



Management of symptomatic sacral perineural cysts with microsurgery and a vascularized fasciocutaneous flap

Parmenion P. Tsitsopoulos¹, Niklas Marklund^{1,2}, Konstantin Salci¹, Anders Holtz¹, Maria Mani³

¹Department of Neurosurgery, Uppsala University Hospital, Uppsala, Sweden; ²Department of Clinical Sciences, Neurosurgery, Lund University, Skåne University Hospital, Lund, Sweden; ³Department of Plastic and Reconstructive Surgery, Uppsala University Hospital, Uppsala, Sweden

Contributions: (I) Conception and design: PP Tsitsopoulos, N Marklund; (II) Administrative support: PP Tsitsopoulos, N Marklund; (III) Provision of study materials or patients: All authors; (IV) Collection and assembly of data: All authors; (V) Data analysis and interpretation: PP Tsitsopoulos, N Marklund, K Salci, M Mani; (VI) Manuscript writing: All authors; (VII) Final approval of manuscript: All authors.

Correspondence to: Parmenion P. Tsitsopoulos. Department of Neurosurgery, Uppsala University Hospital, 75185 Uppsala, Sweden.

Email: ptsitsopoulos@auth.gr.

Background: The optimal treatment of symptomatic perineural (Tarlov) cysts is controversial. Numerous surgical techniques have been proposed with conflicting results. A series of Tarlov cysts treated with a novel surgical approach is presented.

Methods: Patients with surgically treated symptomatic perineural cysts during 2013–2016 were included. The main indications for surgery were persistent radicular symptoms, pelvic pain, urinary and/or bowel disturbances. At surgery, the cyst was opened and fenestrated. The cyst wall was then closed with packing, fibrin glue and a pedicled vascularized fasciocutaneous flap rotated into the area for obliteration of the dead space. Patients were followed-up with clinical visits and repeat magnetic resonance imaging (MRI) scans.

Results: Seven consecutive patients were included. The mean age was 50.3 years (range, 25–80 years) and the mean duration of symptoms was 49.3 months (range, 3–130 months). With one exception, all patients had urine and/or bowel problems (incontinence) preoperatively. A lumbar drain was inserted in five patients. The mean follow-up period was 15.4 months. Symptoms improved in 4/7 patients, in two cases no clinical difference was noted while one patient deteriorated. In two cases, a spinal cord stimulator was eventually implanted. In all seven cases, a significantly decreased cyst size was noted on MRI.

Conclusions: Cyst fenestration and the use of a vascularized fasciocutaneous flap successfully obliterated all cysts, with satisfactory clinical efficacy. Larger and comparative studies are warranted to clarify the long-term effects of this surgical technique in patients with symptomatic Tarlov cysts.

Keywords: Perineural sacral cysts; Tarlov cysts; surgical management; muscle flap; fasciocutaneous flap; outcome; complications

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Introduction

Perineural or Tarlov cysts were initially described in 1938 during cadaveric observations of terminal filum specimens (1-3). Their origin is largely unknown (3-6). According to the classification by Nabors *et al.*, Tarlov cysts are considered type II meningeal cysts defined as “extradural meningeal cysts with spinal nerve roots” (7). Most are

discovered incidentally on magnetic resonance imaging (MRI) scans and approximately 13% are considered symptomatic (3,6,8,9). The most common complaints attributed to them are low back pain, sciatica, perineal discomfort, pain or numbness and urinary/bowel problems (3,8,10-13).

There is no general agreement on the optimal management of symptomatic Tarlov cysts. Conservative

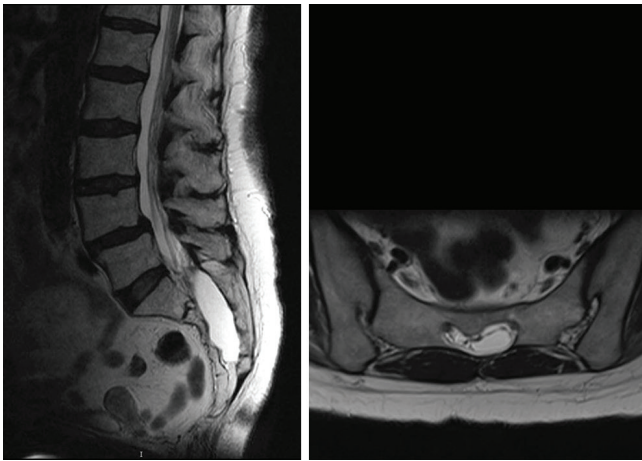


Figure 1 Sagittal (left) and axial (right) magnetic resonance imaging (MRI) of the lumbosacral spine showing a large Tarlov cyst in an 80-year-old female patient extending from S1 to S3 level.

