Eur Spine J (1997) 6:298–303

ORIGINAL ARTICLE

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Cavernous angiomas of the spinal district: surgical treatment of 11 patients

Received: 6 May 1996
Revised: 19 October 1996
Accepted: 7 January 1997

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Abstract Cavernous angiomas, also called cavernous malformations or cavernomas, are vascular hamartomas accounting for 3–16% of all angiomatous lesions of the spinal district. Although histologically identical, these vascular anomalies may exhibit different clinical behavior and radiological features, depending on their location, hinting at different managements and therapeutic approaches. The authors report 11 cases of symptomatic spinal cavernous angiomas diagnosed and surgically treated over the past 18 years. Age of patients ranged from 15–75 years; males outnumbered females. Three patients had vertebral cavernous malformations, secondarily invading the epidural space; two had pure epidural lesions; two patients had intradural extramedullary lesions, and four intramedullary lesions. Surgical removal was completely achieved in four patients with intramedullary lesions, in two with subdural extramedullary lesions, and in one with a pure epidural lesion. Subtotal excision of another one epidural and three vertebral cavernous angiomas was followed by radiotherapy. There was no morbidity related to surgery; the mean follow-up was 2 years. The outcome was excellent in two cases, good in six, and unchanged in the other three. The authors discuss the different modalities of treatment of these vascular lesions variously placed along the spine.

Key words Cavernous angioma · Cavernous malformation · Cavernoma · Spine · Surgical treatment

Introduction

Cavernous angiomas, also called cavernous malformations or cavernomas [3, 9–11, 23–25, 28], are vascular hamartomas that can occur throughout the central nervous system (CNS) [15, 27]. During the last decade their recognition as incidental or symptomatic CNS vascular anomalies has been facilitated by MRI [7, 25, 26]. However, in the spinal district cavernous angiomas remain uncommon lesions that may show different clinico-radiological pictures and prognoses, depending on their location [2, 11]. Owing to their different patterns of peculiar characteristics, spinal cavernous angiomas have been often classified in the neurosurgical literature as separate pathological entities, similar to cranial cavernous angiomas of calvaria, dural sinuses, and brain [4]. This separation is not justified, since all spinal cavernous angiomas are histologically identical [11], although, depending on the location, they may require different modalities of treatment [1–3, 5, 8, 10–14, 20].

We present a surgical series of 11 patients treated for symptomatic cavernous angiomas located in the vertebral, extra- and intradural spinal compartments.

Materials and methods

Eleven patients with symptomatic spinal cavernous angiomas underwent surgery at our institution between 1977 and 1995. Age

Table 1 Characteristics of 11 patients with spinal cavernous angiomas

Case no.	Age (years)	Sex	Lesion location	Clinical onset		Symptoms and signs (duration)
				Acute	Insidious	
1	39	M	Epidural C3–C4	+	–	Cervicalgia; tetraparesis; urinary retention (1 day)
2	75	M	Epidural T3–T4	–	+	Paraparesis; control of sphincters maintained (2 years)
3	15	F	Vertebral T8 with epidural extension	–	+	Paraparesis; sensory loss below T8 level; urinary retention (6 months)
4	53	M	Spinal cord T9–T10	+	–	Paraparesis; sensory loss below T10 level; paralysis of sphincters (3 days)
5	31	M	Subdural L2 (cauda equina)	–	+	Intermittent radicular symptoms (4 months)
6	35	M	Spinal cord T10	–	+	Weakness of inferior limbs; sensory deficits below T10 level (3 months)
7	49	M	Vertebral L2 with epidural extension	–	+	Syndrome of the cauda equina (5 months)
8	54	F	Subdural-extramedullary C2–C3	+	–	Severe headache with cervicalgia; no deficits (1 day)
9	70	F	Vertebral T10 with epidural extension	–	+	Paraparesis; sensory loss below T10 level; paralysis of sphincters (1 year)
10	50	F	Spinal cord T1	–	+	Syndrome of Brown-Séquard below T1 level (6 months)
11	48	M	Spinal cord C5	–	+	Left hemiparesthesias (4 months)

