



CASE REPORT

## Surgical intervention for a pediatric isolated intramedullary spinal aneurysm

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### Abstract

**Purpose** To report the case of a pediatric patient with intramedullary spinal aneurysm.

**Methods** A 9-year-old boy presented with low back pain and subsequent gait disturbance. He had no history of trauma. After admission, MRI revealed an intramedullary spinal cord mass lesion surrounded by hemorrhage at the cervical–thoracic junction. Initial treatment was started with intravenous methylprednisolone and bed rest. Neurological deficit disappeared under careful observation for a few months. Surgical intervention was applied for diagnosis and resection of the mass lesion to prevent recurrent hemorrhage.

**Results** Intraoperative ultrasound sonography helped to diagnose the lesion as a spinal cord aneurysm, prior to midline myelotomy. Monitoring of transcranial muscle evoked potentials helped to avoid spinal cord damage during surgery. There has been no evidence of spinal aneurysm on MRI for 3 years after surgery and no neurological deterioration.

**Conclusion** To our knowledge, this is a first report of an intramedullary spinal cord aneurysm at the cervical–thoracic junction in a pediatric patient. Careful observation after initial symptoms followed by surgical intervention was favorable in this case.

**Keywords** Spinal aneurysm · Pediatric · Surgery · MRI · Ultrasound

### Abbreviations

MRI Magnetic resonance imaging  
TcMEP Transcranial muscle evoked potential

### Introduction

Among intracranial arterial aneurysms in adults, 3.2% develop without comorbidity [1] while this rate for pediatric intracranial arterial aneurysm is 0.5–4.6% in large series [2, 3]. Spinal aneurysm in childhood is rare and treatment has not been established. Spinal aneurysm are classified as those with a coexisting congenital arterial condition, such as arterial malformation or coarctation of aorta; and those isolated without a vascular comorbidity [4]. Here, we report an isolated spinal aneurysm in a pediatric case with intramedullary hemorrhage that was successfully treated surgically.

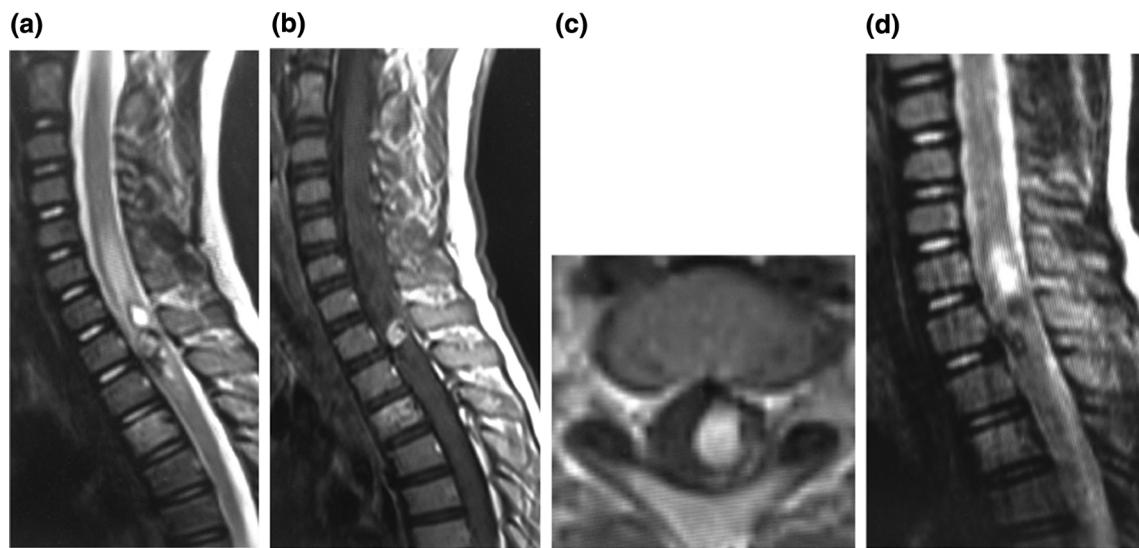
### Case

A 9-year-old boy presented with mild back pain and subsequent gait disturbance. He was admitted to hospital for assessment. Paralysis was present in both legs suggesting myelopathy. MRI revealed an enhanced mass lesion at C7/T1 in a swollen spinal cord. Intramedullary hemorrhage extended from the rostral to caudal side from the lesion center (Fig. 1a–c). The patient was diagnosed with intramedullary tumor with related hemorrhage and prescribed bed rest. Methylprednisolone was administered to offset secondary damage related to hemorrhage.

