

Histopathology of a symptomatic Tarlov cyst – Case report and review of the literature

J.P. Warnke^{a,*}, V. Chanamoglu^a, C. Mawrin^b

^a Department of Neurosurgery, Paracelsus-Hospital Zwickau, Germany

^b Department of Neuropathology, Otto-von-Guericke University Magdeburg, Germany

ARTICLE INFO

Keywords:

Leptomeningopathy
Tarlov-cyst
Histopathology
Tarlov cyst

ABSTRACT

Here we describe the case of a 63-year old woman with a sacral perineural cyst presenting with different symptoms such as headache, dizziness, and lower back pain with radiation and neurological claudication. The patient was treated by endoscopic assisted cyst resection and sacrum reconstruction. Histopathological examination of the cyst material revealed a cystic lesion with degenerative changes and hemorrhages. Postoperative course was uneventful, and the patients is free of symptoms three years after surgery. This case illustrates that symptomatic Tarlov cysts with extensive degenerative tissue changes can successfully managed by cyst resection.

1. Introduction

Tarlov cysts, a Type-2 meningeal cysts according to the Nabors classification [1], are nerve root cysts filled with cerebral spinal fluid (CSF) and most frequently located in the spinal canal of the S1-S5 region. They arise between the covering layer of perineurium and the endoneurium near the dorsal root ganglion. The incidence of Tarlov Cysts is rare most common in women, and most of them are asymptomatic, usually detected as incidental findings on MRI, while symptomatic Tarlov cysts seem to be rare.

Tarlov cysts were first described in 1938 by Isadore Max Tarlov [2] as an incidental finding during autopsy. Tarlov described a case of symptomatic perineural cyst and recommended its removal since then few cases has been reported [3]. Tarlov cysts are one of many mimics of discogenic radiculopathy with a reported incidence of 4.6% in back pain patients. The patients may present with lower back pain, sciatica, coccydynia, or a cauda equina syndrome. The cysts as cause for the various symptoms are frequently overseen [2,3]. The cysts are usually diagnosed on MRI which reveals the lesion arising from the sacral nerve root near the dorsal root ganglion.

Different treatment options for symptomatic Tarlov cysts have been reported. Over the past decades, many authors advised extensive surgery with sacral laminectomy and excision of the cyst along with the nerve root [4,5,6]. Recently, microsurgical excision of cyst has been advocated, combined with duraplasty or placcation of cyst wall [1]. We have observed that beside closure of the fistulas point, reconstruction of

the dorsum of sacral bone is essential in preventing reoccurrence of the condition (own observation in 52 cases, unpublished data). Here we report a case of symptomatic Tarlov cyst presenting as lower back pain and demonstrate the histopathological changes associated with a symptomatic Tarlov cyst.

2. Case description

A 61 years old female presented with radiating pain on her dorso-lateral legs with numbness off and on for 5 years. She further complained about headache, dizziness and vertigo. The pain was not associated with specific time, posture or activity. She had Peridural anesthesia 25 years ago at her 4th pregnancy, after that she had CSF leak symptoms for few days and later there are no symptoms for 12 years. Twelve years before symptoms had started with lower back pain, and during the past 5 years the intensity of pain increased gradually. The pain had progressed to the sacrum and bilateral upper thigh till ankle. Moreover, the pain became worse during activity, prolonged standing, and walking. Finally, the patient complained about rest pain as well.

The MRI of spine revealed fluid filled cystic lesions mainly from S2 and S3 nerve root Ganglion with thinning of sacrum i.e; 2 × 2.5 cm in size and the other is of 4.0 × 2.9 cm in size (Fig. 1A).

To treat the cystic lesion, endoscopic assisted microsurgical cyst resection and sacrum reconstruction was performed while retaining the nerve root. The cyst wall was sent for histopathological investigation. The operation was carried out in prone position under general

* Corresponding author.

E-mail address: jan-peter.warnke@pkd.de (J.P. Warnke).

<https://doi.org/10.1016/j.inat.2021.101426>

Received 7 October 2021; Received in revised form 8 November 2021; Accepted 8 November 2021

