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ORIGINAL ARTICLE

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Extradural spinal cavernous angiomas: report of seven cases

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Abstract The authors describe seven cases of extradural spinal cavernous angioma. Although cavernoma itself is not rare, the extradural spinal localization is uncommon and makes preoperative differential diagnosis difficult. Routine MRI investigation has aided neurosurgeons in evaluating the true incidence of these vascular malformations, which was understimated in the past. The data published so far have not entirely clarified the treatment of choice for these lesions. Considering their rarity in this site, their presenting symptoms and the difficulties involved in neuroradiological diagnosis, the authors discuss the role of surgery as the principal form of treatment and review the relevant literature. Seven patients (4 male, 3 female) were admitted to our Institute of Neurosurgery between 1992 and 2004, with a 5–6 month history (range=2–365 days) of low back pain or radicular pain, sometimes associated with paresthesia. All patients had a CT scan, as well as MRI with gadolinium when possible, which detected an extradural roundish lesion: differential diagnosis was very difficult, especially between neurinoma and cavernoma. Treatment was always surgical and resection of the lesion radical. Postoperatively, all patients presented complete regression of clinical symptoms. In all cases histological diagnosis was cavernous angioma. Postoperative MRI with gadolinium or CT scan with IV contrast, performed before discharge, confirmed radical removal of the vascular malformation in all cases. Our experience confirms that surgery should be the treatment of choice for these lesions, in view of both their tendency to bleed and their straightforward surgical removal.

Keywords Cavernous malformations · Epidural cavernous angioma · Spinal cord · Magnetic resonance imaging · Surgery

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Introduction

Spinal cavernous angiomas account for about 3–16% of all vascular spinal malformations [1, 5, 7, 12, 22, 24, 26, 34, 35, 38, 39, 42, 45, 46, 50]. They usually involve the vertebral body with rare pure epidural extension. Pure spinal extradural cavernomas are approximately 75 cases reported in the world literature accessible to us [2–4, 6, 8–11, 13–21, 23, 27–33, 36, 37, 40, 41, 43–45, 48–50]. The segment most frequently affected is the thoracic one, whereas cervical involvment is rare [1, 17].

The etiopathogenesis of this pathology resembles that of a congenital malformative lesion, and its tendency to progressively enlarge often leads to micro-and/or macro-hemorrhages which are the principal cause of clinical symptoms [49]. Growth of these lesions has been attributed to several mechanisms such as hemorrhage, dilation of the capillary bed, thrombotic-like processes, angioblastic proliferation or fusion of the vascular walls leading to the formation of real lacunae [35].

Although clinical evolution of these lesions is usually chronic and characterized by a protracted history of mild pain [16, 23], on occasion there may be a rapid deterioration of symptoms. The most frequent diagnostic tools are CT and MRI. MRI has made it possible to assess the true incidence of these malformations which was undoubtedly underestimated in the past years. However, preoperative differential diagnosis of this lesion, especially with neurinoma, continues to pose considerable difficulties. Diagnosis of cavernous angioma is made easily on pathologic

examination but awareness of their characteristics may facility the treatment of these lesions.

Materials and methods

Between February 1992 and November 2004, we treated seven patients with pure spinal extradural cavernoma. All these patients, four males and three females, presented a history of sporadic lumbar pain lasting between 2 days and 12 months, which did not respond to conventional anti-inflammatory medication and worsened in the days prior to admission. All these patients had a lumbo-sacral CT scan or MRI before and after surgery and were neurologically negative on discharge.

Case 1

A 34-year-old woman was referred to us in October 1995 with a 1-year history of low back pain, with nocturnal accentuation, associated with paresthesias of the left leg. A few days before admission she also complained of sciatic pain. On admission, neurological examination showed absence of the knee jerk reflex on the left. CT scan (Fig. 1) detected an expansive intraforaminal left extradural lesion at L3–L4. The lesion was roughly oval and produced a hypo-isodense signal with moderate and inhomogeneous contrast enhancement. A left L3–L4 interhemilaminectomy was performed. Demolition of the medial articular facet was necessary in order to follow the root of L4 into the conjugate foramen. The intracanalar portion of the root was not easy to identify because a lobulated mass, which presented a clear plane of cleavage with the root, was surrounded by a hemorrhagic component.

