Clinical and radiological presentation of spinal epidural haemangiomas: clinical series in a tertiary care centre during a 10-year period

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Abstract

Purpose Haemangiomas are very frequent benign spinal tumours. However, pure epidural location is extremely rare. At present, only 52 cases have been reported in the literature during the last 10 years. We proposed to analyse clinical and radiological features of this rare entity treated in a tertiary care centre over the last 10 years.

Methods A study of a retrospective surgical series (2002–2012) was conducted. The clinic's electronic database was searched for “spinal” and/or “vertebral haemangiomas”, which were treated by surgery and/or vertebroplasty. Clinical, radiological and histopathological data were analysed.

Results In total, the series comprised 30 spinal haemangiomas. There were 6 epidural (20 %), 17 vertebral (57 %) and 7 intradural lesions (23 %). There were four men and two women, mean age 28.3 years, with epidural lesions. One patient presented with localised back pain only, two with radiculopathy and focal neurological deficit, two with radiculopathy only and one with isolated focal neurological deficit, respectively. The onset of symptoms was progressive in four cases over weeks to months and sudden in two cases. Preoperative MRI imaging revealed features of meningioma, neurinoma or metastasis.

Conclusion Epidural haemangiomas are extremely rare spinal lesions. They may mimic more common spinal

tumours clinically and radiologically. The usual treatment is gross total resection confirming the diagnosis histologically.

Keywords Diagnosis · Epidural haemangioma · Spine tumours · Surgical resection

Introduction

Haemangiomas are considered to be of either dysembryogenic or hamartous origin [1], developing throughout the whole body. In the central nervous system, they are observed within the brain or the spinal cord. Within the spine they may be localised frequently in the vertebrae or rarely in the epidural and intradural space [2], while frequently having a long history of growth. Especially, vertebral haemangiomas are often asymptomatic and frequently diagnosed as incidental findings on radiological examinations. Vertebral haemangiomas are common primary spine tumours [2]. They are benign vascular lesions of endothelial origin with an estimated incidence of 10–12 % in all patients at autopsy [3, 4]. Haemangiomas are histologically classified as capillary (50 %), cavernous (28 %) or mixed type (22 %) [5]. Vertebral haemangiomas may have soft tissue expansion [4]. However, pure epidural location (i.e. >90 % tumour volume) is extremely rare with only 50 cases reported in the last 10 years.

Here, a review of cases of spinal haemangioma operated in the Department of Neurosurgery, University Clinics Düsseldorf, Germany over the last 10 years with special focus on epidural haemangiomas is presented. The clinical presentations, including time of diagnosis, diagnostic features, treatment options and the prognosis of this rare entity, are evaluated. The presented series is supported by a

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thorough literature review of the English literature about epidural haemangiomas.

Materials and methods

A retrospective, single-centre series from the Department of Neurosurgery of Heinrich-Heine-University, Düsseldorf, Germany from 2002 to 2012 was analysed. The patients were identified by an electronic documentation system, when coded as spinal or vertebral haemangioma. Inclusion criteria for the study were a surgically resected epidural haemangioma which was histologically confirmed by pathology. No nonoperated cases were found. The review of literature was done via Pubmed search for 2002–2012.

Results

A database search of the diagnosis-documentation system and neuropathological archives of our clinics to identify all cases of spinal haemangioma found a total of 30 patients with symptomatic spinal haemangiomas over the last 10 years. The most common location was “vertebral body” (17 patients), of which three showed extension to the extradural space or the neuroforamina. The second most frequent location was “intramedullary” (7 patients). Six lesions were “epidural” with “epidural” being defined as more than 90 % of the tumour volume located in the epidural space. There was no “intradural extramedullary” or exophytic lesion. Histopathological diagnoses were either “capillary” (5 patients), “cavernous” (8) or “mixed type” (1), or without any histological confirmation (16). Those without histological confirmation were those with a pure “vertebral” location.

