

Case Report

Giant Iatrogenic Lumbar Pseudomeningocele: A Case Report and Literature Review

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ABSTRACT

Post-discectomy iatrogenic lumbar pseudomeningoceles are an uncommon complication. This pathology is an extradural, encapsulated, cerebrospinal fluid collection which develops at the site of previous surgery as the consequence of an unnoticed or unrepairable dural tear. A pseudomeningocele is defined as giant if it grows beyond 8 cm in length. Giant iatrogenic pseudomeningoceles with detailed information are quite rare in the literature with only 21 reported cases including the current case. Herein, we present a middle age woman with a giant pseudomeningocele which had developed subsequent to L4-L5 discectomy.

Keywords

Iatrogenic lumbar pseudomeningocele; Pathology; Discectomy.

INTRODUCTION

Post-discectomy pseudomeningocele was reported first in 1947 by Swanson and Fincher.¹ Three-years later in 1950, Winkler et al² reported two additional cases of a pseudomeningocele after a lumbar discectomy. In 1968, Miller et al³ reported 10 new cases and classified pseudomeningoceles into congenital, iatrogenic and traumatic. Congenital pseudomeningoceles have been described in the patients with Marfan disease and neurofibromatosis mostly in the thoracic and lumbosacral regions respectively.^{4,5}

Majority of the traumatic pseudomeningoceles develop subsequent to blunt traumatic events and are mostly seen in the cervical spine, but seldom in the lumbar region or with penetrating injuries.⁶⁻⁸

Iatrogenic pseudomeningoceles may occur in the lumbar, cervical or thoracic regions in decreasing frequency.⁹⁻¹³ In the lumbar region, they are mostly seen following the laminectomy for lumbar disc herniation or canal stenosis.

The cause of these cystic cerebrospinal fluid-containing lesions are incidental dural tears.⁹⁻¹³ The growth of the iatrogenic lumbar pseudomeningoceles is limited but in rare occasions it

may continue to grow, till the pressure of the cyst's contents and the surrounding tissues reach to equilibrium.⁹⁻¹³ Most of the pseudomeningoceles remain relatively minute (below 5 cm in size) and a small number will grow above 5 cm in size which are classified as large. Very rarely this pathologic sac may grow beyond 8 cm being classified as giant subtype.¹⁴⁻¹⁶

In 1963, Miller et al³ were the first to report 3 cases with giant iatrogenic pseudomeningocele and since then several cases with detailed information have been published so far.¹⁷⁻²⁷ Herein, we present a middle age woman with giant pseudomeningocele which had developed after L4-L5 discectomy. With consideration of the previously published cases of giant pseudomeningocele with detailed information, the current case will be the 21st in the literature. The detailed information of 21 cases with iatrogenic giant lumbar pseudomeningoceles including the current case are presented separately (Table 1).

CASE PRESENTATION

A 49-year-old female who had undergone a lumbar laminectomy for a central L4-L5 disc herniation eight months earlier was referred to our facility after the appearance of a large subcutaneous midline lumbar mass which was associated with severe low back

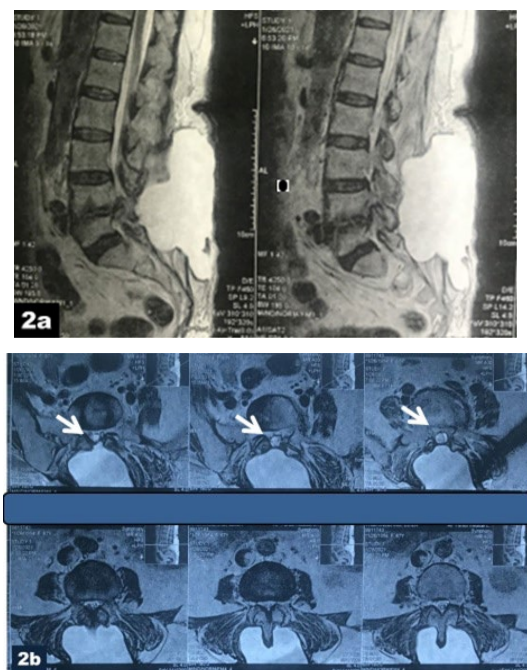
Table 1. Data on the Surgical Hardware Used in the Course of Surgical Treatment of Patients with Fracture-Dislocations of the PH

