



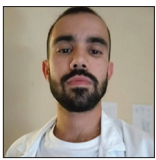
Case Report

Extradural hemorrhagic spinal cavernous angioma in a paucisymptomatic child: A rare case with review of the current literature

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ABSTRACT

Background: Cavernous angiomas, also referred to as cavernous hemangiomas or cavernomas (CMs), are vascular malformative benign neoplasms that may develop in any part of the central nervous system. Spinal CMs are uncommon (overall incidence rate of 0.04–0.05%). Pure epidural CMs account for 1–2% of all spinal CMs and 4% of all spinal epidural tumors. Diagnosis is extremely rare in the pediatric age. To the best of our knowledge, only 10 cases have been described so far. The treatment of choice is microsurgical resection.

Case Description: We describe here the rare case of a cervicothoracic hemorrhagic spinal epidural cavernoma in a paucisymptomatic, 8-year-old female Bangladeshi child. C7–T2 laminectomy with excision of a scarcely defined, capsulated dark red lesion was performed with good recovery.

Conclusion: Spinal epidural cavernomas are rare. Childhood presentation is even rarer. The reason could be found in a greater “compliance” and to a rarer occurrence of acute bleeding in children, thus resulting in a delayed diagnosis. Surgical excision is the gold standard of treatment.

Keywords: Pediatric spinal cavernous angioma, Pediatric spinal cavernous hemangioma, Pediatric spinal epidural cavernoma

INTRODUCTION

Cavernous angiomas, also known as cavernous hemangiomas or cavernomas (CMs), are vascular malformative lesions consisting of a dense bundle of dilated capillary-like channels lacking intervening neural parenchyma. They may be found anywhere in the central nervous system (CNS), mostly in the brain where they are associated with an increased risk in symptomatic intracerebral hemorrhage, seizures, and focal neurological deficits. Spinal CMs are rare, particularly pure epidural ones. These are also extremely rare in the pediatric population, with only few cases described in the literature.

We report here the case of a child affected by a cervicothoracic hemorrhagic spinal epidural cavernoma in whom emergency surgery at diagnosis was performed. We also reviewed the available literature (using *PubMed*, *Scopus*, and *Cochrane Library* databases) to delineate the clinicoradiological features of the disease and the most frequently applied treatment options.

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METHODS

Study design

The present paper consists of a case report and a systematic review of the literature conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement.

Eligibility criteria

All written papers about pediatric extradural spinal CMs reporting demographical and clinical data, diagnostic workflow, treatment protocol, histological findings, potential postoperative complications, and prognosis were considered for eligibility. Articles lacking single-patient information were excluded together with surgical and radiological technical notes, abstracts from scientific meetings, and unpublished reports. Articles discussing anything other than spinal epidural CMs were considered “not eligible,” along with those in which the disease was reported in an adult (>18 years).

Papers in languages other than English were considered for eligibility through the analysis of the abstract (only in cases in which it was available and English written).

Information sources and search strategy

The systematic review of the literature was conducted on three different online medical databases (PubMed, Scopus, and Cochrane Library), using as search terms “child,” “childhood,” “children,” “spine,” “spinal,” “extradural,” “epidural,” “cavernous malformation,” “cavernous angioma,” “cavernous hemangioma,” “cavernoma,” and combined with the Boolean operators “OR” and “AND” ([Title/Abstract]). The last search was conducted on December 31, 2021, and went back as far as data were available. The search strategy is summarized in [Figure 1].

Data collection process

Abstracts and full texts were independently screened by two authors (A.K.S. and M.G.), and any discordance was solved by consensus with a third senior author (M.P.).

CASE REPORT

An 8-year-old female Bangladeshi child was referred to the pediatric emergency department of our institution with a 7-day history of thoracic pain with progressive nontraumatic cervicodorsal discomfort and constipation in the past 48 h. A medical history was unremarkable except for a precocious puberty under evaluation. Physical examination on admission showed a marked pain-induced limitation in neck movements with tenderness on palpation of cervicodorsal muscles.