or minimally invasive methods such as cyst aspiration and cerebrospinal fluid (CSF) diversion have provided conflicting results since symptomatic cyst recurrence and aseptic meningitis can occur (3,5,14-16). Surgical techniques have evolved over the years and include procedures such as laminectomy or laminotomy and decompression, cyst fenestration, cyst resection, cyst imbrication (wall fixed to neighboring structures), sacroplasty and removal of the proposed ball-valve mechanism (5,10-13,17-22).

In recent years, mobilization of muscle flaps to augment cyst closure, minimize cyst recurrence and wound related complications has been increasingly used during surgery (10,11,23). Here, we present our operative series of symptomatic Tarlov cysts with microsurgery and use of a vascularized fasciocutaneous flap.

Methods

Patients

Patients surgically treated from October 2013 to November 2016 for symptomatic sacral perineural/Tarlov cysts were included. The cysts were identified on MRI (*Figure 1*). Patients considered candidates for surgery were typically primarily subjected to a diagnostic cyst puncture. A transient relief of symptoms followed by symptom relapse and associated with cyst recurrence on MRI led to the decision to proceed to surgery. Moreover, patients with persistent radicular symptoms, pelvic pain and sphincter problems (urine and bowel incontinence) refractive to conservative

treatments including anti-nociceptive treatment or physical therapy were also considered candidates for surgery, given that complaints were unrelated to other processes such as degenerative spine disease, tumors, infections and traumatic lesions excluded by MRI and medical history.

Medical records were retrieved to identify major symptoms, radiological characteristics, neurological deficits, surgical technique and outcome.

The study was approved by the regional ethics committee of Uppsala University Hospital. Due to the retrospective nature of the study, no informed patient consent was deemed necessary for the analysis of clinical, radiological and surgical characteristics.

Surgical technique

Patients were placed in prone position on a Wilson frame. A midline incision at the level corresponding to the cyst was undertaken. After opening the lumbosacral fascia, subperiosteal dissection of the surrounding muscles from the spinous processes and laminae was done. Next, a laminectomy at the level of interest (usually L5–S2) was performed to expose the cyst. The cyst was then opened and excised under the microscope. The area was inspected and the nerve roots were identified. Whenever possible, the nerve roots were released from the cyst wall and surrounding tissues using microneurosurgical techniques and neuromonitoring. When present, communication between the cyst and the subarachnoid space was identified and sealed with TachoSil packing and fibrin glue if neural elements were not included. When possible, the remaining cyst wall was sutured with a running non-absorbable suture but even in these cases, watertight closure could not be accomplished. Packing with Surgicel® (Ethicon, USA) and/or TachoSil® (Kaneda, Japan) followed and fibrin glue (Tisseel®, Baxter, USA) was placed over the cyst wall (*Figure 2*).

A plastic and reconstructive surgeon (MM) mobilized a local perforator fasciocutaneous flap to carefully cover the area after cyst evacuation. Specifically, a lumbar perforator was identified with a handheld Doppler lateral to the spinal column. The perforator was included in the flap once designed and marked pre-operatively. The flap was either designed as a broad-based V to Y fashion with one edge transposed into the space created by cyst opening/ excision or as a flap isolated on the specific perforator and rotated into the defect (named a propeller flap). The tissue positioned into the defect was de-epithelialised and anchored with 3.0 Monocryl sutures. A drain was placed in the donor-site area and the

