



Case Report

Giant Tarlov Cyst presenting as pelvic mass: Often doing less is better

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ABSTRACT

Background: Tarlov cysts are sacral perineural cysts arising between the peri and endoneurium of the posterior spinal nerve root at the Dorsal Root Ganglion and have a global prevalence rate of 4.27%. These are primarily asymptomatic (only 1% with symptoms) and typically arise in females between the ages of 50–60. Patients' symptoms include radicular pain, sensory dysesthesias, urinary and/or bowel symptoms, and sexual dysfunction. Non-surgical management with lumbar cerebrospinal fluid drainage and computerized tomography-guided cyst aspiration typically provide only months of improvement before recurring. Surgical treatment includes a laminectomy, cyst, and/or nerve root decompression with fenestration of the cyst and/or imbrication. Early surgery for large cysts provides the longest symptom-free periods.

Case Description: A 30-year-old male presented with a very large magnetic resonance-documented Tarlov cyst (Nabors Type 2) arising from bilateral S2 nerve root sheaths with marked pelvic extension. Although he was initially treated with a S1, S2 laminectomy, closure of the dural defect, and excision/marsupialization of the cyst, he later required placement of a thecoperitoneal shunt (TP shunt).

Conclusion: A 30-year-old male with large Nabors Type 2 Tarlov cyst arising from both S2 nerve root sheaths required a S1-S2 laminectomy, dural closure/marsupialization, and imbrication of the cyst, eventually followed by placement of a TP shunt.

Keywords: Nabor cyst, Pelvic cyst, Spine, Tarlov cyst, Theco-peritoneal shunt

INTRODUCTION

We present a 30-year-old male whose lower extremity radicular and sphincteric complaints attributed to an magnetic resonance (MR)-documented Type 2 Tarlov cyst arising from both S2 nerve root sheaths. Although he underwent a laminectomy, closure of the bilateral dural fistulas with imbrication/marsupialization of the sacral cysts, he ultimately required placement of a thecoperitoneal (TP) shunt.

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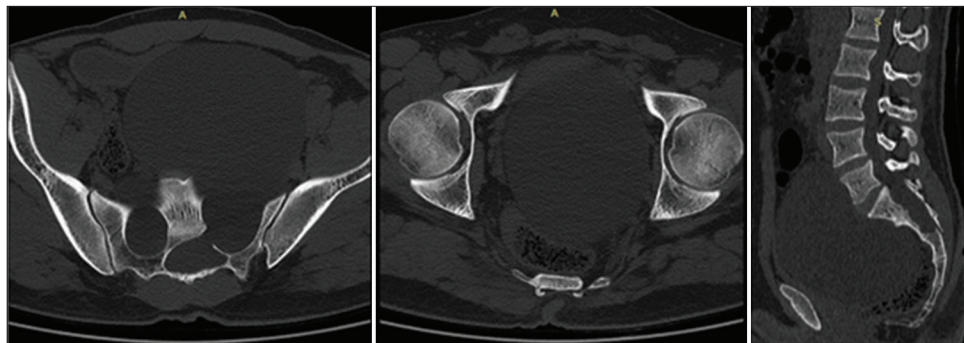


Figure 1: Axial and sagittal computerized tomography scan of Lumbo-Sacral (LS) spine. Hypodense Expansile cystic lesion is seen filling the pelvic cavity reaching almost up to the anterior abdomen wall. It displaces the bladder and rectum posteroinferior with bony erosions of the sacrum from S2-4. Note the simultaneous spinal canal expansion at the S2-4 level.

CASE DETAILS

A 30-year-old male presented with posterior left thigh pain of 18 months' duration and 1 year of urinary frequency/urgency, constipation, and scrotal/perineal pain. The lumbosacral MR imaging (MRI) revealed a Tarlov cyst (Nabors Type 2) arising from bilateral S2 nerve root sheaths with marked pelvic extension.

