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CASE REPORT

Anterior cervical intradural arachnoid cyst, a rare cause of spinal cord compression: a case report with video systematic literature review

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Abstract
Purpose Mostly seen at the thoracic level, arachnoid cysts are a very rare cause of cervical spinal cord compression. Generally treated by laminectomy and cyst fenestration, this approach does not allow removing the cyst in its entirety without manipulating the weakened spinal cord. The aim of this report is to present the case of a cervical intradural arachnoid cyst surgically removed by an anterior approach with corporectomy.
Methods Here is the case of an 18-year-old amateur boxer presenting with a voluminous cervical intradural anterior arachnoid cyst, extending from C2 to C5. Symptoms were cervical pain, quadriparesis, and clumsiness of both arms which had appeared just after a traffic accident. An anterior approach was chosen, through a C5 corporectomy.
Results The patient totally recovered from his sensitive symptoms at discharge and from his motor symptoms 6 weeks later. Early as well as 3-years post-operatively, MRI confirmed expansion of the spinal cord without any centro-medullar signal. The patient remained asymptomatic 3 years after surgery. Since the first report in 1974, 16 cases of symptomatic cervical intradural arachnoid cysts were treated via a posterior approach, one by MRI-guided biopsy, and one was re-operated on through an anterior approach. For 14 patients, their conditions had improved, while one died of pneumonia, one presented a condition worsened, and one had a stable neurological status.
Conclusion Using an anterior approach is a safe procedure that allows resection of a cervical arachnoid cyst without any manipulation of the weakened spinal cord, while giving the best possible view.

Keywords Anterior approach · Arachnoid cyst · Cervical corporectomy · Intradural · Spinal cord

Introduction

Arachnoid cysts are rarely symptomatic and represent a rare cause of spinal cord compression [1]. They correspond essentially to the type 3 of Nabors' classification (intradural cyst). Only 17 cases of spinal cord compression by cervical anterior intradural arachnoid cysts have been reported. Since they are symptomatic, these cysts must be treated. Three different options have been described: two surgical and one percutaneous under MRI guiding. Surgically, most authors performed a posterior approach. After the laminectomy, the cyst, anterior to the cord, was punctured and partially resected. Only one case of an anterior approach with complete resection has been reported.

We present the case of a young patient operated by anterior approach, with a C5 corporectomy. He experienced spinal cord compression due to a cervical anterior arachnoid cyst. This report is followed by a systematic literature review.

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Case report

History of the disease

An 18-year-old man, amateur boxer, was admitted to the emergency department for cervical pain, diffuse paresthesia, and weakness of both arms. History shows that he had been suffering from cervical pain for 3 months, poorly relieved by step 2 analgesics. Two weeks before, he presented diffuse paresthesiae and clumsiness of both hands just after a traffic accident. At that time, standard X-rays of the cervical spine had been interpreted as normal.

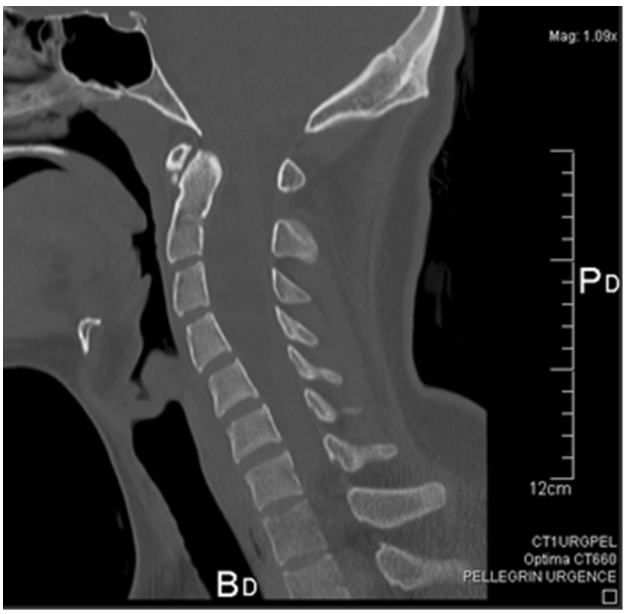


Fig. 1 Cervical CT-scan, sagittal. Bony window

Clinical examination and pre-operative investigations

The clinical examination showed a deficit in the profound sensibility of both legs. He had a clumsiness of both hands and a deficit quoted at 4/5 of the right thumb and index. He had no sign of pyramidal irritation.