Author	Year	Age	Sex	Lumbar Procedure	Clinical Picture	Outcome
Miller et al ³	1963	F	40	Low lumbar laminectomy	Low back and sciatic pain	Good
Miller et al ³	1963	M	35	L5- S1 laminectomy	S1 radiculopathy	Good
Miller et al ³	1963	M	31	L4-L5 laminectomy	Headache & LBP relieved by lying down	Good
Rinaldi et al ¹⁷	1969	50	M	L3-L4 hemilaminectomy	Radiculopathy	Good
Rinaldi et al ¹⁷	1969	42	F	L5-S1 discectomy	LBP, bilateral S1 radiculopathy	Good
Rinaldi et al ¹⁸	1970	49	M	Subsequent to discectomy for recurrent L4-L5 disc	LBP, Unilateral, radiculopathy	Good
Schumacher et al ¹⁹	1988	39	M	L3-L4 hemilaminectomy+ discectomy,	Radiculopathy	Good
Lee et al ²⁰	1992	18	M	Right L4-L5 discectomy+ PLIF	Persistent LBP, Right radiculopathy	Good
Lee et al ²⁰	1992	25	F	Right L4-L5 hemilaminectomy	Palpable mass, L5 Paresthesia	Good
Pavlou et al ²¹	2005	59	F	L4-L5 discectomy	Weakness of dorsiflexion	Good
Hamilton et al ²²	2009	51	M	3 time surgery for L3-L4 spondylolisthesis	Retropertitoneal mass on left ureter	Good
Weng et al ²³	2010	26	F	L4-L5 laminectomy discectomy	Not described	Good
Liu et al ²⁴	2011	40	M	L4-L5 laminectomy& Screw-rod fixation	Radiculopathy & Postural headache	Good
Alvarez et al ²⁵	2018	39	M	L4-L5 interlaminar-Laminectomy + discectomy	LBP & radiculopathy	Good
Alvarez et al ²⁵	2018	92	M	L3-L4 & L4-L5 laminectomy	LBP, Right leg radiculopathy	Good
Eneke et al ¹⁵	2018	64	F	L4 & L5 laminectomy, L4-L5 discectomy	LBP, Radiculopathy, N claudication	Fair
Hamdan et al ²⁶	2018	26	F	L5-S1 discectomy	Episodes of black out while lying	Good
Rahimizadeh et al ¹⁴	2019	30	F	L5-S1 interlaminar laminectomy+ discectomy	LBP & left S1 radiculopathy	Good
Rahimizadeh et al ¹⁴	2019	52	F	Multilevel laminectomy	Neurogenic claudication	Good
Jah	2021	38	M	L4-L5Inter- laminectomy, discectomy	LBP & subcutaneous lump	Good
Current case	2021	49	F	L4-L5Inter- laminectomy, discectomy	LBP & Left L5 radiculopathy, Lump	Good

Figure 1. A Photograph of the Lumbar Region; Shows a Giant Subcutaneous Lump



Figure 2. (a) T2 Weighted Sagittal MRI, Shows a Giant Pseudomeningocele Extending from L2 to S4, (b) T2 Weighted Axial Images Showing the Narrow Stalk of the Pseudomeningocele (white arrows)



pain for a duration of 3-months (Figure 1). Neurological examination of motor and sensory nerves was normal. With the diagnosis of a pseudomeningocele, magnetic resonance imaging (MRI) was performed and displayed a very large pseudomeningocele at the site of the previous surgery. The pseudomeningocele was extend-

ed from L2 to S4, with a total length of 13 cm (Figure 2).