Computerized tomography (CT) and MR findings

A Nabors-2 Tarlov cyst was diagnosed based on both lumbosacral CT and MR studies. The CT showed a hypodense lesion filling the pelvic cavity origination from bilateral S1 sacral roots, accompanied by chronic bony erosion of the sacrum at multiple levels (i.e., specially from S2 to 4) [Figure 1]. The MR showed cerebrospinal fluid [CSF] signal cystic lesions with air-fluid levels, communicating with the thecal sac bilaterally, involving bilateral S1 and S2 neural foramina, and demonstrating anterior extension [Figure 2]. The right lesion contained the S2 nerve root (measured $4.5 \times 3.3 \times 3.7$ cm [Antero-Posterior (AP) x Transverse (TR) x Cranio-caudal (CC)]), while the left lesion ($11 \times 9.5 \times 9.7$ cm [AP x TR x CC]) contained the left S2 nerve root. The extremely large cyst displaced the urinary bladder and adjacent bowel loops inferolaterally. Notably, the cyst additionally contained irregular T2 hypointense septae. Further, the dural sac herniated bilaterally through the posterior lamina of the S1 and S2 vertebrae.

Surgery

Following a S1, S2 laminectomy, dural fistula closure, and excision/marsupialization of the cyst, he remained asymptomatic for a few months. However, 6 months later symptoms recurred at which point he underwent, lumbar drainage that relieved his urinary complaints; a TP shunt was then placed. At surgery, two separate large tense cysts were encountered extradurally surrounding the bilateral S2; these cysts extended ventrally into the pelvis. Another small cyst

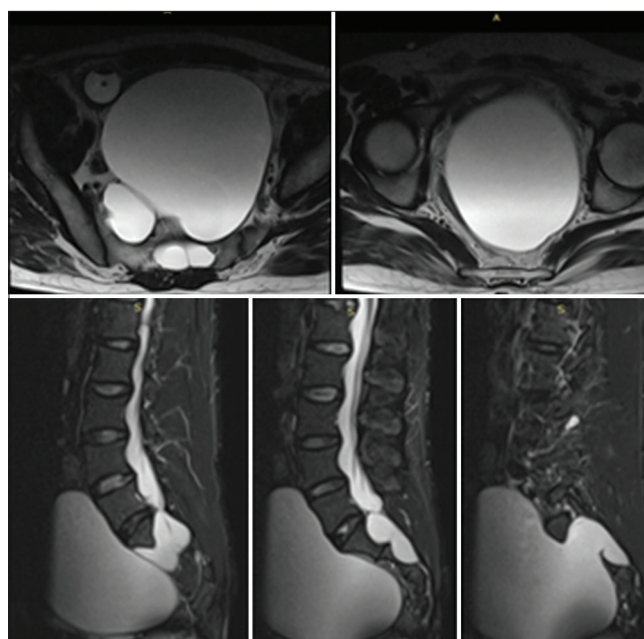


Figure 2: Preoperative axial and sagittal T2-W magnetic resonance imaging scan Lumbo-Sacral (LS) spine. The T2-hypointense lesion originates from bilateral neural foramen at the level of S2. Appreciate the septa between two sides of the cavities within the pelvis. Bony elements are seen missing at levels S2-4.

compressed the left S1 root as it exited the canal. The three cysts were incised and drained (i.e., around 1.5 L of Cerebro-Spinal Fluid (CSF)). Biopsies were taken from the cyst walls, and the cysts were marsupialized. In addition, gel foam was placed over the neck of the bilateral cysts bilaterally in conjunction with the application of fibrin glue (i.e., over the dura and cyst necks).

Postoperative MR imaging

The postoperative spine/pelvic MR revealed air-fluid levels in both cysts which were reduced in size vs. the preoperative scans.

