postoperatively and recovered within 1 week by keeping the supine position and drainage. At final follow-up, the radiological images showed that the internal implant was in good position. No loosening or breakage of instrumentations was detected. No affected vertebra re-fracture and adjacent vertebral fracture were developed, and none of the patients experienced recurrence of tumor.

Discussion

VHs are histologically spinal benign vascular malformations consisting of newly formed blood vessels [1]. Occasionally, VHs can become aggressive to present as resorption of underlying bone, resulting in pathological compression fracture and then neurological deficit. The mechanism of aggressive VHs causing vertebral pathological fracture is that hemangiomas might cause diffuse bone infiltration, erode the horizontal trabeculae, and weaken bone mineral density [19]. Furthermore, the enlargement of the affected vertebra and the consequently compression fracture can yield spinal canal narrowing and spinal cord compression. The diagnosis and treatment of these lesions are challenging. But there is little literature focusing on the aggressive VHs with compression fracture and neurological deficits because of the rarity of these cases. In this report, we reviewed the patients who had aggressive VHs with compression fracture in our department from 2011 to 2016. We aimed to share our experience in the diagnosis and treatment of these patients.

Recently, Wang et al.[12]conducted a radiological analysis of 95 cases with aggressive VHs and reported more than 1/3 of aggressive lesions exhibit at least 1 atypical feature. Among these atypical findings, approximately 17% caseshad a vertebral compression fracture sign; the author also indicated that some patients may have atypical MRI signals (e.g., hypointense to isointense in T2-weighted images). Atypical VHs have more vascular but fewer fatty content so that these lesions can appear atypical MRI signals [9]. Therefore, the radiological diagnosis of compression fracture by aggressive VHs is difficult because atypical lesions can mimic other conditions, such as osteoporotic vertebral compression fracture, primary spinal malignancy, or metastatic disease [5, 9]. The misdiagnosis of aggressive VHs may cause uncontrollable intraoperative hemorrhages. In our study, CT-guided percutaneous biopsy was conducted in all patients before surgery (Fig. 2). Previous studies reported CT-guided biopsy has high value in the diagnosis of suspected atypical spinal malignancy and the overall diagnostic accuracy of CT-guided biopsy is up to 89% [20]. Our findings showed only one case had negative pathological diagnose, the false negative rate was in accordance with the literature.In this case, a diagnosis of VHs was still suspected because only osteocyte and blood cells were detected on biopsy, and postoperative pathological diagnosis of VHs was finally confirmed.

Surgery is still the primary but the most radical treatment for aggressive VHs, but the choice of surgery is still debated.The surgical method mainly involved decompression surgery, intralesional vertebrectomy, and total en-bloc spondylectomy[2-4, 6, 7, 9, 17]. Decompression surgeryby laminectomy is widely used because it is relatively simple and less technically demanding, but this procedure is reported to have higher recurrence rate.Combined with the intraoperative vertebroplasty, decompressive laminectomy can achieve less incidence of recurrence and complication, but also less blood loss[7, 11]. Vertebroplasty can provide immediate strong spinal column stabilization, shrink the blood vessels and cause sclerosis of the VH lesions. According to our experience and the literature review (Table 3)[5, 11, 15-17, 19, 21-29], the indication of surgical decompression is that cases with severe or rapidly developing neurological deficits. In a multicenter study conducted by Goldstein et al.[3], no recurrence of VHs was found in patients after en bloc resection and decompression with an average 3.9 years follow-up. The author suggested en bloc resection is not required because symptomatic VHs are benign spinal tumor, the excellent local control rate of and long-term survival can be due to aggressive intralesional resection during index surgery. In our clinical practice, we used preoperative trans-arterial embolization and intraoperative injection of gelfoam to further reduce blood loss.After injection of gelfoam and cement, the visualization of operative field was clearer, and the tumor tissue can be removed more effectively and thoroughly. We previously found the intraoperative injection of absorbable gelfoam is associated with fewer blood loss and surgery duration [30]. Because the size of gelfoam particlesare small, the communicating blood vessels of the tumor body can be embolized by the gelfoam particles, and the mixed contrast medium injected at the same step can demonstrate the effect of embolization. Besides, injecting gelfoam before vertebroplasty may effectively minimize the cement leakage into the spinal canal and paraspinal soft tissues. The gelfoam mixed contrast offers a relatively closed space for the diffusion of the bone cement, which can prevent the cement leaking into the spinalcanal, especially for patients with the destruction of the posterior vertebral body. In our series, the mean blood loss was observed to be 908.3 ml, with no bleeding- and cement-related complications occurred.

