Extradural Sacral Arachnoid Cyst - An Institutional Experience from Rajasthan, India

Jitendra Singh Shekhawat[1], Arvind Sharma[2], Jagdish Chaudhary[3], Ashish[4]

[1, 3, 4] Department of Neurosurgery, Sawai Man Singh Medical College, Jaipur, Rajasthan, India.
[2] Department of Surgery, Government Medical College, Churu, Rajasthan, India.

ABSTRACT

BACKGROUND
Arachnoid cyst of spinal cord (ACS) is a very uncommon lesion of the spinal cord. The problem with these lesions is that they can stimulate the pain of prolapsed intervertebral disc pathology and patient may be misdiagnosed for the same. In this study we wanted to diagnose and establish a surgical treatment in such patients.

METHODS
This study is a case series and comprised of 12 patients which was conducted in Department of Neurosurgery, Sawai Man Singh Medical College, Jaipur, in which all patients having arachnoid cyst of spinal cord were operated and follow-up for reliving of symptoms or development of new symptoms.

RESULTS
Our study demonstrates that surgical result is good when spinal arachnoid cyst is better. All patients who underwent surgery were having significant improvement in symptoms and quality of life.

CONCLUSIONS
Surgical management is primary treatment modality in patients of arachnoid cyst of spinal cord and the results of surgery in such patients are good.

KEYWORDS
Spinal Arachnoid Cyst, Lumber Pain, Radiculopathy

Corresponding Author:
Dr. Arvind Sharma,
 # 22, Sunder Nagar, Civil Lines,
Ajmer Road, Jaipur – 302006,
Rajasthan, India.
E-mail: dr_arvindsharma@yahoo.com

DOI: 10.18410/jebmh/2021/303

How to Cite This Article:
Shekhawat JS, Sharma A, Chaudhary J, et al. Extradural sacral arachnoid cyst - an institutional experience from Rajasthan, India. J Evid Based Med Healthc 2021;8(21):1603-1607. DOI: 10.18410/jebmh/2021/303

Submission 08-10-2020,
Peer Review 18-10-2020,
Acceptance 08-04-2021,
Published 24-05-2021.

BACKGROUND

Spinal arachnoid cyst is a rare lesion constituting less than 1 % of all spinal tumors.[1-5] Arachnoid cysts are out pouching's from spinal meningeal sac and can be intra or extradural in location. Spinal arachnoid cysts are commonly located in thoracic region (65 %) with very scant literature on cysts outside the thoracic cord.[3,5] These cysts can be congenital or secondary i.e. after trauma, infection, haemorrhage, spinal operative procedures and after spinal anaesthesia. ACS mimics prolapsed intervertebral disc in its presentation and hence without a spinal magnetic resonance imaging is often treated with analgesics and muscle relaxants. Once detected, its severity can be assessed using the Oswestry disability index (ODI) scale and this can also be reliably used in deciding further line of management.[6] Laminectomy with excision of cyst and repair of dural defect is the standard surgical practice.[3] Being an extremely rare lesion and with good neuro-surgical outcome, a closer look at this cystic lesion is warranted, herein we report a study comprising of 12 cases with extradural sacral arachnoid cyst. We also briefly discuss the aetiopathogenesis and various available treatment options.

METHODS

This study is case series in nature and was performed in the Department of Neurosurgery, Sawai Man Singh Medical College, Jaipur for a period of five years from January 2015 to December 2019. The study compromised of 12 patients, 8 males and 4 females. The mean age of the male patients was 46 years and the same for the female group was 36 years. The mean duration of presentation of symptoms in males were 2 years while in female group it was 2.2 years. In all these patients there was no history of previous spinal trauma or spinal surgical procedure nor history of any infection. All these patients were not involved in heavy work. In 9 patient symptoms were involving the right side of lower limb radiculopathy while rest of the patients were having in left lower limb. In all the patients the presentation was insidious in onset and gradually progressing.

Assessment of clinical outcome was done by Oswestry disability index scores and visual acuity scale (VAS), which were recorded for the clinical outcome pre-operatively and 6 months post-operatively.

Inclusion Criteria

1. Patients in between 30 to 60 year of age.
2. Patients having symptoms of radiculopathy in either of the lower limb
3. Patients having spinal arachnoid cyst in magnetic resonance imaging (MRI) lumber spine

Exclusion Criteria

1. History of previous spinal trauma
2. History of previous spinal surgical procedure
3. History of previous spinal infection.
4. On MRI if any disc pathology

In all patients, pain was dull aching, brought on walking, bending forward and relieving on rest. There were no sensory symptoms except pain, no weakness of lower limb and no suggestion of dysautonomia. There was no tenderness on palpation over lumbosacral spine and overlaying skin was normal in colour and texture. Further examination revealed hypoesthesia in perianal region in 4 patients with normal sphincter tone, superficial (anal and plantar reflex) and deep tendon reflexes (knee and ankle jerk). Mean ODI score was 42 % in males and 44 % in females and mean VAS score was 8 in both the groups.