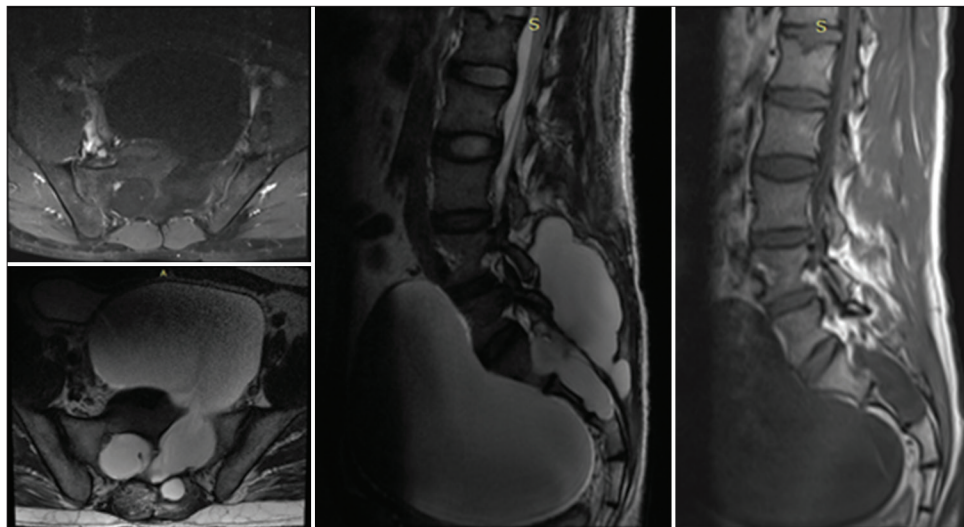


Figure 3: Axial and sagittal magnetic resonance imaging scan of Lumbo-Sacral (LS) spine 6 months after index surgery. T1-hypointense and T2-hyperintense lesion is seen reappearing in the pelvic cavity. Posteriorly, the collection of cerebrospinal fluid can be noted at the level of surgery/laminectomy.

Postoperative symptoms

Postoperatively, the patient had severe headaches that resolved within the 1st postoperative day by keeping the head of bed flat. By postoperative day 2, there was complete resolution of the deep pelvic pain and no further urinary symptoms. The remainder of the postoperative course was uneventful, and the patient was discharged on postoperative day 5.

Recurrent symptoms requiring placement of a TP shunt

Within 6 postoperative months, the patient presented with recurrent symptoms (i.e., pelvic pain and urinary dysfunction). The lumbosacral MR spine now revealed a posterior dural sac-CSF collection, measuring $4 \times 4.4 \times 10$ cm (AP \times TR \times CC) with recurrent bilateral presacral tarlov cysts communicating with the thecal sac (i.e., through the neural foramina of S1-S2, right Cyst $4.6 \times 3.3 \times 4.2$ cm, left cyst $12.3 \times 12 \times 15.4$ cm [AP \times TR \times CC]) [Figure 3]. Cysts extended into the pelvis, causing compression of the urinary bladder/rectum, and were notably larger than the initial cysts, but did not contain the previously noted septae. A lumbar drain was inserted at the L3-L4 disk space, that improved the patient's symptoms, and was followed by application of a TP shunt at the L4-L5 level [Figure 4]. The postoperative course was uneventful, the patient remained asymptomatic up to the 1-year follow-up examination.

DISCUSSION

Tarlov cysts are CSF-filled cavities contained by an epithelium first described in cadaveric surgery by Tarlov.^[5,7] With a global prevalence rate of 4.27%,^[3,14] they form

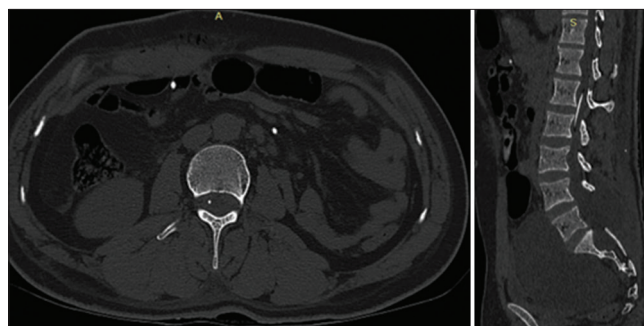


Figure 4: Axial and sagittal computerized tomography scan revealing Theco-peritoneal (TP) shunt tip in the cavity of the spinal canal at the level of L1 vertebral body and reduction in the size of the cyst within the pelvic cavity. The white dots of the TP shunt can be seen within the peritoneal cavity.

between perineurium and endoneurium at dorsal nerve roots (DRG) and mostly at sacral levels.^[16] They are reported as incidental findings 99% times because they are asymptomatic at the same percentage. If symptomatic, radiculopathy and perineal pain is the usual presentation along with urinary disturbances. Symptomatic patients constitute only 1% of the cases and can present with radicular pain, sensory dysesthesias, urinary and/or bowel symptoms, and sexual dysfunction.^[4,5,10] Rare presentation of Tarlov cyst being mistaken for hydrosalpinx had also been reported.^[1,29] Non-surgical management with lumbar CSF drainage and CT-guided cyst aspiration is scarcely reported to provide months of symptom-free periods before recurrence.^[20] Neurosurgical interventions include laminectomy, cyst and/or nerve root excision, and fenestration of cyst and imbrication.^[19,22,26,28] Interestingly, a double crush syndrome of concomitant Tarlov

Table 1: Literature Review on Tarlov Cyst with emphasis on Treatment, Recurrence, and special observations.