A few days later, motor function recovered and there were no signs of neurological deficit on admission to our hospital. Preoperative MRI showed reduced spinal cord

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**Fig. 1** **a–c** MRI a few days after onset of symptoms showed a contrast-enhanced mass lesion at the C7/T1 vertebral level, surrounded by hemosiderin. Spinal cord edema appeared to be extended in the cervical swollen spinal cord. A cystic lesion was identified on

swelling (Fig. 1d). The mass lesion showed contrast enhancement and was suspected to be a hemangioblastoma or cavernous hemangioma. A more thorough MRI investigation confirmed the absence of an intramedullary tumor. A surgical procedure was planned for definitive diagnosis of the mass lesion and to prevent recurrence of paraplegia due to recurrent hemorrhage.

We planned to resect the mass lesion posteriorly. C7–T1 laminectomy was performed and the dura was exposed. Prior to dura opening, intraoperative Doppler ultrasound suggested a mass lesion with an excessive color effect was identified in the spinal cord (Fig. 2b) and diagnosed as spinal aneurysm. When the dura was opened, there was no vascular malformation and no evidence of color change in the dorsal surface of the spinal cord (Fig. 2a). Via midline myelotomy, the aneurysm was seen in the center of the spinal cord (Fig. 2d). We gently detached the aneurysm from gray matter. Careful inspection revealed a blood supply to the aneurysm via arteries on the cranial and caudal sides (Fig. 2c, e). After the arteries were cauterized, the aneurysm was resected. During surgery, transcranial muscle evoked potential (TcMEP) monitoring was performed to prevent permanent neurological deterioration [5]. After detachment of the right side of the aneurysm from gray matter, the TcMEP amplitude declined and a different wave pattern appeared in all muscle. The spinal cord was rested for a moment and surgery was continued with gentle handling.

Hematoxylin–eosin staining of the resected specimen revealed an aneurysmal wall of elastic fibers with a few infiltrating inflammatory cells. Epithelial cells lined the

the cranial side of the tumor. **d** At 6 months after initial symptoms, spinal cord edema was obscure, but hemosiderin was still present close to the tumor