Because of the rarity of this lesion, the focus in this work was on patients with epidural haemangioma. There were four men and two women with a mean age of 28.3 years (range 9–47 years) at the time of surgery (Table 1). In four cases, the onset of symptoms was slowly

progressive over weeks to months, while two patients showed sudden onset. The clinical presentation may be classified into three groups: localised pain, radiculopathy and focal neurological deficit. One patient presented with localised pain only (lumbalgia), two patients showed radicular pain (dermatome Th12 and dermatome C8), one patient had isolated neurological deficits (gait disturbance and upper limb paraesthesia) and the remaining two patients presented with combined radicular pain and focal neurological deficits (numbness and motor weakness). In comparison, the clinical features of vertebral and intra-medullary haemangiomas were local pain and isolated neurological deficit, respectively. At operation, laminotomy was done in two cases, hemilaminectomy in two, fenestration in one and, due to difficult reachability, a corpectomy with cage and ventral stabilisation in one case. Only in one case, there was considerable blood loss. All patients had good improvements of their initial symptoms.

The search of the English literature revealed that epidural haemangiomas were rare (Table 2). In fact, during the last 10 years only 52 cases have been reported. Note that of our present series, three patients were reported as single case reports beforehand [6, 7]. There was a predominance of cavernous haemangioma (46 cases). Capillary epidural haemangiomas (2 cases) and mixed-type lesions (4 cases)—such as the presented case—were very rare. With regard to the nomenclature, we defined “epidural location” as more than 90 % of the lesion located in the epidural space. The distribution of epidural haemangiomas over the spinal levels showed a predominance of thoracic (43 cases), followed by cervical (10), lumbar (5) and sacral (4) location. Extension over more than one level is possible. Remarkably, about 16 % of the patients were admitted with acute to hyperacute epidural or intralesional bleeding of the haemangiomas (8 of 51; 1 not reported). The majority of patients reported in literature underwent surgery (49 patients, 1 conservative, 1 with radiosurgery and 1 without information) with laminectomy (11 patients), hemilaminectomy (10 patients) and laminotomy (7 patients). There was also one open biopsy, one fenestration and one far

Table 1 Summary of patients with epidural haemangioma in our clinic over the last 10 years

Age	Sex	Clinical presentation	Location and extension	Histological type	Treatment	Outcome
14	Female	Radicular pain	D12/L1 epidural and neuroforamen	Cavernous	Resection	Good recovery
37	Male	Gait disturbance and upper limb paraesthesia	C5 epidural	Cavernous	Resection	Moderate to good recovery
37	Male	Cervicobrachialgia	C7/D1 epidural	Capillary	Resection	Good recovery
47	Male	Lumbalgia	L1 epidural and pedicle and neuroforamen	Mixed type	Resection	Good recovery
27	Male	Pain, paraesthesia, numbness	S1/S2 epidural and neuroforamen	Cavernous	Resection	Good recovery
9	Female	Pain, paraesthesia, weakness	C7/D1 epidural and neuroforamen	Cavernous	Resection	Good recovery

lateral extraforaminal approach. Seven cases were reported with considerable blood loss during operation and one during biopsy, while partial resection was only reported in one case and no other intraoperative complications were mentioned. Recovery of initial neurological symptoms was good in most of the patients (45 patients, 2 with improvement of the initial symptoms, 6 not reported).

Illustrated clinical case

A 47-year-old male presented with chronic back pain. At the time of admission, there was neither radiculopathy nor any neurological deficit. MRI showed a well-defined intraspinal, epidural mass at the left neuroforamen of Th12/L1 with a widening of the foramen and erosion of the

pedicle (Fig. 1). Differential diagnoses included neuroinoma, meningioma, metastasis and haemangioblastoma.

The lesion was resected via a Th12 laminotomy. Intraoperatively, the tumour was highly vascularised; compression of the spinal cord and extension into the neuroforamen was observed. The cortical bone of the facet joint and pedicle showed abnormal consistency and were partially drilled. There was no infiltration of the nerve root or the thecal sac. Histopathological examination of the intraspinal mass showed dilated cavernous vessels with fibrotic walls and interjacent capillary vessels. Neither inflammatory nor malignant cells were observed. In conclusion, the intraspinal mass was histopathologically diagnosed as a mixed type of haemangioma (Fig. 2).