Abdominal distension was also detected. Neurological examination was otherwise negative. The child was admitted to the pediatric unit. Despite oral analgesia, the pain gradually increased. After 3 days of essentially normal investigations, a magnetic resonance imaging (MRI) of the cervicodorsal spine [Figure 2] revealed an extramedullary blood collection at C7–T2 level, extending craniocaudally for about 35 mm, within the left posterolateral portion of the vertebral canal, displacing the medullary cord to the right. An offshoot of this collection also extended to the left T1–T2 neural foramen. After gadolinium injection, no enhancement was appreciated. Within 1 h, the child was brought to the operating room where she underwent emergency C7–T2 laminectomy with excision of a scarcely defined, capsulated dark red lesion. Part of the capsule extended anteriorly to the spinal cord between the left C7 and C8 nerve roots. This portion of the capsule was removed all the same, following a partial foraminectomy with release and decompression of the roots. By the end of the procedure, the spinal cord appeared “aligned” and well decompressed. The patient was transferred to the ICU for immediate postoperative multiparameter monitoring. She came back to the ward on postoperative day 1 and was mobilized with soft collar on postoperative day 2. A check-up MRI performed on postoperative day 4 [Figure 3] ruled out further bleedings and demonstrated realignment of the medullary cord. The patient was discharged without neurological deficits on postoperative day 10. Histopathological examination of the specimens was compatible with the diagnosis of a hemorrhagic CM [Figure 4].

RESULTS

After duplication removal, 148 papers were screened for this systematic review. Forty-six papers were excluded for the following reasons according to our exclusion criteria: 38 since “not pertinent,” while eight because “not retrieved;” 102 full-text papers were, therefore, evaluated. Among these, 93 papers were excluded according to our inclusion criteria (76 since discussing “adult presentation,” 17 due to scarcity of data), and three “foreign” papers were similarly excluded lacking a complete, English written abstract. Six papers (reporting eight cases) were finally included and analyzed. Two studies (each reporting one case) were found through further search (references/Google) and were similarly included.

Findings related to the reported cases are shown in [Table 1] and thoroughly discussed in the following section.

DISCUSSION

Spinal epidural hematoma (SEH) is a relatively rare, but potentially disabling disease. First described by Jackson in 1869,^[15] it was successfully surgically treated for the