S. No.	Author, Year	Location	Age gender	Presenting symptom	Single/Multiple	Sacral erosion	Treatment	Follow-up	Outcome	Remarks
1.	J M Voyadzis, ^[35] 2001	S2	46 M	Back, leg pain	Single	Present	Microscopic cystectomy	22 months	Complete Resolution	Associated popliteal fossa Baker cyst.
		S1	38 M	Back, leg pain	Single	Present	Microscopic cystectomy	18 months	Complete Resolution	
		S1	45 F	Bowel and bladder dysfunction	Single	Present	Microscopic cystectomy	3 months	Complete Resolution	Associated spina bifida, tethered cord
2.	Curtis W Slipman, ^[31] 2003		47 F	Abdominal pain, bowel and bladder dysfunction, back, leg pain	Single	Present	Conservative treatment	NR	Persistent symptomatology	Retroperitoneal extension
3.	Hefli and Landolt, ^[10] 2006		55 M	Bowel and bladder dysfunction, impotence, back, leg pain and	Multiple	Present	Resection and imbrication	6 months	Complete resolution	Marfan syndrome
4.	Masato Tanaka, ^[33] 2006	S3	64 F	Bowel and bladder dysfunction, back, leg pain	Single	Present	Imbrication	6 months	Improvement	Cerebellar bleeding
5.	Dongsheng Guo, ^[9] 2007		44 M	Motor disturbances, back, leg pain	Multiple	Absent	Resection and imbrication	156 months	Improvement	
			36 M	bowel and bladder dysfunction	Multiple	Absent	Resection and imbrication	3 months	Worsening Persistent symptomatology	Post-operative CSF leakage
			40 F	Motor and sensory disturbances, bowel and bladder dysfunction, back, leg pain	Single	Absent	Resection and imbrication	30 months	Improvement	
			36 F	Motor and sensory disturbances, bowel and bladder dysfunction, back, leg pain	Single	Absent	Resection and imbrication	28 months	Improvement	
			34 F	Motor and sensory disturbances, bowel and bladder dysfunction, back, leg pain	Single	Absent	Resection and imbrication	2 months	Improvement	
6.	Kazuhiko Ishii, ^[13] 2007		29 F	Back, leg pain, sensory disturbances, bowel and bladder dysfunction	Single	Present	Ligation	1 month	Improvement	Associated infertility
7.	M W C H'ng, ^[12] 2009	S1-S2	29 F	Abdominal pain, motor disturbances	Multiple	Absent	Conservative treatment	NR	Improvement	Mimicking adnexal mass
		S1	29 F	motor disturbances	Single	Absent	Conservative treatment	NR	NR	Mimicking adnexal mass
		S2	26 F	Abdominal Pain Asymptomatic	Multiple	Absent	Conservative treatment	NR	NR	Mimicking adnexal mass

(Contd...)

Table 1: (Continued).