Table 2 Diagnostic investigations performed in 11 patients with spinal cavernous angiomas (*CSF* cerebro-spinal fluid; *SSEPs* somato-sensory evoked potentials)

Case no.	Investigations	Preoperative diagnosis
1	CSF examination, plain radiography, myelography, angiography	Epidural angiomatous lesion
2	CSF examination, plain radiography, myelography	Epidural tumor
3	Plain radiography, myelography, CT, MR	Vertebral angiomatous lesion
4	CT, MR, angiography	Spinal cord cavernous angioma
5	Plain radiography, CT, MR	Tumor of the cauda equina
6	Myelography, CT, MR, angiography	Spinal cord cavernous angioma
7	Plain radiography, CT, MR, angiography	Vertebral angiomatous lesion
8	CSF examination, MR, angiography	Subdural-extramedullary cavernous angioma
9	Plain radiography, CT, MR	Vertebral tumor
10	Plain radiography, CT, MR	Spinal cord cavernous angioma
11	MR, SSEPs	Spinal cord cavernous angioma

ranged from 15 to 75 years; the male : female ratio was 7 : 4. The lesions were located in the vertebral bodies with epidural extension in three cases; two were in the epidural space; two were placed in the subdural extramedullary space, and four were within the spinal cord (Table 1). Eight patients (73%) complained of a slowly progressive symptomatology, while three (27%) presented an acute clinical onset; spinal cord dysfunction was observed in the majority of patients. Symptoms and signs ranged in duration from 1 day to 2 years before admission (Table 1). Diagnosis was obtained with several radiological investigations and, in some cases, with cerebro-spinal fluid (CSF) examination; somato-sensory evoked potentials (SSEPs) were also employed in a patient with a spinal cord cavernoma during the pre-, peri- and postoperative course (Table 2). Among the diagnostic investigations, plain roentgenograms of the spine and myelography detected only vertebral and epidural lesions, better delineated by CT in their extra- or intrathecal extension. Angiography was performed in five patients, showing abnormal vascular supply only in two patients with epidural and vertebral lesions. MR imaging was performed in nine patients and proved to be the most sensitive tool in the detection of the lesions (Fig. 1). However, preoperative diagnosis of cavernous angioma was considered in only five patients of this series, while in the remainder an aspecific angiomatous growth, or a spinal tumor, was suspected (Table 2).

Surgical treatment

All the surgical procedures included laminectomy centered on the lesion level; intradural approach was performed for extra-and intramedullary cavernous angiomas (Table 3). Myelotomy with high magnification was performed for the removal of spinal cord le-

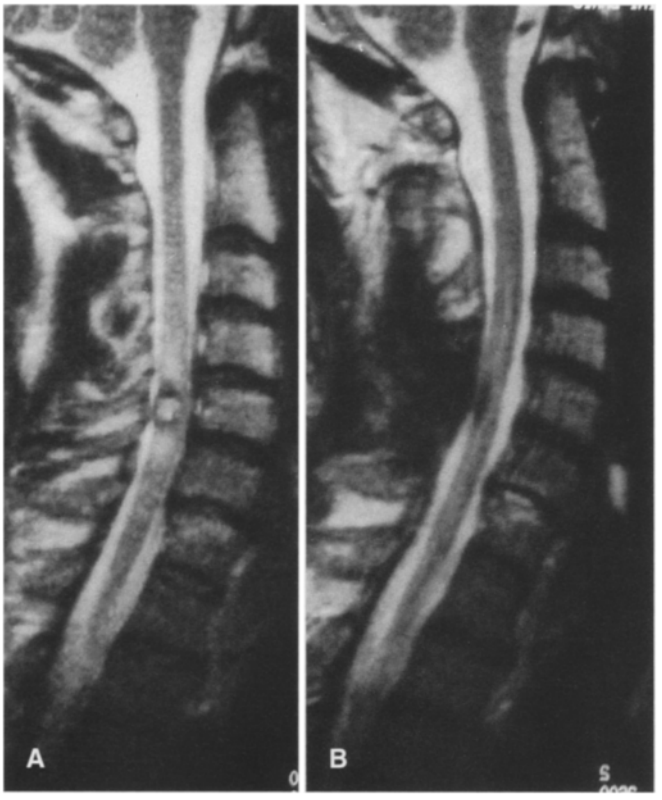

Fig. 1 **A** Preoperative and **B** postoperative T2-weighted MR images of a cervical intramedullary cavernous angioma

