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Case report

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Diagnosis and treatment of spinal extradural arachnoid cysts: A chronic traumatic case report with review of the literature

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Abstract

Arachnoid cysts are formed by duplication of the arachnoid membrane between the arachnoid and the pia mater. Although it is very common in intracranial localization, those with spinal location are rare. Extradural arachnoid cysts of the spinal canal are extremely rare pathologies regarded as either congenital or acquired. These cysts, which can develop idiopathic, post-traumatic, and after arachnoiditis, are often detected incidentally. They present with weakness in the extremities, neuropathic pain, paresthesia, or myelopathy. Here we describe the case of a 17-year-old male patient with a history of chronic spinal trauma who attended our clinic with severe low back pain for 7 months.

Keywords: arachnoid cysts, spinal, extradural, trauma

Introduction

Spinal arachnoid cysts are rare lesions that are mostly idiopathic and congenital. Intradural arachnoid cysts are more common, although they can be seen in intra-dural or extradural localization [1]. They usually develop after arachnoid herniation through a small dural defect and progressively expand with increased cerebrospinal fluid (CSF) pressure. This increased wall tension causes counterpressure that contributes to the closure of the communication pedicle. Changes in the arachnoid trabecula after a trauma or arachnoiditis can also cause intradural spinal arachnoid cysts to appear [2-4].

Although spinal arachnoid cyst is mostly seen in the thoracic region, it can also be seen more rarely in the cervical and lumbar regions [5]. It is frequently seen in the middle and lower thoracic region and men. Although they are usually asymptomatic, the ones with anterior localization often present with weakness and myelopathy, and those with posterior localization are presented with neuropathic pain and paresthesia [6].

Spinal magnetic resonance imaging (MRI) is the method for detecting spinal arachnoid cysts that cannot be seen with conventional myelography due to their posterior localization [7]. Treatment involves surgical excision of the cyst. In cases that undergo early surgery, the prognosis is good [1]. Here we describe the case of a 17-year-old male patient who attended our clinic with a history of chronic spinal trauma, complaining of severe low back pain for 7 months.

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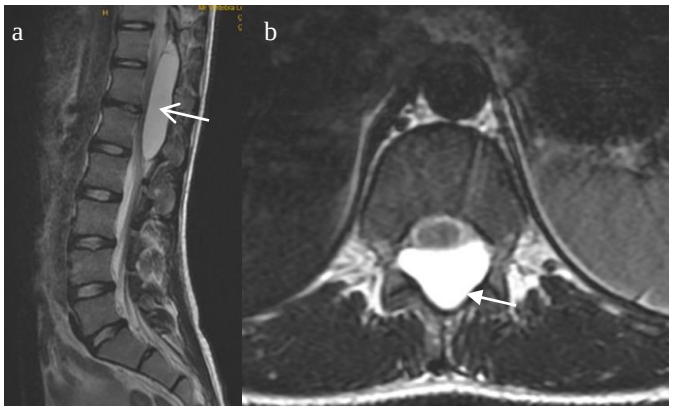
Case presentation

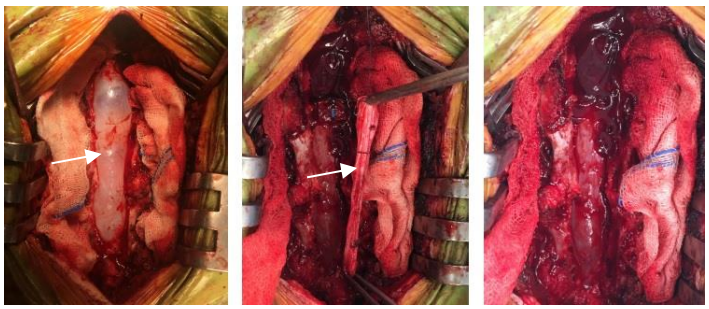
A 17-year-old male patient, whose written consent was obtained from his legal guardian before the surgical procedure, had complained of severe back and low back pain for 7 months. The patient, a professional football player, had no known diseases or history of infections, according to his medical history. In his neurological examination, his motor strength was normal, below T11 was hyposthenic, and the sphincter tonus was intact. On contrast-enhanced thoracolumbar MRI, an isointense cystic lesion of 18 × 32 × 90 mm in size at T11-L1 level was detected with CSF that did not show contrast enhancement. The cystic lesion in the epidural area at this level caused significant narrowing of the thecal sac, and the conus medullaris was pressed (Figure 1).

