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Pain pattern in multiple vertebral hemangiomas involving non-adjacent levels: report of two cases

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Abstract Hemangioma of the bone is a benign tumor usually involving the spine and the bones of the skull and pelvis. It may be either a single lesion or part of a generalized multifocal disease. Multiple lesions involving non-adjacent vertebrae are rare. Two cases of multiple vertebral hemangiomas at non-adjacent levels with different pain patterns are presented at various stages of follow-up in order to emphasize the fact that multiple vertebral hemangiomas may present with different clinical characteristics over a long period of time. The change in the location and pattern of the initially presented pain in both patients suggested the possibility of multiple level involvement. In-

vestigation revealed multiple hemangiomas involving three non-adjacent vertebrae in the first patient and four in the second. We stress the fact that the existence of multiple non-adjacent lesions may remain undiagnosed for a considerable period of time and may be responsible for even longer-term recurrent episodes of pain. Multifocal location of back pain in patients with a known vertebral hemangioma may be considered a relative indication for the presence of multiple non-adjacent level lesions.

Key words Adult · Case report · Spinal neoplasms · Hemangioma · Spine

Introduction

Hemangioma is a benign tumor consisting of blood vessels. There are two types: generalized and localized. In the generalized type there is involvement of multiple organs such as the liver, skin and bone [7]. Localized hemangiomas are the most common. In the skeleton, the most common site is the spine, followed by the skull and pelvis. Hemangiomas are often asymptomatic and discovered accidentally, but they may cause back pain or spinal cord compression in some cases. Radiographic and CT evidence of the hemangioma are characteristic. Multiple lesions of the spine are rather uncommon. Management of the patient depends on the severity of the symptoms, and may be either conservative or surgical.

We present the cases of two patients with multiple hemangiomas of the spine involving multiple non-adjacent

vertebrae and we comment on the possible clinical significance of their presentation.

Case reports

Case 1

A 68-year-old man initially presented at the outpatient clinic because of persistent low back-pain. Radiographs revealed hemangiomas of the L3 and L4 vertebrae (Fig. 1). Conventional radiographs of the upper spine were normal. The patient was treated by conservative means. He was given non-steroidal anti-inflammatory drugs (NSAID) and a course of physiotherapy, and he was soon symptom free. At a follow up examination 1 year later, he was symptom free and was discharged. He was advised to attend annual follow-up examinations but did not.

The patient came back to the clinic 15 years later complaining of suffering acute back pain localized in the area of T8–T10 during the previous 3 months. Common pain-killers gave him no relief. On examination, there was no evidence of neurologic involvement,

Fig. 1 Case 1. Lateral radiograph of the lumbar spine. Typical radiographic appearance of hemangiomas (L3 and L4 vertebrae). Vertical striations in the vertebrae give the so-called “corduroy cloth” appearance

and the only finding was localized tenderness over the spinous processes of the T8 and T9 vertebrae. Radiographs revealed moderate osteoporosis with no evidence of any other pathology. He was given a course of NSAID and physiotherapy, with a little improvement.

The interesting feature of this clinical presentation was the alteration of the pain pattern. In contrast to the low back pain he had complained of, on initial presentation, the location of the pain 15 years later was in the lower thoracic region. Computed tomography (CT) showed a hemangioma of the T9 vertebra (Fig. 2) and the lesions of the L3 and L4 vertebrae (Fig. 3) that had been already diagnosed the first time. We continued conservative treatment. Two months later the patient was almost pain free and he has remained well for the past year. We arranged a follow-up examination for 1 year's time.

Case 2

The second patient, a 56-year-old woman, initially presented with an incidence of acute low back pain. She had no neurologic deficits, and the only clinical finding during the examination was moderate tenderness over the lumbar spine. Radiographs revealed minor pathology of the lumbar spine and, by chance, the presence

of a hemangioma of the T11 vertebra. Six months later she suffered another episode of acute pain, localized over the thoracic region of the spine. As in the previous case, the change in the pattern of the pain led us to suspect, among other possible causes of recurrent localized pain, a multi-level involvement. A CT examination revealed hemangiomas in the vertebral bodies of T10, T11, T8, and L1 (Fig. 4), and in the transverse processes of the T10 vertebra (Fig. 5). There was no evidence of spinal cord compression, and the width of the spinal canal appeared to be normal. We gave the patient a course of NSAID and physiotherapy and explained to her the natural history of the lesion. She visited the outpatient clinic again 6 months later, reporting minor symptoms and we intend to see her again in 1 year's time.

