# Original Article

# Symptomatic extradural spinal arachnoid cyst: More than a simple herniated sac

# ABSTRACT

**Introduction and Study Design:** Spinal arachnoid cyst is an uncommon entity, presenting with radiculopathy or paraparesis. These cysts are usually found in intradural extramedullary region; and the extradural region is a rare location. The exact pathogenesis in the existence of these cysts in the extradural region is still debated. In this article, we have retrospectively analyzed the clinical profile of the rare extradural arachnoid cyst (EDAC).

**Methodology:** In this study, 19 patients of EDAC operated at our institute between January 2006 to June 2016 are analyzed. All patients with the Oswestry disability index score of >20 were managed surgically (open laminectomy and cyst excision). The clinical outcome was assessed at using 5-point satisfaction scale and McCormick grading.

**Results:** All 13 operated patients had EDACs with communication with the intradural compartment. In 11 (84.6%) patients, cyst wall was excised completely, in 2 (15.3%) patients underwent partial excision of cyst wall; dural communication was closed in all patients (n = 13). None of the patients had clinical deterioration or radiological recurrence till the last follow-up. Mean follow-up was 52.2 months (range 1–160); all patients were satisfied after surgery (median score was 3).

**Discussion:** Symptomatic EDACs account <2% of all spinal tumors. The EDACs have communication with the intradural compartment. In our article, we have discussed the approach and management of EDAC, including minimally invasive percutaneous procedures.

**Conclusion:** One should aim for preoperative or intraoperative localization of dural communication and try to disconnect the extradural cyst from the intradural connection to prevent recurrence. Surgical treatment is complete excision of the cyst.

**Keywords:** Arachnoid cyst, ball valve mechanism, Core Outcome Measures Index, extradural, minimally invasive approach

#### INTRODUCTION

Arachnoid cyst in extradural location is a rare etiology of compressive myelopathy. Most of these cysts are asymptomatic and detected incidentally. The lower thoracic spine is the most commonly reported site; although, rare cases have been reported from lumbar, lumbosacral, and cervical region also. Most of these lesions are considered congenital in origin. Other etiologies of trauma, inflammation, and infection have also been proposed. The exact pathogenesis of these cysts is still debated; they are believed to arise as herniation of arachnoid from small defect in the dura. Then, gradual enlargement of these cysts forms symptomatology of cord compression. Extradural arachnoid cyst (EDAC) is of

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two types (may have a pedicle or sessile connection it to subarachnoid space). With few such cases reported in the literature, exact incidence is difficult to comment on. In this

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article, we present our institutional experience of EDAC and review management options and surgical nuances to prevent recurrence.

### **METHODOLOGY**

The article is written according to the Strengthening the Reporting of Observational Studies in Epidemiology Statement guidelines. Individual consent from the patient to use clinical and radiological details for publication was taken as per our department policy.

#### **Patient spectrum**

We have retrospectively evaluated 19 patients of EDAC presented at our institute between January 2006 and June 2016. The data regarding demographic profile, symptomatology, McCormick disability scale, the location of the cyst, surgical findings, and postoperative complications were recorded from hospital records. Individual consent from all patients was obtained for surgery and publication of clinical details.

#### **Study parameters**

"Primary cyst" was defined as the presence of cyst in cases operated for the first time, whereas "Recurrent cyst" was defined when the patient was operated for second or third time. The dural communication was termed "superior" if the position lies above midway along the length of cyst and "inferior" when the position lies below midway along the length of the cyst. The cyst was sent for histopathology examination and any evidence of blood was noted. None of the patients had hemorrhagic cyst as content, and hence, evidence of capillary proliferation was considered as a marker of previous trivial trauma.

#### **Inclusion criteria**

Patients were categorized on the basis of the Oswestry disability index (ODI). Minimally symptomatic patients with ODI <20 were managed conservatively, whereas symptomatic patients with ODI >20 were managed surgically. Patients underwent laminectomy and excision of the cyst with the closure of communication of cyst with the intradural compartment.