Available online 23 November 2021

2214-7519/© 2021 Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

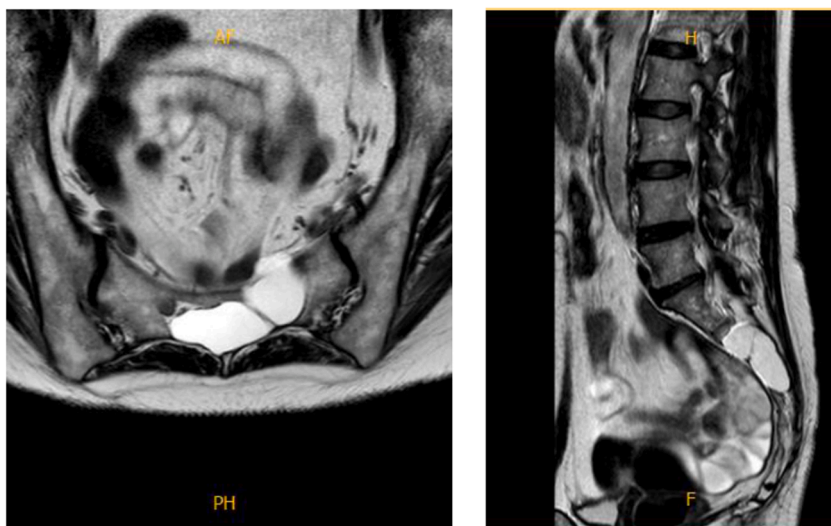
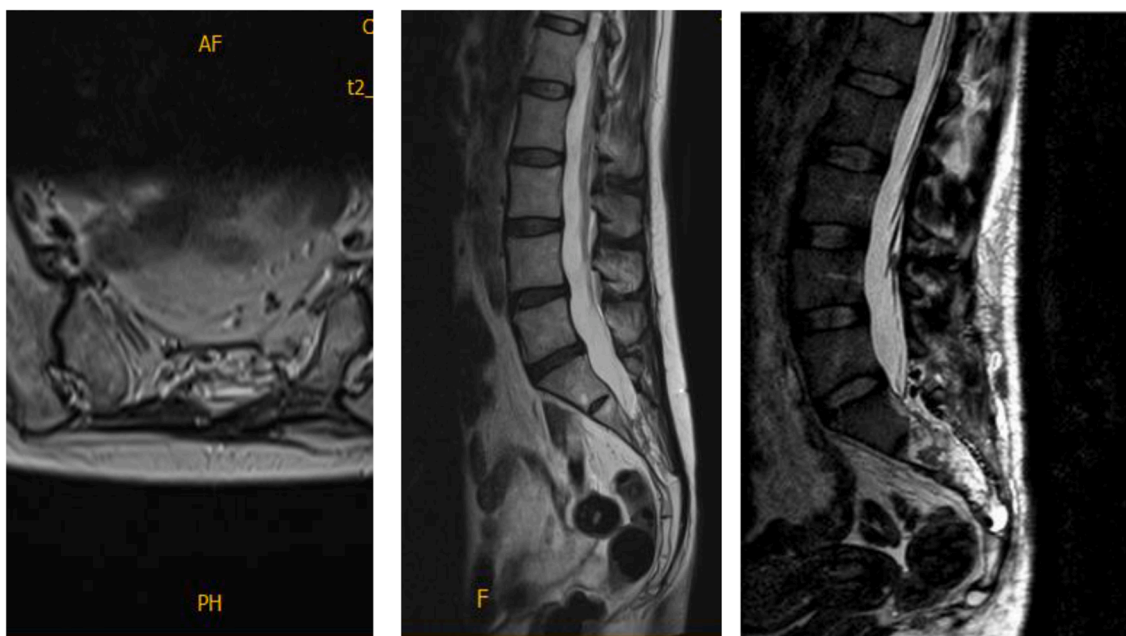
A**B**

Fig. 1. A [T2 weighted sag. Ax. MRI picture preop]. B [T2 weighted sag, ax MRI post-op].

anesthesia (Fig. 2A-C). The endoscope (Karl Storz, 12 Fr steerable, flexible) was inserted after a 4 mm trepanation of the L3 lamina, right side into the lumbar subarachnoid space. Observing the course of the nerve-roots, there position at the entrance into the cyst at the end of the subarachnoid space is detected to avoid damage later. Having the endoscope in place the surface of the dorsal sacrum was exposed using a midline skin incision under microscope. Adequate length of approached is secured by fluoroscopy. The sacrum was opened in an “open door”

technique. The cyst was evacuated, and the wall separate from surrounding nerves and venous plexus under both, the endoscopic and microsurgical view. Close of the cyst was achieved by Tachosil wrapping. Subcutaneous fat tissue was placed instead the former cyst-volume and the sacrum reconstructed using the bone flap secured by microplates and screws. Representative postoperative MRI is shown in Fig. 1B.