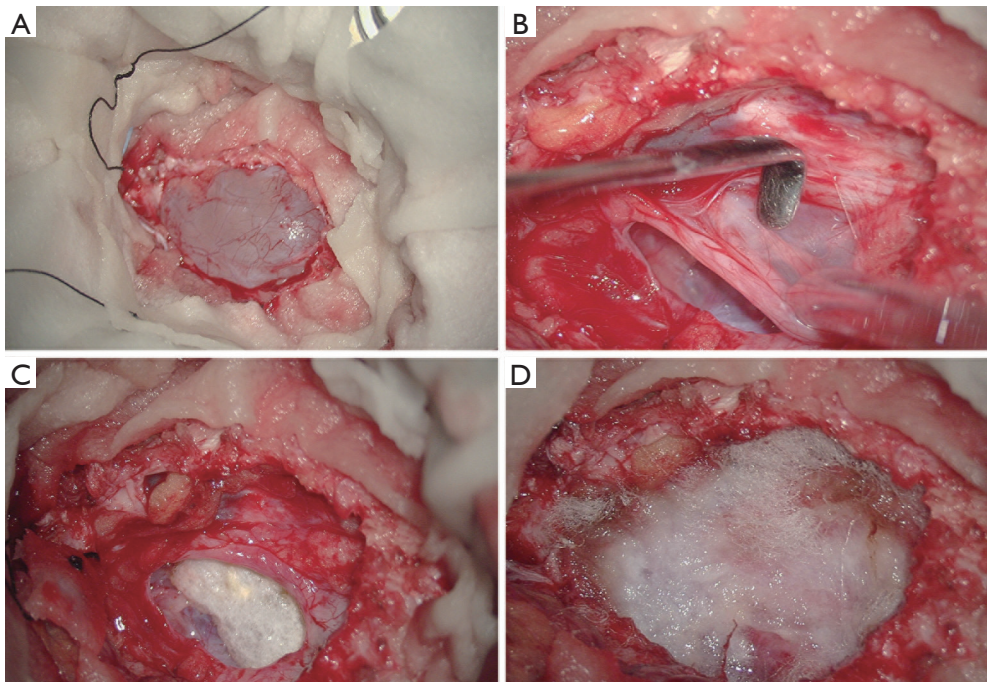


Figure 2 Intraoperative images. (A) Cyst exposure at S1–S2 level following posterior approach and laminectomy; (B) opening of the cyst wall, inspection and decompression of nerve roots; (C) placement of sealant (fibrin glue) between the communication of cyst with the subarachnoid space; (D) packing of the cyst cavity with Surgicel and fibrin glue.

wound was closed in an ordinary three-layer fashion (*Figure 3*). In patients with large cysts, large dural defects and unsecure closure, a lumbar drain was also placed following surgery.

Postoperative care and follow-up

The wound drain was removed 24 hours postoperatively. The lumbar drain typically drained for 3–5 days, then it was clamped, the wound was inspected for CSF leakage and removed if no leakage or symptoms of intracranial hypotension were observed. Patients were followed-up by consecutive visits to the treating physicians where a detailed neurological examination was undertaken, and post-operative MRI scans. Possible relapse of symptoms, complications, reoperations and cyst recurrence were recorded.

Results

Baseline characteristics

Seven patients were included (*Table 1*). The mean age was 50.3 years (range, 25–80 years) and the mean duration of symptoms was 49.3 months (range, 3–130 months). The

major complaint was back pain, present in all patients. One patient exhibited decreased motor strength in the lower limbs. All but one patient presented with urine and/or bowel incontinence. All cysts had a maximum diameter ≥ 2 cm. No patient had a coexisting spina bifida.

Surgical results

Prior to the present surgery, all but one patient received surgical treatment either minimally invasive by cyst puncture or open surgery. Intraoperatively, a CSF fistula was identified in one patient and the communication was successfully sealed. In only two patients, the cyst wall was closed with a running suture. In the remaining patients, the cyst was packed with Surgicel and TachoSil and fibrin glue was also applied. A V-Y muscle flap was mobilized in five patients while a propeller flap was used in the remaining two. A wound drain was inserted in 4/7 patients and a lumbar drain in 5/7 patients (*Table 2*).

Postoperative course and complications

The follow-up period was 15.4 (range, 4–32) months.