Histological diagnosis was cavernous angioma. Preoperative clinical symptoms regressed after surgery and gross total removal of the lesion was confirmed by a control lumbo-sacral MRI (Fig. 2).

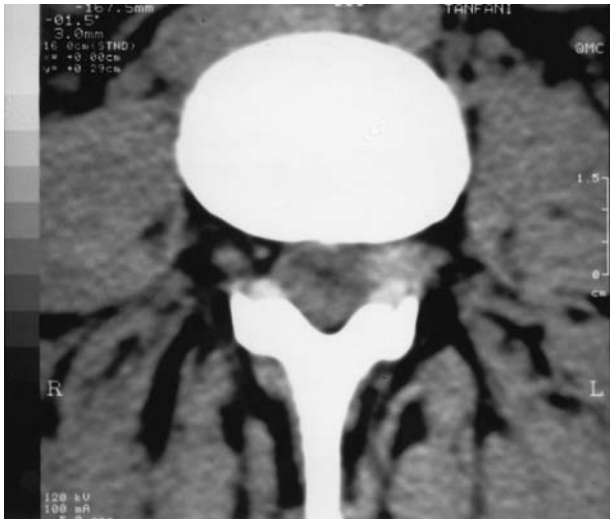

Fig. 1 CT scan with enhancement shows an expansive intraforaminal left extradural lesion at L3–L4 with a hypo-isodense signal with moderate and inhomogeneous contrast enhancement

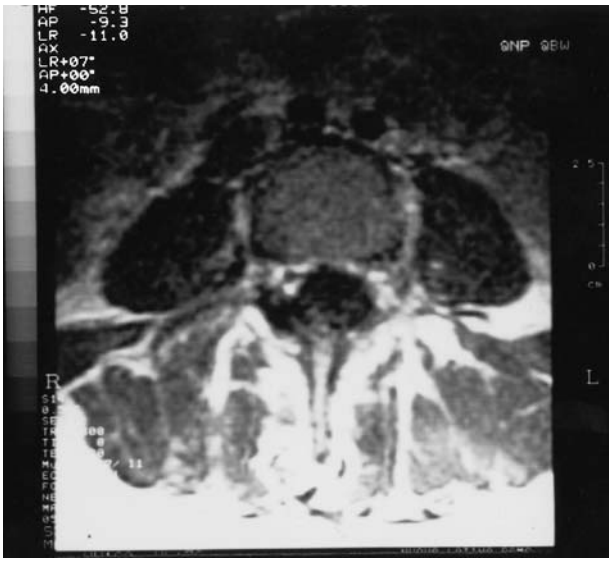

Fig. 2 MRI, T1 weighted images after Gadolinium administration. The examination, performed postoperatively, confirms the complete removal of the lesion

Case 2

A 66-year-old man was referred to us for a 5-month history of progressive left radicular pain resistant to conservative treatment. On admission, neurological examination was negative. MRI showed an expansive lesion in the extradural space at L5–S1. The lesion was roughly oval with a maximum diameter of 2 cm and adhered to the articular facet. The mass was hyperintense on both T1 and T2 weighted images and did not enhance after IV gadolinium administration. Surgery consisted of an left L5 interhemilaminectomy and partial facectomy. An oval, cystic neoformation in the lateral extradural space could be clearly seen compressing the left root of S1; its content were partly hematic, partly solid and brownish in colour. The lesion was totally removed after foraminectomy. Postoperatively, the clinical symptoms completely regressed. Lumbosacral MRI, performed on day 2 after operation, confirmed total removal of the lesion.

Case 3

A 72-year-old man, was referred to us in February 1996 for the onset of a right radicular pain 9 days earlier. On admission, neurological examination revealed a positive Lasegue sign at 45°.

A lumbo-sacral CT scan detected a roundish lesion in the extradural space at the level of the right L4–L5 lateral recess. This lesion was hypo-isointense and inhomogeneously hyperintense after IV contrast medium administration.

The patient underwent surgery. Via a right L4–L5 interhemilaminectomy, a purplish lesion presenting a clear plane of cleavage with the surrounding structures was totally removed. Histological diagnosis was cavernous angioma. After surgery, there was immediate and complete regression of the painful symptoms.

Case 4

A 68-year-old male patient, was admitted in May 1998 for a worsening of a 3-month history of right radicular pain. A lumbo-sacral CT scan with contrast medium showed a round, expansive lesion at L5–S1 level and a clear bony erosion of the lateral recess. The lesion was interpreted as a neurinoma.

