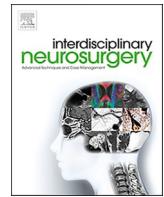




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Technical notes & surgical techniques

Combined endoscopic and microsurgical treatment of Tarlov cysts

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ABSTRACT

Tarlov cysts are a rare condition. While they appear asymptomatic mostly, some patients develop severe clinical symptoms. Tarlov cysts can grow over time and may result from disturbances within the spinal CSF system. Due to the rare character, the disease can be a burden for both patients and physicians. Treat or not to treat, and if yes, how to treat is a relevant question. Here, a retrospective, observational study on 49 patients suffering from symptomatic Tarlov cysts treated with a combined endoscopic-microsurgical approach was performed. The microsurgical resection was combined with thecaloscopy inside the lower lumbosacral subarachnoidal space to improve the CSF flow and pressure regulation capability. The purpose was to prove that the surgical method was able to prevent recurrence and gives a major relief of symptoms. Our data suggest that combined interventional treatment of symptomatic Tarlov Cysts is helpful to treat the long-standing symptoms. In all patients we found signs for inflammatory processes at the level of the lumbar leptomeningeal sheets, involving the pia mater and the arachnoid. We found evidence that patients suffering from a slow inflammatory process within the leptomeninges might have increase risk of developing Tarlov cysts. We hypothesized that local CSF flow and pressure regulation mechanism become invalid by the morphological changes at the arachnoid and lose the capability to regulate CSF pressure down by pressing minimal volumes of CSF out of the dural sac at the nerve root exits.

1. Introduction

Tarlov cysts (TC) usually arose from increasing cerebrospinal fluid (CSF) collections between the endoneurium and the perineurium of sacral spinal nerve roots [1,2]. Despite the relatively high prevalence in the adult population of nearly 5 % most TC are asymptomatic and usually be incidentally discovered during radiological examination of the lumbosacral spine [3,4]. It is undisputed that asymptomatic cysts do not require treatment. In some cases, however, TC may lead to severe symptoms like intractable radicular pain, bladder/bowel and sexual dysfunction [5]. Despite an increasing body of evidence TC are subject of an ongoing debate about their clinical importance. In fact, TC are often considered as irrelevant despite its typical clinical presentation [6,7].

A broad variety of interventional approaches were presented to treat TC ranging from microsurgical procedures to percutaneous approaches and glue administration [8–10]. Among the surgical interventions

different approaches are suggested varying from shrinking, fenestrating, resecting and imbricating, clipping, and shunting the cysts; with and without sacral laminoplasty [5,11,12]. However, complication rates of up to 39 % and recurrence rates of up to 50 % were reported [8]. Therefore, many authors attributed the development of TC to inflammatory processes within the nerve root sheath and an increased CSF pressure [13,14]. As indicated by Naderi [15] this broad variety of treatments reflects rather a lack of knowledge of the optimal treatment and the precise pathophysiology of this condition [16,17].

In this study we present the clinical and pathophysiological results of an observational study on surgical treatment of symptomatic TC. All patients with TC underwent a microsurgical treatment for the TC in combination with an endoscopically inspection of the spinal subarachnoid space to prove for signs of inflammatory processes at the level of the lumbar leptomeningeal sheath. Additionally, internal shunting from subarachnoid space to subdural subdural space by fenestrating the

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arachnoid sheath was performed.

2. Patients and surgical procedures

Over the last 10 years we included 49 patients suffering from symptomatic Tarlov cyst. All prior alternative treatment attempts such as physical therapy and/or, medication using anti-neuropathic drugs were insufficient. Patients had to have a significant subjective reduction of quality of life. In all patients alternative treatments such as physical therapy or, medication using anti-neuropathic drugs were insufficient. Patients had to have a significant subjective reduction of quality of life. Clinical data on patients were obtained by retrospective chart review. Including criteria were symptomatic Tarlov cysts, unsatisfactory pain situation uncontrolled by pain medication, symptoms or neurological deficits corresponding to the morphology of cysts. Excluding criteria for the study were previous surgical treatment or the absence of above-mentioned criteria. All patients underwent usual neurological examination, history exploration, MRI and CT investigation, as well as testing of bladder function. In conformity to our institutional experience all patients showed signs of more or less far advanced stages of an aseptic leptomeningeopathy [18]. Therefore, we performed in all patients suffering from symptomatic Tarlov cysts as first therapeutic step an endoscopic inspection of the lumbar subarachnoid space applying thecaloscopy. The degree of arachnoiditis was classified according to Hofmann [19,20]. Simultaneously, fenestration of the ventral arachnoid and the reopening of the arachnoid nerve root perforation by balloon dilatation was performed to release CSF into subdural space for eased CSF circulation within the spinal canal to give more pressure regulation ability. In the second step, a microsurgical resection of the sacral cysts were performed. The follow up of the patients with a at least 12 months comprised of clinical follow up examination and lumbosacral MRI. The median age was 58 years (range: 33 to 79 years) at the date of treatment. 42 females (85.7 %) and 7 males (14.3 %) were included. **Table 1** presents the patient's demographic data und symptoms.