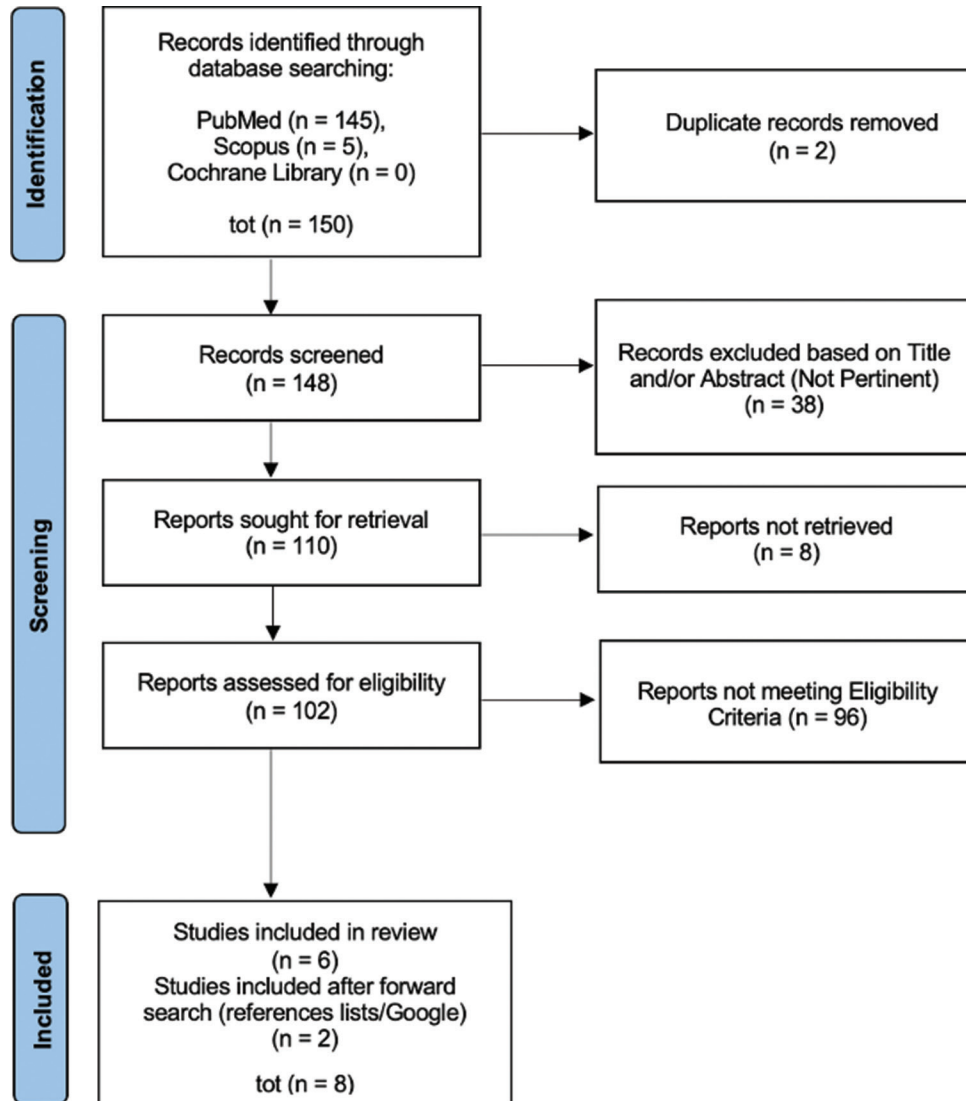


Figure 1: Pediatric extradural spinal cavernous angiomas: Preferred Reporting Items for Systematic Reviews and Meta-analyses flowchart.

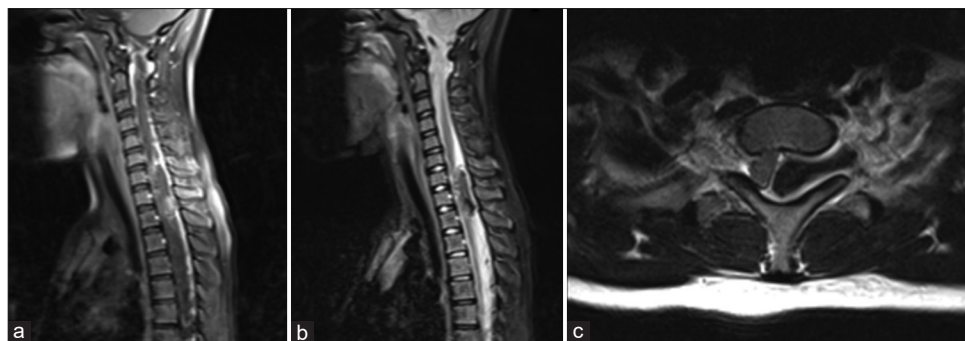


Figure 2: Preoperative MRI. (a) Sagittal section, C+ T1WI showing a nonenhanced iso-hyperintense epidural mass compressing the thecal sac from behind. Note the edema of the paravertebral soft tissues and the straightening of the cervical spine from antalgic contracture. (b) Sagittal section, T2WI showing hypointense signal of the lesion. (c) Axial section, T2WI showing T1–T2 foraminal extension of the lesion.