A cervical spine CT-scan was performed in first intention. It showed an enlargement of the spinal canal, from C2 to C4, and on parenchymal windows, a hypodense collection extending from C2 to C5, laying back the spinal cord flat against the laminae (Figs. 1, 2).

The diagnosis of anterior cervical intradural compressive arachnoid cyst, with the spinal cord pressed against the laminae, was obtained on performing a complementary MRI exploration (Fig. 3).

Surgery

In this case, a surgery was indicated. We opted for an antero-lateral approach, to avoid manipulating this thin weakened spinal cord and to give us a better view.

After a standard anterior cervical spine approach, C4–C5, C5–C6 discectomies and C5 corporectomy were performed under operative microscope. The anterior longitudinal ligament was thoroughly resected from C4 to C6 and the dura mater was vertically opened (Fig. 4). The cyst, which was under pressure into the intradural compartment, immediately came out of the dural incision, which facilitated its wall dissection (Fig. 5). Extraction was possible with a gentle traction of the cyst's wall just after having released all adherences and opened the cyst to empty it (Fig. 6). The last arachnoid bridles were coagulated and sectioned, so that we could easily visualize the emptied

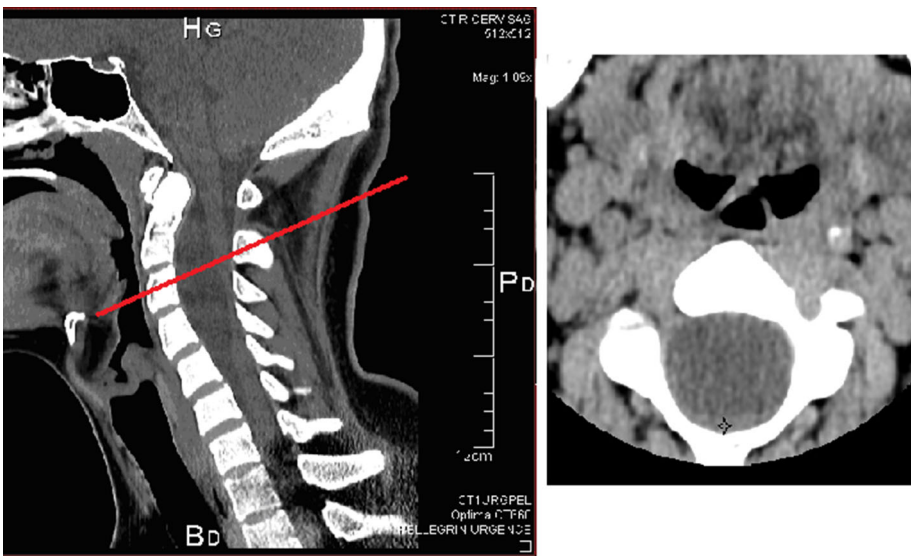


Fig. 2 Cervical CT-scan. Sagittal (**a**) and axial (**b**) at C3 level. Parenchymal window. Note how the spinal cord is thin and compressed at C3–C4 level (*star*)