At surgery, the abnormally thick wall of the sac was widely opened. After evacuation of cerebrospinal fluid and at the depth of the cavity; a small breach with slow flow of cerebral spinal fluid

(CSF) was found (Figure 3). The defect was subsequently enlarged and closed tightly with interrupted silk sutures. This was subsequently covered with free fat graft being reinforced with suturing of the lower part of the pseudomeningocele's fibrotic wall. This was followed by a placement of a drain and closure of the wound in three layers. The pathological result of the wall of the pseudomeningocele was composed of connective tissue, mainly

fibroblasts being aggregate with foamy macrophages (Figure 4). The patient recovered well-during the 4-days hospital stay and all her complaints had ceased at the time of the one month follow-up encounter. The patient had complete relief of her back pain and was doing well 5-months post-operatively. Successful excision and closure of the pseudomeningocele was confirmed in a MRI taken at six months follow-up (Figure 5).

Figure 3. (a) An Intraoperative Photograph Showing a Breach is Demonstrated at the Bottom of the Surgical Scene. (b) An Intraoperative Photograph showing a Pediatric Nasogastric Tube Inserted intrathecal via the Breach Only to Show its Communication with Thecal Sac, so was Later Removed. (c) After Closure of the Breach, Reinforced with a Second Layer Composed of the Fibrotic Cyst's Wall

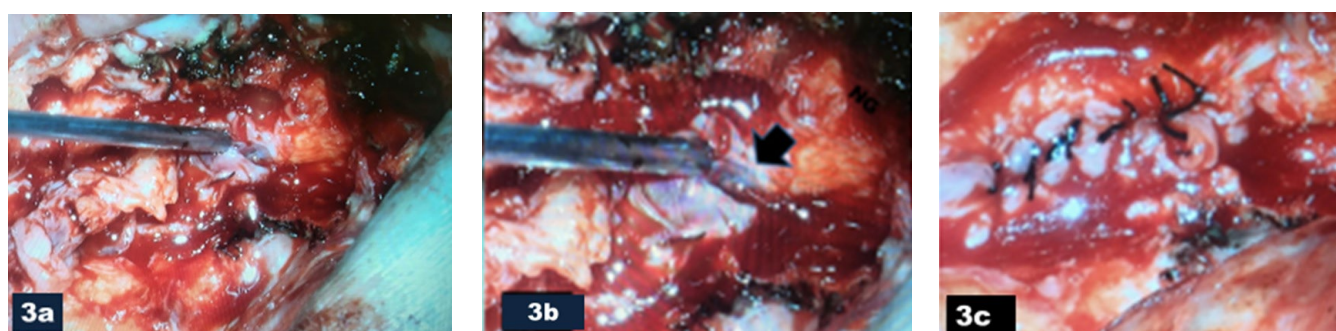


Figure 4. Histopathologic Result of the Cyst's Wall (a) Note an External Layer being Composed of Fibroblasts. This Layer is Aggregated with Foamy Macrophages. No Epithelial Lining is Seen. (b) Histopathologic Result of the Cyst's Wall with Higher Magnification