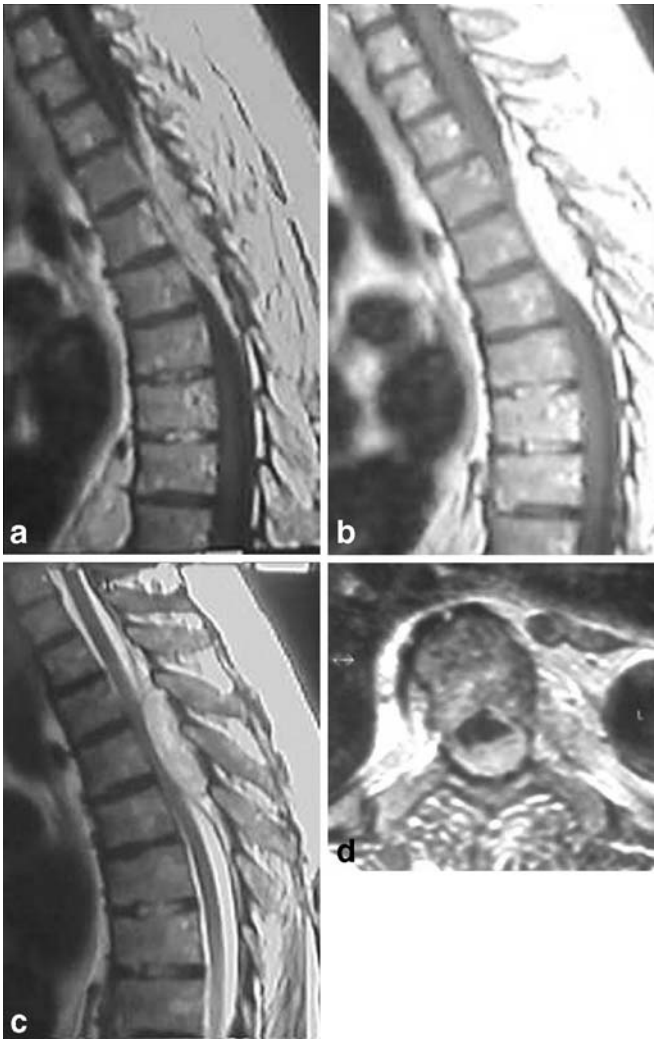

Fig. 3 MRI examination that shows the presence of an epidural cavernous angioma at D8–D10 in sagittal T1 weighted images (**a**), in sagittal T1 weighted images with gadolinium enhancement (**b**), in sagittal T2 weighted images (**c**) and in axial T1 weighted images with gadolinium enhancement (**d**)

