

## Case Reports &amp; Case Series

## Multiple vertebral hemangiomas of the thoracic spine with atypical radiological features and aggressive behavior causing myelopathy: A case report

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## ABSTRACT

**Background:** Vertebral Haemangiomas (VHs) are frequent and generally asymptomatic benign tumors, involving the spine, usually incidentally found on computed tomography and magnetic resonance. Despite being usually asymptomatic and quiescent lesions, VHs can occasionally manifest aggressive features, leading to clinical manifestations such as back pain and neurological deficits.

**Case report.**

We report a case of a 54-year-old man, presented with 5 months history of pain, associated with lower limbs paraesthesia and weakness, gait disturbance and episodes of accidental falls. Radiological evaluation by spine pre- and post-contrast MRI indicated multiple vertebral hypervascular lesions, compatible with haemangiomas, involving from T3 to T11 levels, showing several different features (typical and atypical); aggressive haemangioma radiological pattern may be valuable at T3 and T4 vertebrae. A thoracic spine pre- and post-contrast computed tomography confirmed the radiological diagnosis of multiple aggressive haemangiomas.

**Discussion:** Aggressive VH consists in a very rare subset of vertebral haemangiomas characterized by a greater tendency in being symptomatic. They may show atypical radiological features, that make their diagnosis very complex. In the recent years, many strategies for treatment of symptomatic or aggressive VHs have been developed, but the optimal treatment strategy is still controversial.

**Conclusion:** Although aggressive VHs being extremely rare, recognizing radiological features of these lesions is mandatory to achieve a correct diagnosis and appropriate therapeutic targets.

### 1. Introduction

Vertebral haemangiomas (VHs) are well described lesions of the spine which comprise almost 12% of all benign tumours affecting the general population. VHs consist in a pathological proliferation of normal thin-walled capillary, cavernous or venous blood vessels within the marrow space and the bony trabeculae of the vertebra. Their most frequent location in the spine is at the thoracic and lumbar column [1], even if they can also involve multiple locations in the same patient. VHs are usually asymptomatic, incidentally discovered on imaging studies and do not require any treatment. [Table 1](#).

Among all VHs cases, just 0.9–1.2% of them shows an aggressive

behaviour to the point of becoming symptomatic.

Clinical manifestations can vary from back pain (55%) to neurologic deficits (45%) [2] and from myeloradiculopathy to paralysis, due to spinal cord or root compression, both resulting from bone expansion, erosion through cortex, pathologic fractures and hematomas.

Although rare, aggressive VHs represent a challenge for radiologist.

In order to obtain a correct diagnosis, recognizing the typical radiological features of VHs comes to be mandatory.

### 2. Clinical case

The patient was a 54 years old man presented with severe pain in

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**Table 1**

Overview of the literature: outcomes of patients with aggressive vertebral haemangiomas causing neurological deficit treated by only radiotherapy.

Authors	N of cases	Outcomes
Asthana et al. (1990) [33]	9 (on 17 total)	66% complete regression 11% partial response 22% no response
Aksu et al. (2008) [34]	1	Improvement of neurological status
Grau et al. (2009) [35]	1	Complete regression
Aich et al. (2010) [36]	7	Improvement of neurological status
Heyd et al. (2010) [37]	24 (on 84 total)	79% complete regression 21% partial response
Sewell et al. (2016) [38]	1	Improvement of neurological status
Wang et al. (2019) [31]	20	65% complete regression 35% worsened or no response

superior extremities and shoulder girdle of 5-months duration, slightly responsive to conservative treatments, including over-the-counter analgesics. During the four weeks prior to the admission, the clinical picture worsened with the appearance of lower limbs paraesthesia, gait disturbance and episodes of accidental falls. The patient's neurological examination revealed ataxo-spastic gait, mild weakness (Medical Research Council grade 4) involving the lower limbs and normal strength in the upper extremities. He denied urinary/bowel incontinence and perineal anaesthesia.