Discussion

Hemangiomas are often diagnosed accidentally. According to Junghanns [22], there was a prevalence of 10% of vertebral hemangiomas in 10,000 radiographs of the spine. Toepfer [41] reports a prevalence of 12% in 2154 spines. Junghanns [22], in an autopsy study of 409 spinal lesions, found 66.5% single and 32.8% multiple lesions. Cutaneous lesions at the same dorsal segment as the deep lesion are present in some cases [5]. The lesion consists of abnormal vessels and it causes bone resorption, producing a honeycomb appearance [30, 33, 43].

Conventional radiography and CT are the most valuable diagnostic tools, because of the characteristic appearance of vertebral hemangiomas [3, 18]. CT is the procedure of choice for patients with symptomatic hemangiomas, as it determines the extent of the lesion and the site of compression, if present, and it reveals lesions in other vertebrae that an X-ray may have failed to demonstrate [26, 32, 38]. Magnetic resonance imaging (MRI) is useful, especially in differentiating a hemangioma from other tumors; it is valuable in pre-operative planning and it can provide additional information regarding the aggressiveness of the lesion [14, 27]. Spinal angiography is a useful diagnostic tool, especially in pre-operative planning, since it can demonstrate both the vascularity of the lesion and the origin of the feeding vessels [1, 11, 17, 30].

Hemangiomas of the bone are usually single lesions. Multiple hemangiomas of the bone are more common in generalized diseases involving the spine, skull and pelvis as well as the liver and other organs [7, 23, 43]. Reports of multiple hemangiomas of the spine are rare in the literature [3, 23, 34]. Reeves [34] reports a case of hemangioma in the T1, T2 and T3 vertebrae, which was apparent on a simple radiograph. Blankstein et al. [3] reported a patient with a hemangioma of T10, which was diagnosed by plain radiographs. In this patient, CT revealed similar lesions in the T8 and T9 vertebrae. Blankstein believes that multiple hemangiomas in the spine may be much more common than previously believed, and that CT may help show this. Djindjian et al. [8] reviewed the role of various imaging modalities and options for therapeutic intervention and presented a case of multiple involvement with progressive neurologic involvement. More recently,

Fig.2 Case 1. Computed tomographic (CT) scan shows a hemangioma at the T9 vertebra

Fig.3 Case 1. CT scan shows a hemangioma at the L4 vertebra

Fig.4 Case 2. CT scan demonstrates the lowest location of the hemangioma at the level of the L1 vertebral body

Fig.5 Case 2. CT scan depicts hemangioma involvement of the vertebral body and the transverse process of the T10 vertebra

Bremnes et al. [4] reported on the role of radiotherapy in the treatment of symptomatic multiple-level vertebral hemangiomas. Kulshreshtha and Byrne [25] presented multiple primary hemangiomas of bone mimicking vertebral metastases, while Duprez et al. [9] reported multiple aggressive vertebral hemangiomas in an adolescent. Our review of the available literature has failed to locate any other reports of similar cases. Fox and Onorrio [14] reported six cases of multiple lesions (two to five affected vertebrae), but there is no information regarding pain pattern or alterations in clinical presentation between the various stages of follow-up. In addition, we found no report regarding the prognosis of the patient with multiple involvement. Finally, we found no other reports referring to multiple lesions in non-adjacent vertebrae. Blankstein et al. [3] suggest that the lesion in adjacent vertebrae is in fact a single tumor, expanding

through the intervertebral disks. Unfortunately, they present no evidence to support this. On the contrary, CT scans of our cases show normal disks.

Hemangiomas are usually asymptomatic, but in the spine they may become symptomatic because of the enlargement of the affected bony structures and the narrowing of the spinal canal. Sometimes the lesion may directly invade the epidural space. The lesion weakens the vertebrae, and this may lead to compression fractures. The incidence of symptomatic hemangiomas is unknown. Fox and Onorrio [14] reported 13 patients out of 58 with symptomatic lesions. Patients with hemangiomas of the spine may remain asymptomatic for many years [30]. Hemangiomas are often discovered accidentally during the diagnostic evaluation of neck or lumbar pain. In these patients, the cause of pain or neurologic deficit may be irrelevant to the hemangioma and rather due to spondylosis, disk herniation or other musculoskeletal disease [18]. It is therefore important for the clinician to exclude any other pathology before considering hemangioma as the cause of the patient's complaints. In both our patients, investigations and clinical examination failed to locate any other cause of pain, and since pain and tenderness were localized over the affected vertebrae, we surmised that the hemangiomas were responsible. The clinical significance