#### Follow-up

The clinical outcome at last follow-up was assessed using 5-point scale (satisfaction score) and McCormick grading. Patients were asked two questions similar to Core Outcome Measures Index (COMI) question 10 and 11: "In the perspective of your back problem, how satisfied were you with the treatment?" and "As compared to previous medical treatment, how much the operation had helped the problem overall." Table 1 shows the total satisfaction score of both questions after surgery. Last follow-up was noted telephonically and as outpatient clinic basis.

#### **Statistical analysis**

The Statistical Package for the Social Sciences version 22.00, International Business Machines Corporation (IBM), Armonk, New York, USA, was used for statistical analysis.

#### RESULTS

#### **Patient spectrum**

Out of 19 patients, 6 patients with ODI scores of < 20, both at first visit and subsequent follow-up, were managed conservatively. Twelve patients with ODI more than 20 were managed surgically; 1 patient with initial ODI of 10 but subsequently worsened (ODI = 24) in the course of follow-up was operated [Figure 1]. Therefore, we hereby discuss all

Table 1: Total satisfaction score calculated by	asking two questions
(similar to core outcome measures index ques	stion 10 and 11)

Question	Score
In perspective of your back problem, how satisfied	
were you with the treatment?	
Very satisfied	1
Somewhat satisfied	2
Neither satisfied nor dissatisfied	3
Somewhat dissatisfied	4
Very dissatisfied	5
As compared to previous medical treatment, how much the operation had helped the problem overall	
Helped a lot	1
Helped	2
Helped only little	3
Did not help	4
Made things worse	5
Total satisfaction score calculated by adding score	
of both questions	
Maximum satisfaction score	10
Minimum satisfaction score	2



Figure 1: Representative axial (a) and sagittal (b) magnetic resonance imaging T2-weighted image of a patient with extradural arachnoid cyst at D4 level. The patient was initially managed conservatively but later on needed laminectomy and excision of cyst

<i>n</i> =12	Age/gender	Level	Oswestry disability index score	McCormick scale	Bladder	Radiculopathy	Cyst/type	Excision	Follow-Up in months	COMI satisfaction score	Position of communication	Histopathology
Patient 1	60/male	L4-L5	28	2	S	Yes	One/Pr	Total	160	2	Dorsal aspect (inferiorly)	Focal proliferation of capillaries surrounded by mild mixed inflammatory cells
Patient 2	14/male	D10-L2	24	1	No	No	One/Pr	Partial	124	м	Dorsal aspect (inferiorly)	Dense fibrocollagenous wall
Patient 3	16/male	L5-C0	24	2	Yes	No	One/Pr	Partial	106	2	Dorsal aspect (midway)	Dense fibrocollagenous wall
Patient 4	18/male	D7-D9	28	т	Yes	Yes	Two/Pr	Total	92	м	Dorsal aspect (superiorly)	Dense fibrocollagenous wall with congested blood vessels
Patient 5	15/female	L5-S4	24	2	Yes	Yes	One/Pr	Total	64	м	Dorsal aspect (superiorly)	Dense fibrocollagenous wall with attenuated lining epithelium
Patient 6	40/female	L5-S4	24	2	Yes	Yes	One/Rec	Total	36	ω	Dorsal aspect (inferiorly)	Fibrocollagenous wall
Patient 7	18/female	D8-L1	30	4	Yes	Yes	One/Rec	Total	30	2	Dorsal aspect (superiorly)	Fibrocollagenous cyst wall along with hemorrgahe
Patient 8	27/male	D10-L3	30	4	No	Yes	One/Pr	Total	25	വ	Nerve root slleve	Fibrocollagenous wall with congested blood vessels
Patient 9	20/female	D9-D12	24	2	No	Yes	Two/Pr	Total	-	4	Dorsal aspect (inferiorly)	Dense fibrocollagenous wall
Patient 10	53/female	D9-D12	28	т	Yes	No	One/Rec	total	2	4	Two communiation	Fibrocollagenous wall with congested blood vessels
Patient 11	17/male	D2-D6	28	4	No	No	One/Pr	Total	1	Q	Nerve root slleve	Dense fibrocollagenous wall
Patient 12	17/male	D2-D6	28	4	No	No	One/Pr	Total	-	4	Nerve root slleve	Dense fibrocollagenous wall
Patient 13	20/male	D10-L2	30	4	Yes	No	0ne/Pr	Total	36	4	Dorsal aspect (superiorly) at D11	Fibrocollagenous tissue displaying few entrapped congested blood vessels and mild