1st time by Jonas in 1911.^[16] In the largest series published on this topic (Domenicucci *et al.*),^[10] the etiology concerned

mainly iatrogenic factors (i.e., drugs, spinal puncture, or acupuncture^[7]), rather than noniatrogenic factors (i.e.,

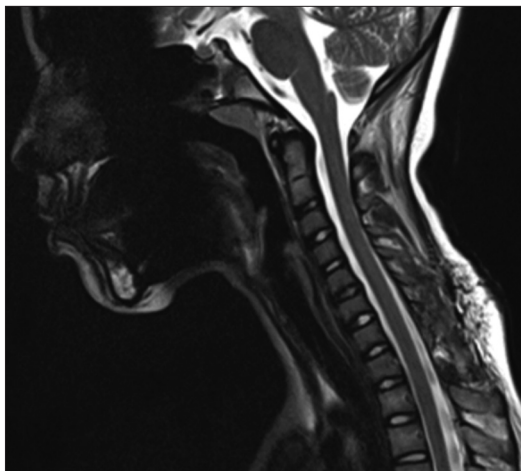


Figure 3: Postoperative MRI. Sagittal section, T2WI showing no further bleedings and a realigned medullary cord. Note the partial restoration in cervical lordosis.

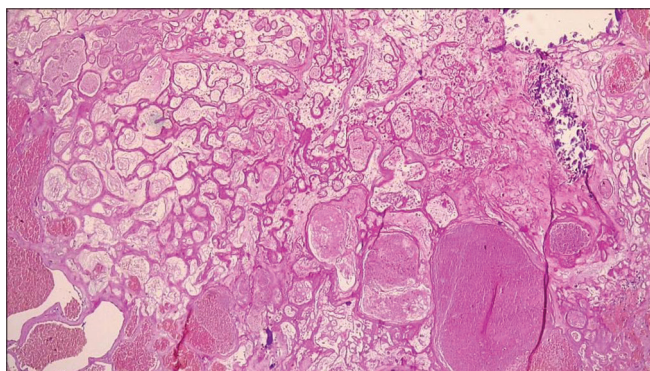


Figure 4: Histological findings. Note the architecture of the lesion, made up of multiple closely packed vascular channels lined with endothelium within a fibrous stroma, with signs of hemorrhage.

trauma, genetic or metabolic coagulopathy, and pregnancy). Spontaneous SEHs are an infrequent clinical entity with an incidence of 0.1/100 000/year. SEHs related to vascular malformations such as CMs are notably uncommon.^[4]

CMs are vascular malformative benign neoplasms that may affect any part of the CNS. They consist of closely packed endothelium-lined vascular channels within a collagenous stroma. Some of these vessels are partially or completely thrombosed and contain hemorrhage in different stages of evolution. Most of these lesions occurs in the brain, with 63–90% in the supratentorial compartment and 7.8–35.8% located infratentorially.^[13,22] Spinal CMs are rare with an overall incidence rate of 0.04–0.05%.^[28] They account for 3–16% of all spinal vascular malformations. Although they usually originate from the vertebral body, other locations are described (i.e., intramedullary, intradural-extramedullary, or in the epidural space).^[26] Pure epidural CMs are rarer,

accounting for 1–2% of all spinal CMs and 4% of all spinal epidural tumors.^[3,5] It has been hypothesized that anomalous vascular elements from the primordial peridural plexus may be the source of postnatal development of these lesions; an association with cutaneous vascular nevi has also been described.^[24]

Less than 100 cases have been reported.^[17]

Spinal CMs are frequently diagnosed in women between the third and the sixth decades. Diagnosis is extremely rare in the pediatric age (<18 years), though they are fundamentally congenital. To the best of our knowledge, only 10 cases have been described hitherto [Table 1].^[1,6,12,18,20,25,27,29] The reason for this could be attributed to a greater “compliance” and to a rarer occurrence of acute bleeding presentation in children, leading to a delayed diagnosis in adulthood.^[27] From the review of previously described cases and based on our personal experience, the following data were extrapolated.