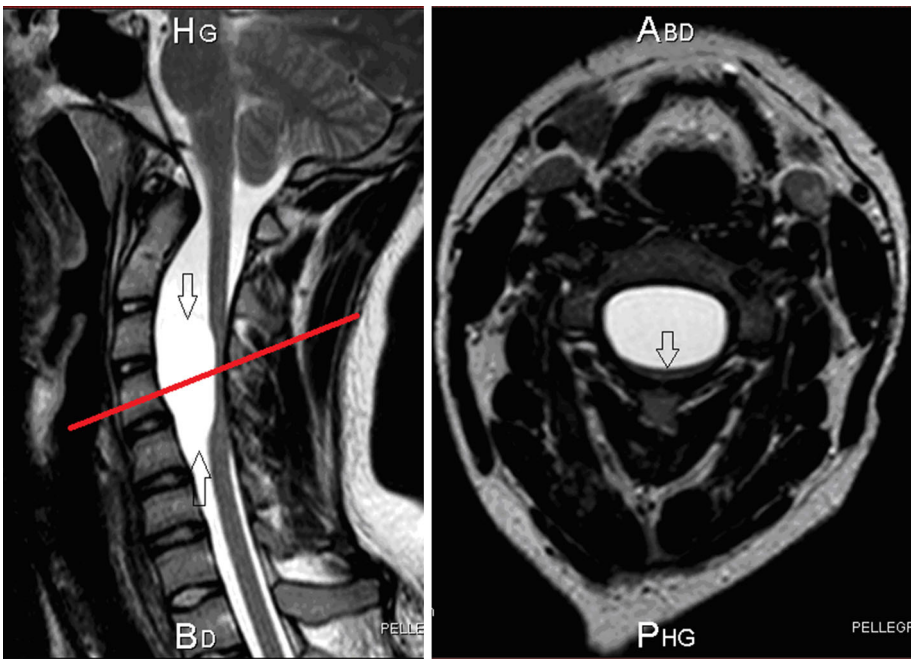


Fig. 3 Cervical MRI T2-weighted images, sagittal (**a**) and axial (**b**) at C4 level. The cyst appears in T2 hypersignal (same signal as CSF) and is intradural, with hyposignal at the superior and inferior poles (flow turbulences). Note how the spinal cord is thin and compressed at C3–C4 level (*arrow*)

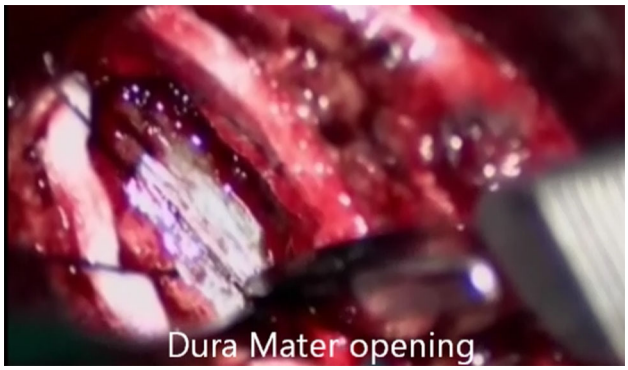


Fig. 4 Dura-mater opening

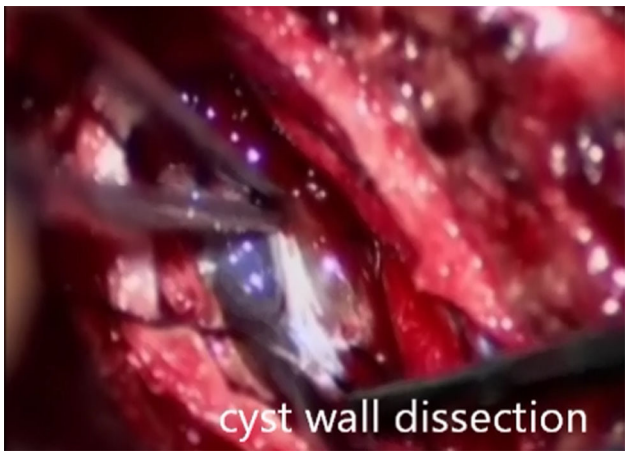


Fig. 5 Cyst wall dissection

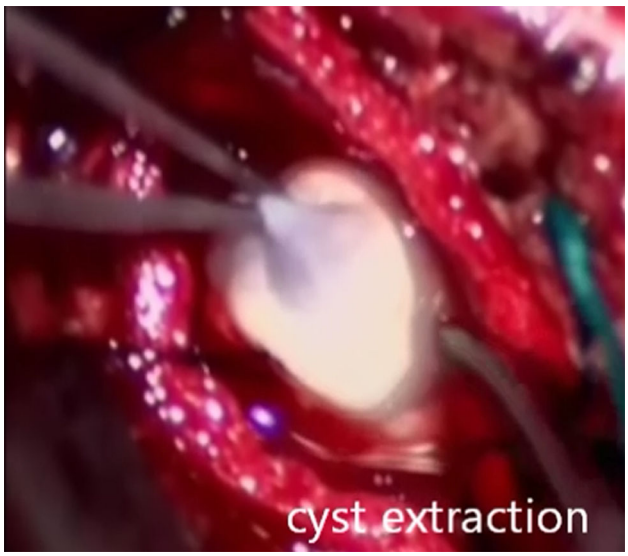


Fig. 6 Cyst extraction

sub-arachnoid space with a good flow of the cerebrospinal fluid in front of the newly decompressed spinal cord (Fig. 7). Closure of the dura was performed with interrupted silk sutures reinforced with biological glue (Fig. 8). Reconstruction was performed using an iliac graft (Fig. 9) with a plate. Further information about surgery is available in the video (see video).