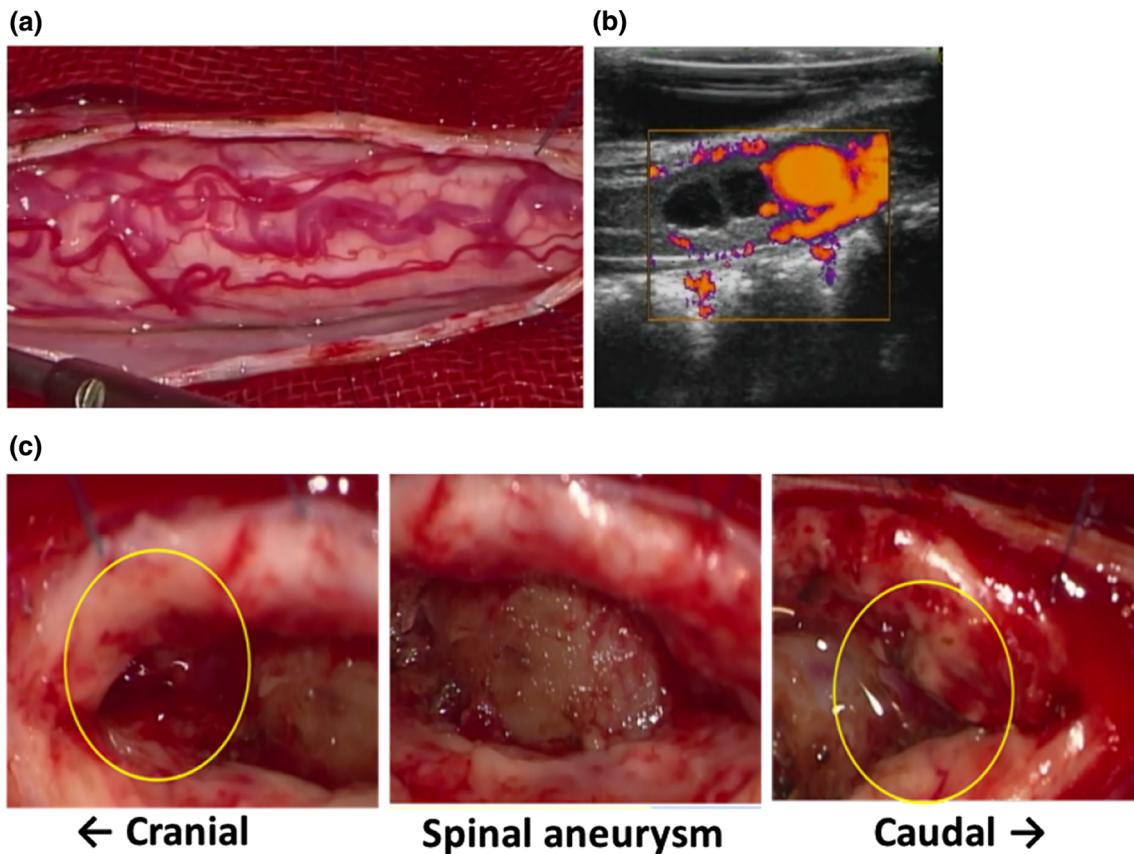
luminal side of the aneurysm. Hemosiderin was found around the aneurysm, suggestive of intramedullary hemorrhage (Fig. 3).

Postoperatively, transient paraplegia and bladder disturbance persisted for a few days, but then improved markedly. The patient could walk independently without support after two months. Subsequently, there has been no evidence of neurological deficit or recurrence of spinal aneurysm on MRI in follow-up of 3 years.

## Discussion

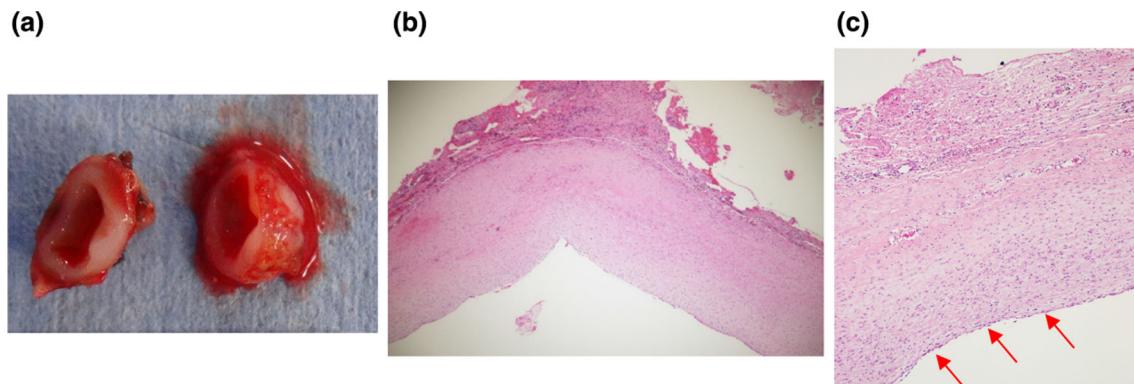
Spinal aneurysms can be divided into those with and without vascular malformation. In a systematic review of 123 cases, Madhugiri et al. [4] found a mean age of 38 years old and only 3 isolated spinal aneurysms in patients <20 years old [6–8]. Due to this rarity of isolated pediatric spinal aneurysm, management and treatment have not been established.