Table 2 Literature review of epidural haemangiomas since 2002

References	Year	Cases	Location	Treatment	Outcome	Histopathological type
[24]	2002	1	Thoracic	Surgery	Good recovery	Cavernous
[9]	2003	1	Thoracic	Surgery	n/a	Cavernous
[25]	2003	1	Thoracic	Surgery	n/a	Capillary
[26]	2003	1	Thoracic	Surgery	Good recovery	Cavernous
[27]	2004	2	Thoracic (2)	Surgery	Improvement (1) + n/a (1)	Cavernous
[28]	2004	1	Thoracic	Surgery	Good recovery	Cavernous; +Schwannoma
[29]	2005	1	Thoracolumbar	Conservative	Improvement	Cavernous
[30]	2005	1	Cervicothoracic	Surgery	Good recovery	Cavernous
[31]	2006	1	Thoracic	Surgery	Good recovery	Cavernous
[32]	2006	1	Cervical	Surgery	Good recovery	Cavernous
[33]	2007	3	Cervical (1); thoracic (1); sacral (1)	Surgery	Good recovery (3)	Cavernous
[34]	2007	9	Cervical (1); cervicothoracic (3); thoracic (5)	Surgery	Good recovery (9)	Cavernous
[35]	2007	1	Thoracic	n/a	n/a	Mixed type
[36]	2008	1	Thoracic	Surgery	Good recovery	Mixed type
[37]	2009	1	Thoracic	Surgery	Good recovery	Cavernous
[38]	2009	1	Thoracic	Surgery	Good recovery	Cavernous
[19]	2009	1	Thoracic	Radiotherapy post-surgery	Good recovery	Cavernous
[39]	2009	6	Thoracic (2); thoracolumbar (1); lumbar (1); sacral (2)	Surgery	Good recovery (6)	Cavernous
[7]	2010	2	Thoracolumbar (1); cervicothoracic (1)	Microsurgery	Good recovery (2)	Cavernous
[6]	2010	1	Sacral	Surgery	Good recovery	Cavernous
[40]	2010	3	Cervicothoracic (2); thoracic (1)	Surgery	Good recovery (2) + n/a (1)	Cavernous
[41]	2010	1	Thoracic	Surgery	Good recovery	Cavernous
[42]	2010	1	Thoracic	Surgery	Good recovery	Cavernous
[43]	2011	1	Thoracic	Surgery	n/a	Capillary
[18]	2012	9	Thoracic (8); lumbar (1)	Surgery	Good recovery	Cavernous (7); mixed type (2)

Fig. 1 MRI and CT scans. The *upper row* shows MRI-T1-sequences with gadolinium; note the enhancement of the lesion by contrast agent. The *lower row* shows bony erosion in the CT scans

Postoperative MRI of the lumbar spine showed complete resection of the haemangioma. Clinically, the patient was seen at 2 months and 1.5 years in the outpatient clinic: the neurological examination was normal and no residual back pain was reported.

Discussion

Spinal epidural haemangiomas are rare lesions representing 5–12 % of all spinal vascular abnormalities [2]. Most of them are solely located in the bone, with some of them expanding slightly into the paravertebral tissue, e.g. the neuroforamina. Malignant degeneration is unknown [8].

An epidural location, i.e. more than 90 % of the tumour volume being epidural, is extremely rare. In a tertiary care centre, we only encountered 6 cases over the last 10 years and a literature research identified another 52 cases. The capillary type was solely seen epidurally, while the cavernous type may present with bony involvement. There may be a relationship between the histopathological type and growth pattern, which could not yet be confirmed due to the small number of cases.

On comparing the clinical presentation of epidural and vertebral haemangiomas, there is a clear relationship between the location and extension of the lesion. At our centre, vertebral haemangiomas often presented with local pain (11 patients), while focal neurological deficits or radiculopathy was rare and due to an extraosseous extension (3 cases). Though only a small number of vertebral

Fig. 2 Histopathological examination (hematoxylin–eosin staining; magnification $\times 100$). Note the capillary component (*star*) and the cavernous component above (*arrow*)

haemangiomas have been reported, it does not indicate the real number, as many of them are incidental findings without any clinical relevance. In epidural haemangioma, sudden onset of pain (sometimes due to haemorrhage or thrombosis) and focal neurological symptoms are much more likely due to the proximity of neural structures. Even a case of sudden onset and consecutive death of the patient were reported [9]. In Aoyagi's series, 18 patients had a history of symptoms for less than 1 year [9]. This is similar to spinal metastases, where pain is followed by rapidly