Early post-operative evaluation

Post-operative MRI showed expansion of the spinal cord without any centro-medullar hypersignal. The cerebrospinal

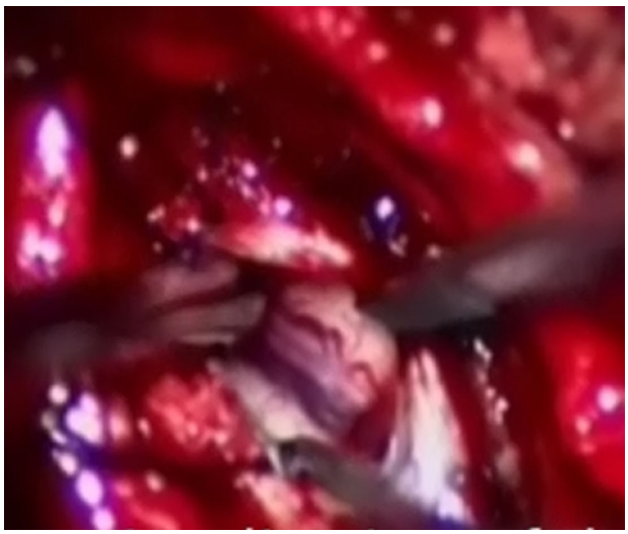


Fig. 7 Visualization of the emptied sub-arachnoid space and newly decompressed spinal cord

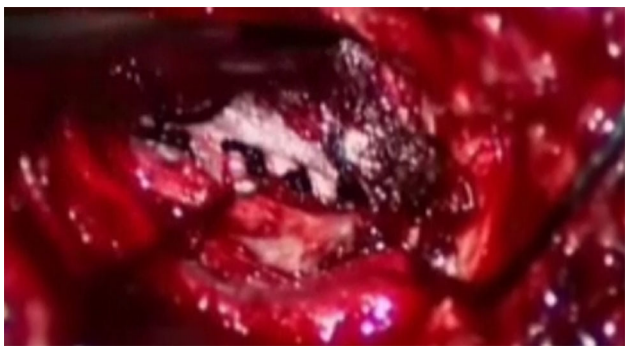


Fig. 8 Dura-mater closure

Fig. 9 Cervical X-rays face/profile showing the osteosynthesis C4–C6 with iliac graft

fluid's flow was recovered at the anterior part of the canal, as shown by flow MR sequences (Fig. 10). Histopathological analysis confirmed the diagnosis of benign arachnoid cyst.

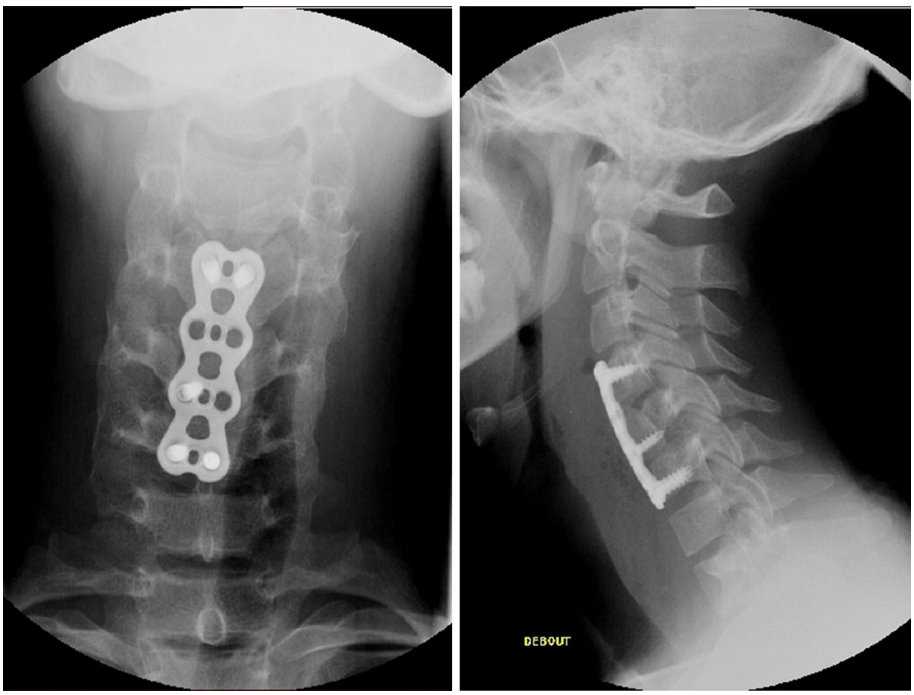
The post-operative phase was marked by a regression of the paresthesia at discharge. Complete motor recovery was reached 6 weeks later. The patient presented a post-operative Claude Bernard Horner syndrome, which completely regressed within 3 months.