Figure 1: Preoperative thoracolumbar MRI, T2 contrast section, sagittal (a) and axial T2 (b) imaging; at the T11-L1 level, a cystic lesion (arrow) of 18 × 32 × 90 mm in size is located in the epidural area without contrast enhancement, and a marked narrowing of the lesion in the thecal sac, and compression of the conus medullaris are seen



The patient was taken into operation with intraoperative neuromonitorization. T11, T12, and L1 posterior elements bilateral end block were removed. On the dura, a cystic lesion was seen (Figure 2). Using bipolar biopsy forceps and a director in the craniocaudal plane, the cystic lesion seen from the cranial and caudal ends of the dura was then dissected and totally excised. A serial Valsalva maneuver was performed, and it was seen that there was no connection between the cyst and the subarachnoid space. T11, L1, and L2 laminoplasty was performed with the miniplate screw system after it was observed that the spinal cord was relieved from the posterior.

Figure 2: a, b: Cystic lesion (arrow) after laminectomy at T11-L1 level in the perioperative photograph, c: Intact appearance of the dura after en bloc removal of the cyst



Control thoracolumbar MRI was performed on the patient who was taken to the service without any postoperative deficit, and it was seen that the lesion was totally excised (Figure 3). The patient was mobilized on the first postoperative day and was discharged on the fourth day without any problems. No newly developed complaints or radiological findings were detected in the 1-year clinical follow-up of the patient whose histopathological examination was reported to be compatible with an arachnoid cyst (Figure 4).

Figure 3: Postoperative thoracolumbar MRI, contrast-enhanced T2 sagittal (a) and axial (b) imaging; it is seen that the extra-axial cystic mass (arrow) at the T11-L1 level was totally excised, and there was no evidence of residual/recurrence.

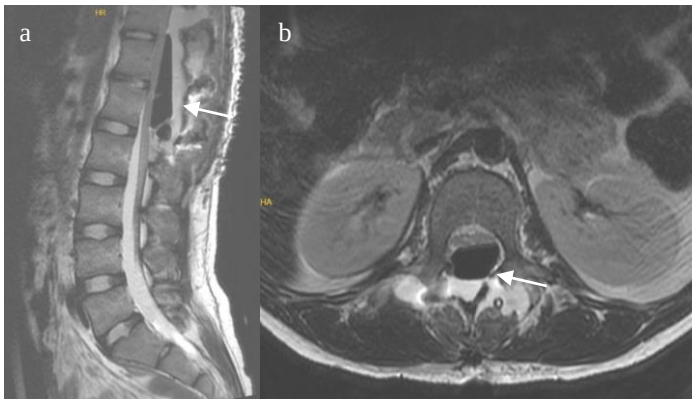


Figure 4: Postoperative 1st year, thoracolumbar X-ray; sagittal (a) and axial (b) imaging; deterioration of the patient's thoracolumbar axis and newly developed kyphotic deformity are not observed.