Following operation, patient was treated with pain relieve medication and bed rest for 3 days due to CSF loss. Slowly the patient

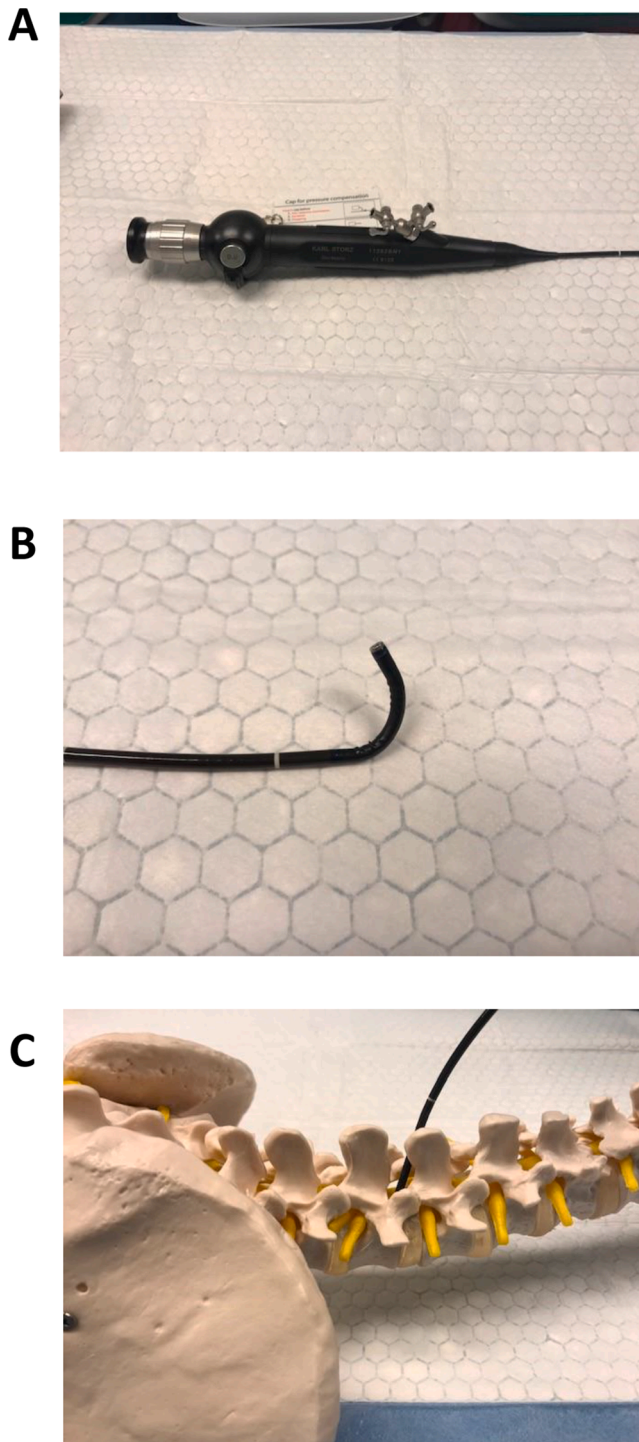


Fig. 2. Instruments and operation procedure. A flexible endoscope (A & B) is inserted into the L3 subarachnoid space (C).

appreciated relief of pain after the surgery. Post-operative MRI did not show any evidence of remaining cyst parts.

Histopathological examination of the resected cyst material revealed a cystic lesion with extensive fibrotic changes, fresh and old hemorrhages, as well as dense infiltration by macrophages (Fig. 3). Occasionally, macrophages appeared as multinucleated structures which were highlighted by CD68 immunostaining. Lymphocytic infiltrations were sparse. Neuronal tissue or nerve fibers were not detected by immunohistochemistry.

3. Discussion

It is still under debate whether Tarlov cysts are variants of anatomy, or the result of an active ongoing pathological process. Assuming an active process, different causes including inflammatory processes inside nerve roots up to the level of the Ganglia followed by fluid trap are discussed [2–4].

It remains speculation what this type of cyst makes its growing. Doubtless plays the pulsatile force of the CSF coming from the above existing CSF space a crucial role. Over years it comes to an erosion of sacral bone structures. It appears to be logic, that bigger cysts grow faster than smaller over a defined time period. Losing more and more the surface tension of the perineural structures the surfaces contact by big volumes is larger and therefore more powerful. This leads to a chronic inflammatory reaction of the perineural sheets. However, description of the histopathological changes are rare. In order to contribute to the knowledge of the genesis of perineural cysts we used the results of our histopathological findings in this case. Zheng et al. [5] described a model what demonstrates the way of CSF from the lumbar subarachnoid space between endo- and perineurium forming the cyst driven by the pulsatile force of the CSF. The pathomechanism leads to a chronic inflammatory process at the level of the cyst. Our results confirm that chronic pressure inside this CSF space extension produces the histopathological findings demonstrated above.

Hemorrhages into the cysts seem to occur [6]. In one case, the cyst wall was described consisting of perineural cells, without evidence of degeneration or hemorrhage [7]. In other cases, fibrous tissue with blood and fibrin deposition, but without signs of inflammation was reported [8]. Calcification along with granulomatous inflammation was reported in a patient following lumbar interbody fusion [9]. Fibro-collagenous, membranous connective tissue seemed to be the most frequent histopathological finding [4,10–12]. Rarely, an inflammatory component can be observed [13].