25 patients had a single cyst within the sacral spinal canal, 26 patients suffered from multiple cysts. The largest cysts were found at the sacral level of S1 and S4 with an extension of about 24 mm followed by the level S3 (19 mm) and S4 (18 mm).

2.1. Description of surgical procedure

In all cases the treatment was an endoscopic assisted microsurgical sacrotomy, cyst-resection, fistula closure, autologous fat tissue augmentation of the sacral canal and bony reconstruction of sacral canal using patient's bone or autologous materials if the bone was eroded from the cysts.

As depicted in **Fig. 1** the surgical procedure started with a thecaloscopy, as an inspection of the lumbar sacral subarachnoid space (A). Signs of chronic, aseptic leptomeningeopathy such as calcification of the arachnoid layer were always visible (B). The thecaloscopy with a flexible endoscope allows the reconstruction of the lumbar CSF flow regulation

Table 1
Patients characteristics and clinical presentation.

Characteristic	Number (%)
Number of patients	49
Mean age of operation, range	58 (33-79y)
Female	42 (85.7)
Length of history in years	5.2
Low back pain	27 (55.1)
Sacrococcygodynia	31 (63.3)
Radicular pain	30 (61.2)
Sensory disturbances of sacrococcygeal region	14 (28.6)
Sensory disturbances lower extremities	25 (51.0)
Bowel disturbances	10 (20.4)
Bladder disturbances	17 (34.7)
Affection of sexual function	38 (77.6)

capacity by re-opening of the arachnoid nerve root perforations or simple fenestration of the ventral arachnoid layers (C). For the reopening we used balloon dilatation techniques. Despite refilling the CSF space with saline, the reduced lumbar CSF amount seemed to reduce the pressure load on the closed fistula points of the cysts in the first days. In addition, the endoscopic view allowed to determine the position of lower sacral nerve-roots as they are tightly attached to the wall of giant cysts. As the sacral nerves are often thinned and cannot morphologically be detected even under high magnification this technique serves for better identification.

After thecaloscopy in all cases a microsurgical approach the resect the sacral Tarlov cysts was performed. Therefore, as **Fig. 2** describes, a usually rectangular sacrotomy was performed using a micro-drill to open the spinal canal (A). After discovering and dissection the cysts were sharply fenestrated and shranked (B).

As far as the closure of the fistula is concerned, different methods were applied. Depending on individual anatomy, the involved nerve tissue and the wideness of the fistula point, 3 principal methods were used (C).

1. Wrapping of the nerve using autologous cyst wall-tissue and fixation by micro suture secured by fibrin coated gel foam (Tachosil), (36 patients, 73.4 %)
2. Wrapping of nerve using autologous cyst wall-tissue surrounded by fibrin coated gel foam, (12 patients, 24.5 %)
3. Shrinking and tailoring of the cyst wall using bipolar or micro stiches, (3 patients, 6.1 %)

As final step, the reconstruction of the bony sacral bony defect was performed by replacing the bone flap or implanting alloplastic material after fat augmentation of the sacral canal (D). **Fig. 3** shows an example of the results of the surgical approach.

After surgery all patients were treated with bed rest for 3 days before mobilization. The aim was to reduce the intradural pressure by avoiding the water column to press upon the lower end of the subarachnoidal space. For better comfort patients had a urinary catheter. Following the 3rd postoperative day patients were mobilized under physiotherapeutic control and were usually able to leave hospital on 7th day.

The value of this combined endoscopic-microsurgical approach to patients suffering from Tarlov cysts needs to be evaluated for both, the immediate and the long-term result. This group of patients usually have a long history with gradually slowing impairment. We wanted to demonstrate that using this method the reoccurrence of cysts can be avoided, the symptoms can be improved or eradicated and this for the immediate postop time as well as for the long-term period. Observation data for up to 96 months were available.