The acute symptoms of spinal aneurysm are associated with intramedullary or subarachnoid hemorrhage that may result in neurological deterioration. Atraumatic intramedullary hemorrhage presents with conditions such as primary spinal cord tumor, vascular malformation, and the other etiologies [9–16]. Abnormal vascular lesions in the spinal cord are major causes of spinal intramedullary hemorrhage, including neoplastic vascular lesions, spinal aneurysm, and arteriovenous malformation [17]. In our case, intramedullary hemorrhage was identified on initial MRI. Contrast-enhanced MRI is useful to distinguish



**Fig. 2** Intraoperative view after spinal cord exposure and images obtained from ultrasound sonography. The left side is cranial in all images. **a** There were no color changes of the spinal cord and no vascular malformation on the posterior surface of spinal cord. **b** Intraoperative ultrasound sonography revealed two mass lesions

inside the spinal cord with/without a color Doppler effect. **c** After midline myelotomy, a spinal aneurysm was found at the center of the spinal cord. Communicating vessels (yellow circle) were identified on the cranial and caudal sides of the aneurysm



**Fig. 3** Gross and micropathology of the resected aneurysm using hematoxylin–eosin staining. **a** The aneurysm had a 7-mm diameter and hemosiderin deposition was observed around the aneurysm. **b**,

**c** Epithelial cells lined the inside of the aneurysm (red arrow) and infiltrating inflammatory cells were present in the elastic wall of the aneurysm

vascular malformation from a solitary mass lesion, and we thought that the solitary lesion was an intramedullary tumor.

The prevalence of intramedullary rebleeding in pediatric spinal aneurysm has not been established. In pediatric

cranial aneurysm, 89/114 cases (78%) presented with subarachnoid hemorrhage [18]. Intramedullary cavernous malformations are vascular hyperplasias with a high risk of recurrent bleeding [15], with a rate of 66% per patient/year [19]. Careful monitoring of neurological status is clearly

**Table 1** Published cases of pediatric isolated intramedullary spinal aneurysm (compared with this case)

Author and year	Age (years)	Sex	Level	Treatment	Outcome	Remarks
Decker et al. [7]	1	F	L1	Intraoperative aspiration	Improved	Endovascular treatment was needed due to recurrence at 14-year old
Handa et al. [6]	3	F	C2	Surgical resection	Improved	Uneventful after operation
This case	9	M	C7/T1	Surgical resection	Improved	Symptom free without recurrence on MRI for 3 years after operation

needed, but the optimal timing for surgery is unclear. In general, early surgery is recommended for a case with progressive neurological deterioration. However, for a good outcome, surgical intervention should be postponed until neurological recovery in the case of cavernous hemangioma [15]. In our case, motor deficit improved following initial intramedullary hemorrhage, so we chose a wait-and-see strategy, and spinal cord swelling decreased on MRI 6 months after symptom onset.

Surgical resection or endovascular therapy are used to treat spinal aneurysm, depending on size, location (intra- or extramedullary), presence of feeding arteries, and comorbid vascular malformation. Definitive treatment for an isolated pediatric spinal aneurysm has not been described. To the best of our knowledge, there are only 2 cases published previously on pediatric isolated intramedullary aneurysm (Table 1). In one such intramedullary case with aneurysm at the thoracolumbar junction, Decker et al. [7] performed needle aspiration of the spinal aneurysm because a giant aneurysm was compressing the spinal cord. This achieved size reduction of the aneurysm, but flare-up occurred a few years later. Embolization of the Adamkiewicz artery was performed to obliterate the aneurysm [7]. In this case, endovascular intervention was applied because diagnosis was established in initial surgery. In our case, MRI could not distinguish between spinal aneurysm and intramedullary tumor. We planned surgery to diagnose and resect the solitary lesion to prevent progression of neurological deterioration due to additional intramedullary hemorrhage. At 3 years after surgery, there has been no neurological deterioration or further evidence of symptoms. Follow-up MRI revealed no recurrent aneurysm.

## Conclusion

This is the first report of pediatric isolated intramedullary spinal aneurysm at the cervical–thoracic junction that was successfully treated with surgery. Intraoperative ultrasound sonography helped with diagnosis of spinal aneurysm

before myelotomy. Preoperatively, observation until recovery of neurological deficit after intramedullary hemorrhage is favorable and associated with a better postoperative outcome.

## Compliance with ethical standards

**Conflict of interest** None of the authors has any potential conflict of interest.

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