Later post-operative evaluation: 3-year follow-up

The patient underwent a clinical check-up every year after surgery and post-operative imaging at 3 years. At this time, he still remained asymptomatic, and X-rays and MRI performed 3 years after surgery showed a well-positioned reconstruction plate, with no signs of pseudo-arthrosis and upper or lower disc degeneration (Figs. 11a, 12a). However, T2 CISS sequences showed a small cyst residuum probably communicating with the subarachnoid spaces, but non-compressive, with no mass effect on the spinal cord and no centro-medullar hypersignal (Fig. 12a, b). Moreover, there was still a good cerebrospinal fluid's flow at the anterior part of the canal and the cyst did not seem to be excluded from the CSF circulation, as shown by flow MR sequences (Fig. 12c).

Discussion and literature review

The majority of spinal arachnoid cysts have a posterior and thoracic topography [2], only 15 % are located at the cervical level. Consequently, only 17 cases of spinal cord



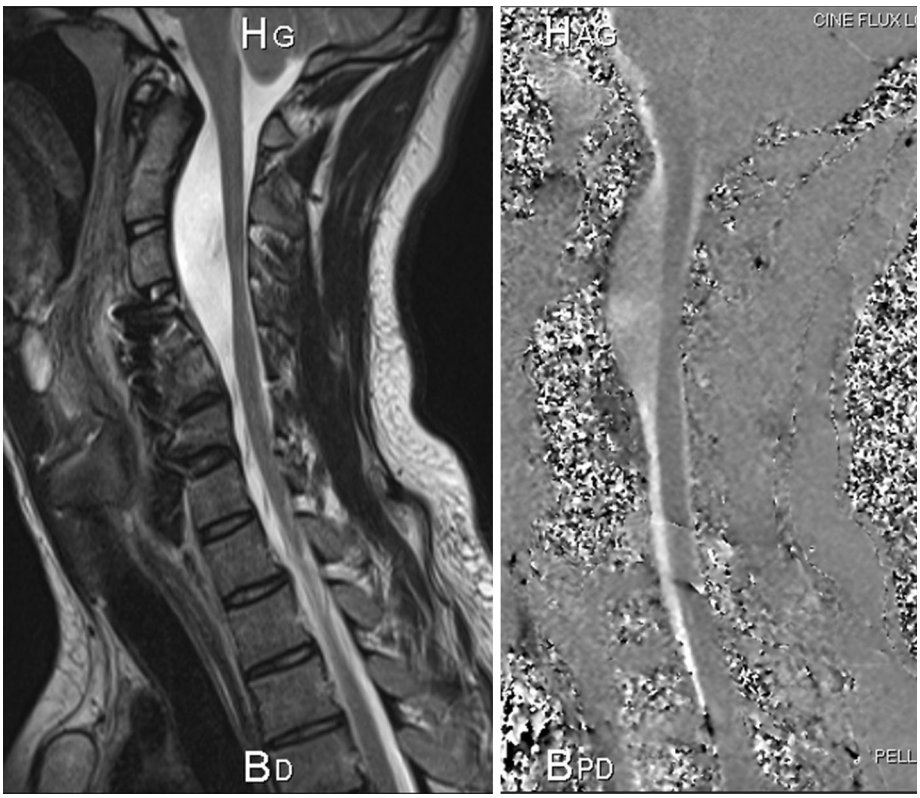


Fig. 10 Post-operative sagittal MRI. *Left* T2-weighted image; *right* flow sequence