Pre- and post-contrast magnetic resonance (MRI) showed multiscal protrusions at C3-C4 and C6-C7 levels, with spinal canal stenosis due to pseudohypertrophy of the yellow ligament, and associated myelopathy. Moreover, the MRI also revealed multiple typical vertebral haemangiomas involving the dorsal spine from T3 to T11 levels.

T3 and T4 vertebral bodies showed the atypical aggressive haemangiomatous radiological pattern, with inhomogeneous signal on T1w and high intensity on T2w and STIR T2w sequences, involving the pedicles, transverse processes, vertebral laminae and part of the spinous processes in both vertebrae. The lesion also presented a mild inhomogeneous post-contrast enhancement. Starting from T3 posterior wall, it moved forward posteriorly as a soft vascular tissue, occupying the anterior epidural space and the T3-T4 right intervertebral foramen, resulting in spinal cord and radicular compression respectively (Fig. 1). Indeed, an hyperintensity signal in T2w was also found at T3 level, that can be reported to post-compressive myelopathy. The same angiomas

lesion also involved the other intervertebral foramen T3-T4, on the left, and both T4-T5 intervertebral foramina, but to a lesser extent.

T5, T6, T7, T8, T9 and T10 levels were just partially involved by similar haemangiomas. In particular, to be infiltrated were the external portion of the body, the pedicles and articular processes, on both sides (Fig. 2).

In T11 vertebral body, instead, was depicted another typical haemangioma with not aggressive morphology and an ovoidal shape (Fig. 3).

A thoracic spine pre- and post-contrast computed tomography was also performed, confirming the presence of a vertebral haemangioma, involving the T3 and T4 vertebral bodies, laminae and transverse processes, showing the pathognomonic pattern of trabecular pseudoepithelioid ("polka-dot sign"). The portion of this lesion located behind T3 vertebral body extended posteriorly, reaching the T3-T4 right lateral epidural space, compressing the spinal cord and showing a humble post-contrast enhancement (Fig. 1).

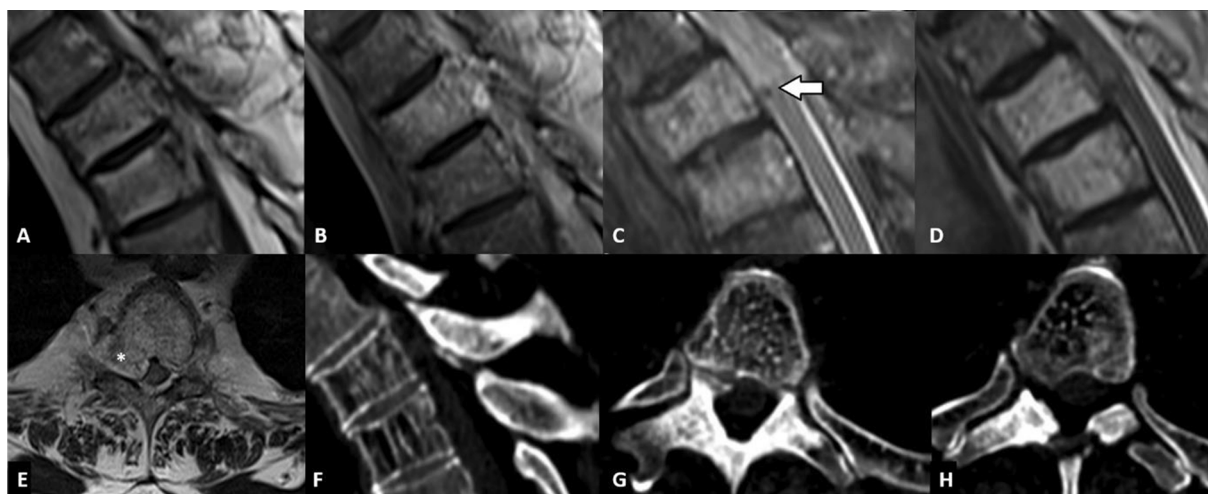
For a further in-depth investigation, the patient was evaluated by a Fluorodeoxyglucose-labelled Positron Emission Tomography (FDG-PET). On positron emission tomography-CT, VH scan show variable degrees of 18-FDG and 68-Ga DOTATATE uptake, manifesting either as a "cold" or "hot" lesion [12]. The exam didn't show any bony area of increased uptake, confirming the ametabolic nature of the lesion (Fig. 4).