sions. Complete excision was obtained in seven patients, one with an epidural lesion, two with subdural extramedullary lesions, and four with intramedullary lesions. Incomplete removal was achieved in one patient with epidural cavernous angioma with intrathoracic extension, and in another three patients with vertebral lesions. At surgery, subdural extra- and intramedullary cavernous angiomas presented with typical mulberry-like growth appearance; epidural lesions were less firm in consistency than intrathecal lesions, and vertebral cavernous malformations presented as poorly defined, hemorrhagic bone lesions. Completely removed angiomas ranged in size from 1–3 cm in diameter. They were composed of vascular spaces made of collagenous walls, flattened by endothelial cells without mitotic activity; no neural tissue was found between the vascular spaces (Fig. 2). Pathological diagnosis of cavernous angioma was obtained in all cases in this series.

Mortality and morbidity from surgery were absent in all 11 patients. Postoperative radiotherapy was delivered in four patients with incomplete surgical excision of cavernous angiomas (Table 3).

Results

The outcome was excellent in two patients: one showed complete regression of preoperative paraparesis after subtotal removal and radiotherapy of an epidural thoracic cavernous angioma; the other remained neurologically intact after complete excision of a cervical subdural extramedullary lesion (Table 3). Good results were observed in six patients, including one with incomplete removal of a vertebral cavernous angioma. All showed improvement of preoperative clinical manifestations. Unchanged clinical picture after surgery was seen in three patients: two with incomplete excision of vertebral cavernous angiomas and one with a completely removed spinal cord lesion.

Table 3 Treatment and results in 11 patients with spinal cavernous angiomas

Case no.	Surgical procedure	Removal		Adjuvant therapy	Result	Follow-up (years)
		Complete	Incomplete			
1	Laminectomy C3–T1	+	–	Not done	Good	5
2	Laminectomy T3–T6	–	+	Radiotherapy (40 Gy)	Excellent	3
3	Laminectomy T7–T9	–	+	Radiotherapy (35 Gy)	Good	3
4	Laminectomy T9–T11, intradural approach and myelotomy	+	–	Not done	Unchanged	2
5	Laminectomy L1–L2, intradural approach	+	–	Not done	Good	2
6	Laminectomy T8–T10, intradural approach and myelotomy	+	–	Not done	Good	3
7	Laminectomy L1–L2	–	+	Radiotherapy (40 Gy)	Unchanged	2
8	Laminectomy C1–C4, intradural approach	+	–	Not done	Excellent	2
9	Laminectomy T9–T10	–	+	Radiotherapy (35 Gy)	Unchanged	1
10	Laminectomy C7–T1, intradural approach and myelotomy	+	–	Not done	Good	1
11	Laminectomy C4–C6, intradural approach and myelotomy	+	–	Not done	Good	1

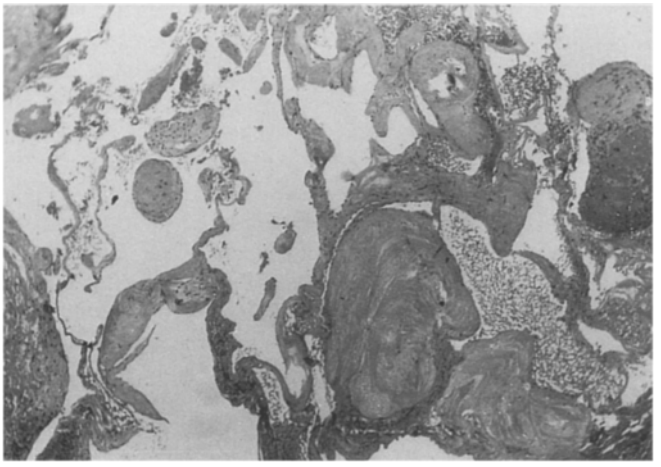

Fig. 2 Histologically, all the lesions in this series shared similar features. The angiomas were composed of vascular channels, lined by flat endothelial cells; no neural tissue was discovered between vascular spaces, some of which contained thrombi (hematoxylin and eosin, × 40)

Follow-up period ranged from 1 to 5 years (mean: 2 years). Recurrence of clinical symptoms in four patients with incomplete excision of cavernous malformations was not noted; however, follow-up exceeding 2 years was available only for two of these patients (Table 3).