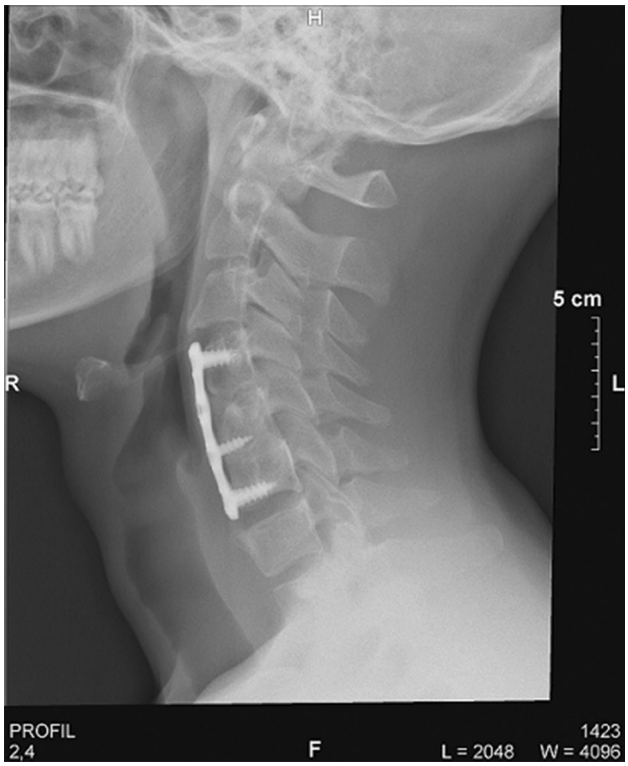


Fig. 11 Post-operative X-ray at 3 years

compression by anterior intradural cysts have been reported. The pathophysiology of the genesis of these cysts remains unclear. They are essentially intradural (type 3 of Nabors' classification). Regarding their aetiology, most are considered to be idiopathic. Cases of post-traumatic cysts, or more rarely post-infectious (meningitis) and congenital cysts were described. Many pathophysiological hypotheses were considered, like a dura-mater defect with trapped cerebrospinal fluid and the effect of anti-reflux valve [3], but these hypotheses mainly explain type 1 cyst formation. For type 3, another possible explanation could be the pathological proliferation of arachnoid trabeculae and arachnoiditis, leading to partitioning of the subdural space (mainly seen after meningitis).

The analysis of the cases reported in the literature (Table 1), [4–14] helped us isolating the following data.

Concerning epidemiology, it is a rare pathology (17 cases in the literature since 1974) with a sex ratio clearly in favour of male (12 men for 7 women), affecting young subjects (median age of 16.7 years old).

Twelve cases were considered as idiopathic; history of cervical trauma was noted in three cases and another associated malformation in four cases (for instance Chiari, myelomeningocele, and other forms of spina bifida). In our case, the neurological symptoms appeared just after a