Taking into consideration all these findings, a radiological diagnosis of atypical vertebral haemangioma was possible.

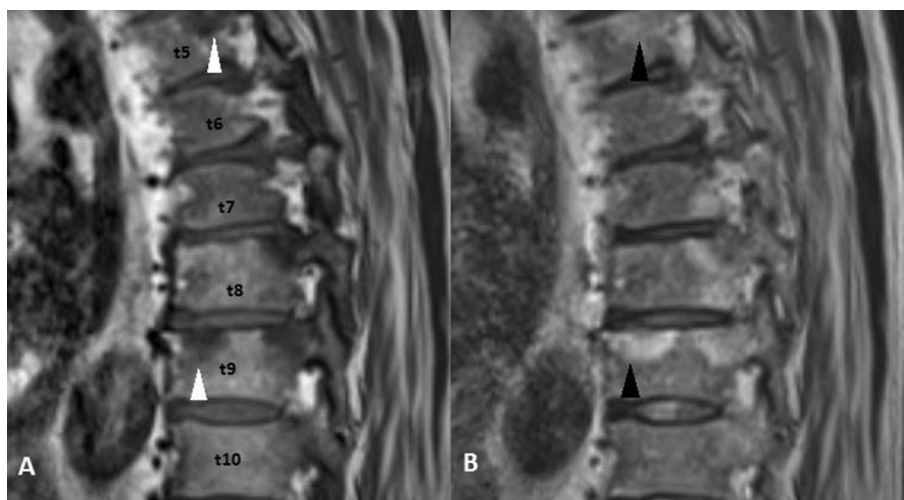
Although surgical treatment was suggested to the patient, he refused this possibility. So, we recommended him radiotherapy as a suitable option.

### 3. Discussion

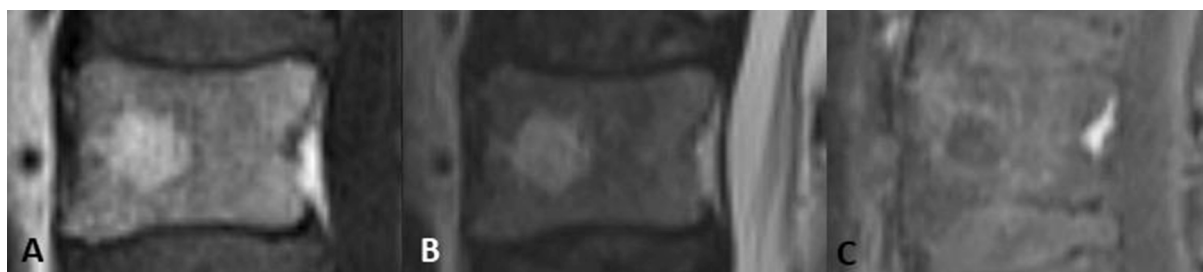
First described by Virchow in 1863 [3], Vertebral Haemangiomas (VHs) are benign neoplasms [4] involving the spine that arise from the endothelial cells that grow within marrow space in vertebral bone. Thus, they are able to penetrate and remodel surrounding bony trabeculae. Even if they are generally considered as neoplasms, it is hard to call them tumours, due to the lack of clear histopathological features. For this reason, some authors have referred to them as hamartomas or vascular malformations [5,6], even if at least one genetic correlated mutation has



**Fig. 1.** a-e: sagittal T1w (a,b), STIR-T2w (c), TSE-T2w (d), and axial TSE-T2w (e) MRI images of aggressive VH of the T3 and T4 vertebrae. Signal alteration of the spongy bone of T3-T4 vertebral bodies with superior endplates fractures and bone marrow oedema. Lesion appears to be variably hyperintense, particularly in T2w images (c, d). Post-contrast T1w MRI shows a slightly dishomogeneous enhancement, more valuable in the posterior portion extending into the epidural space (b). An hypointense hemosiderin nucleus may be appreciated in the posterior side of T3 vertebral body (c, arrow). The lesion extends posteriorly in epidural space, involving especially the right pedicle (e, asterisk). f-h: Sagittal (f) CT scan shows the typical vertical striation due to the thickened trabeculae; polka-dot sign in axial T3 (g) and T4 (h) CT scan can be easily recognized.