3. Follow-up and outcome

Neurological symptoms varied from radicular pain, sensory disturbances, anogenital pain, disturbances of bladder and bowel control to dizziness, ear noise as symptom of CSF loss. Patients suffering from giant cysts with massive destruction of the sacral bone were suffering from CSF loss syndrome while getting into upright position.

No correlation was found between size of cyst or cysts and severity of symptoms. Patients with giant cysts had the same complaints as patients with smaller cysts. Exceptional was the CSF loss syndrome, which we only found in giant cysts.

Out of 49 patients, 20 (40.9 %) reported about preoperative bowel and bladder control disturbances, 38 patients (77.6 %) about anogenital pain and 35 patients (71.4 %) about radicular symptoms as pain and sensory disturbances. Although only 20 patients reported about bladder and bowel control problems, 42 (85.7 %) patients showed urinary retention in the functional bladder ultrasound testing.

For the immediate postoperative period we observed both the improvement and the temporary worsening of symptoms (**Table 2**). 5



Fig. 1. Intraoperative findings of thecaloscopy: A – Intraoperative view of the lumbosacral spinal canal using a flexible endoscope, B – Calcificartions as sign of chronic leptomenigopathy, C – Fenestration of the arachnoid sheath using a balloon catheter.

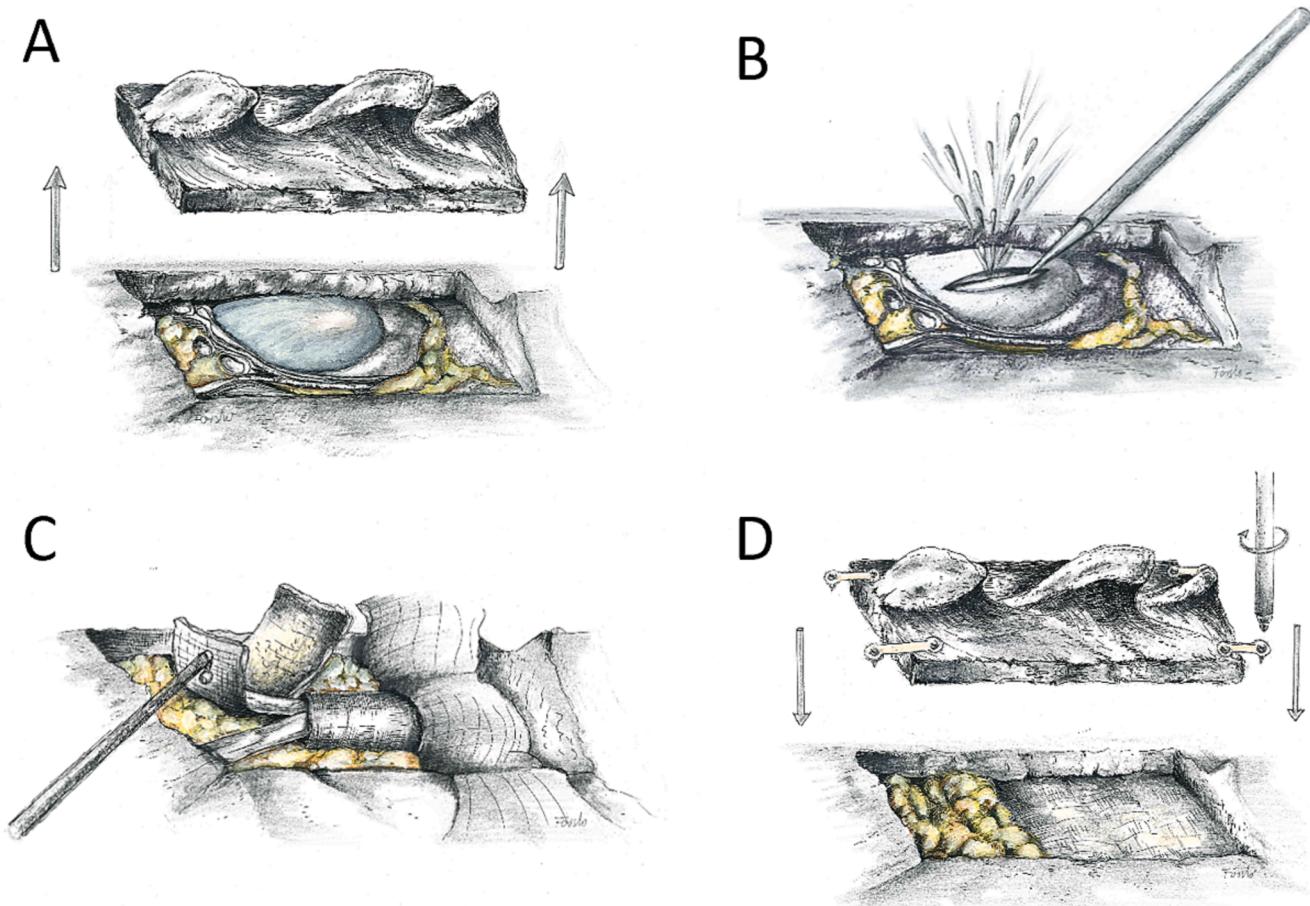


Fig. 2. Illustration of the microsurgical resection of a Tarlov cyst.

patients (10.2 %) complained about worsening and 44 (89.8 %) about improvement of symptoms. Even temporary new symptoms could be seen in the first days following surgery in 6 patients (12.2 %). New pain or other symptoms never lasted more than 15 days following surgery. In 4 patients we detected through postop MRI a CSF collection above the sacral bone. One of those patients suffered from CSF loss symptoms such as headache and photophobia. The symptoms resolved within 7 days. In another patient we observed severe CSF loss syndrome for 13 days. In

the MRI within the first week there was no reoccurrence to be expected and not seen in all patients. In the long-term follow up no cyst reoccurred. There was no difference between autologous or foreign-material sacral reconstruction as far as the cyst-reoccurrence concerned. We did not observe either any difference in the later outcome using the different methods of fistula closure, as long as the closure procedure was efficient enough to avoid reoccurrence. Worsening of preoperative existing neuropathic pain and temporary impairment of bladder control was the



Fig. 3. Pre- and postoperative MRI of sacral Tarlov cysts after combine approach of treatment.

Table 2
Long term results of combined approach (at least 12 months).

	Number (%)
Number of surgeries	49
Combination of microsurgery and endoscopy	49
Confirmed Leptomeningeopathy by endoscopy	49
Degree of Inflammation according to Hofmann	
1	2 (4.1 %)
2	26 (53.1 %)
3	20 (40.8 %)
4	0
Improvement of symptoms	48 (98.1 %)
Specific complications of thecaloscopy	0
Complication after combined approach	2 (4.1 %)
Internal CSF fistula	
Temporary worsening of bladder symptoms	10 (20.4 %)