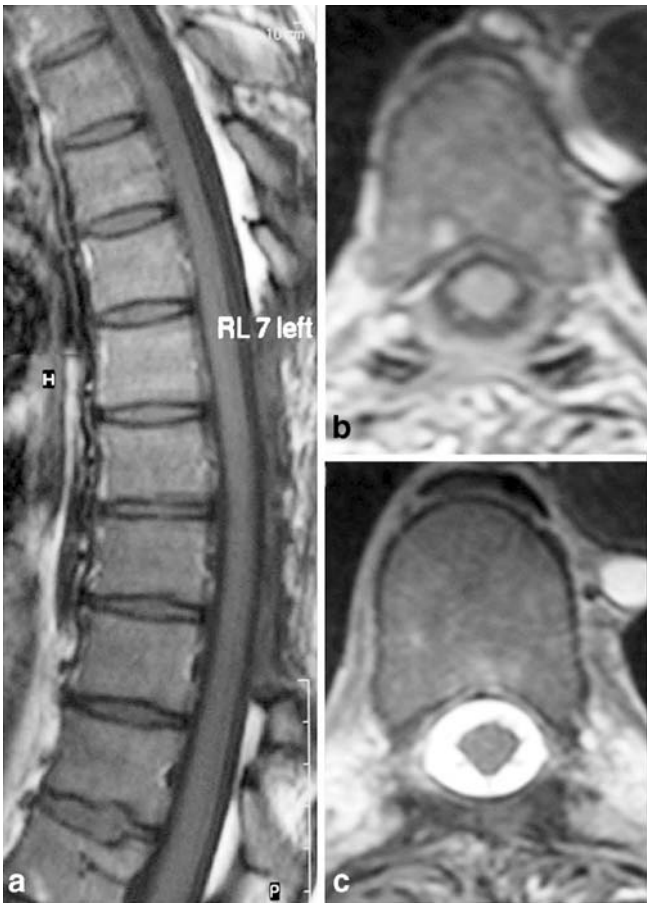

Fig. 4 MRI examination that shows the complete removal of the cavernous angioma at D8–D10 in sagittal T1 weighted images with gadolinium enhancement (**a**), in axial T1 weighted images with gadolinium enhancement (**b**) and in axial T2 weighted images (**c**)

At operation an L5–S1 hemilaminectomy was performed. A well-encapsulated, brownish-red mass suggesting cavernoma was found in the extradural space. The lesion was easily removed “en bloc”.

Postoperative CT scan with IV contrast confirmed total removal of the lesion. Histological diagnosis was cavernoma.

Case 5

A 80-year-old woman was referred to us in November 2001 with a 5-month history of right radicular pain. The MRI examination showed an extradural right lesion at L4–L5 level. The mass was hypointense on T1 weighted images and hyperintense on T2 weighted images and did not enhance after IV gadolinium administration. Via a right L4–L5 interhemilaminectomy, a purplish lesion presenting a clear plane of cleavage with the surrounding structures was totally removed. Postoperative CT scan with IV contrast confirmed total removal of the lesion. Histological diagnosis was cavernoma. Postoperatively, the clinical symptoms completely regressed.

Case 6

A 39-year-old woman was referred to us in November 2002 with a clinical history of 1 month of worsening paraparesis. An MRI examination performed with gadolinium highlighted the presence of an extradural mass at D8–D10 (Fig. 3a–d). A D8–D10 laminectomy was performed. A well-encapsulated, brownish-red mass suggesting cavernoma was found in the extradural space. The lesion was completely removed. Histological diagnosis was cavernoma. A MRI examination was performed after surgery and confirmed complete removal of the lesion (Fig. 4a–c).

Case 7

A 55-year-old man was referred to us in November 2004 with a history of 2 days of acute onset and worsening of right radicular pain with a significant deficit of plantar flexion of the right foot. There was no right ankle-jerk reflex. An MRI examination showed the presence of

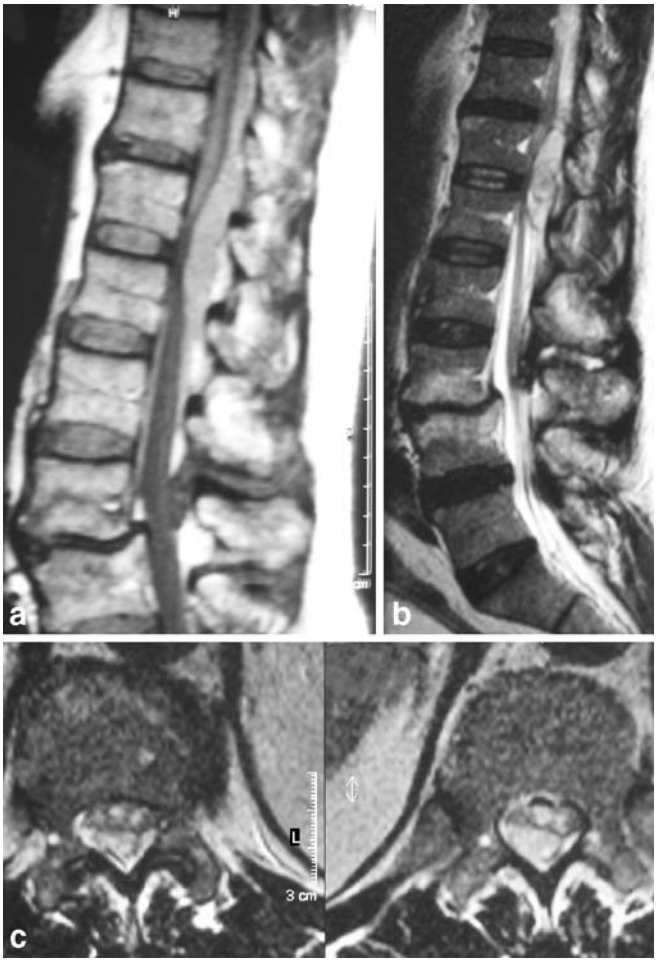

Fig. 5 MRI examination showing the presence of an epidural hematoma at D12–L1, due to the presence of a bleeding epidural cavernoma in sagittal T1 weighted images (**a**), in sagittal T2 weighted images (**b**) and in axial T2 weighted images (**c**)