**Fig. 2.** Atypical MRI features of vertebral haemangiomas of the dorsal column. Sagittal MRI shows low-signal intensity due to low adipocyte content in T1-w images (a - white arrowhead) and hyperintensity in the corresponding T2-w images (b - black arrowhead).



**Fig. 3.** Typical MRI features of asymptomatic vertebral haemangioma of T11. In sagittal sequences the lesion appears well-circumscribed and hyperintense in T1w (a) and T2w (b) images; the corresponding sagittal contrast enhanced fat-saturated T1-w MRI shows the near-total reduction of the high signal intensity (c).

been detected, resulting in the fusion gene *EWSR1-NHATC1*, supporting the tumoral hypothesis [7].

VHs are considered the most common benign tumours of the spine [6]. Indeed, their incidence ranges from 10% to 15% of the general population [8], according to several anatomical and imaging studies, resulting especially common among adults (40–50 y.o.) [9] and rare among elderly and children [10], with a preference for female sex (estimated male to female ratio 2:3) [11].

Classically, in spine radiographs VH appears as an isolated well-defined lesion, involving only one vertebral body, although multiple lesions can be present as well. The base of implantation is usually localized in one of three anatomical sites: on the periosteal surface, within the cortex or within the medullary canal [3].

VH radiological findings just reflect their histological composition and are related to the amount of adipocytes, vessels and interstitial oedema.

Typical VHs diagnostic features include, on axial TC scan, the “polka-dot” sign, consisting in a spotted appearance due to the presence of sparse thickened hyperdense trabeculae, surrounded by hypodense stroma [12]. On sagittal and coronal CT scan, instead, the “corduroy cloth” results to be pathognomonic. It consists in the demonstration of the same thickened hyperdense trabeculae, but presented in a vertical orientation and no more imaged in cross section [12]. On MRI as well [12], VHs show a specific radiological appearance, consisting in the presence of serpentine vascular channels with secondary reactive phenomena associated, including fat overgrowth and bone trabeculae thickening. These lesions appear hyperintense on T1-weighted MRI, due to the predominance of fatty tissue within the haemangioma, and on T2-weighted images too, due to increased water content [13]. For this reason, VHs are also slightly hyperintense on fluid-sensitive sequences