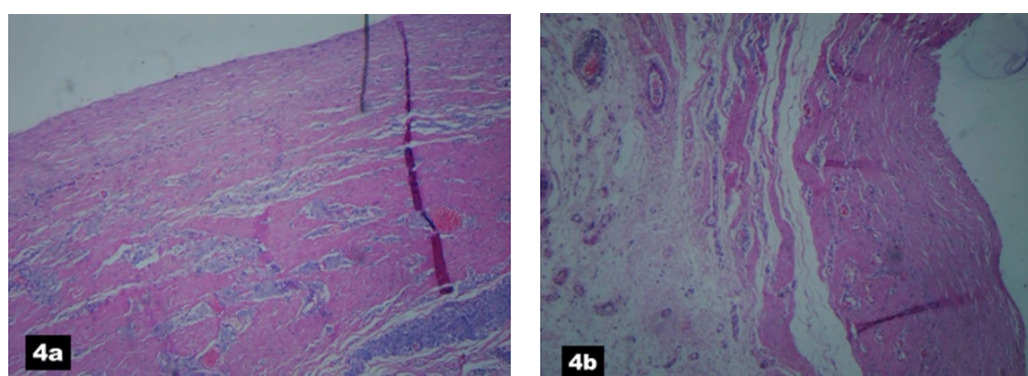
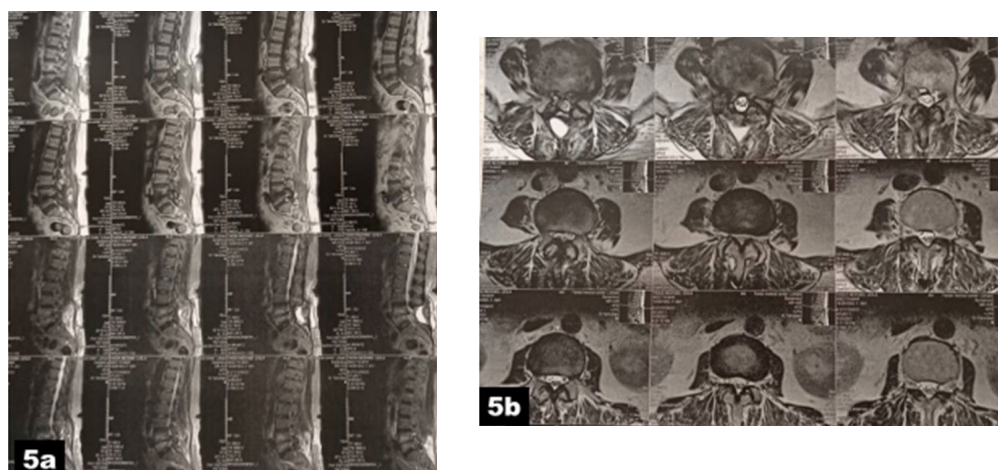


Figure 5. Post-Operative Sagittal and Axial Lumbar Spine MRI at 6-months Follow-up, (a & b) both T1 and T2-Weighted Sagittal and Axial Images after Excision of the Pseudomeningocele, the Small Hyperintense Mass is Probably Free Fat Graft



DISCUSSION

Lumbar pseudomeningoceles are uncommon complication of lumbar disc surgeries which develop as a consequence of an incidental or unrepairable dural tear. The true incidence of post-discectomy lumbar pseudomeningocele is difficult to ascertain because many remain asymptomatic.¹⁶ However, it is estimated to be between 0.1 and 2%.^{14,19}

A small dural tear with concomitant arachnoid layer perforation results in a gradual extradural accumulation of the cerebrospinal fluids.^{14,16,28} According to one theory, the small dural tears leads to a higher probability of pseudomeningocele formation with respect to ball-valve mechanism.^{28,29} With subsequent reactive fibrosis, the fluid will be enveloped resulting in a cystic mass with false walls or pseudomeningoceles.¹⁶⁻²⁷

Unnoticed dural tear with intact arachnoid and a ball valve mechanism will result in the development of a true cyst lined with arachnoid.²⁹ This type of iatrogenic cyst is called true meningocele where surrounding connective tissue might reinforce the arachnoid capsule with time.^{14,16,28}

Higher frequency of the pseudomeningoceles in the lumbar region in comparison to thoracic and cervical region is due to the relatively higher CSF pressure in the caudal thecal sac and the more frequency of lumbar disc surgeries.

The size of the lumbar pseudomeningoceles in majority remain minute, but in a small number of the cases, in a time frame which varies from a few months to a year after laminectomy, the pathology continue to grow till it reaches to an equilibrium. In rare occasions, a pseudomeningocele might grow beyond 8 cm.

Weng et al²³ have attributed the giant size of a pseudomeningocele to high body mass index. However, we believe that intra-operative extensive dissection of the paravertebral muscles and fatty degeneration of these muscles might be the other predisposing factors.