COMI - Core outcome measures index, Pr - Primary ; Rec - Recurrent

surgically managed (n = 13) in this study [Table 2]. Mean age was 25.76 years (range 14–60; n = 3 with age >40 years) (8 M: 5 F). Out of 13 patients, 7 (53.8%) patients had lower dorsal region cyst (below D6), 2 (15.3%) patients had upper dorsal cyst (above D6), and 4 (30.7%) patients had lumbosacral cyst. The dural rent was found in all patients, 3 (23%) had communication along nerve root sleeve and remaining 10 (76.9%) had communication at dorsal surface.

#### **Clinical assessment**

Mean ODI of 13 patients was  $26.9 \pm 2.4$  (24–30) (median 28); mean McCormick scale was 2.8 (median 3). 3 (23%) had flaccid paraparesis and 6 (46.1%) had spastic paraparesis. Six (46.1%) patients had severe motor deficit (power below 3/5), 5 (38.4%) had mild weakness (power 3–4/5) and 2 (15.38%) patients had no motor weakness. Six (46.1%) patients had sensory deficit, whereas remaining 7 (53.8%) had no sensory involvement. Similarly, 7 (53.8%) patients had radicular pain, whereas 6 (46.1%) had diffuse, dull aching back pain. In our series, only 2 (15.3%) patients had overall severe neurological deficit with a severe motor deficit, sensory and bladder involvement.

Three (23%) patients in our series presented as "Recurrent cyst." On retrospective analysis of previous operation notes, we found that all three patients underwent cyst excision without closure of dural communication. During resurgery, we found the dural communication at the dorsal surface and closed it. These three patients had motor deficit (n = 2, 0-1/5 and n = 1, 3/5). All three patients were female (2 had lower dorsal region cyst and 1 had lumbosacral cyst). These patients also had bladder involvement at presentation, which persisted in the postoperative period also.

Eleven patients underwent complete excision of the cyst and in two patients' partial excision was done (due to dense adhesions). These partially excised cysts were located at lower dorsal in the first and lumbosacral region in second). Dural communication was closed in all patients.

#### Surgical outcome and follow-up

None of the patients had clinical deterioration or radiological recurrence till last follow-up. Mean follow-up was 52.2 months (range 1–160) with three patients <3 months FU. On histopathology, in the majority of patients, we found the fibrocollagenous wall. Four patients had proliferation of capillaries, 2 patients had hemorrhage, 2 patients had inflammatory infiltrates, and 1 patient had epithelial proliferation. Among 3 patients with recurrence, 2 had proliferation of capillaries.

(median 3). There was a trend suggesting less satisfaction score in patients with poor McCormick Score and more ODI score.

# DISCUSSION

Spinal symptomatic EDACs are a rare clinical entity, commonly located in the lower thoracic region.<sup>[1,2]</sup> It accounts for <2% of all spinal tumors, presents as an uncommon etiology of compressive myelopathy.<sup>[3]</sup> Nabors classifies EDAC as type IA spinal meningeal cyst.<sup>[4]</sup> Dorsally located cysts are more common than ventral, either single level or multiple level involvements. None of the 13 cases in our series had ventral located cyst. In the series of seven cases, Kulkarni *et al.* reported a rare case of extradural cyst located anterolateral.<sup>[2]</sup> The presence of the dural defect in EDAC, as one-way communication with intradural space, needs necessary identification of defect along with the excision of the cyst.

The exact etiology is still debatable; traumatic or congenital (mutation in the FOXC2 gene) being most accepted. Contrary to our belief, none of the patient in our series had previous history of arachnoiditis or spinal surgery. In addition, only two patients had a history of trauma. None of our patients had curvature anomaly including juvenile dorsal kyphosis or Scheuermann's disease; although both of these are reported in the literature. The age distribution in our series further decreases the possibility of congenital origin. We believe idiopathic origin to be most likely etiology.