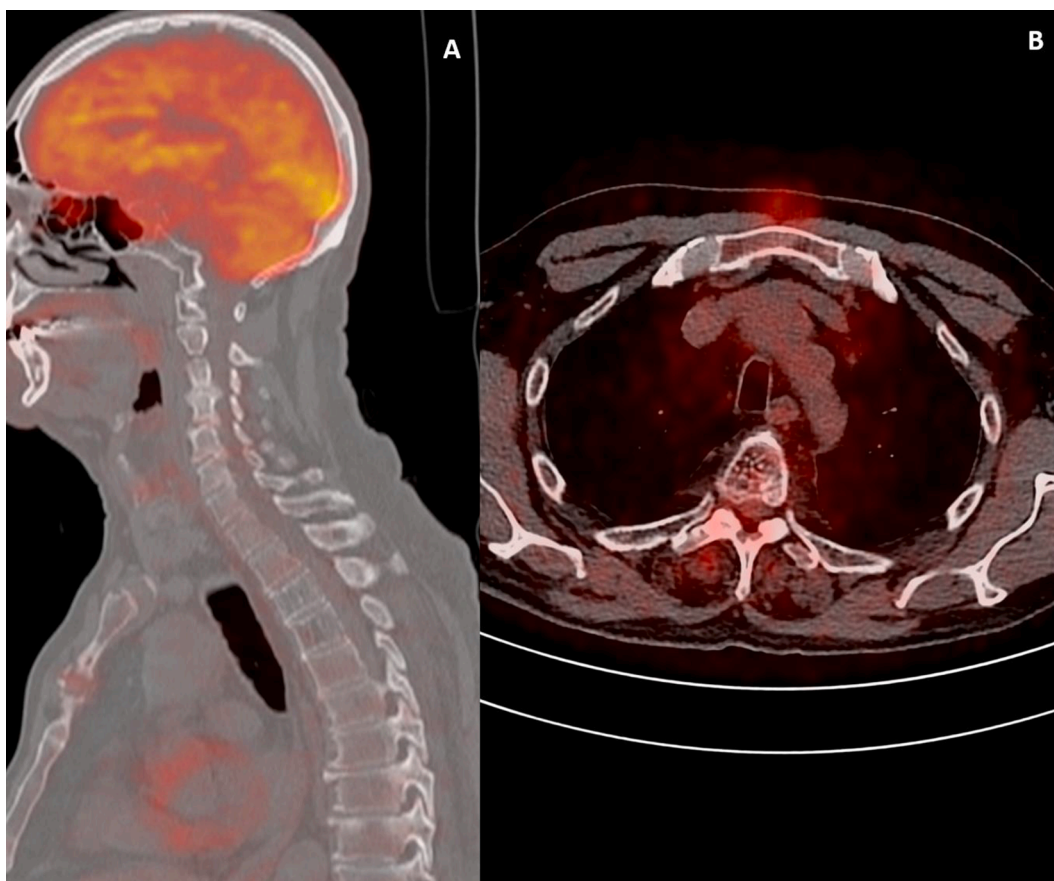
(i.e., short-tau inversion recovery or fat-saturated T2-weighted images), considering the rich presence of vascular components.

However, in some cases, VHs may show a pattern which moves away from this description, configuring themselves as atypical VHs. A VH is defined atypical when it exhibits one or more of these features: isohypointense signal, rather than hyperintense, on T1w sequences; very hyperintense signal on T2w ones; variable enhancement, after gadolinium administration; and better visualization on fat-suppressed images [12].

Finally, but most important, even if they usually preserve an easily recognisable pattern on CT, in atypical VHs the pathognomonic thickened trabeculae may be more difficult to see, presenting, instead, an irregular “honeycomb pattern”. Anyway, according to the literature [12], when coarsened trabeculae are found on CT, they result crucial to guide the correct diagnosis of atypical VHs. All these radiological differences are explainable by the major vascular contribution, at the expense of a less fatty one, to the lesional mass composition.

Regardless of the radiological aspects, typical and atypical VHs, both seldom cause clinical symptoms, without needing for treatment or even surveillance. Whether a VH remains asymptomatic or not, it depends on the extent of growth and compression of adjacent neural elements related to lesion itself.

As reported in literature, pregnancy is an important risk factor in inducing VHs to become symptomatic, especially during first trimester of gestation [14], most commonly involving the thoracic spinal levels. Vena cava compression, applied by the enlarged uterus, and hormonal changes, seems to be the two main factors implied in the development of a rapid onset of symptoms from these normally asymptomatic lesions. The former causes venous obstruction and increased abdominal pressure, leading to blood flow volume expansion through the vertebral



**Fig. 4.** (a) Sagittal and (b) axial images at T4 level: A fluorodeoxyglucose (FDG) positron emission tomography (PET)-computed tomography (CT) scan performed to characterize the vertebral lesions at T3-T4 level which have the classic “salt and pepper” appearance at basal CT showed no increased uptake on bone scan. These finding is typical of vertebral hemangiomas, benign neoplasm with ametabolic pattern at FDG PET examination.

venous plexus and so enlargement of these lesions. The latter, instead, is thought to be able to produce structural changes within the vessel wall [15,16].

Around 0,9 to 1,2% of affected patients show clinical symptoms, which consist in back pain alone, in 55% of cases, while in the other 45% of cases, the VHs extension into the spinal canal can cause compression of the spinal cord and/or of the nerve roots leading to radiculopathy and myelopathy [17–21].

The main drawback of our case is that there is no pathological proof of the diagnosis. Nevertheless, it seems that after all imaging techniques the only possible diagnosis is aggressive haemangioma.