Discussion

Spinal cavernous angiomas represent an inhomogeneous group of vascular lesions regarding their incidence, clinical picture, and radiological features [1–3, 5, 8, 10, 11, 20, 23, 28]. Vertebral and extradural lesions arise within mesodermally derived elements, frequently display rich vascularity on diagnostic investigations, and present as hemorrhagic growths at surgery [1, 5, 6, 8, 10–14, 16, 17, 22, 23, 26, 31]. In contrast, intradural and spinal cord cavernous angiomas arise within neuroectodermally derived tissues, are avascular lesions, and are usually surgically resected with relative ease [1–3, 7, 9, 11, 18–20, 23, 24, 28]. This peculiar variability of spinal cavernous angiomas has led to their classification in the neurosurgical literature as separate pathological entities, with extradural lesions being compared to vascular tumors, while intradural lesions are considered true hamartomas [4]. This separation is not histologically justified, since embriologically, immunohistochemically, and electronmicroscopically, vertebral, extra-, and intradural cavernous angiomas are identical [11]: they arise from the localized arrested development of blood vessel progenitors, are composed of dilated vascular spaces walled by thin endothelial cells, and there is no evidence that they may grow by mitotic activity [11, 15, 27]. As a consequence, all spinal cavernous angiomas are developmental vascular hamartomas, and should be considered as a single pathological entity [11]. Undoubtedly, they may require various modalities of treatment, depending on the spinal location [2].

Vertebral cavernous angiomas, also called hemangiomas, are considered a rare clinical entity, since they seldom cause neurological manifestations; they account for 2–12% of all vertebral growths found at necroscopy [5, 8, 10, 12, 15]. They occur mostly in the thoraco-lumbar tract of spine, more commonly in women after the fourth decade of life; familial form has not been reported in the literature [5, 8, 11]. Symptomatic lesions may present only with local pain or with neurological signs due to the epidural extension and the compression of the meningoneural structures, with or without fracture of the involved vertebral body [5, 8, 11]. Neurological impairment may also result from sudden hemorrhage into the epidural space, as reported sometimes in pregnant women [31]. Diagnosis of asymptomatic lesions is commonly an incidental finding on spinal radiological investigations. However, the features displayed on angiography, CT, or MR imaging may differ between asymptomatic lesions, which possess normal or slightly increased vascularity, and symptomatic lesions, which show moderate to intense hypervascularity [16, 26]. Due to their increased blood flow, symptomatic cavernous malformations may expand in vertebral bodies, producing bony erosion in some areas and reactive vertical trabeculation in others. On plain radiographs, vertebral bodies may display coarse vertical striations [12, 16], while on CT they present a "polka dot" appearance [8]. Angiography is essential in the diagnosis of vertebral cavernous malformations, and MR imaging may delineate their extraosseous components better than CT [28]. However, the radiological picture of these cavernous malformations is not as typical as that of other intra-axial lesions [7, 11, 25], and their angiographic and MR images may sometimes suggest a preoperative diagnosis of vertebral vascular tumor [2].

Therapy for vertebral cavernous angiomas causing only pain and without fracture should be conservative: radiotherapy of these lesions alone has been reported to improve the clinical picture [5, 8, 11, 12]. The treatment of vertebral lesions symptomatic from compression of neural structures should include their removal through laminectomy, with postoperative radiotherapy for residual lesions [2, 5, 8, 11]. Since vertebral cavernous malformations have a vascular supply, preoperative embolization may prevent blood loss during operative procedures [11]. Cavernous angiomas causing symptoms from vertebral fractures require a more complex therapeutic management, including lesionectomy, vertebrectomy, spinal stabilization, and postoperative radiotherapy [5, 8, 11, 12]. Clinical stabilization of patients is often considered the goal of this treatment.