#### Etiopathogenesis of extradural arachnoid cyst

The position of the spinal cord with canal is maintained by dentate ligament and posterior trabeculae. The posterior fibrous trabeculae from the posterior fissure to duramater are called septum posticum. Degeneration of this septum subsequently leads to the fibrous band and entrapped cyst within subarachnoid space. If the fistulous connection remains open, the arachnoid membrane can protrude out through defect. Gradually, cerebrospinal fluid (CSF) accumulates within the protruded arachnoid via ball valve mechanism, leading to progressive enlargement.<sup>[3,5]</sup>

There are cases reported in the literature with EDAC and split cord malformation or other dysraphism simultaneously in the same patient, suggesting the congenital nature of these cysts. The congenital origin is also supported by the occurrence of familial syndrome, including multiple spinal arachnoid cysts, lower-extremity lymphedema (milroy disease), and distichiasis (double rows of eyelashes).<sup>[4,5]</sup>

#### Postoperative patient satisfaction

Mean postoperative COMI satisfaction score was 3.2

#### Progressive enlargement of cyst size

One patient (with ODI <20) was initially managed

conservatively, but in due course of follow-up; symptoms worsened and we had to operate him. There are various theories in literature explain possible reasons for cysts expansion. Seung *et al.* observed that one nerve rootlet was stuck in dural defect prohibiting spontaneous closure of cyst. He believes this could be a factor responsible for the progression of cyst size.<sup>[4,6]</sup> In our series, we appreciated similar "*stuck*" in two patients. Other explanations being:

- a. Rohrer ball valve mechanism: Once the arachnoid protrudes from dural rent to extradural space, CSF accumulates by ball valve mechanism into cyst leading to progressive enlargement<sup>[6]</sup>
- Osmotic enlargement of cyst: Gradual absorption of the water by hyperosmolar cyst fluid and secretion of CSF by the arachnoid lining of the cyst's wall may lead to progression of EDAC
- c. Kulkarni *et al.* proposed that the cyst wall itself may be responsible for the production of CSF or absorption of CSF from the subarachnoid space.<sup>[2]</sup>

These cysts usually present with backache or radicular intercostal thoracic or leg pain.<sup>[1]</sup> Pain is progressive and associated with paraesthesia of related region limbs and subsequently worsens to spastic or flaccid paraparesis or quadriparesis. The cyst present in the thoracic region becomes symptomatic early in comparison to lumbar region cyst due to the narrower canal in the dorsal region. The motor weakness is usually more common than sensory or urinary involvement. Cases presenting with acute paraplegia have been reported due to herniation of the spinal cord through the dural defect into the EDAC; although all our cases had benign progression [Table 3]. Lumbar region cyst attains a larger size and usually presents with low back pain due to persistent pressure of growing cyst on nearby pain generators. Recurrent cysts tend to present more often with bladder symptoms. Sensory numbness may be present on examination. Sometimes, the progression in waxing and waning type and backache fluctuates with coughing possibly due to ball valve mechanism at the dural defect.

One interesting finding in our study is that five patients presented in McCormick grade 4 and 1 patients in McCormick grade 3. The severity of myelopathy is difficult to correlate with the nature of the disease. We believe that long-standing duration of disease resulted in chronic compressive myelopathy. Wang *et al.* and Garg *et al.* also found similar clinical findings. They reported pain and myelopathy as the most common symptoms. About 30%–40% of patients had bladder involvement in their series.<sup>[10-12]</sup>

The EDAC are usually misdiagnosed or diagnosis is delayed. The duration of symptoms varies with the location of the