Overall mean age was 8.9 years (range 1.7–14 years). Females were slightly more frequently affected than males (F/M ratio = 1.8:1). The lesions extended between 1 and 3 vertebrae in 8 cases (72.7%); the extension was greater in the remaining 3 cases (27.3%), with, in one case, 12 vertebrae involved. Cervicothoracic junction was the preferred site ($n = 6$, 54.5%). Pure thoracic location was found in 2 cases (18.2%), while thoracolumbar and sacral locations were “extraordinary.” This may be specific of the pediatric age since spinal CMs usually occur in the thoracic and lumbar spine in adulthood.^[5] Location of the neoplasms within the vertebral canal was “posterior” or “posterolateral” to the spinal cord in mine of the 11 analyzed cases (partial “anterior” extension of the lesion was, however, found in one case); this piece of information was not available for two patients. Extension through intervertebral foramina was noticed in 7 cases (63.6%). These findings were consistent with what literature reports concerning spinal CMs in adults.^[5]

Clinical status at the time of first observation and follow-up was assessed using a modified Neuro-Grade (NG) scale previously reported in other studies.^[8,9] This scale defines clinical status according to four grades: Grade 0, no deficits; 1, back pain, mild sensorimotor, and/or sphincteric deficits; 2, para- or tetraparesis; and 3, para- or tetraplegia. On admission, six patients (including ours, 54.5%) had an NG score of 1, 3 patients (27.3%) an NG of 2, while in 2 (18.2%) patients NG score was 3. Clinical course is, indeed, usually chronic with a prolonged history of pain both in children and adults. Acute or subacute onset with neurological deficits is not unusual in adulthood, depending on the location and biological behavior of the malformation.^[26] In childhood, however, it is – as formerly stated – rarer.^[27] Three of the 10 previously

Table 1: Pediatric extradural spinal cavernous angiomas. Our case and systematic review of literature.

Case No.	Author and year	Age (yrs)	Sex	Spinal level	Location	Clinical presentation	MR Imaging Characteristics	Type of surgery	Timing	Complications	Outcome	Risk factors for bleeding	Histological description
1	Zeygariadis, et al., 1998 ^[29]	13	M	T6-T7	Posterior/ (right) intraforaminal	Midthoracic back pain radiating into the right T6 dermatome (NG 1)	T1WI: iso, T2WI: hyper. C+ T1WI: heterogeneous enhancement	Right hemilaminectomy at T6-T7 and transversectomy	NA	None	NG0/F-up 6-month MRI negative	None	Well-circumscribed cavernous hemangioma composed of thin-walled sinusoidal vascular spaces of varying sizes lined with a single layer of endothelial cells. Surrounded by a fibrous capsule, which enclosed a small portion of a spinal ganglion. NA
2	Alvarez Sastre et al., 1999 ^[1]	1, 7	F	C7-T4	Right posterolateral	Progressive tetraparesis and established paraplegia 8 h before diagnosis. (NG3)	T1WI: hyper. No other information available.	T1-T4 plastic laminotomy	13 h	None	NG0 at 3 weeks	None	Multiple closely opposed vascular channels lined with endothelium without any neural tissue in-between. Blood vessels were separated by intervening hyalizing stroma.
3	Cho et al., 2006 ^[6]	1, 9	M	C5-T3	Posterior (with partial anterior extension)	Acute paraplegia (NG 3)	T1WI: iso/hyper, T2WI: hyper. C+T1WI: heterogeneous enhancement	C6-T3 laminectomy	NA	Postoperative epidural hematoma requiring reintervention	NG1 at 2 weeks	None	Multiple closely opposed vascular channels lined with endothelium without any neural tissue in-between. Blood vessels were separated by intervening hyalizing stroma.

(Contd...)

Table 1: (Continued).