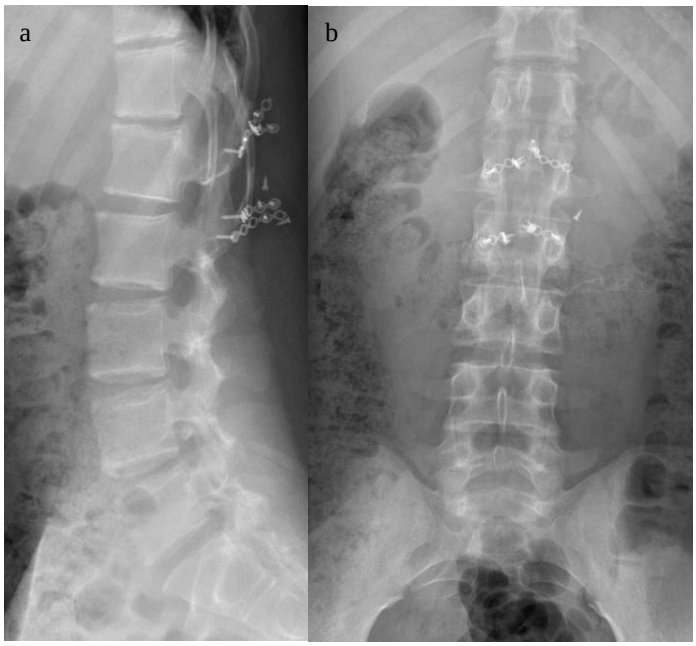


Table 1: Comparative data analysis of previous series reported in literature

Author& Year	No of Pts	Mean Age (yrs)	Female Sex	Presentation (% pts)	Imaging	Location	Surgical treatment (% pts)	Follow-up (months)	Outcome (%)
Wang et al., 2003 [6]	21	52	38%	Pain (76%), sphincter (24%), myelopathy (52%)	MRI only	10 dorsal thoracic, 4 ventral thoracic, 2 dorsal cervical, 2 ventral cervical, 3 dorsal lumbar	Laminectomy w / cyst fenestration & radical cyst wall resection, plus syrinx-subarachnoid shunting	17	No cyst recurrences during the FU period. Symptoms of weakness (100%), hyperreflexia (91%), & incontinence (80%) were more likely to improve than neuropathic pain (44%) & numbness (33%).
Bassiouni et al., 2004 [19]	12	43.6	52%	Pain (50%), myelopathy (62%)	MRI & CTM in 11 cases	12 dorsal thoracic	Laminectomy/laminoplasty w/ total excision of cysts <5 levels, otherwise generous fenestration	38.4	Follow-up MRI: no recurrence of the cyst was observed in any patient.
Netra R et al., 2011 [23]	18	34.6	22.2%	Post-traumatic back pain (10%)	MRI only	7 dorsal thoracic (one of them post traumatic patient), 8 dorsal thoraco lumbar, 3 dorsal lumbar	Unspecified	Unspecified	Unspecified
Funao et al., 2012 [18]	12	39.7	42%	Pain (42%), weakness (33%), gait ataxia (58%), paresthesia (67%), sphincter (50%)	CTM & cine MRI	12 dorsal thoraco lumbar	Laminectomy w/ total resection (58%), closure of dural defect w/o cyst, resection (42%)	56	No recurrence of the SEAC during the FU period. Improvement in the mJOA score.
Bond et., 2012 [22]	11	9.6	54.5%	Back pain (27%), loss of BLE function (36%), loss of sensation (18%), gait instability (27%), limited hip flexion (9%), incidental MRI finding (%9)	MRI only	3 dorsal thoracic, 4 dorsal thoraco lumbar, 1 dorsal lumbo sacral, 3 dorsal sacral	Total resection (100%)	19.1	No recurrence of the SEAC during the FU period
Kong et al., 2013 [14]	1	65	0%	Progressive paraparesis from 15 years following trauma, mild motor weakness of bilateral legs, urinary incontinence, muscle atrophy of both lower extremities	MRI only	Dorsal thoraco Lumbar	Laminectomy+ total resection	5	No recurrence of the SEAC during the FU period
Tokmak et al., 2015 [29]	10	50	60%	Hypoesthesia (20%), back pain (70%), post-traumatic back pain (10%), paraparesis (40%), radicular pain (20%), monoparesis (10%)	MRI only	7 dorsal thoracic, 1ventral thoracic, 2 dorsal thoraco lumbar (one of them post traumatic patient)	Hemilaminectomy/ total resection (30%), Laminectomy/ total resection (20%), Laminoplasty/ total resection (40%)	26.2	Follow-up MRI: no recurrence of the cyst was observed in any patient, incomplete recovery (20%)
Garg et al., 2016 [21]	9	29.7	22%	Weakness and sensory loss (44%), Urinary incontinence (33%), backache (55%), weakness and pain (33%)	MRI only	3 dorsal thoracic, 1 ventral thoracic, 3 dorsal thoraco lumbar, 1 ventral lumbo sacral, 1 ventral cervico lomber	Excision (88%), marsupialization (11%)	19.5	Follow-up MRI: no recurrence of the cyst was observed in any patient.
Viswanathan et al., 2017 [25]	14	52.1	36%	Weakness (79%), gait ataxia (100%), paresthesia (86%), sphincter (28.6%), myelopathy 71.4%	MRI only	12 dorsal thoracic, 1 dorsal cervico thoracic, 1 dorsal thoraco lumbar	Cyst wall fenestration & partial resection	22	Median improvement in mJOA score of 2.0 (1.3E3.0) (P<0.001) w/ respect to the preop scores.
French et al., 2017 [15]	10	60	66%	Pain (10%), gait ataxia (90%), paresthesia (60%), sphincter (20%)	MRI & cinemode bSSFP MRI in 3 pts	Unspecified	Fenestration (60%), complete excision (40%)	4.4	Follow-up MRI & subjective symptom assessment.
Fam et al., 2018 [20]	16	57	75%	Pain (63%), falls (31%), paresthesia (6%), weakness (44%), gait ataxia (50%)	MRI & CTM in 5 pts	10 dorsal thoracic, 2 ventral thoracic, 1 dorsal cervical, 1 ventral cervical, 1 dorsal lumbar, 1 ventral lumbar	Total cyst excision (79%), fenestration/marsupialization only (14%), fenestration & ligation (8%)	8.2	Improvement in SF-36 parameters across all quality-of-life parameters.
Singh et al., 2019 [24]	10	27.4	50%	Radiculopathy (60%)	MRI only	10 dorsal	Total cyst excision (80%), partial cyst excision (20%)	65.1	No one of the patients had clinical deterioration or radiological recurrence till last follow-up.
Our case	1	17	0%	Post traumatic back pain	MRI only	Dorsal thoraco lumbar	Laminectomy/ total cyst excision	12	Follow-up MRI: no recurrence of the cyst was observed in patient.