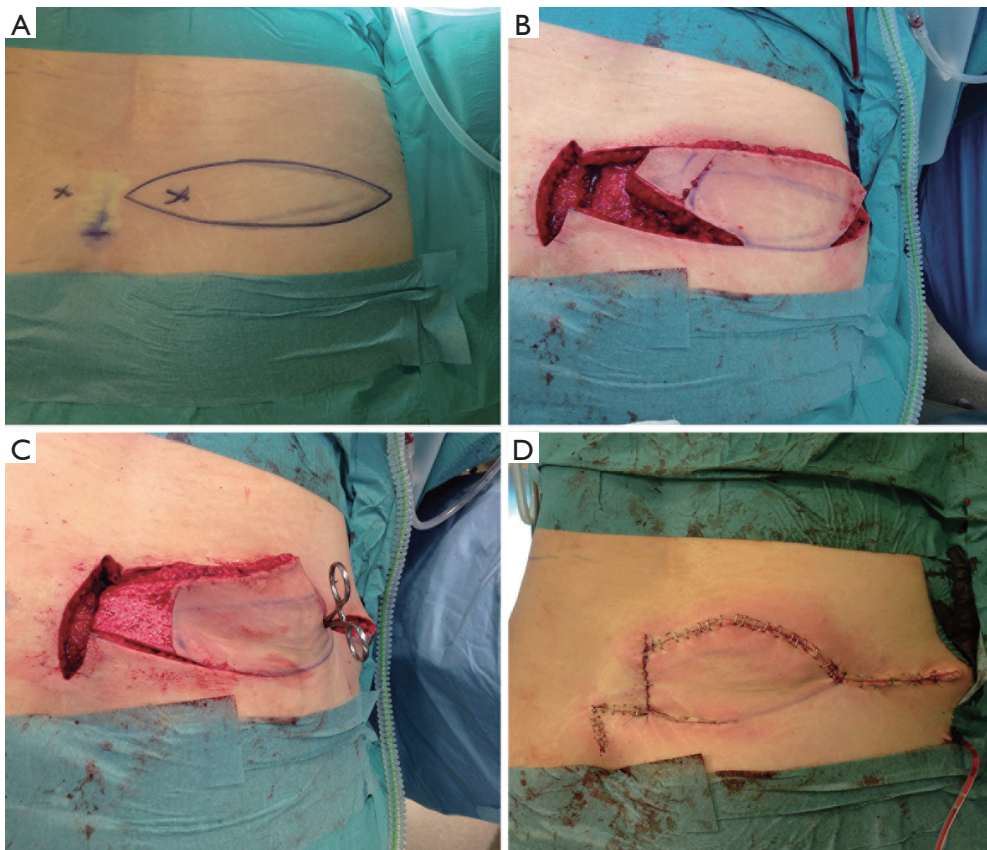


Figure 3 Intraoperative images. (A) A lumbar perforator was identified with a handheld Doppler lateral to the spinal column. The perforator was included in the flap once designed and marked pre-operatively. (B) The flap was raised based on the specific perforator and rotated into the defect. (C) The tissue positioned into the defect was de-epithelialised and anchored with 3.0 Monocryl sutures. (D) The flap was sutured in place and the donor site was closed without tension.

Symptoms improved in four patients, two patients experienced transient relief but symptoms later recurred while one patient had deteriorated clinically at long-term. One patient developed meningitis, postoperative hematoma and wound rupture and one was reoperated for removal of excess skin tissue. In two cases, a dorsal column stimulator was implanted due to persisting neurogenic pain. In half of the patients (2/4), urine incontinence improved. In addition, 4/7 patients returned to normal activities of daily life. Bowel disturbances did not improve. In all seven cases, a significant decrease in cyst size was noted on MRI (*Table 3, Figure 4*).

Discussion

In the current patient cohort of surgically treated sacral perineural (Tarlov) cysts, the use of microsurgical

technique and a fasciocutaneous vascularized flap resulted in significant resolution of symptoms in more than half of the patients. Only one patient deteriorated following surgery. Half patients with urine incontinence experienced improved bladder control. Serious complications were rare and observed in only one patient. The use of a vascularized flap aided in the significant reduction in cyst wall size in all operated cases.