Case No.	Author and year	Age (yrs)	Sex	Spinal level	Location	Clinical presentation	MR Imaging Characteristics	Type of surgery	Timing	Complications	Outcome	Risk factors for bleeding	Histological description
4	Feng et al., 2009 ^[12]	14	M	S1	Lateral/(right) intraforaminal	Right lower limb numbness, pain (NG1)	T1WI: hypo, T2WI: hyper, C+T1WI: homogeneous enhancement	NA	NA	None	NG0	NA	Many blood vessel cavities with large size and thin wall. Somewhat red blood cells, as well as somewhat chronic inflammatory cells. Local thrombosis.
5	Sarikaya-Seiwert et al., 2010 ^[27]	13	F	T12-L1	Posterolateral/(left) intraforaminal	Radicular pain radiating along the left T12 dermatome. 3-week history of back pain (NG1)	T1WI: iso, T2WI: hyper+dark rim. C+ T1WI: homogeneous enhancement	Left T12-L1 hemilaminectomy +electrophysiological monitoring of the somatosensory evoked potentials	NA	None	NG0	None	Large, blood-filled venous vessels with thin walls, located next to each other, surrounded by fibrous tissue.
6		9	F	C7-T1	Posterior/(right) intraforaminal	Acute severe neck pain. Mild right biceps and severe triceps weakness. Decreased sensation in right C6, C7, and C8 dermatomes (NG1)	T1WI: iso, T2WI: hyper+dark rim. C+ T1WI: homogeneous enhancement	Right C7-T1 hemilaminectomy + electrophysiological monitoring of the somatosensory evoked potentials	< 1 h	None	NG0	None	Several closely apposed, thin-walled, ectatic vascular channels.
7	Khalatbari et al., 2013 ^[18]	13	F	T6-T8	Posterior	Acute severe midthoracic back pain. Paraparesis. Urinary retention. (NG2)	T1WI: slightly hyper, T2WI: hyper. C+ T1WI: homogeneous enhancement	Bilateral T7 and partial T6 and T8 laminectomy	NA	None	NG1 at 3 months. No recurrence at 5-yr F-up	None	NA

(Contd....)

Table 1: (Continued).

Case No.	Author and year	Age (yrs)	Sex	Spinal level	Location	Clinical presentation	MR Imaging Characteristics	Type of surgery	Timing	Complications	Outcome	Risk factors for bleeding	Histological description
8	Mühmer et al., 2014 ^[20]	9	F	C7-T1	Foraminal extension. No other information available	Pain, paresthesia, lower limb weakness (NG2)	NA	NA	NA	None	Good recovery. No other information available	NA	NA
9		14	F	T12-L1	Foraminal extension. No other information available	Radicular pain (NG1)	NA	NA	NA	None	Good recovery. No other information available	NA	NA
10	Roman et al., 2015 ^[25]	1, 7	M	C7-T11	Right posterolateral	Progressive Paraparesis (NG2)	Heterogeneous on T1-weighted images as well as on T2-weighted images	Extensive plastic laminotomy	NA	None	NG1 (no mention of long-term F-up)	Congenital coagulopathy	Cavernous space lined with endothelial cells and median to large cavernous hemangioma vessel space filled with hemorrhagic substrate.
Ours	Scafa et al., 2021	8	F	C7-T2	Posterolateral/ (left) T1-T2 intraforaminal	7-week history of thoracic and cervicodorsal pain. Constipation. (NG1)	T1WI: iso/hyper, T2WI: heterogeneous hypo. C+ T1WI: no enhancement	C7-T2 laminectomy	<1 h	None	NG0	None	See Figure 4

h: Hour; hyper: Hyperintense; hypo: Hypointense; iso: Isointense; F: Female; F-up: Follow-up; M: Male; MRI: Magnetic resonance imaging; NA: Not available information; NG: Neuro-Grade scale*;
T1WI: T1-weighted image/C+T1WI: Contrast-enhanced T1-weighted image; T2WI: T2-weighted image; yr/yr: Year/years. * NG0: No deficits; NG1: Pain, mild sensorimotor, and/or sphincter deficits; NG2: Para- or tetraparesis; NG3: Para- or tetraplegia

described cases (30%) showed a truly acute debut of symptoms. Presentation was subacute in our patient who showed, among other things, a “singular” symptom, that is, chest pain, probably related to a “referred” pathogenetical mechanism (to the best of our knowledge, this is the first case reported in the literature).