Aggressive VH consists in a very rare subset of vertebral haemangiomas that, despite being histologically benign, is able to progress and enlarge, causing disturbance of the blood flow and occasionally compressive fractures, even resulting in neural compression [8]. This type of haemangiomas are pathologically classified as Enneking Stage 3 lesion, by Enneking’s musculoskeletal neoplasms staging system [22].

In spite of sharing many common radiological presentation elements with VHs in general, aggressive vertebral haemangiomas also present some unique features. First of all, it is more frequent to find them involving thoracic vertebrae (T3 - T9) [1,23], rather than lumbar ones. Secondly, they more often involve the entire vertebral body, with extension to the posterior arch, pedicles and lamina. Finally, a consistent soft tissue mass that extend into the extradural space is associated to these lesions in the majority of cases, leading to spinal cord compression [15]. In rare cases, an extradural hematoma and compression fractures can be also detected, due to the lesion-induced bony structure subversion.

It is important to highlight that aggressive VHs may show atypical

radiological features, that make their diagnosis very complex. Indeed, various aggressive spinal tumours (such as solitary bone plasmacytoma, lymphoma, spinal chordoma, epithelioid hemangioblastoma and metastases) can mimic an aggressive VHs and should be considered in the differential diagnosis [24–28]. In these cases, CT-guided biopsy is indicated, giving the correct diagnosis in over 80% of all patients [29]. Nevertheless, it is also a possibility that biopsy can lead to false negative results, due to insufficient tumour tissue collection within the biopsy sample [30]. For this reason, we performed a PET that showed an ametabolic lesion, whereas CT showed typical honeycomb and corduroy cloth patterns, pathognomonic for VH.

There is no consensus regarding which is the best treatment modality for symptomatic vertebral haemangiomas. It is generally accepted that surgical intervention (laminectomy, corpectomy or partial tumour removal) is the first choice in patients with symptomatic VHs, presenting neurological deficits or rapid worsening of the neurological status. In fact, surgery consents to obtain spinal cord decompression and to stop symptoms progression. Nonetheless, the major drawback of surgical treatment is the high risk of spinal instability, which can lead to fusion and instrumentation needing, especially when it comes to VHs extended through multiple levels [19], as in our reported case. In addition, the high risk of unintentional damage of the deformed vessels, resulting in an intraoperative haemorrhage, has to be considered.

In the recent years, many strategies for treatment of symptomatic or aggressive VHs have been developed, including vertebroplasty [23,31], embolization or sclerotherapy or a combination of them. Radiation therapy [16,31–38] has produced good results and is considered an effective therapeutic option for symptomatic VHs by the 1930. Indeed, RT is helpful in reducing the lesion and in controlling the pathologic

vessels due to a direct and controlled induced vascular endothelial damage. On the other hand, radiation has no radiographically demonstrable effect on the involved bone tissue.

Although being generally reserved just for painful VHs, some authors have described RT as first line of treatment also for aggressive VHs, especially for the ones causing neurological deficit, actually reporting a satisfactory percentage of success (tab. 1). Far from advice RT as the first choice of treatment, however, we consider RT suitable in case of aggressive haemangiomas involving no more than two vertebral bodies, with a consistent soft-tissue mass, but a vertebral canal encroachment ratio <40%, causing mild or slow progressive neurological deficit, and for patients refusing to undergo surgery or displaying contraindications for surgery and other treatments. However, because of the rarity of this pathology and the lack of long-term follow-up reported in literature, the optimal treatment strategy is still controversial.

#### 4. Conclusion

Aggressive VHs are rare benign spinal neoplasm characterized by bony involvement and extraosseous extension. They have no metastatic potential and are not associated with mortality, but can cause transitory or irreversible neurological deficit due to spinal cord compression.

Because of the infrequency of the lesions, the absence of characteristic radiological findings can lead to a misdiagnosis.

In case of atypical MRI aspect, CT can be crucial in detecting the aggressive hemangioma peculiar features, so reaching a correct diagnosis.

#### Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

#### Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.inat.2020.100954>.

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