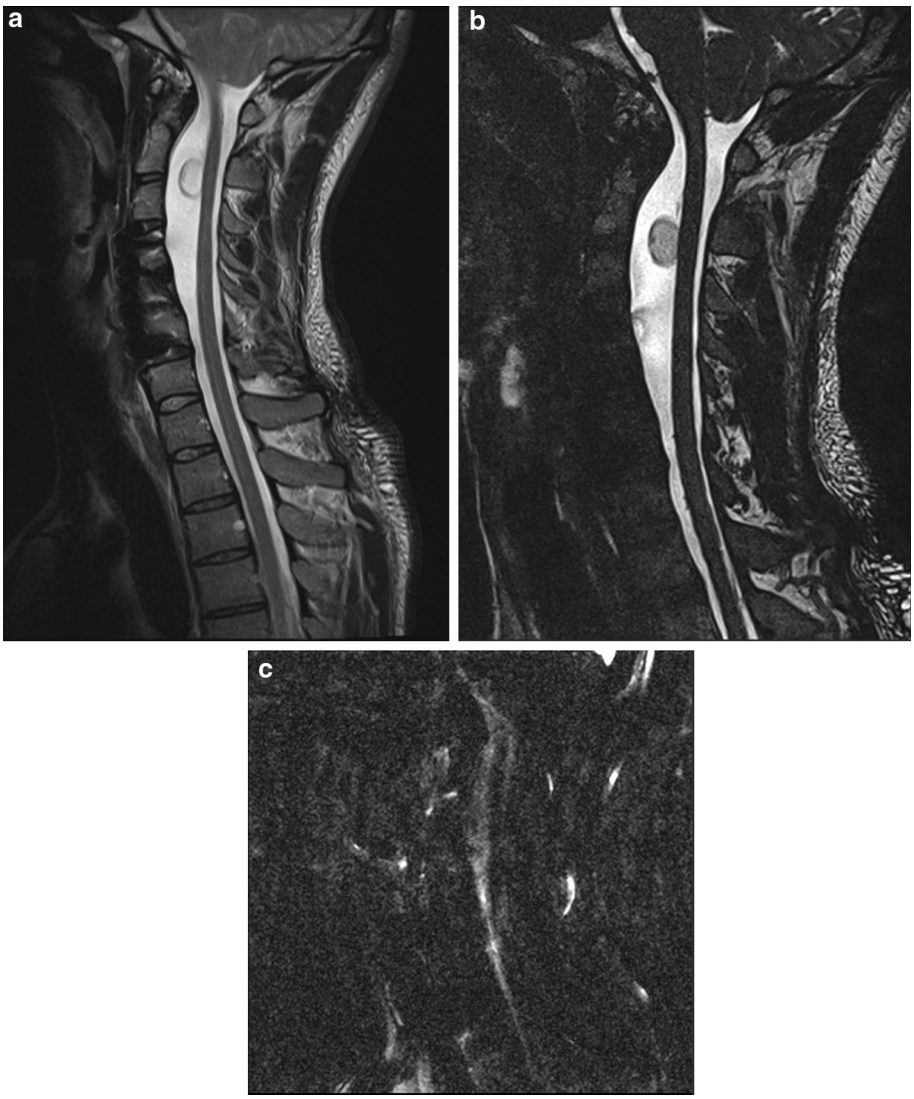


Fig. 12 Post-operative sagittal cervical MRI. **a** T2-weighted sequence, **b** T2-weighted CISS sequence, **c** flow sequence

traffic accident with cervical trauma. However, we agreed that this is a decompensation of a pre-existing cyst and not a post-traumatic cyst. Indeed, the great enlargement of the spinal canal and the tolerance of such a spinal cord compression proved that the cyst was there before this recent accident and had been developing for many years. The same remark might apply to the cases describing a cyst detected just after a cervical trauma.

Clinically, the great majority [15] of the cysts were revealed by motor symptoms, such as progressive para- or quadriparesis, which seems to be logical given the topography of the cysts. In our case as in Yair et al.'s, sensitive symptoms led to the discovery of the cyst. At last, in three cases, non-specific signs such as headaches, cervical pain, or vertigo were noted.

No preferential localization was found; all the levels between C0 and T1 can be affected.

Many treatments were described. The most common treatment was the surgery by posterior approach (laminectomy) leading to an incomplete resection in 16 cases and to a complete resection in one case. As the result of many recurrences in four cases, two cysto-peritoneal shunts, one cysto-pleural shunt, and one subcutaneous reservoir were necessary. Only one anterior approach has been described before ours, leading to a complete resection. Finally, one case of percutaneous puncture, guided by MRI, was published, without any recurrence. In our case, early post-operative MRI did not show any sign of a remaining cyst, whereas late post-operative MRI (at 3 years) showed a small cyst residuum with no compression on the spinal cord. Two authors described a complete excision of an arachnoid cyst in this location. However, Palmer et al. described the complete excision in 1974 [4] without a post-operative imaging as sensitive as MRI, and Muhammedrezai et al. [14] more