Author	Number of cases/mean age (years)	Myelopathy	Radiculopathy	Standard laminectomy/ laminoplasty	Minimally invasive approach	Focus
Rahimizadeh A <i>et al.,</i> 2013	4/39	2	2	4	0	All four cyst were in thoracic region
Funao H <i>et al.</i> , 2012	12/39.7 6±3.5	6 patient had BBD	-	7	5	Patients with long duration of symptoms had poor outcome; significant difference in postoperative kyphosis between patients treated with standard laminectomy and minimally invasive approach
Liu JK <i>et al</i> ., 2005	1/11	1	0	1	0	No dural defect or fistulous connection
Choi <i>et al</i> ., 2013 <sup>[3]</sup>	2/55	0	2	2	1	In one case, only cyst fenestration was done due to moderate bleeding while dissection
Woo JB <i>et al.</i> , 2016	2/55.5	0	2	2	0	
Kong <i>et al</i> ., 2013 <sup>[7]</sup>	1/65	0	1	1	0	History of trauma present
Netra R <i>et al</i> ., 2011	18/34.6	-	-	-	-	Nabors types I and II meningeal cysts; One case had a previous history of trauma
Quillo-Olvera <i>et al.</i> , 2017 <sup>[8]</sup>	1/67	1	0	0	1	-
Tokmak M <i>et al</i> ., 2015	10/50	5	4	10	0	<pre>n=1 cyst was anterolateral; n=6 total excision was done</pre>
Muthukumar 2001 <sup>[9]</sup>	1/25	0	1	1	0	Sacral EDAC; should be included in differential diagnosis of sacral meningocoele
Kulkarni <i>et al</i> ., 2004 <sup>[2]</sup>	7/17.5	7 had paraparesis, 1 had urinary incontinence	1	7	0	Radical resection of the walls leads to a lasting cure from extradural arachnoid cyst

Table 3: Comparative data analysis of previous series reported in literature

BBD - Bowel bladder dysfunction; EDAC - Extradural arachnoid cyst

cyst with overall slow progression. The thoracic cysts present earlier when compared to lumbar cysts. In our series, patients with cyst located at thoracic level had mean 154  $\pm$  20 standard deviation (SD) days of clinical presentation, whereas patients at lumbar level had mean 320  $\pm$  84 SD days of presentation. Funao *et al.* also showed a similar pattern of presentation in their series.<sup>[10]</sup>

Magnetic resonance imaging is the investigation of choice for these cysts. Magnetic resonance imaging (MRI) shows characteristic CSF like intensity in both T1WI and T2WI [Figure 2]. It can also highlight surrounding bony changes and relation of cord or cauda with EDAC. We advise contrast sequences to see for cyst wall enhancement. With advancement in MRI sequences, now we can ascertain size, number and even exact level of dural defect. Miyamoto et al. and Doita et al. have demonstrated dural defects by using myelogram and Kinematic MRI sequences.<sup>[13,14]</sup> Neo et al. suggested that cine MRI and dynamic sequences are not always helpful and one may need computed tomography myelography sequences to localize the defect.<sup>[15]</sup> Nakagawa et al. recently proposes constructive interference in steady-state imaging as a better tool to localize.<sup>[16]</sup> We did not have such special sequences MRI in our series, being retrospective analysis. On MRI absence of extradural fat, cord atrophy or myelomalacia are other rare features to be searched for. Parasitic cyst including cysticercosis or hydatid



Figure 2: T1-weighted sagittal section of thoracic spine magnetic resonance imaging with extradural cystic lesion dorsally placed and pushing cord ventrally (a). The (b) T2 weighted sagittal section of thoracic spine magnetic resonance imaging shows same cyst with cerebrospinal fluid like intensity and ventrally displaced cord. The (c) axial section magnetic resonance imaging T2 weighted image confirms the extradural nature of cyst with thin hypointense dural wall seen. Intraoperatively (d) the cyst can be separated from normal dura and (e) postoperative specime of same cyst

cyst must be kept in differential diagnosis especially in patients of the Indian subcontinent. The signal within the cyst may appear hyperintense compared with the CSF in the spinal canal because of the higher protein content of the cyst fluid.<sup>[17]</sup> EDAC are benign expanding lesions, which widens the spinal canal, thinning surrounding bone and characteristically present with scalloping the posterior plate of the vertebral body and increased interpeduncular distance. It displaces the spinal cord and cauda roots anteriorly or laterally.