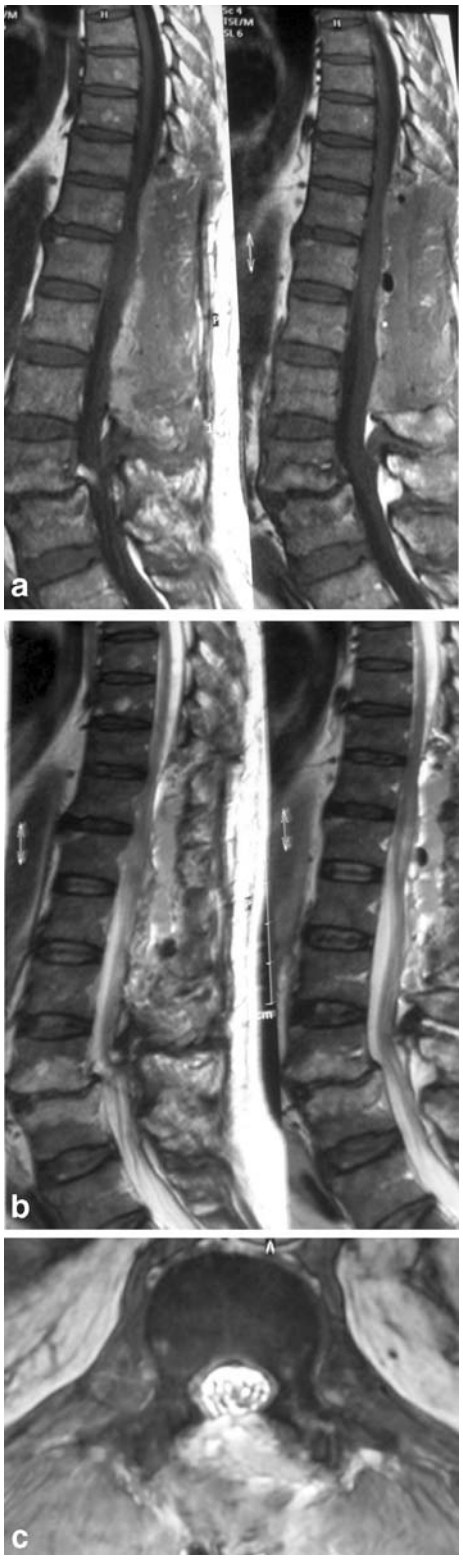

Fig. 6 Postoperative MRI examination showing complete removal of the epidural hematoma at D12–L1 in sagittal T1 weighted images (**a**), in sagittal T2 weighted images (**b**) and in axial T2 weighted images (**c**)

a hemorrhagic extradural mass at D12–L1. A D12–L1 laminectomy was performed, an epidural hematoma was found and completely removed. Histological diagnosis showed the presence of an epidural cavernoma. A MRI examination was performed after surgery and confirmed the complete removal of the lesion. The patient, after 1 week of physiotherapy, completely recovered from the preoperative deficit (Figs. 5 and 6).

Discussion

Although the histological characteristics of cavernomas are well established, a variety of names are used to describe it (cavernous angioma, hemangioma, venous angioma) [1, 47]. This subdivision has no histological significance, with the exception of angiolipoma, which many workers consider to be a distinct variant. In their study, Graziani and coworkers [19] affirmed that these denominations, including angiolipoma, describe a single pathology which varies only in terms of its adipose component, emphasizing that this variant principally depends on the site of the lesion.

However, it normally appears to the naked eye as a purple red, lobulated, well-circumscribed lesion which varies in size from a few millimeters to several centimeters. Under magnification, cavernoma is made up of dilated capillaries with a thin wall and a simple endothelial lamina with thin adventitia, indistinguishable from the lamina of teleangectasias [47]. In typical cases, the cavernoma does not contain parenchyma of the organ in which it is situated between the vascular channels and the elastic fibers are absent in the walls of the vascular spaces themselves. Very often, residues of previous hemorrhages are visible, with fibrous scar tissue, hemosiderin calciufication and even ossification, particularly in larger lesions. As far as the name "cavernoma" is concerned, Yasargil considers it to be "a purely descriptive term, indicating the gross microscopic appearance of lesions which are composed of sinusoidal vascular space" [47]: he maintains that the term "cavernous venous malformation" is preferable.