Table 3
Summary of the treatment of patients with vertebral fracture caused by aggressive hemangiomas

Authors	No. of pts.	Medical History	Tumor types	Site	Treatment	Results	Follow-up (month)	Complications
Graham et al. (1984) [21]	1	Three intestinal operations	AVHs	T12	PE and vertebral resection	Symptoms relief	15	NO
Schwartz et al. (2000) [22]	1	After parturition	SVHs	T11	Decompression (anterior approach) and RT	Symptoms relief	-	NO
Hadjipavlou et al. (2007) [23]	1	-	SVHs	L1	Balloon kyphoplasty	Symptoms relief	36	NO
Vinay et al. (2011) [24]	1	After loading a heavy	AVHs	L1	Decompression and VP	Symptoms relief	6	NO
Armaganian et al. (2013) [25]	1	Traffic accident	AVHs	L1	Percutaneous osteosynthesis, embolization and kyphoplasty	Symptoms relief	12	NO
Haque et al. (2013)[26]	1	NO	AVHs	L4	PE and vertebral corpectomy	Neurogenic claudication relief	12	NO
Liu et al. (2013) [16]	2	-	AVHs	L4:1 Not mentioned:1	VP	Symptoms relief	Mean 15.8	Cement leakage with no clinical symptoms
Jiang et al. (2014) [5]	5	-	AVHs	T2:1 T5:1 T11:1 T12:1 L3:1	RT:1 RT and decompression:1 Decompression:1 Decompression, VP and RT:1 Decompression and VP:1	Symptoms free	Mean 51.1	NO
Cloran et al. (2015) [27]	1	2-year after RT	AVHs	L5	VP	Symptoms relief	48	NO
Li et al. (2016) [11]	28	-	Murphy type IV VHs	T7:1 T8:1 T11:4 T12:9 L1:7 L2:4 L3:2	Decompression:14 Decompression and VP:14	Decompression plus VP is associated with better outcomes and few complications.	Mean 24	Cement leakage occurred in 2 cases in Decompression and VP group.
Gajaseeni et al. (2017) [28]	1	NO	AVHs		PE and total T10 resection	Symptoms relief	12	NO
Wang et al. (2018) [19]	2	Pregnancy	AVHs	T7	Decompression, VP and RT	Symptoms relief	38	NO
		Pregnancy	AVHs	T5	Decompression and VP	Symptoms free	15	NO
Wang et al. (2018) [15]	1	-	AVHs	T4	RT and VP	Symptoms relief	108	NO
Wang et al. (2019)[29]	1	4-day after delivery	AVHs	T5	Decompression and VP	Symptoms relief	6	NO
Ji et al. (2019) [17]	7	-	AVHs	T9:2 T10:1 L2:2 L3:2	Total en bloc spondylectomy	Neurological improved and pain relief	Mean 51.4	NO
No. number, Pt patient, AVHs aggressive vertebral hemangiomas, SVHs symptomatic vertebral hemangiomas, VHs vertebral hemangiomas, PE preoperative embolization, RT radiation therapy, VP vertebroplasty								

We performed laminectomy decompression and internal fixation in the last step, the cement escaped into the spinal canal can also be removed during the decompression, which can also decrease the complication of cement leakage. On the other hand, the instrumentations can offer stability to protect the affected vertebra from re-fracture and decrease the trend of adjacent segment fracture following vertebroplasty [31]. Clinical outcomes assessed by the

VAS score and Frankel grade were significantly improved after surgery, without severe complications. The ABH, MBH and kyphosis angle of the fractured vertebra were all significantly corrected at final follow-up. The results indicated that this multimodality surgery is an effective and safe technique.