Lumbosacral Tarlov cysts are uncommon lesions, occurring in approximately 1–5% of the general population and located at the junction of the dorsal ganglion and the posterior nerve root between the endoneurium and the perineurium (5,12,15,24). Although still unclear, a valve-like mechanism between the subarachnoid space with the dura leading to CSF inflow is considered the most probable pathogenetic theory of their formation (5,7,25). Genetic factors may also contribute to their pathogenesis (4).

Table 1 Baseline characteristics of the seven patients with symptomatic perineural (Tarlov) sacral cysts obliterated by microsurgery and mobilization of a fasciocutaneous flap

Patient	Age/ sex	Chief symptoms/signs	Duration of symptoms (months)	Motor Deficits	Sensory deficits	Sphincter disturbances	Cyst size (mm ³)	Side	Level
1	80/F	LBP, radiating pain in both legs, incontinence	58	No	No	Bowel/ bladder	60×15×45	Left	S1–S3
2	61/M	LBP and anal pain	25	No	Yes	Bowel	20×20×45	Bilateral multiple	S2–S3
3	31/F	LBP, radiating pain in both legs, gait disturbance	33	Yes	Yes	Bladder	39×17×22	Right	S2–S3
4	25/F	LBP, radiating pain in the right leg, incontinence	3	No	Yes	Bladder	12×18×28	Right	S2–S3
5	76/F	LBP, abdominal pain, radiating pain in the left thigh	130	No	No	Bladder	17×14×32	Left	S1–S2
6	35/M	LBP, radiating in the right leg pain, bowel problems	82	No	Yes	Bowel	29×43×72	Right	S1–S3
7	44/F	LBP, burning pain in the left leg	14	No	Yes	No	16×27×35	Left	S2–S3

F, female; M, male; LBP, low back pain.

Table 2 Management characteristics of patients with Tarlov cysts. Previous treatment attempts and surgical details are shown

Patient	Previous treatment	CSF fistula*	Cyst wall closure	Muscle flap	Fat graft	Fibrin glue	Wound drain	Lumbar drain	Lumbar drain/days
1	Twice partial open resection of cyst	Yes	TachoSil packing	V-Y	No	Yes	Yes	Yes	9
2	Twice cyst puncture, spinal cord stimulation	No	TachoSil packing	V-Y	No	Yes	Yes	No	N/A
3	Cyst puncture	No	Surgicel packing	Propeller	No	Yes	No	Yes	6
4	Cyst puncture, local anesthetic	No	Running suture plus packing	Propeller	No	Yes	No	No	N/A
5	Cyst puncture	No	Running suture plus packing	V-Y	No	Yes	No	Yes	6
6	None	No	TachoSil packing	V-Y	No	No	Yes	Yes	7
7	Cyst puncture	No	TachoSil packing	V-Y	No	Yes	Yes	Yes	5

*, communication of the cyst with the subarachnoid space evidenced either preoperatively or intraoperatively. CSF, cerebrospinal fluid; N/A, not applicable.

Table 3 Complications and clinical outcome following surgery for Tarlov cysts

Patient	Follow-up (months)	Postoperative complications	Clinical outcome	Reoperation	Type of reoperation	Return to normal activities
1	10	No	Improvement	No	N/A	Yes
2 [†]	30	No	No change	No	N/A	No
3	32	Meningitis, postoperative hematoma, wound dehiscence	Deterioration	Yes	Wound revision with hematoma drainage, neuroma exploration, spinal cord stimulator	No
4	5	No	Improvement	No	N/A	Yes
5	4	No	No change	No	N/A	Yes
6	16	Excess tissue removed	Improvement	Yes	Excision of excess tissue	Yes
7	11	No	Improvement	Yes	Spinal cord stimulator	No

[†], died of unrelated cause 3 years after surgery. N/A, not applicable.