S. No.	Author, Year	Location	Age gender	Presenting symptom	Single/Multiple	Sacral erosion	Treatment	Follow-up	Outcome	Remarks
8.	Jane E Hirst, ^[11] 2009	Sacral	76 F	Back, leg pain, bowel and bladder dysfunction	Multiple	Absent	Microscopic cystectomy	1 month	NR	Inadvertent iatrogenic injury to tarlov cyst mistaken for a adnexal mass
9.	Smith ZA, ^[32] 2011		24 F	Back, leg pain, Motor and sensory disturbance	Single	Absent	Microscopic fenestration	16 months	Improvement	
10.	Shunji Asamoto, ^[4] 2013	S3	29 F	Back, leg pain, sensory disturbance	Single	Present	Microscopic cystectomy	72 months	Improvement	Sacrifice of left S3 root
11.	Bonnie Wang, ^[36] 2014	S2	67 F	Back, leg pain, bowel and bladder dysfunction	Single	Present	Clipping	3 months	Improvement	Marfan syndrome
12.	Suchan Lee, ^[17] 2015	S1-S3	41 F	Dysmenorrhea	Single	Absent	Laparoscopic cystectomy	6 months	Worsening	
13.	P Ostojic, ^[25] 2015	Sacral	46 F	Back, leg pain	Single	Present	Microscopic cystectomy	NR	Improvement	
14.	Mayur Sharma, ^[30] 2015	L4 L5-S3 L5-S1	42 M 48 F 28 F	Cauda Equina Cauda Equina Sciatica	Single Single Single	NR Present NR	Microscopic cystectomy Marsupialization Complete excision	NR NR NR	Complete resolution Complete resolution Complete resolution	Post-operative cerebrospinal fluid cerebrospinal fluid leak
15.	Ahmadi and Akhbari, ^[1] 2017	S2-S3	45 F	Chronic pelvic pain	Single	NR	Conservative management	3 months	Improvement	Tarlov cyst misdiagnosed as hydrosalpinx
16.	Cyril Andrieux, ^[3] 2017	L5, S1 and S2	57 F	Chronic low back pain, acute groin and buttock pain	Multiple	Present	Conservative treatment	6 months	Improvement	
17.	Aljuboori et al., ^[3] , 2017	T1	39 F	Left upper extremity pain, burning sensation, weakness in the left hand	Single	Absent	Microscopic cystectomy	NR	Complete resolution	Thoracic perineural (Tarlov) cysts are rare but they may present in a similar fashion to nerve root sheath tumors.
18.	Zhang et al., ^[39] , 2018	S2	21 M	Perineal and left lower extremity pain.	Multiple	Absent	Endoscopic resection	6 months	Improvement	First case report to describe a novel percutaneous endoscopic minimally invasive surgical strategy for Tarlov cysts.

(Contd...)

Table 1: (Continued).

S. No.	Author, Year	Location	Age gender	Presenting symptom	Single/Multiple	Sacral erosion	Treatment	Follow-up	Outcome	Remarks
19.	Sunday Patrick Nkwerem, ^[24] 2018	S2	40 M	Right lower extremity pain and right S2 dermatome	Single	Absent	Microscopic cystectomy	6 months	Complete resolution	-
20.	Semih Gungor, ^[8] 2018	T12-L5 S1-S3	64 F	Chronic lower back and leg pain	Multiple	Present	Interlaminar epidural steroid injection Fibrin glue injection	6 weeks	Complete resolution	-
21.	Kieran P Murphy, ^[21] 2019	S2	65 F	Left labial and left perirectal pain along S2 dermatome.	Multiple	Present	Intravenous injections of steroids Lumboperitoneal shunt	18 months	Improvement	-
22.	Keitaro Yamagami, ^[38] 2019	S1 S2	48 M	Urinary dysfunction, paresthesia of left S2 and S3 dermatomes.	Multiple	NR		2 months	Complete resolution	-
23.	Konstantinos Paterakis, ^[27] 2019	S2	33 F	Right flank pain, hydronephrosis	Multiple	Present		12 months	Complete resolution	-
24.	Nishimura et al., ^[23] 2020	S2	42 F	Perianal pain, urinary symptoms	Single	NR	Imbrication	2 Year	Complete resolution	Double crush syndrome by concomitant tarlov cyst and epidermoid tumour
25.	Tokgöz et al., ^[34] 2021	16 cervical, 13 thoracic, and 10 lumbar spine	54 F	Dorsalgia	Multiple	Absent	Conservative treatment	2 Years	Persistent symptoms	-
26.	Serratrice et al., ^[29] 2022	L5-S3	33 F	Painful sciatica following delivery	Multiple	Absent	CT-guided puncture and aspiration	12 months	Complete resolution	Delivery-induced intracystic bleeding
27.	J P Warnke, ^[37] 2022	S2 S3	63 F	Headache, dizziness, and lower back pain, neurological claudication.	Multiple	Present	Endoscopic assisted cystectomy and sacrum reconstruction	3 years	Complete resolution	-
28.	Abhishek Mehan, 2023 (Present Report)	S2 S3 S4	30 M	Deep Pelvic and perineal pain, urinary frequency, and Sacral radiculopathy	Multiple	Present	Laminectomy and marsupialization Recurrence at 6 months, managed with TP Shunt	1 year	Complete resolution	Largest Sacral Tarlov cyst in literature, presenting as pelvic mass in a male patient Emphasizes the role of TP shunt in management of Tarlov cyst

NR*: Not reported, TP shunt: Theco-peritoneal shunt, CSF: Cerebrospinal fluid, M: Male, F: Female

Table 2: Analysis of literature review with exceptional reports.