Pts: patients, FU: Follow-up

Discussion

Terminology and classification

Spinal arachnoid cysts were first identified by Schlesinger in 1893 and were first published by

Spiller et al. in 1903 [8]. In the literature, the terms "arachnoid diverticulum", "leptomeningeal cyst", "localized adhesive arachnoiditis", and "serous spinal meningitis" have been used by various authors to define SAC based on different pathological components [9]. Nabors classifies extradural spinal arachnoid cysts as type IA spinal meningeal cysts [10]. Dorsal cysts are more common than ventral cysts. The cyst localization was 120 dorsal and 14 ventral in the 134 patients presented in the literature (Table 1). Our case was also located dorsally, which is compatible with the literature.

Pathogenesis

Various theories have been proposed that claim that the formation mechanism of spinal arachnoid cysts is multifocal. Elsberg et al., who presented the first theory, suggested that spinal arachnoid cysts develop after arachnoid membrane herniation from a congenital diverticulum or congenital dural defect. Neural tube defects further support this theory, and a familial disposition has been noted in some patients.

According to certain theories, the reasons stated in most cases lead to a defect in the meninges membranes, which results in the herniation of the arachnoid membrane [11]. It is thought that if the cysts are associated with the subarachnoid space, they expand with the "valve-like" mechanism, and if not, they expand with H_2O osmosis from the cyst wall or active fluid secretion from the epithelial cells lining the cyst and eventually become symptomatic [9,11]. Most cases of spinal arachnoid cysts are idiopathic, but those of traumatic origin are particularly rare.