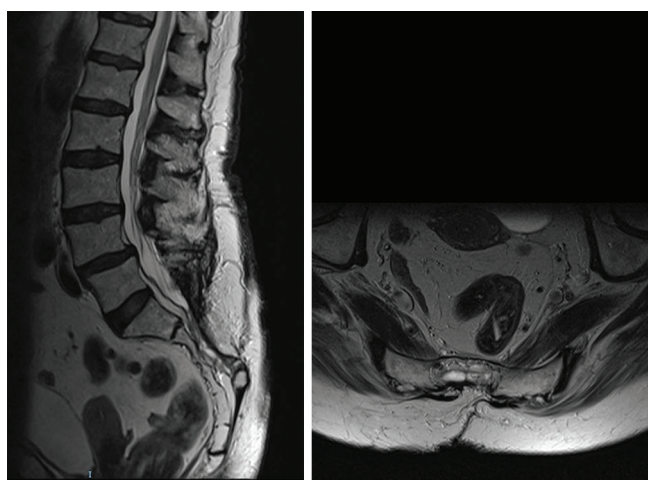


Figure 4 Sagittal and axial postoperative magnetic resonance imaging (MRI) of the lumbosacral spine showing complete obliteration of the cyst following microsurgical opening and the use of a fasciocutaneous flap in the patient shown in *Figure 1*.

Even in symptomatic cysts, the indications to proceed to surgery have not been defined and most published reports describe a limited number of patients (6,12,13,18,20-22,25-27). The following surgical indications for symptomatic patients with Tarlov cysts were recently suggested: (I) multiple cysts on MRI; (II) symptoms related to the location of the cyst; (III) symptoms appeared in the past 6 months; and (IV) single, unilateral cyst over 1 cm in size (10). In particular, younger patients with single unilateral cysts with medically refractory symptoms have mostly benefited from surgery (10).

We suggest that patients with progressive and/or refractory lumbosacral or radicular pain, urogenital pain and persistent urine incontinence attributed to the presence of the cyst are appropriate candidates for surgery (5,10,26).

Numerous more or less invasive surgical techniques have been proposed for the treatment of Tarlov cysts (5,8,10-12,17-21,28). The overall goal is alleviation of symptoms with radiological evidence of decrease in cyst size and prevention of further sacral bone erosion. The results have been conflicting with the best clinical outcomes observed following the use of direct microsurgical approaches (5,8,10). From the surgical perspective, neural decompression is important, and equally important is a meticulous closure and obliteration of the cyst wall and the overlying layers with the aim to avoid postoperative cyst and symptom recurrence and CSF leakage. Outcome following surgery for symptomatic Tarlov cysts has been recently reviewed. Of the 646 patients that were included in the analysis, 32% reported complete alleviation of symptoms, 50% partial resolution and 16% no improvement or clinical deterioration. Large cysts were not associated with complete resolution of symptoms (8). In the current series, cyst size did not influence the postoperative results.

Recent reports have used paraspinal muscle flaps to assist in wound closure and prevent cyst recurrence (10,23). In 35 patients with symptomatic Tarlov cysts, after microsurgical opening of the cyst and repair of the cyst wall, a multifidus muscle pedicle flap was mobilized to fill the cyst cavity. The muscle flaps were rotated into the sacral cavity and occasionally sutured to the

ligamentum flavum. Using this technique, 93% reported improvement at some time postoperatively. Nevertheless, symptoms recurred in half patients, and three patients required reoperation due to pseudomeningocele. It should be noted, however, that uniform muscle atrophy was frequently seen on postoperative images attributed to impaired innervation caused by the functional reconstruction of the muscles (11).

The use of vascularized fasciocutaneous flaps, as in the present study, reduces the risk of progressive muscle atrophy and provides better conditions for a long-lasting obliteration of the cyst cavity. This was also evident in the current case series. As such, fasciocutaneous flaps may offer better chances for an effective surgical treatment of Tarlov cysts in a long-term perspective.

Conclusions

Cyst fenestration and the use of a vascularized fasciocutaneous flap enabled the obliteration of the cyst in all patients with Tarlov cysts. Although incomplete remission of symptoms or persistent complaints were not infrequent after surgery, the overall clinical outcome was favorable. Careful patient selection remains the most important factor determining outcome following surgery for sacral perineural cysts. Although we suggest initially a cyst puncture and in case of symptom regression, cyst excision and mobilization of a fasciocutaneous flap over the cyst wall, larger series are justified to clarify the long-term effects of this combined surgical approach in patients with symptomatic lesions.

Acknowledgements

None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

Ethical Statement: The study was approved by the regional ethics committee of Uppsala University Hospital (Spinal Research No. 400/2012). All procedures performed in studies involving human participants are in accordance with the ethical standards of the institutional and/or national research committee, and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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