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## **Case Report**

## Resection and imbrication of symptomatic sacral Tarlov cysts: A case report and review of the literature

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## Abstract

**Background:** Symptomatic Tarlov cysts are extremely rare, and there is no consensus regarding their optimal surgical management. Here, we encountered a patient with a symptomatic sacral Tarlov cyst and reviewed the appropriate literature.

**Case Description:** A 40-year-old male presented with right lower extremity pain and hypoesthesia in the right S2 dermatome. The lumbosacral MR demonstrated a right S2 Tarlov cyst compressing the S2–S3 perineural sheaths. After the patient underwent microscopic cystectomy with obliteration of the subarachnoid connection to the cyst, the patient's symptoms resolved. Here, we reviewed our operative approach, and others proposed in the literature for the surgical management of these lesions.

**Conclusions:** Here, we present a 40-year-old male who was symptomatic from a right S2 Tarlov cyst compressing the S2–S3 perineural sheaths and successfully underwent microscopic cystectomy with obliteration of the subarachnoid connection. Additionally, the appropriate Tarlov cyst literature was reviewed.

**Key Words:** Cyst resection, perineural cyst, recurrence, surgical treatment, Tarlov cyst



## **INTRODUCTION**

Tarlov cysts, first described by Tarlov in 1938, are perineural cysts often seen in the extradural segment involving the posterior sacral nerve roots.<sup>[13]</sup> They are caused by dilation of the nerve root sheaths and are composed of abnormal sacs filled with cerebrospinal fluid (CSF) thatin; some cases become symptomatic (e.g., result in progressively radiculopathy). They are incidentally seen in ~4.6% of lumbosacral magnetic resonance imaging (MRI) studies. Sacral meningeal cysts are also reported on MR studies in 6.9% of Japanese females, but typically only 1% are asymptomatic.<sup>[12]</sup> Asymptomatic cysts should be managed conservatively; only rarely is surgery indicated for symptomatic patients.<sup>[1,3,4,6,7]</sup> Although multiple different surgical approaches to Tarlov cysts have been described, here, we report our experience with patient, whose right S2 Tarlov cyst compressed the S2–S3 perineural sheaths, requiring imbrication and resection.

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## **CASE REPORT**

#### History

A 40-year-old male presented with a 3-year history of sensory changes involving the S2–S3 distribution in the right leg (e.g., hypoesthesia without motor weakness or dysuria). The lumbosacral MRI showed a noncontrast enhancing right-sided cystic mass at the S2–S3 level; it had the same intensity as CSF on both the T1- and T2-weighted images [Figure 1]. Coronal fat-suppressed T2-weighted images revealed the mass likely originated from the right S3 nerve root and was additionally compressing the right S2 nerve root [Figure 2].

### **Operation**

A laminoplastic laminotomy was performed at three levels using an ultrasonic bone curette. At surgery, the S3 nerve root was enveloped within cyst wall and the S2 nerve root was clearly compressed [Figure 3a and b]. Partial resection of the cyst wall and imbrication of residual tissue was performed [Figure 3c]. An inlet from the subarachnoid space was identified, and its obliteration

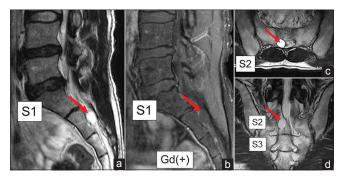


Figure 1: Lumbosacral magnetic resonance images. Sagittal (a and b), axial (c), and coronal (d) view of lumbosacral magnetic resonance imaging showing the cyst (red arrow) at the S2 vertebra. Preoperative magnetic resonance images showing a less-enhanced tumor (b). The contents of the cyst have the same intensity as the cerebrospinal fluid on both T2-weighted images

was confirmed by a Valsalva maneuver [Figure 3d]. This subarachnoid connection was sealed with adipose tissue and fibrin glue [Figure 3e]. Plication of the cyst wall was performed with nonpenetrating titanium clips (Vascular Clip System; LeMaitre Vascular Inc., Burlington, MA) [Figure 3f]. No postoperative CSF leakage occurred, and spinal lumbar drainage was not warranted. The patient's preoperative sensory disturbance resolved. The postoperative MRI showed a reduction in the cyst's size [Figure 4a and b] and no residual compression of the S2 nerve root [Figure 4c]. Histopathological examination confirmed collagen connective tissue without nerve fibers, findings consistent with a Tarlov cyst [Figure 5]. The patient remained asymptomatic 6 months later, and the 6-month postoperative sacral MRI demonstrated no cyst recurrence.

## DISCUSSION

Tarlov cysts are meningeal dilatations commonly found between the endoneurium and perineurium in the spinal nerve root sheaths at the S2 and S3 levels. They commonly communicate with the subarachnoid space.