All these patients were evaluated with first X-Ray of the lumbosacral region and it was inconclusive. Patients were further evaluated with MRI of lumbosacral spine with all the requisite sequence. MRI showed cerebrospinal fluid (CSF) intensity lesion on T1 and T2 weighted. No disc degeneration, or ligamentum flavum hypertrophy and no listhesis were seen. Tarlov cyst was ruled out as the lesion showed no extension along the nerve roots, leaving us with the possibility of extradural sacral arachnoid cyst.

Surgical Procedure

After evaluation, these patients planned for surgery. After explaining all the pros and cons of surgical outcome, consent was taken. All the patients underwent general investigation for the fitness for surgery and pre-anesthetic check-up was done. All the patients were operated in prone position with placing bolsters, one below upper part of chest and another one at lower end of abdomen with taking care of positional chest compression. All the pressure points were padded with cotton and a pillow was inserted underneath knee to prevent iatrogenic nerve injury. A standard midline incision made over lumbosacral region, the length of which depends upon the level involved. Dorsolumbar fascia was incised and paravertebral muscle was dissected subperiosteally to prevent unnecessary blood loss. Cyst was excised with repair of dural defect and was reinforced with fibrin glue. Drain was inserted in all these patients. Post-operative period of all the patients was uneventful. There was no CSF leak from the drain or from surgical wound after taking our drain. Patients improved in post-operative period with no radiculopathy in lower limbs.

RESULTS

The surgical outcome in these patients was remarkable. All patients were relived of radiculopathy symptoms on second post-operative day. The mean OSD score was 25 in males and 26 in female group. All these patients were followed up every month and after 6 months the mean ODI score was 16 in both male and female group.