Clinically, most of the lumbar pseudomeningoceles remain small and asymptomatic, this is in contrast to symptomatic ones which are relatively large. Large pseudomeningoceles might present a large subcutaneous lump. Nonetheless, in symptomatic cases, low back pain (LBP) which characteristically tends to be aggravated with straining and Valsalva maneuver is the most frequent clinical feature.³⁰ If a rootlet is extruded through the breach and trapped within, radiculopathy may coexist.^{16,31-33} Such manifestation is clinically quite similar to a recurrent lumbar disc herniation.^{16,31-33} Rarely, in those with anterior dural breach, some rootlets might be trapped in corresponding collapsed intervertebral disc space.³⁴ Occasionally, lower limbs motor dysfunction and incontinence and even *cauda equina* syndrome might occur.³⁵ Headache as well as syncope and hypotension may be caused by compression of the subcutaneous lump of the pseudomeningocele. Positional headache has been also described in an iatrogenic lumbar pseudomeningocele.³⁶ Positional syncope is another rare presentation of pseudomeningoceles which has been reported in a case report.

The patient's symptom disappeared following the excision of the pseudomeningocele.³⁷ Headache due to chronic subdural hematoma is another rare complication of the pseudomeningoceles.³⁸ Clinical features of meningitis have been reported in an infected pseudomeningocele.^{39,40}

An abdominal mass due to retroperitoneal growth of a pseudomeningocele is another rare presentation of the scenario.⁴¹ Hydronephrosis secondary to ureteral obstruction caused by a retroperitoneal growth of a giant pseudomeningocele has been reported in a rare case report.²²

On plain radiographs, erosion of the surrounding bones might be seen in long standing cases.⁴² Ossification of the cyst's wall is an infrequent scenario.⁴³⁻⁴⁷

The degree and extent of ossification can be best demonstrated in reconstructed computed tomography (CT) images.⁴⁷

Computed tomography myelography can detect the small pseudomeningoceles, even in those tiny ones that grow intraosseously.⁴⁸

Magnetic resonance imaging remains the most useful diagnostic tool for the demonstration of a pseudomeningocele and its short fistulous tract. A pseudomeningocele displays low signal intensity in T1-weighted and high signal intensity in T2-weighted MRI images.⁴⁹ Furthermore, the measurement of the length of the pseudomeningoceles and their classification to minute, large and giant became possible with the aid of MRI.¹⁶⁻²⁷ Generally, this specific CSF-containing mass is located posterior to the dural sac; although in rare instances it might grow into the intervertebral disc space and even progress into retroperitoneal space.^{22,50}

Owing the high chances of spontaneous regression, conservative treatment is recommended for asymptomatic cases.⁵¹ Spontaneous regression may occur within 3-months to a few years following the diagnosis.⁵¹ According to Solomon et al⁵¹ healing of the dural defect with the gradual resorption of cerebrospinal fluid is the possible mechanism for the spontaneous resolution of pseudomeningoceles.^{16,52} Surgery for a symptomatic lumbar pseudomeningocele starts with widely opening the cyst and closure of the dural breach itself.¹⁴⁻²⁰ For closure of the breach, both interrupted suture and titanium U shape clips can be used.^{14-16,52,53} In the case of a radiculopathy where entrapment of a rootlet is responsible; reduction of the rootlet into the thecal sac through the breach is the key to adequate treatment.^{16,31-33}

Following the closure of the breach, lumbar myofascial flap was introduced by Misra et al.⁵⁴ Myofascial flap which can be achieved with advancement of lumbar paravertebral muscles has been advocated in those with large dead space.

CONCLUSION

Incidental dural tears with CSF leakage during lumbar laminectomy should be properly addressed. Lumbar pseudomeningocele should be suspected in patients with a delayed reappearance of

lower back pain or radiculopathy within a few months to several years after the initial laminectomy. Appropriate surgical intervention should be decided upon and undertaken once the diagnosis is reached. There remains no difference in the management of large and giant pseudomeningoceles.

CONSENT

The authors have received written informed consent from the patient.

CONFLICTS OF INTEREST

The authors declare that they have no conflicts of interest.

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