Regardless of the etiology, the underlying pathology results from herniation of the arachnoid through a defective or fragile dura mater. In traumatic cases, the defect is frequently found on the dorsal plane of the dura. When the literature was reviewed, we found that only four of the 144 cases presented had a history of trauma (Table 1). In these four cases of extradural arachnoid cysts that developed after trauma, the cyst localization was on the dorsal surface.

The fact that our case was a professional football player made us think of chronic trauma exposure, and the patient was evaluated as having a case of a post-traumatic arachnoid cyst. Additionally, consistent with the literature, the cyst location was on the dorsal surface.

Localization

It has been reported that cystic lesions are mostly in the thoracic regions (69–80%), followed by the cervical region (15–20%) and lumbar region (5–7%) [12]. Most patients with spinal arachnoid cysts of congenital origin present clinically in adolescence or early adulthood, and diverticula tend to be located in central regions. Cysts that extend along several vertebral segments and connect with the subarachnoid space via a small space are often located in the thoracic region [1,6]. Our case was also a young man, consistent with the literature. The lesion was in the form of a thoracic arachnoid cyst connected to the subarachnoid space in several areas.

Clinical presentation

In the pediatric age group, symptomatic spinal arachnoid cysts are rare lesions. Cysts that appear are frequently seen together with neural tube defects, such as meningomyelocele and diastometomyelia [13]. The mean age in the literature is 41.3 years (Table 1). Kong et al. [14] presented a case of a dorsal thoracolumbar spinal extradural arachnoid cyst that developed after a trauma at 65 years old, which is the most advanced age case in the literature. It is frequently seen in the middle and lower thoracic region and men. The literature shows an average of 59.2% male dominance (Table 1). Consistent with the literature, our case was a 17-year-old male patient. Although spinal arachnoid cysts are often asymptomatic, symptomatic cases present with slowly developing myelopathy findings [6]. The most common findings, according to research, are pain and myelopathic symptoms. Other common findings are radiculopathy, weakness, ataxia, and urinary incontinence. Post-traumatic back and low back pain is the most common symptom in patients with a history of trauma (Table 1). French et al. reported the incidence of pain as 10% in a study of 11 patients, which is contrary to the literature [15]. Kong et al. [14] presented a case with progressive paraparesis that developed after trauma from 15 years ago in addition to back pain. The only symptom seen in other post-traumatic cases presented in the literature, including our case, is back pain.

Imaging findings

As seen in most series, spinal extradural arachnoid cysts are more common than intradural cysts [16]. In the diagnosis, myelography, post-contrast computed tomography (CT), and MRI, a non-invasive and effective method, provide sufficient information about the lesion's width, volume, and structure [17]. MRI shows characteristic CSF-like density in both T1WI and T2WI (Figure 2). Additionally, it may highlight surrounding bone changes and the association of the cord or cauda with spinal extradural arachnoid cysts. The signal within the cyst may appear hyperintense compared with the CSF in the spinal canal due to the higher protein content of the cyst fluid. Contrast series is recommended to see if there is healing in the cyst wall. With the progress in MRI series, the size, number, and even the exact level of the dural defect can now be determined. Other rare features to look for in MRI include the absence of extradural fat, cord atrophy, and myelomalacia. Other imaging methods used in diagnosis include myelography, CT myelography, and cinematic MRI [15,18-20]. These imaging methods can show the location of the communication zone between the dura and the cyst cavity. Parasitic cysts, including cysticercosis or echinococcal cyst, should be included in the differential diagnosis, especially in patients from Anatolia.