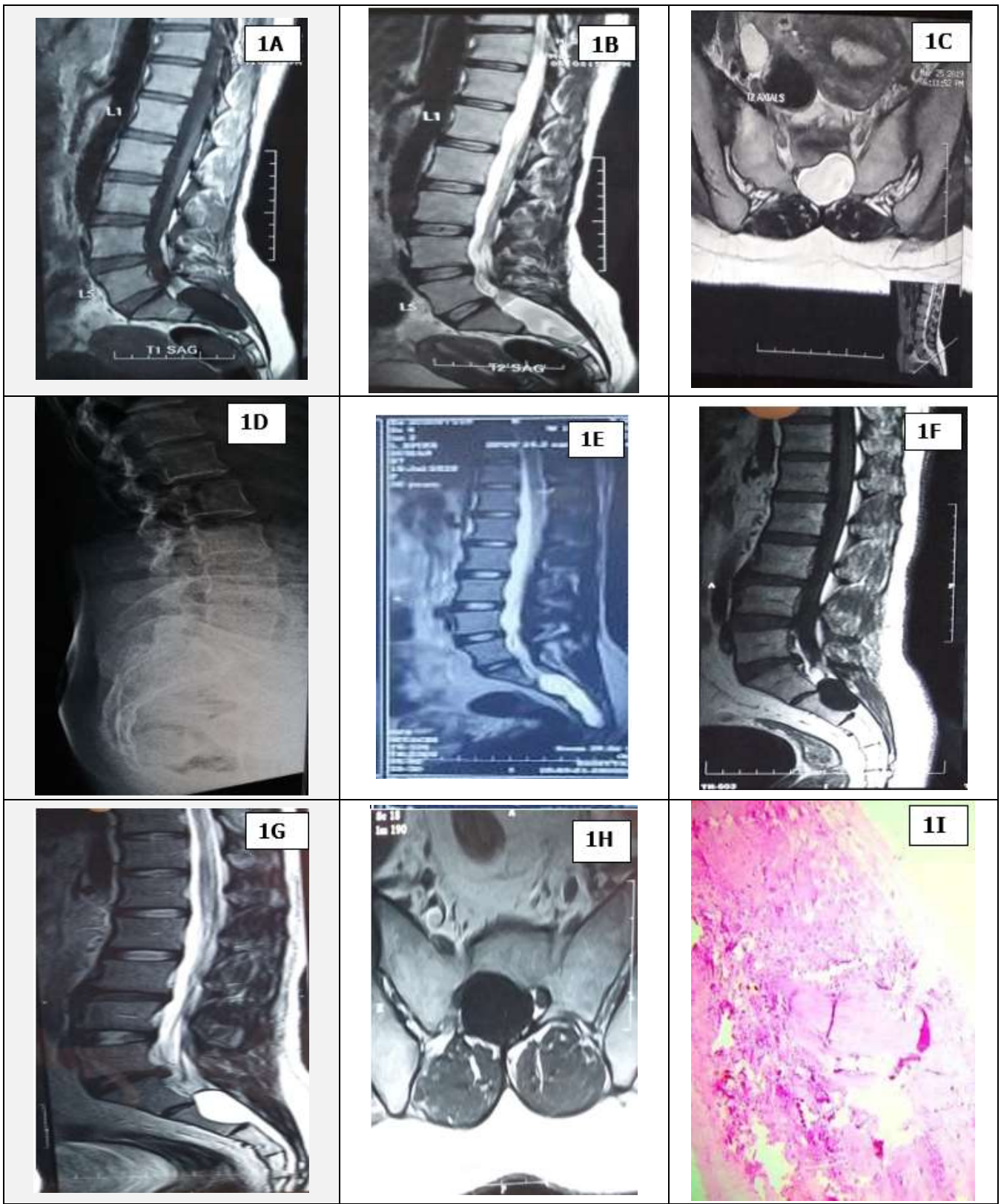

Figure 1A, 1B, 1C - Case 1: MRI Lumbosacral Spine (Sagittal T1, T2 and Axial T2) Shows a 3.2 cm X 2.6 cm Cerebrospinal Fluid Intensity Lesion Extending Dorsally from S1-S3 with Thecal Sac Compression and Mechanical Scalloping Effect on S1 -S3 Vertebrae.

1D- X Ray of Lumbosacral Spine (Lateral View) Showed Mild Erosion of S1-S2 Vertebrae.

1E- Case 2: MRI Lumbosacral Spine (Sagittal T2) Shows Extradural CSF Signal Intensity Cystic Lesion Measuring 3.2 cm X 6.0 cm in Sacral Spinal Canal Extending Over Dorsal S1 to S4 Vertebrae with Thecal Sac Compression and Mechanical Scalloping Effect on S1 to S4 Vertebra.

1F, 1G, 1H- Case 3- MRI Lumbosacral Spine (Sagittal T1, T2 and Axial T1) Shows Well Defined Extradural Cystic Lesion with CSF Intensity 2.8 cm X 2.2 cm Over Dorsal S1- S2 with Thecal Sac Compression with Mechanical Scalloping Effect on S2 Vertebra and No Extension along Nerve Roots.

1I- Haematoxylin and Eosin Stained Slide (100X) Shows Thin Arachnoid Layer with Meningothelial Cells Lining the Cyst Wall, Suggestive of Arachnoid Cyst

DISCUSSION

Arachnoid cyst of spinal cord was first described in 1903 by Spiller. Nabors et al. classified spinal meningeal cysts into three types.[7]

- Type 1, extradural cysts with nerve root fibres;
- Type II, extradural cyst without nerve root fibres;
- Type III, intradural cysts or Tarlov cyst.

Type 1 is further sub classified into type IA (extradural arachnoid cyst) and type IB (sacral meningocele). Type 1 and Type II extradural cysts are uncommon compared to Type III intradural cysts. Dentate ligament and septum posticum maintain position of spinal cord in the canal. It is hypothesized that degeneration of septum posticum can lead to entrapped cyst formation in sub arachnoid space.[3]

Different theories have been put forward to explain the cyst expansion. The more widely accepted theories are mentioned below:

a. Rohrer ball valve mechanism: The protrusion of arachnoid from dural rent to extradural space allows the CSF to accumulate by ball valve mechanism into cyst leading to its progressive enlargement.[5]
b. Osmotic enlargement of cyst: The hyperosmolar cyst fluid is capable of gradual absorption of water. The arachnoid lining of the cyst's wall can further secrete CSF and together these two processes contribute to progressive enlargement.[3]
c. Kulkarni et al.: Cyst wall can produce CSF or absorb CSF from the subarachnoid space resulting in cyst expansion.[2]
d. Seung et al. observed that stuck nerve rootlet(s) in dural defect prohibit spontaneous closure of cyst and could contribute to increase in its size.[8]

Arachnoid cyst can be congenitally associated with split cord malformation or dysraphism. Associations with familial syndromes such as Milroy's disease and distichiasis have also been reported.[3]

MRI remains investigation of choice for arachnoid cyst. All our patients had typical arachnoid cyst MRI findings, that is CSF like intensity on T1 and T2 weighted sequences with thecal sac compression and mechanical scalloping effect on vertebrae with widening of spinal canal and thinning of bony elements. Parasitic cyst such as cysticercosis and hydatid cyst were ruled out as arachnoid cysts are homogenous and show CSF intensity in all sequences.