Indication of surgery in the asymptomatic cyst is controversial. We managed all patients according to ODI. The curable nature of disease intrigues surgeon to explore various therapeutic options. For symptomatic cyst, laminectomy with excision of cyst followed by localization and ligation of the dural defect is standard of care. The lesser invasive approach of laminoplasty seems better option, especially in the younger population. Previous studies suggest that there is the lesser incidence of postoperative kyphosis after laminoplasty, but Lee *et al.* suggested that laminoplasty is difficult to at the thoracic and lumbar level because the pedicles have a tendency to fracture due to their anatomical structure. Lee *et al.* reported kyphosis deformity even after laminoplasty.<sup>[18]</sup>

Some authors even propose conservative symptomatic management with radiological follow-up, especially in pediatric cases due to additional risk of post-surgical deformity. The risk of spontaneous cord herniation leading to acute paraplegia must be weighted with conservative plan. Laminoplasty is alternative in young patients although we did laminectomy in all our patients and none had any complication. The authors believe that postoperative kyphosis may occur even after laminoplasty of more than five levels. In our series, there were two patients with five or more level laminectomies and none had such spinal deformity in follow-up. In cases of postoperative progressive kyphosis, arthrodesis is advised. A prospective study with longer follow-up, might conclude on the need of laminoplasty over laminectomy.

Previously, the cystoperitoneal shunt was one of the management options. There is a high risk of recurrence with such shunting either with standard open laminectomy or even with CT or MRI guidance percutaneous. With increasing armamentarium in minimally invasive spine surgery, selective laminectomy and closure of dural defect are emerging option.<sup>[10]</sup> Partial excision of cyst has also been reported as the option with low rate of recurrence. Lee *et al.*, in their study, inferred that recurrence is related to inadequate closure of dural defect, rather complete or partial excision.<sup>[18]</sup> Out of three recurrent cases in our series, the dural communication

defect was not closed in primary surgery in all three. The importance of dural defect identification and closure has been highlighted in many studies. Kumar *et al.* reported no recurrence in their series with the closure of dural communication. Sometimes in long-standing or recurrent cyst total excision of cyst wall with dural closure is necessary as cyst wall can thick bands around thecal sac, which will prevent expansion of thecal sac. In our series, there was also no recurrence during the 42-month follow-up.<sup>[19,20]</sup> In terms of patient perspective, postoperative satisfaction questionnaire highlighted that majority of patients were "very satisfied" and surgery "helped" them in regaining normal life.

On histopathology examination, the secretory epithelium could not be appreciated. We want to emphasize the fact that there is no difference between intradural or EDAC. The proliferation of capillaries and inflammatory infiltrates may be indirect evidence for the history of trauma or infective etiology, but the conclusory remark is difficult. We also found the presence of epithelium (n = 1) and hemorrhage in two patients. We could not find any difference between primary and recurrent cyst. The cyst wall may be single layered or double layered. Outer layer in the fibrous sheath and the inner layer is arachnoid lining. The inner arachnoid lining may be absent.<sup>[6]</sup> Other studies found similarly, that the cyst walls contain only calcified tissue and layered collagenous fibers without evidence of the glands or secreting tissues.<sup>[21]</sup> Other findings of spinal arachnoid cysts usually reveal a thick fibrous connective tissue of the outer and inner wall of the arachnoid membrane. The hemosiderin, detected in the cyst wall may suggest a traumatic origin. However, we did not analyze such changes in the secretory properties of the tissues in our series.

#### CONCLUSION

The symptomatic EDAC presents with spectrum of symptoms ranging from compressive dorsal myelopathy to radiculopathy. The dural communications should be searched and separately sutured to prevent recurrence. Preoperative radiological localization of dural rent is helpful. Laminectomy with complete excision is standard care, but the trend toward selective closure of dural communication without even cyst excision is emerging.

#### Highlights

- 1. Extradural arachnoid cyst is rare clinical entity
- 2. Complete excision along with dis connection of dural communication is necessary to prevent recurrence
- 3. The cyst, although extradural, may present with clinical features of myelopathy

- 4. Minimally invasive percutaneous techniques are upcoming changing paradigm of management
- 5. Prognosis is good in these patients
- 6. COMI suggests satisfactory outcome.

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#### **Conflicts of interest**

There are no conflicts of interest.

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