of both these cases was the change in pain characteristics. Both patients presented with localized pain over the affected region of the spine. But the painful spine segment was different at various stages of follow-up. There is no explanation for why the clinical manifestations of the disease, such as pain and tenderness, present in the lumbar or thoracic spine at different times in the course of the disease. We do not know why one spine segment with vertebral hemangiomas may not show any clinical manifestations for a long period of time, while another does. We do not know the reason for the recurrence of pain in a different spinal segment of the first patient 15 years later. The patient did not report any aggravating factor or any other cause that could easily explain this change. There were no major reported symptoms or complaints in the time between the two pain episodes. It seems to us that, provided there is no other cause of spinal pathology, when we are presented with a change in pain pattern and location of a known vertebral hemangioma, this may be indicative of a multiple-level lesion even in non-adjacent or at remote levels. Pain pattern modification in a known vertebral hemangioma in the absence of a neurologic deficit seems to suggest the possibility of multiple-level disease. The specific pain pattern depends on the affected spinal segment and is different in the lumbar or thoracic spine. If this is the case, we believe that a CT scan is the diagnostic procedure of choice, because it provides valuable information about possible spinal canal encroachment and the integrity of the posterior elements. Further modification of therapeutic management, conservative or surgical, can be based on this information.

Although our observations are derived from only two cases, we suggest that these clinical findings will provide a new perspective for the interpretation and management of this rare disorder.

Patients with symptomatic lesions may develop neurologic deficits or fractures, and therefore they must be closely observed [3]. According to the literature, intraosseous hemangioma with no involvement of the posterior elements of the spine is a low-risk lesion with a low rate of neurologic deterioration [14]. When the lesion is in the thoracic spine and there is posterior element involvement, the potential for fracture appearance or neurologic deficit is higher, especially in young females [35]. It has also been reported that asymptomatic lesions may become painful or develop a neurologic deficit during the last weeks of pregnancy [14, 28, 39]. We could not find any reference in the literature to multiple involvement and the potential of these lesions to produce pain or neurologic deficits, although we

believe that multiple lesions may increase the risk of pain, especially in elderly, osteoporotic patients.

The management of hemangioma depends on the severity of the symptoms. Observation, radiation or embolization [10, 12, 24, 31, 44] and surgical decompression [13, 16, 20, 24, 30, 32, 36, 37] are the various stages of management, and they are employed according to the patient's situation. Surgical decompression must be performed when there is a progressive neurologic decline [1, 30, 32]. The greatest risk is excessive blood loss during the resection of the lesion, or postoperative epidural hematoma [13, 16, 20, 24, 37]. To avoid this, radiation or embolization may be used preoperatively [2, 11].

Recent advances in the management of vertebral hemangiomas are the intralesional injection of absolute ethanol [19, 29] and percutaneous vertebroplasty [6, 21]. Vertebroplasty is an effective new radiologic procedure consisting of the percutaneous injection of a biomaterial, usually methyl methacrylate, into a lesion of a vertebral body. This technique allows marked or complete pain relief and results in bone strengthening in most cases. The principal indications for vertebroplasty are osteolytic metastasis and myeloma, painful or aggressive hemangioma, and osteoporotic vertebral collapse with debilitating pain that persists despite correct medical treatment.

Conclusions

We present two patients with multiple non-adjacent vertebral hemangiomas. This is a rather rare entity. Such patients may be or remain asymptomatic. Diagnosis of all involved levels is difficult in the first instance. Both our patients had lesions that had been overlooked in the initial examination. A thorough investigation is necessary in these patients in order to locate lesions that might be missed on plain radiographs. The main indication for this procedure is a significant change in the pain characteristics of vertebral hemangiomas during follow-up examinations. CT appears to be the investigation of choice. Although it was not the case in our two patients, there is the possibility of an involvement of the neural elements, meaning that missed lesions could cause future problems for the patients. Multiple symptomatic lesions must be treated according to the severity of their symptoms. Moderate pain can be treated by conservative means, as in our patients. We believe, however, that these patients must be closely observed, because multiple involvement may increase the risk of compression fractures and the development of neurologic deficits.

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