It may realistically be assumed that the incidence of spinal cavernomas, extradural or otherwise, has so far been underestimated. Technological advances in diagnostic imaging techniques have made it possible to detect pathologies previously considered rare [17, 46]. However, extradural spinal cavernoma is still an uncommon lesion of which approximately 75 cases have been reported in literature [2–4, 6, 8–11, 13–21, 23, 27–33, 36, 37, 40, 41, 43–45, 48–50]. Epidural cavernomas represent about 4% of all epidural tumors and 12% of all spinal cavernomas [7, 45, 49, 50]. Clinical onset usually occurs during the 3rd to 6th decades of life and does not present any sex prevalence: although the published data is not concordant on this point, no statistically significant case-series has yet established a greater incidence of one sex or the other [1, 15–17, 19, 22, 23, 46, 49]. In this context, it is interesting to note that some workers have suggested that these lesions may be hormone-dependent, which would explain why hemorrhage often occurs during pregnancy [35]. Moreover, when a spinal cavernoma is detected, a brain scan should also be performed to exclude other localizations; in fact, cavernous angiomas may be associated with similar lesions affecting other organs or within the CNS itself [45].

Clinical symptoms usually progress in a fairly insidious fashion and consist of sensory deficits, accompanied by radicular pain and/or paraparesis.

However, an acute or subacute onset is not rare and may be attributed to microhemorrhages or actual hematomas [1, 17, 19, 23] and the resulting motor deficits may be severe.

A peculiarity of this series is the fact that five of the seven patients had the cavernoma located in the lumbar region, although the thoracic segment is the most frequent extradural site [1, 17]. All of them were medically treated for the radicular pain. In fact, presurgical diagnosis of these lesions may be difficult. It is often complex to make a clinical and radiological differential diagnosis between the other causes of radicular pain, particularly with neurinoma, disc herniation, angiolipoma, ependymoma, chordoma, Ewing's sarcoma, lymphoma and meningioma [17, 49]. An indirect marker may be signs of bone erosion, frequent in neurinoma, Ewing's sarcoma and lymphoma, but very unusual in cavernoma [11, 49]. MRI is the imaging modality of choice in detecting cavernous angioma. The cavernoma MRI image is usually hypo-isointense before IV administration of contrast medium on T1 weighted images, usually because of calcifications and iron deposits, while it is hyperintense in angiolipoma and has a heterogeneous signal in vertebral chordoma and ependymoma [17]. On T2 weighted images, the cavernoma signal has a high intensity, just slightly less than that of cerebrospinal fluid, while it is isointense in meningioma [49]. On the other hand, contrast administration may produce enhancement which is generally homogeneous or slightly heterogeneous in cavernomas, more heterogeneous when the lesion is a neurinoma or an ependymoma [11, 17, 48, 49]. In the disc herniation, there is no enhancement. This description is more useful for didactic rather than practical purposes because the neuroradiological aspect of the lesion is susceptible to modifications, which complicate differential diagnosis, regardless of whether or not there are calcifications and/or recent or past hemorrhages. However, the obtaining of the correct preoperative diagnosis is important in order to plan and perform the best operation, also bacause intraoperative diagnosis of an epidural cavernoma is difficult, due to the limited exposure in most disc surgery and the epidural bleeding.

Conclusion

This case series re-emphasizes the need to consider cavernomas in the workup of the spinal extradural mass lesion. The treatment of choice of spinal extradural cavernous angiomas is always the total surgical removal. In fact, regarding radiotherapy, we agree with other studies [1, 7, 35, 45, 50] that it plays a secondary role and is mainly

useful in multiple lesions, those which are difficult to approach and in asymptomatic patients. The surgical treatment is preferable because total resection of the lesion is usually possible, due to the almost constant presence of a plane of cleavage. This is true even when the lesion is an occasional finding, particularly because the natural tendency of these lesions to hemorrhage may result in even severe deficits.

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