Spinal extradural arachnoid cysts are often located in the thoracic area. In the literature, Wang et al. [6] reported 17 extradural and four intradural cyst cases in their series of 21 patients. In the series of Fam et al. [20], 12 of 16 patients had extradural cysts, and only four had intradural cysts. However, the same situation may not be true in children, as seen in several pediatric patients with intradural cysts in 58% of patients [21]. The authors suggest that this may be due to the high incidence of associated congenital central nervous system malformation in children in their series, known to be associated with intradural cysts [22]. In the reviewed articles, none of the patients had

intramedullary cysts described in the literature, albeit rare. In our case, thoracic MRI was used because it is a non-invasive method to define the lesion, and a dorsal cystic lesion with an isointense mass effect without contrast enhancement was detected.

The thoracic region is the most common site of spinal extradural arachnoid cysts [6,19,20,23-25]. In our case, similar findings were observed with thoracolumbar involvement. This could be because the dorsal column is the longest segment and/or because arachnoid cysts in the spinal extradural of the dorsal region are almost always symptomatic due to the narrow dorsal spinal canal.

Treatment and outcomes

There is no standard treatment protocol for the management of spinal arachnoid cysts. The usual practice is the excision of the cyst with the closure of the dural defect in extradural cysts, especially in symptomatic cases. Fenestration of the cyst is usually performed in intradural cysts, particularly ventral to the cord. Other surgical treatments include shunting the cyst to the peritoneum, pleural space, and right atrium with wide fenestration. Patients who undergo surgery before experiencing neurological symptoms have a better postoperative prognosis [1]. Wang et al. [6] recommend total cyst excision for spinal intradural arachnoid cysts. Simple cyst aspiration is not recommended because it does not meet adequate treatment. Despite a dural defect, some surgeons have also performed partial excision of the cyst wall. Hatashita et al. [26] stated that if the cyst is excised, it is not important whether there is a dural defect or not. This conclusion is based on cases when the dural defect area is sometimes not found or where there is no CSF accumulation if the entire cyst is removed without closure of the dural defect.

The large laminectomy required for complete cyst wall excision can cause complications, such as kyphosis and instability. To prevent these complications, laminoplasty is increasingly recommended instead of total laminectomy. In addition, another approach aims to limit the required laminectomy distance by closing only the dural connection instead of excision of the entire cyst wall [18,27,28]. To limit the size of laminectomy to maximum levels, Endo et al. advocate using endoscopes in spinal extradural arachnoid cyst management [28]. After partial hemilaminectomy/laminectomy, an endoscope is inserted into the cyst cavity after partial resection of the cyst wall through the bony window and moved cranially and caudally for fenestration of the cyst wall. Thus, communication of the cyst cavity with the subarachnoid space is ensured. It was observed that cyst recurrence rates were not higher in patients who underwent partial cyst excision after partial laminectomy and endoscope use than in patients who underwent radical laminectomy and complete cyst excision. However, kyphotic changes were observed in the spine in two patients in the total laminectomy group, while kyphosis did not develop in any patient in the partial laminectomy group, but this was not statistically significant.

A limitation of the study was that the mean follow-up time in the limited laminectomy group was significantly lower than the other group. Neurological improvement in these patients was the same as in patients with complete excision of the cyst without recurrence at a mean follow-up of 4.7 years. However, the degree of postoperative kyphosis in patients where only the dural connection is closed was significantly less than in patients who underwent a wide laminectomy to completely excise the cyst wall.

To prevent total cyst excision and possible kyphotic deformity, we applied T11-L2 laminoplasty on our patient, who had no other active complaints other than chronic back pain, and whose neurological examination was normal. No new complaints or radiological findings were detected in the follow-up of the patient, who was discharged without any postoperative neurological deficit or additional complaints.

Conclusion

The etiology, pathogenesis, and treatment of spinal extradural arachnoid cysts are not well defined. Neurological recovery appears to depend on the size of the cyst and the degree and duration of spinal cord compression. They are benign cysts that can show complete improvement in neurological findings when diagnosed with necessary radiological examinations and treated with early appropriate surgical methods before compression findings occur. Although early surgery is satisfactory, the rate of neurologic recovery decreases as the duration increases.

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