Editorial

Vertebral hemangiomas: common lesions with still many unknown aspects

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Histology of vertebral hemangiomas (VHs)

Histologically VHs are not considered vascular neoplasms, but rather hamartomas or malformation of the microcirculation. Basing on the predominant type of vascular channel they are classified in: capillary, cavernous, arterio-venous and venous malformations. Most of spinal epidural hemangiomas belongs to the cavernous type (1). Numerous terms have been introduced and different classification systems. Unfortunately, none of these has been accepted due to the lack of accepted histological criteria and limited correlation with clinical outcome. Vascular tumours of bone originate from endothelial cells resulting in variable expression of endothelial markers, but none of these markers is useful to discriminate between benign and malignant lesions. Verbeke and Bovée (2) proposed the classification of vascular tumours of bone according to the WHO classification scheme for soft tissue tumours dividing entities into benign, intermediate and malignant category. Hemangioma is the most commonly recognized benign lesion. Epithelioid hemangioma was better defined over the past few years. Based on its locally aggressive behaviour and occurrence of lymph node metastases, its classification within the intermediate category could be considered. Angiosarcoma is the only accepted term for high-grade malignant vascular tumour of bone and epithelioid hemangioendothelioma is the only accepted low-grade malignant vascular tumour of bone. It is still unclear whether other low grade malignant vascular tumours of bone (e.g., hemangioendothelioma) truly exist. Molecular/genetic studies of vascular tumours of bone which might support the proposed classification are very sparse.

Pregnancy and VHs

Some authors report pregnancy as determining physiologic changes able to induce a rapid onset of symptoms from normally asymptomatic VHs. By the seventh month of gestation, the gravid uterus begins to compress the vena cava causing obstruction or functional closure. Venous obstruction and increased intra-abdominal pressure cause redistribution and increased blood flow volume through the vertebral venous plexus, resulting in the expansion and growth of previously existing VHs. Perhaps the hormonal changes taking place during pregnancy have been considered promoting a growth effect on an already existing VH, mainly through structural changes within the vessel wall (3). Maternal progesterone may increase the venous distension. The endothelial growth-effect of oestrogen may contribute to an increased size of a pre-existing VH. Relaxin may have an effect on the vessel walls (4).

Radiological appearance

VHs are frequent, incidental findings on spinal CT and MRI. When their imaging appearance is "typical" they show coarsened vertical trabeculae on radiographic and CT images and hyper-intensity on T1- and T2-weighted MRI. Nonetheless VHs might also display an "atypical" appearance because of their histological features composed by fat, vessels and interstitial oedema (5). The hyperintense areas represent fat, while the hypointense areas represent flow voids (6). It has been illustrated that aggressive VHs usually produce a low signal on T1-weighted images and a high signal on T2 (7,8). Low signal intensity on T1-weighted MRI indicates a hypervascular lesion with the potential to compress the spinal cord. Aggressive VHs can present a quickly growing with extension beyond the vertebral body invading the paravertebral or epidural space

with possible compression of spinal cord or nerve roots. The vertebral endplates are usually preserved, but the extension into disc spaces or neighboring ribs has been described (9).

Treatment proposals

Surgery

Dang *et al.* (6) affirmed that decompression is recommended in cases with rapid progressive or severe myelopathy and they recommend embolization of feeding vessel as a pre-surgical adjuvant treatment with the aim to reduce the intraoperative bleeding. The aim of surgery includes bony decompression and excision of the soft tissue components of the tumour that compress the neural elements. Different surgical techniques have been reported: laminectomy, corpectomy, spondylectomy and total en bloc spondylectomy.

Radiotherapy

Rades *et al.* (10) considered radiotherapy as the most common treatment for painful lesions. Heyd *et al.* (11) affirmed that radiotherapy is easy and effective, surgery is recommended if radiotherapy misses in 3 months. They reported the complete recovering of neurological symptoms after radiotherapy in 79% of their cases. The optimal dose, fraction protocol and time period necessary for a therapeutic effect of radiotherapy are controversial. The problem of the dose-response relationship for painful VHs is widely discussed. Previously the most used total dose was 20–24 Gy in 2 Gy fractions. In recent publications, an increasing of the total dose has been highlighted. Miszczyk *et al.* (12) described the superiority of a total dose >34 Gy over a total dose <34 Gy. The effective dosage was 30–40 Gy over a 3–4 weeks' period with a rare rate of associated complications such as radionecrosis, radiation-induced myelitis and secondary malignancy. Nevertheless, some authors report the risk of vertebral fracture after radiotherapy. Jiang *et al.* (13) reported the use of vertebroplasty to reduce this risk.

Embolization

Multiple studies have demonstrated safe and effective results in preoperative embolization of VHs. Goyal *et al.* (14) evaluated the therapeutic efficacy of alcohol ablation for treating VHs. 14 patients were treated by injection of absolute alcohol into the lesion via the percutaneous trans-pedicular route under CT guidance. On 14 patients, three complications were reported: 1 recurrence, 1 abscess due to intracanal contrast leaking during the test injection, 1 vertebral collapse after the procedure.

Another complication was reported by Yadav *et al.* (15) reporting a case of cardiorespiratory reflex with bradycardia, hypotension and temporary apnea after intravenous ethanol injections in the right atrium.

Smith *et al.* (16) demonstrated the safety and effectiveness of trans-arterial embolization of symptomatic VHs using polyvinyl alcohol in 8 cases. In 2013, Yao *et al.* (17) described 4 cases of trans-pedicular n-butyl cyanoacrylate (NBCA) embolization of VHs as a preoperative adjunct, facilitating the resection of these high vascular tumours. Hurley *et al.* (18) were the first to document the use of onyx trans-arterially for the treatment of 2 cases of aggressive VHs. In onyx embolization, the advantages include a more controlled injection due to gradual precipitation in a centripetal fashion, allowing a more accurate injection and the penetration in very small vessels.

Vertebroplasty or kyphoplasty

Jankowski *et al.* (19) considered patients with pathologic vertebral fractures and persistent pain eligible for percutaneous vertebroplasty or kyphoplasty but they stated that vertebroplasty is a contraindication if VH expands into the spinal canal. In kyphoplasty, in fact, the risk of major complications as the cement leakage is present. Fourney *et al.* (20) proposed the intraoperative vertebroplasty following laminectomy to achieve direct visualization during the procedure to reduce this risk.

Proposed algorithm of treatment

Dang *et al.* (6) proposed an algorithm for the diagnosis and treatment of aggressive VHs. In their study radiotherapy was proposed for symptomatic or extensive VH and surgery for patients affected by progressive neurological deficit. We reported (21) a case of vertebral fracture following radiotherapy as primary treatment for a large thoracic VH and we proposed a new algorithm of treatment basing on the size of VHs suggesting surgery for wide or symptomatic lesions and radiotherapy for wide asymptomatic ones.

Conclusions

Nowadays we still have concerns about the management of wide and asymptomatic VHs, is it indicated any treatment

or just a radiological follow-up?

We conclude that VHs are common lesions with still many unknown aspects. There is no universal consent regarding the management and treatment of these lesions, perhaps all the proposed treatments present some pitfalls.

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Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

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