Certain MRI features may be useful for the differential diagnosis. T1-weighted images usually show a homogeneous iso- or hypointense signal intensity (though hypersignal has also been described, like in cases 2, 3, 7, and in our case [Table 1]), while on T2-weighted images, the signal is high. Enhancement is typically homogeneous or slightly heterogeneous.^[19,29] The unique radiological aspects found in our case (i.e., hypointensity on T2-weighted images, absence of enhancement) were probably related to the presence of hemorrhage in two different stages of development, with methemoglobin and deoxyhemoglobin (“early subacute stage,” i.e., 3–7 days, and “acute stage,” i.e., around 12 h–2 days).

The treatment of choice for spinal CMs is microsurgical *en bloc* resection of the lesions.^[3] Surgery may be difficult due to the frequent formation of dense adhesions between the CM and the nerve roots. In some cases, dissection of the nerve roots (thought to be the origin of the neoplasm) is mandatory. Embolization has no role, and no adjuvant therapy is needed.^[25] “Complete” or hemi-laminectomy seems to be the most frequently performed approaches ($n = 6$, 54.5%). Laminoplasty has also been used ($n = 2$, 18.2%) with excellent results. Anyway, the poor amount of the cases reported does not allow us to recommend one of these treatment options above the others. In the pediatric age, a strong correlation between multilevel, complete, bilateral laminectomy, and postlaminectomy spinal deformities – mainly kyphosis – has been recognized, especially for the cervical and thoracic spine.^[2,23] In the past years, it was suggested that hemi-laminectomy or laminotomy with laminoplasty might reduce this complication; results are, however, still controversial.^[2,11,14,21,23] In our case, the need for an emergency treatment, combined to the difficulty in “dominating” intraoperative bleeding, led us to “adopt” cervicothoracic “complete” laminectomy, excluding other treatment modalities. Timing of surgery was reported only for two cases.^[1,27] We recommend going into surgery as soon as possible, though – of course – it is not always possible to guarantee optimal recovery (e.g., in case of “outdated,” established paraplegia). Neurophysiological monitoring (reported only in two of the analyzed cases^[27]) may be extremely useful in case of nonemergency surgery; further studies are, however, needed to confirm this potentially beneficial role. Only one patient experienced a complication requiring reintervention (new epidural

hemorrhage) in the early postoperative setting; in this case, excision had been incomplete due to intraoperative bleeding and partial ventral extension of the lesion.^[6] Postoperative outcomes, however, were globally good with an NG score of 0 in 6 cases (54.5%) and 1 in 3 cases (27.3%) (this piece of information was not fully available in two cases). Effective and well tolerated, surgery may, therefore, be considered as the gold standard for the treatment of this disease even in children.

CONCLUSION

Spinal epidural cavernomas are uncommon lesions. Diagnosis is rarely made in childhood, and it is important to rule them out even in case of chronic or subacute onset of spinal pain. Surgical excision is the gold standard of treatment.

STATEMENTS

Statement of ethics

This research was conducted ethically in accordance with the *World Medical Association Declaration of Helsinki*. Written informed consent was obtained from the parents of the patient for PUBLICATION OF THIS CASE REPORT AND ANY ACCOMPANYING IMAGES. Ethical approval was not required for this study in accordance with national guidelines.

Data availability statement

All data generated or analyzed during this study are included in this article. Further enquiries can be directed to the corresponding author.

Authors' contributions

A.K.S. conceived and designed the analysis, collected the data, and wrote the paper. M.G.I and M.G.II collected the data with the first author. M.P. critically revised the work.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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