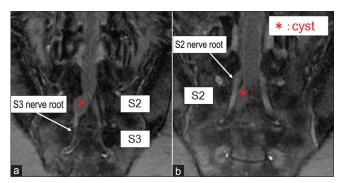


Figure 2: The relationship between the cyst and the nerve roots. (a) Coronal fat-suppressed T2-weighted imaging shows that the cyst (asterisk) likely originated from the right S3 nerve root. (b) The cyst compresses the right S2 nerve root

Author	Year	Number of cases	Treatment	Symptom improvement (%)	Complications	<b>Recurrence</b> (%)
Patel <i>et al</i> . <sup>[9]</sup>	1997	4	Fibrin glue obliteration	4/4 (100)	Meningitis: 3	0/4 (0)
Guo <i>et al</i> . <sup>[5]</sup>	2007	11	Partial cyst resection + imbrication	9/11 (82)	CSF leak: 1 dysuria: 1	1/11 (9)
Smith et al.[11]	2011	18	Cyst fenestration only	10/18 (56)	CSF leak: 2 dysuria: 1	1/18 (6)
Xu et al. <sup>[16]</sup>	2012	13	Partial cyst resection + imbrication	12/13 (92)	CSF leak: 1	1/13 (8)
		2	Conservative	0/2 (0)	None	0/2 (0)
Cantore et al.[2]	2013	19	Cyst wall clipping	12/19 (63)	None	0/19 (0)
Potts et al.[10]	2016	35	Cyst fenestration + paraspinal muscle pedicle flap	32/35 (91)	None	26/35 (74)
Weigel et al.[15]	2016	13	Imbrication + fat graft packing	11/13 (85)	None	2/13 (15)
Present case	2016	1	Partial cyst resection + imbrication	1/1	None	0/1

CSF=Cerebrospinal fluid

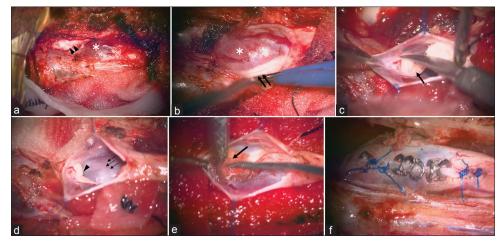


Figure 3: Operative procedure and intraoperative findings. (a) Intraoperative photograph showing that the S2 nerve root (double arrowheads) was compressed by the cyst component (white asterisk). (b) Closer observation revealed that the cyst wall (white asterisk) contains the S3 nerve root (double arrows). (c and d) After excision of the cyst wall, the inlet of the cerebrospinal fluid (CSF) was confirmed around the nerve root (single arrow). The inlet from the subarachnoid space was found (single arrowhead) and the CSF was spontaneously flowing out from it (small triple arrows). A Valsalva maneuver clearly showed the CSF flow from the subarachnoid space to the perineural cyst. (e) After confirmation of the subarachnoid connection, it was sealed with adipose tissue (large arrow) and fibrin glue. (f) Finally, imbrication of the cyst wall was performed with nonpenetrating titanium clips. A repeated Valsalva maneuver showed no CSF leakage

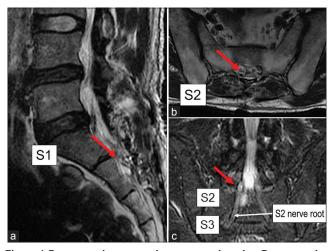


Figure 4: Postoperative magnetic resonance imaging. Postoperative magnetic resonance imaging showed a reduction in cyst (red arrow) size (a and b). There was no compression of the S2 nerve root (c)

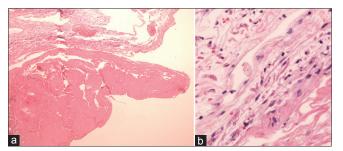


Figure 5: Histopathological examination of the cyst. (a and b) Histopathological specimen from the cyst wall. The histopathological examination indicated that the wall was composed of collagen connective tissue without nerve fibers (H and E staining)

Some authors believe that these are a congenital lesions, whereas others consider them to be acquired.<sup>[8]</sup>

#### Symptomatic Tarlov cysts

Although they are typically asymptomatic, 1% may demonstrate growth and contribute to nerve root compression (e.g., sacral/perineal pain, sphincter dysfunction, radiculopathy, and rarely, infertility).<sup>[1,3,12]</sup> Growth of these cysts and symptoms are typically attributed to the aball-valve effect/net inflow of the CSF from the subarachnoid space, a finding that may be confirmed on myelograph (e.g., delayed filling).<sup>[4,14]</sup> Clinically, Tarlov cysts symptoms may exacerbate in the standing position (e.g., stimulates CSF flow to the cyst); this was seen in the case presented.

#### Surgical options for symptomatic Tarlov cysts

Different surgical treatment options are available for symptomatic Tarlov Cysts [Table 1].<sup>[3,7]</sup> Reducing the size of the cysts often relieves symptoms; this typically requires sealing the connection with the subarachnoid space (e.g., obliteration of the ball-valve mechanism of filling). The aim is to reduce cyst volume and prevent further communication with the CSF pathways.

# Studies demonstrating surgical outcomes of Tarlov cysts

Seven studies (all case series) evaluated the surgical treatment outcomes for Tarlov Cysts.<sup>[2,5,10,11,15,16]</sup> Surgical alternatives included excision and fenestration utilizing different techniques (e.g., cystectomy, imbrication, clipping, obliteration of the CSF fistula, and combined approaches to abolish communication with the subarachnoid space between the dural sac and the cyst). Fibrin glue obliteration and cyst resection are also effective in achieving symptomatic improvement but may lead to postoperative complications such as meningitis. Potts *et al.*<sup>[10]</sup> reported good surgical results after cyst

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fenestration; however, over 70% of patients ultimately suffered a recurrence.<sup>[5,16]</sup> All other studies reported comparatively low rates of recurrence.<sup>[2,5,10,11,15,16]</sup>

#### CONCLUSION

There are several surgical treatment options for treating symptomatic (e.g., 1%) Tarlov cysts. An optimal strategy appears to include direct cyst resection, imbrication, and fat graft packing of the communication between the dural sac to the cyst (e.g., occlude the ball-valve mechanism of refilling).

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#### **Conflicts of interest**

There are no conflicts of interest.

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