major side effect of the operation. We observed bladder dysfunction requiring urinary catheter in two cases for longer than 20 days. Over time showed the neuropathic pain decreased and reached a barable level using medication in all patients. After 12 month the overall pain-level was reduced by 60 %. No recurrence of cysts was observed in any case.

4. Discussion

Within the context of the present study 49 patients suffering from symptomatic Tarlov cysts were treated applying a combined endoscopic and microsurgical approach. A long history of complaints, a broad variety of symptoms in this patient group was frequent. All of the patients reported that the medical system was constantly ignoring the cysts as being a pathology or the disease being existing. Preoperative the patients required high dosages of pain-medication, often using opioids.

It remains still speculative what causes the presence and growth of Tarlov cysts [13]. Located at the lowest end of the CSF column and

surrounded by fat tissue with low resistance against pressure Tarlov cysts are mostly existent inside the sacral spinal canal. These cysts must be distinguished from other cyst-pathologies such as meningocele. The ethological connection to an underlying pathology as a CSF flow disturbance is apparent [14,21,22]. In that case a treatment would need to involve the therapy of the underlying pathological processes. Since we saw no cyst recurrence at all, we hypothesize that the presented approach is able to decrease the risk of reoccurrence and may improve the clinical outcome for patients. It seems to be broad consent that CSF dynamic may play an important role in patients with Tarlov cysts. The most important role seems to be the incapacity of physiological CSF pressure regulation [22]. As described in literature the most common cause is to be seen in the blockade of the CSF outflow mechanism at the rootlet exits inside the root-pockets [13,14]. Any kind of inflammation of the leptomeningeal sheets can lead to such blockade, eventually causing an overpressure inside the subarachnoid space. The CSF driven by its pulsatile force is later responsible for the development of the cysts inside the sacral canal. Once the pressure is irregular high a valve mechanism causes the enlargement of the cysts [13]. Below the mutual end of the dural sac, inside the sacral canal only nerve roots, venous plexus and fat tissue is to be found. The resistant against the pulsatile force at the lowest end of the water column seems to be the reason for the genesis of Tarlov or perineural cysts [23]. Following the theory of the causal connection with leptomeningeopathy and Tarlov cysts the treatment must involve both, the cysts and the leptomeningeopathy. There are more methods to reduce the intraspinal CSF pressure [24]. We performed the thecaloscopy with subarachnoid-perineurostomies and subarachnoid-subdurostomies. A permanent lumbar drainage using a lumbar-peritoneal shunt system would be an alternative option [25]. Other factors to avoid reoccurrence is the fat tissue augmentation and tight bony reconstruction of the sacral canal. As water will always find its way the watertight closure of the fistula point where the cysts are arising needs to be by packing the space with fat tissue graft and keeping all in place by the dorsal reconstruction of the sacral canal.