Currently there are no consensus guidelines on management available. The rarity of the lesion does not give a scope for large studies to be conducted. The different modalities of treatment being followed include:

- Conservative symptomatic management with radiological follow-up.
- Cysto-peritoneal shunt.
- Laminectomy with excision of cyst (partial or complete excision) and closure of dural defect.
- Laminoplasty as an alternative to laminectomy in young patients.

Different schools of thought exist for choosing one option over the other. Some authors favour conservative management with radiological follow up due to post-operative risk of kyphosis. Risk of cord herniation and paraplegia with conservative management raise doubts on this choice. Previously, cysto-peritoneal shunt was favoured by many surgeons. However, cysto-peritoneal shunt with its high recurrence rates is no longer preferred, this is so especially with availability of better surgical options.[3] In current practice, selective laminectomy with excision of cyst and dural defect repair is considered standard choice of care.[3] Partial excision of cyst can be done if a risk of nerve injury exists. In their study on 52 patients with spinal extradural arachnoid cyst Lee et al. noted that recurrence is related to inadequate closure of dural defect and not on the extent of excision.[9]

Similar results were observed by Singh S. et al. in their study on spinal arachnoid cyst conducted over a period of 10 years on 19 patients.[3] Further, Kumar et al. also emphasized on adequacy of dural defect closure and reported no recurrence in their study.[10] Laminoplasty could be considered an alternative to laminectomy in young patients to prevent risk of kyphotic deformity post operatively. However, Lee et al. reported kyphosis even after laminoplasty, raising doubts on which procedure is better [9]. They also mentioned that laminoplasty is technically difficult in lumbar and thoracic spine due to anatomy of pedicle and high risk of pedicle fracture.[9] Singh S. et al. corroborated that kyphosis can occur even after laminoplasty, especially when more than five levels were involved.[3]

All our patients were selected for surgical intervention in view of moderate disability on ODI Score. They underwent laminectomy with excision of cyst and repair of dural defect with fibrin glue and cyst wall was sent for histopathological examination. In the immediate post-operative period all patients had significant pain relief and on 1 year follow up continue to be pain free. Histopathological examination of all the patients showed thin arachnoid layer with meningothelial cells lining the cyst wall, suggestive of arachnoid cyst (Fig. 1I).

CONCLUSIONS

Symptomatic extradural arachnoid cyst can present as low back pain and radiculopathy. Extradural ACS is an extremely rare entity with excellent surgical outcome. Risk-benefit ratio should be assessed before deciding (conservative / surgical) management. ODI score is a helpful tool to quantify the symptom severity and guide management. Through our experience we contribute valuable literature to the existing scant knowledge on extradural sacral arachnoid cysts and demonstrate a complication free excellent surgical outcome following single level laminectomy with excision of cyst and dural defect repair in our patients.

Data sharing statement provided by the authors is available with the full text of this article at jebmh.com.
Financial or other competing interests: None.

Disclosure forms provided by the authors are available with the full text of this article at jebmh.com.

REFERENCES

[1] Choi JY, Kim SH, Lee WS, et al. Spinal extradural arachnoid cyst. Acta Neurochir (Wien) 2006;148(5):579-585.

[2] Kulkarni AG, Goel A, Thiruppathy SP, et al. Extradural arachnoid cysts: a study of seven cases. Br J Neurosurg 2004;18(5):484-488.

[3] Singh S, Bhaisora KS, Sardhara J, et al. Symptomatic extradural spinal arachnoid cyst: More than a simple herniated sac. J Craniovertebr Junction Spine 2019;10(1):64-71.

[4] Rohrer DC, Burchiel KJ, Gruber DP. Intraspinal extradural meningeal cyst demonstrating ball-valve mechanism of formation. Case report. J Neurosurg 1993;78(1):122-125.

[5] Liu JK, Cole CD, Kan P, et al. Spinal extradural arachnoid cysts: clinical, radiological and surgical features. Neurosurg Focus 2007;22(2):E6.

[6] Fairbank JC, Pynsent PB. The Oswestry disability index. Spine (Phila Pa 1976) 2000;25(22):2940-2952.

[7] Nabors MW, Pait TG, Byrd EB, et al. Updated assessment and current classification of spinal meningeal cysts. J Neurosurg 1988;68(3):366-377.

[8] Choi SW, Seong HY, Roh SW. Spinal extradural arachnoid cyst. J Korean Neurosurg Soc 2013;54(4):355-358.

[9] Lee CH, Hyun SJ, Kim KJ, et al. What is a reasonable surgical procedure for spinal extradural arachnoid cysts: Is cyst removal mandatory? Eight consecutive cases and a review of the literature. Acta Neurochir (Wien) 2012;154(7):1219-1227.

[10] Kumar A, Sakia R, Singh K, et al. Spinal arachnoid cyst. J Clin Neurosci 2011;18(9):1189-1192.