Pure epidural cavernous angiomas are less common than vertebral lesions [10, 14, 29]; they have a predilection for thoracic spine, especially in adult males [6, 13, 17, 19, 22]. The clinical course of these cavernous mal-

formations is commonly slow, due to their progressive enlargement with spinal cord compression. However, a sudden increase in size of the lesions following thrombotic occlusion of their vascular spaces, or hemorrhage, may sometimes cause an acute clinical picture [21]. Radiological detection of epidural cavernous angiomas can be achieved with CT and MR imaging [10, 14], and sometimes with myelography [14, 21, 22, 29], although a preoperative diagnosis is usually difficult, since these lesions may have a hemorrhagic pattern like other vascular malformations, or may resemble epidural tumors [10]. Furthermore, though cavernous angiomas are mostly angiographically occult, epidural lesions sometimes exhibit abnormal vascular features on angiography [21], as reported in vertebral cavernous malformations [11].

The treatment of choice for epidural cavernous angiomas is their total removal through laminectomy; when their extension into or out of the vertebral canal makes this impossible, radiotherapy may be delivered as an adjuvant to surgery [11, 13, 17, 22].

About 3% of spinal cavernous angiomas are subdural in location, with extramedullary lesions remaining the most rare [1, 18, 19, 23, 24, 28]. Subdural extramedullary cavernous angiomas occur more commonly in men between the third and fifth decades of life [23], and are more common in the thoraco-lumbar tract [18, 19, 23, 24, 28]. Clinically, these lesions usually cause a progressive myelic dysfunction, or intermittent radicular symptoms [18, 19, 23, 28]; however, they may present with a sudden clinical picture from a subarachnoid hemorrhage [1], or with insidious symptoms of intracranial hypertension from an associated hydrocephalus [24]. Such an unusual clinical picture, in the absence of spinal cord dysfunction, may hint at a wrong diagnosis, even though the increased availability of MR imaging has recently facilitated the detection of subdural extramedullary cavernous malformations [1, 18, 24]. Total removal of these lesions can be safely performed through laminectomy and an intradural approach; microsurgical technique is useful to detach the lesion mass from the spinal cord or a nerve root. Clinical results after surgery are good in the majority of patients [1, 2, 18, 19, 23, 24, 28].

Spinal intramedullary cavernous angiomas are slightly less rare than extramedullary ones; they usually occur in adults, predominantly women [20]. The thoracic spinal cord is affected in more than half of the cases [2, 3, 9, 11, 20]. Clinically, spinal cord cavernous angiomas may present with a slow, progressive myelic dysfunction, or with acute clinical onset, usually due to hematomyelia. The neurological deterioration is sometimes characterized by varying degrees of recovery, although most patients exhibit a gradual clinical decline [20]. Prior to the introduction of MR imaging, the radiological diagnosis of spinal cord cavernous angiomas was difficult, since myelographs and angiographs often appeared normal, and CT examination of all the spine was not feasible. Furthermore, CT is less sensitive than MR imaging in discovering cavernous malformations, the appearance of which is typical on MR images for their pathognomonic intensity pattern [7, 25]. Preoperative SSEPs are useful to establish the clinical assessment of patients [30]. The goal of treatment of spinal cord cavernous angiomas is their total excision with microsurgical technique. Intraoperative SSEPs can guide surgical maneuvers and should be predictive of large spinal injuries, although they may fail to detect focal spinal cord lesions [30]. Cavernous angiomas are usually easily localized on the spinal cord surface by a bluish discoloration [3, 9, 20]: dissection can be started over this area, or directly on the mass lesion if approaching the surface. Ultrasonographic localization can be performed if lesions are deep and not visible. Careful dissection from gliotic, or hemorrhagic, spinal cord is needed to remove cavernous angiomas totally. Incomplete excision may result in symptomatic recurrence, requiring reoperation, since radiotherapy is not advisable in the treatment of spinal cord cavernous malformations [3, 20]. Surgical results are related to preoperative neurological status of the patients: best results have been reported in symptomatic patients who are operated early, before they develop severe or long-standing neurological deficits [2, 3, 9, 20].

As we noted, postoperative SSEPs may contribute to early and later functional follow-up.

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