Whether it is due the combination of thecaloscopy and cyst resection

with sacral reconstruction or not, we experienced no cyst reoccurrence in an up to 8 year follow up.

We see in our observations additional evidence for the causal connection between the inflammatory changes as in leptomeningeopathies as the cyst formations. We did not see a single Tarlov cyst patient without endoscopically proven leptomeningeopathy. Careful closure of the fistulas point using individual techniques, fat augmentation and bony closure of the sacral canal might be mandatory to prevent further development of Tarlov cysts. The size of the cyst does not correspond to the severity of symptoms. This observation is best to be interpreted by the fact that the individual anatomy only makes the space-occupying effect relevant for nervous structures. These results indicate that combined treatment of CSF flow disturbances inside the lower spinal CSF space and the treatment of the Tarlov cysts are a useful mean to achieve permanent absence of cysts. Whether the treatment of the CSF flow disturbances requires a technical complicated procedure, as we performed with a thecaloscopy, or a permanent or temporary lumbar drainage using a shunt system is not clear yet [25]. In our study we preferred the endoscopy as it does make the cyst resection easier as far as the preservation of sacral nerve roots is concerned.

Tarlov cysts are not an individual anatomical variant but the result of a local CSF flow disturbance. Assuming this theory of origin, the causal treatment necessarily requires influencing both, the underlying leptomeningeopathy and the resulting Tarlov cyst. The CSF flow disturbances can be treated by endoscopic manners and the surgery of the cysts requires not only the excision of the cysts but also the avoidance of cyst recurrence including an anatomical reconstruction of the sacral canal with e.g., fat tissue transplantation and securing of the bony reconstruction. Among the potential causes we found in the patient's history traumas involving the spine and head, inflammation as bacterial or viral infections, manifold intraspinal cortisone injections, PDA sometimes via falsa with long standing CSF leak symptoms [26,27]. In most patients no such event could be explored. The average time between event and onset of symptoms was 84 months. Whether this was a coincidental observation, or a causal connection needs to be evaluated by further studies. As Tarlov cysts are rare and symptomatic patients are even more rare it appears almost impossible to demonstrate a causal connection between an event and a later developed pathology, and which might never become clinically relevant.

Even though the presented combined approach seems to be safe, temporary postoperative impairment of bladder control was observed frequently. We observed that patients with large amounts of residual urine prior to surgery tended to develop worsening. The function of the pudendal nerve in controlling the bladder is the crucial moment when irritation of the nerve base nerve-roots is affected by manipulation of the S2 or S3 nerve roots in the process of cyst resection.

5. Conclusion

Tarlov cysts are not an individual anatomical variant but probably the result of a local CSF flow disturbance. Inflammation of the leptomeningeal sheets can lead to such blockade, causing an overpressure inside the subarachnoid space. Following the theory of the causal connection with leptomeningeopathy and Tarlov cysts the treatment must involve both, the cysts and the leptomeningeopathy. Our data suggest that combined interventional treatment of symptomatic Tarlov Cysts is helpful to treat the long-standing symptoms.

CRediT authorship contribution statement

Michael Luchtmann: Validation, Writing – original draft. **Angelika Klammer:** Writing – review & editing, Resources. **Mircea-Alin Iova:** Formal analysis, Visualization. **André Roth:** Formal analysis, Resources. **Christian Mawrin:** Writing – review & editing, Supervision. **Jan-Peter Warnke:** Writing – original draft, Supervision.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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