Parameter	n (%)
Total number of patients	39
Mean age	40.35
M:F	10:29
Number of cysts	
Single	23 (58.97)
Multiple	16 (41.02)
Presenting symptoms	
Lumbar/Sacral radiculopathy	39 (100)
Perineal pain	4 (10.25)
Bladder and bowel symptoms	18 (46.15)
Treatment	
Conservative	7 (17.94)
Microscopic cystectomy	10 (25.64)
Microscopic fenestration	1 (2.56)
Resection and imbrication	8 (20.51)
Marsupialisation	1 (2.56)
Clipping	1 (2.56)
Ligation	1 (2.56)
Aspiration and excision	1 (2.56)
Fibrin glue injection	1 (2.56)
Epidural steroid injection	1 (2.56)
Intravenous steroid injection	1 (2.56)
Theco-peritoneal/ lumbo-peritoneal shunt	2 (5.12)
Endoscopic resection	1 (2.56)
Endoscope assisted cystectomy	1 (2.56)
Computerized tomography-guided puncture and aspiration	1 (2.56)
Exceptional reports	
Largest cyst to be reported	Abhishek Mehan, 2023 (Present Report)
First Report for endoscope assisted treatment	J P Warnke, 2022
Mistaken for hydrosalpinx	Ahmadi and Akhbari, 2017
Delivery-induced intracystic bleeding	Serratrice <i>et al.</i> , 2022
Double crush syndrome	Nishimura <i>et al.</i> , 2020
Retroperitoneal extension	Curtis W Slipman, 2003
Cerebellar bleeding	Masato Tanaka, 2006

M: Male, F: Female

cyst with epidermoid in a 42-year-old woman was reported in year 2020. The case becomes more interesting when it is revealed that the epidermoid is Iatrogenic and acquired.^[23] Although, there are few reports of Tarlov cyst presenting as adnexal mass in females, our case being a young male salpingo-ovarian mass was not a possibility. MRI makes the investigation of choice in diagnosing Tarlov cyst because it gives hyperintense signals to Tarlov cysts as it gives to CSF and helps explore fenestral continuation with subarachnoid space. X-ray, although a financially better investigation, detects Tarlov cysts at advanced stages of bone degradation

and tissue infiltration-paravertebral round shadows.^[14] Laminectomy and marsupialization of sacral Tarlov cyst have shown promising outcomes with low recurrence; thus, our case was managed likewise. Large Tarlov cysts (>3.5 cm) have been reported up to sizes as large as 5 cm (largest diameter),^[16,18,34] while our case presents a rare Giant Tarlov cyst of 12.3 x 12 x 15.4 cm (APXTRXCC). Tarlov cysts are rare at thoracic level with only few case reports.^[2] Authors have done a literature review^[9,11-13,15,17,21,24,25,27] in Table-1 for all cases of Tarlov cysts published with emphasis on clinical parameters, treatment, and recurrence. Total of 39 cases were reported of which only 10 were males. Current case belongs to less common gender of Tarlov cyst. A common differential of Tarlov cyst to be kept in mind is spinal arachnoid cysts. Authors have discussed in detail about this rare spinal pathology in another review.^[6]

Literature review

A review of the literature on Tarlov cysts showed that of the total 39 cases, the majority were females [Tables 1 and 2].^[30-33,35-39] They are attributed to either musculoskeletal defects (supported by Tarlov cysts in collagen disorders like Marfan) that enlarge due to the hydrodynamic forces of CSF (i.e., continuous or pulsatile). In this series, patients averaged 40.35 years of age with a female threefold predominance. While all (100%) have lumbar/sacral radiculopathy, only 50% have bladder and bowel complaints including perineal pain (10%). Over 40% present with multiple Tarlov cysts, only rare of these may be giant lesions extending into the pelvis. Laminectomies, direct dural occlusion of dural/subarachnoid fistulas, imbrication and marsupialization often result in cyst resolution. If and when they recur, TP shunts or lumboperitoneal shunts^[8] may become an option.

CONCLUSION

Tarlov cysts may become extremely large in the presacral region to the point where they contribute to significant pelvic pain and genitourinary symptoms mimicking a pelvic cavity tumor. Treatment typically consists of laminectomy, occlusion of the dural/subarachnoid fistula, and imbrication/marsupialization of the cyst with select cases warranting the placement of TP shunts.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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