Fig. 3 MR images of haemangioma, neurinoma and meningioma of the spinal canal. The *upper row* shows all three entities in T2WI with the haemangioma and neurinoma isointense and the meningioma iso-

to hypointense. The *lower row* exhibits a T1WI with contrast enhancement: all lesions are enhanced. The *white arrow* indicates the lesions

progressive neurological deficits [10]. Localised pain without any neurological deficit is often observed in meningiomas or neurinomas [11, 12]. Typically, radiculopathy or neurological deficits occur at a later stage. One major difficulty of epidural haemangiomas is their diagnostic features in MRI scans. Distinguishing them from meningioma or neurinoma, and also other entities like metastasis or hemangioblastoma, is difficult. We present a synopsis of their typical MR aspect as compared to meningiomas and neurinomas (Fig. 3): epidural haemangiomas often show an isointense signal on T1-weighted images and mostly hyperintense signalling in T2 images. Homogenous contrast enhancement is seen in both sequences as hint for acute haemorrhage or haematoma. Heterogeneous contrast enhancement is observed due to thrombosis. As detailed above, the location may vary from the medulla to the vertebra. Neurinomas also exhibit isointense signal in T1, while on T2-weighted images hyperintensity is observed. The enhancement is very heterogeneous. Similar to the haemangiomas, bone erosion and widening of the foramen may be observed. Their location is typically centred on the foraminal region with possible extensions (Fig. 3). Meningiomas tend to show iso- to hypointensity on T1W1, while T2W1 images exhibit some kind of hyperintensity, especially with the lack of

calcification (Fig. 3) [11, 13]. The enhancement is homogenous. A dural tail sign may be distinguished. The location is most often intraspinal.

Additional spinal imaging techniques like myelography or myelo-CT are less and less frequently used [11]. In some instances, especially for vertebral haemangiomas, angiography may improve the diagnostic accuracy [14]. However, epidural cavernous haemangiomas seem to be angiographically occult [9, 15]. Also, while for intradural capillary haemangiomas, feeding arteries and draining veins have been described [16], none were seen in proximal nerve root capillary haemangiomas [17]. So, the usefulness of angiography remains uncertain. A biopsy is not recommended due to the vascular character and bleeding risk.

Based on the presented series and the literature review, epidural haemangiomas are best managed by open surgery, especially when there are neurological symptoms or pain. This results in good outcome and high recovery rates (Table 1). Acute to hyperacute bleeding before operation (about 16 %) supports a surgical strategy because few intraoperative complications during open surgery have been reported (7 cases with considerable, but controllable blood loss). Piecemeal resection seemed to favour intraoperative bleeding in those cases [18]. First of all, intraoperative monitoring should be routinely used. In our

experience, epidural haemangiomas may be progressively shrunk by bipolar coagulation starting at the part not in contact with the thecal sac. This allows for circumferential preparation and progressive liberation from the dura. Major vessels are not encountered, but smaller feeders have to be coagulated and sharply dissected. Piecemeal resection has to be avoided. Non-stick bipolars are very useful. Theoretically, a cell-saver might be used in patients who do not tolerate even small blood loss (e.g. children), but was not necessary in the cases we had encountered. Radiation therapy has been used as adjuvant therapeutic strategy for partial resected haemangiomas, but without any reliable results so far. One case report used stereotactic radiosurgery for an incompletely resected epidural haemangioma. The reported outcome was good [19]. In contrast to epidural haemangiomas, the clinical management of vertebral haemangiomas is different (radiotherapy, arterial embolisation, vertebroplasty, open surgery and ethanol injection) [20–23]. On comparing Table 2, the outcome of patients with epidural haemangioma who underwent surgery was in 92 % good recovery. Malignant degeneration has never been reported.

Conclusion

Epidural haemangiomas are extremely rare, benign intraspinal tumours. They may present clinically as a malignant, or more frequently as benign, tumours with bleeding rates up to 16 %. Diagnostically they are difficult to distinguish, especially from neurinoma. The natural history is largely unknown. Surgical excision establishes the diagnosis and results in good outcomes. Despite the vascular character, preoperative angiography and/or embolisation seems unnecessary. The prognosis is good and the risk of recurrence appears to be low. Malignant transformation has never been reported.

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Conflict of interest None.

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