Each case of aggressive VHs needs to be evaluated individually, because controversy remains as to the optimal surgical method. We suggest that the limitation of this multimodality surgery may be aggressive VHs caused by circumferential spinal compression or adjacent vertebral extension. For these cases, we still recommend complete tumor resection (total en bloc spondylectomy or intralesional vertebrectomy). Although none of patients in the present study experienced recurrence at final follow-up, the average 49.8 months follow-up period is relatively short to evaluate recurrence of tumor. Other limitations of this study included small number of cases from a single center. Future prospective multicenter studies with large sample sizes are needed to confirm our results.

Conclusions

The diagnosis and treatment of aggressive VHs causing compression fracture with neurological deficits are a challenge for spinal surgeons. Preoperative CT-guided biopsy is of great value for the diagnosis and should be recommended for these patients. The multimodality surgery (preoperative embolization, intraoperative injection of gelfoam mixed with bone cement combined laminectomy decompression) is an effective and safe procedure, and can be considered as an acceptable surgical choice.

Abbreviations

VHs: vertebral hemangiomas; CT: computed tomography; MRI: magnetic resonance imaging; VAS: visual analogue scale; ABH: anterior vertebral body height; MBH: middle vertebral body height; SINS: Spinal Instability Neoplastic Score

Declarations

Ethics approval and consent to participate

Written informed consent was obtained from all patients included in the study; the First Affiliated Hospital of Fujian Medical University Ethics Committee approved the study protocol.

Consent for publication

All patients signed informed consent for publication of their personal details and images in this article.

Availability of data and materials

The data supporting the conclusions of this article is included within the article. Data available upon reasonable request from corresponding author.

Competing interest

The authors declare that they have no conflict of interest.

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Authors' contributions

R-HM (first author): contributed to conception and design the research, and had been involved in drafting the manuscript. L-GS (co-first author): also help contributed to conception and design the research, and did analysis and interpretation of data. L-LM (co-first author): investigated and resolved the questions related to the accuracy of the data within manuscript. H-YM: was responsible for data collection and statistical analysis. X-ZQ (co-corresponding author) contributed to conception and design of the research, processed data and critically revised the manuscript. X-WH (corresponding author): contributed to conception and design of the research, carried out the surgical procedures, and gave final approval of the version to be published. All authors read and approved the final manuscript.

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Figures

Figure 1

A 41-year-old female patient with T12 aggressive vertebral hemangioma.(a-b) Preoperative magnetic resonance imaging demonstrated that the hemangioma extended into the spinal canal and paravertebral space, with T12 compression fracture. (c-d) Preoperative endovascular embolization. (e) The absorbable gelatin sponge (AGS) was scraped into the 1.5 mm × 1.5 mm × 1.5 mm particles and was mixed with contrast agent. (f) Injection of gelatin foam and bone cement during the surgery. (g-h) Postoperative radiography showed good positioning of the internal fixation. (i-j) Postoperative computed tomography revealed T12 vertebra is filled with bone cement and no recurrence of the tumor.

Figure 2

A 44-year-old female patient with T8 aggressive vertebral hemangioma. (a-c) Preoperative computed tomography (CT) demonstrated that an osteolytic tumor with T8 fracture. (d-e) Pathology after preoperative CT-guide biopsy was a hemangioma. (f-g) Postoperative X-ray radiography showed good positioning of the instrumentation. (h-j) Postoperative CT depicted the affected vertebra is filled with bone cement and no recurrence of the tumor.