Table 1 Systematic literature review

References	Age/gender	Trauma?	Comorbidity	Symptoms	Level	Surgery	Outcome
Palmer [4]	19 F	No	No	Tetraparesia	C1–C3	Laminectomy, total resection	Improved
Palmer [4]	3 M	No	Pneumonia	Tetraparesia	C2–C4	Laminectomy, repeated punctures	Death
Herskowitz et al. [5]	28 F	No	No	Tetraparesia	C6–C7	Laminectomy, partial resection	Improved
Chan et al. [6]	37 M	No	No	Arms weakness	C1–T1	Laminectomy, cysto-peritoneal shunt	Improved
Rabb et al. [7]	2 F	No	Myelomeningocele	Tetraparesia	C6–C7	Laminectomy, cysto-pleural shunt	Improved
Chen and Chen [8]	18 M	C2 fracture 9 years before	No	Left hemiparesia	C3–C5	Laminectomy, fenestration	Improved
Jean et al. [9]	14 F	No	Myelomeningocele, Chiari II operated, multiple ventricular shunts	Chronic cephalalgia, vertigo	C0–C6	Cysto-peritoneal shunt	Improved
Jean et al. [9]	9 M	No	Myelomeningocele, Chiari II operated, multiple ventricular shunts	Cervicalgia, vertigo, cephalagia, facial paresia, tetraparesia	C0–C5	Fenestration, subcutaneous reservoir	Stabilization
Kazan et al. [1]	18 M	Cervical trauma 2 months before	No	Tetraparesia	C6–C7	Laminectomy, fenestration	Improved
Kazan et al. [1]	15 M	Cervical trauma 2 weeks before	No	Tetraparesia	C2–C3	Hemilaminectomy, fenestration	Improved
Lee and Cho [10]	4 M			Tetraparesia	C5–T1		
Gezici and Ergun [13]	26 M	No	No	Tetraparesia + C6 sensitive level	C5–T4	T2–T3 laminectomy, partial resection	Improved
Takahashi et al. [11]	13 M	No	Spina bifida occulta C1	Severe posterior cephalalgia to effort	C1–C3	MRI-guided left postero-lateral percutaneous puncture	Improved. Cyst disappearance at 9 months
Muthukumar [2]	4 M			Acute tetraparesia	C3–C4	Laminectomy, fenestration	
Maiuri et al. [12]	43 F			Intermittent tetraparesia	C7	Laminectomy, fenestration	
Gezici and Ergun [13]	25 M	No	No	Cervicalgia, tetraparesia	C0–C7	C3C4 laminectomy, fenestration	Improving of motor symptoms, stabilization of sphincter dysfunction
Muhammedrezai et al. [14]	29 M	No	No	C6 anterior hemi-spinal cord syndrome		Laminectomy C7 corporectomy	Aggravated improved

recently (2008), described a complete resection on the basis of a 1-month post-operative MRI. In our case, with the same post-operative delay, we also assumed that the cyst was entirely removed.

The outcome for patients affected with this pathology is globally good. Thus, the condition of 14 of 17 patients had improved, one died of pneumonia, one presented a condition worsened by the posterior approach surgery and was re-operated on through an anterior approach, and one had a stable neurological function.

In our case, we had a good outcome with total recovery of sensitive and motor symptoms 6 weeks after surgery, with no signs of recurrence at 3 year follow-up. The patient presented a transient post-operative Claude Bernard Horner Syndrome, which completely resolved at 3 months. Although rare (0.1 % of the cases in a large retrospective study including 1015 patients, [15]), it is a well-described post-operative complication after standard anterior cervical approach [15]. Indeed, the orthosympathic tract (i.e. superior cervical ganglia) comes out from the spinal canal from C1 to C4 levels and passes through the anterior paraspinal muscles (longus colli muscles) and the prevertebral lamina, which are detached and pushed aside to perform discectomies and corporectomy.

Although our patient had this transient complication, we believe that this approach is a safe and effective way to remove intradural anterior lesions compressing the spinal cord without taking the risk of a post-operative neurological deterioration by manipulating the weakened spinal cord.

Conclusion

Spinal arachnoid cysts are a rare cause of spinal cord compression. The anterior cervical cysts are even rarer. The great majority are treated by posterior laminectomy with partial resection of the cyst. We have presented the case of a patient operated through an antero-lateral approach without any manipulation of the spinal cord. The patient completely recovered from his sensitive symptoms at discharge and from his motor symptoms 6 weeks later and remained asymptomatic 3 years later despite a small communicating cyst residuum.

Conflict of interest The authors have no conflict of interest to disclose.

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