Taken together, the present case demonstrate how growth of a Tarlov cyst over long time generates extensive tissue degeneration as a contribution to sustained clinical symptoms of affected patients.

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.

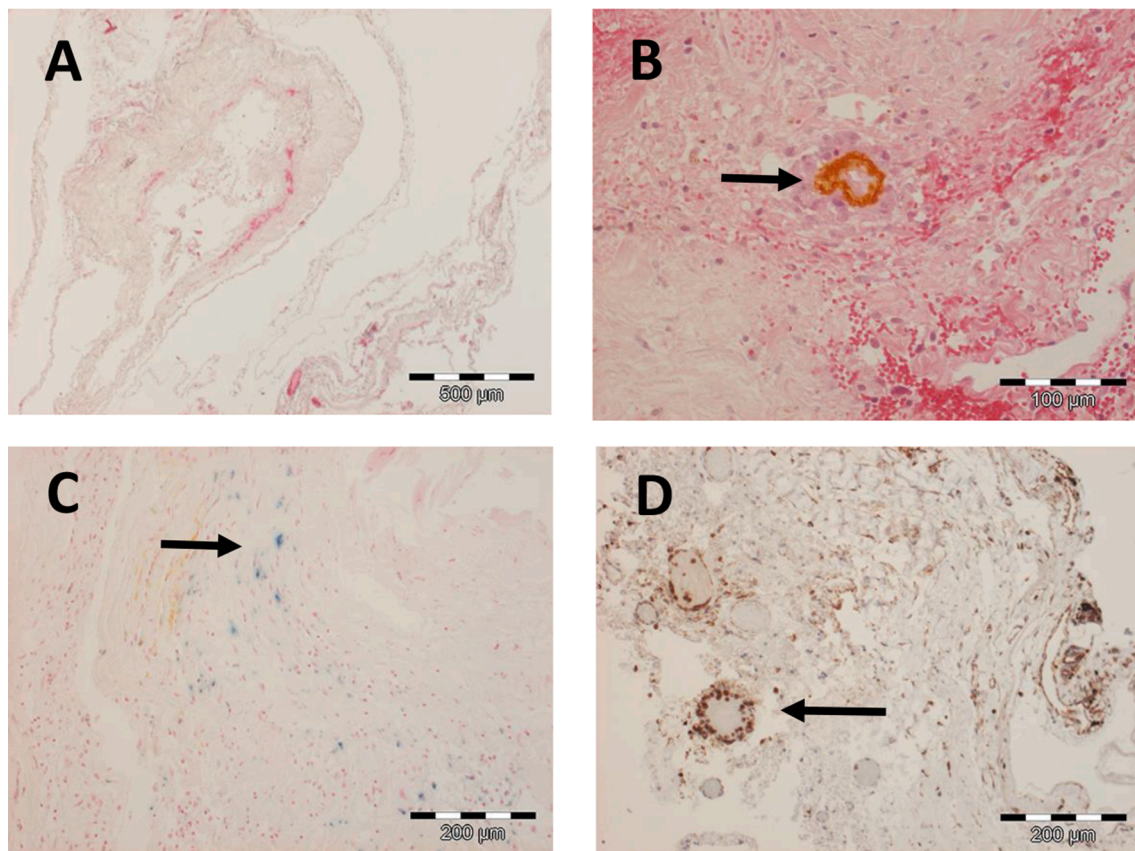


Fig. 3. Histopathological features of the resected cyst. H&E staining shows a cystic lesion with fibrotic changes (A) and several old hemorrhages (B, arrow). Iron staining (C) highlights the previous hemorrhages, while immunohistochemistry for CD68 marks frequent macrophages, some are arranged in a multicellular fashion (D, arrow).

Acknowledgement

We very much thank the Head-Genuit Foundation, Germany for generous support in generating this study. We also thank Heike Vieweger for providing the MRI pictures. Without the precision work of Mrs. Walther and Mrs. Gebhardt this work would not have been possible.

References

- [1] R. Weigel, M. Polemikos, N. Uksul, J.K. Krauss, Tarlov cysts: long-term follow-up after microsurgical inverted plication and sacroplasty, *Eur. Spine J.* 25 (11) (2016) 3403–3410.
- [2] F.L. Acosta, A. Quinones-Hinojosa, M.H. Schmidt, P.R. Weinstein, Diagnosis and management of sacral Tarlov cysts. Case report and review of the literature, *Neurosurg. Focus* 15 (2) (2003) 1–7.
- [3] I.M. Tarlov, Cysts, perineurial, of the sacral roots; another cause, removable, of sciatic pain, *J. Am. Med. Assoc.* 138 (10) (1948) 740–744.
- [4] J.M. Voyadzis, P. Bhargava, F.C. Henderson, Tarlov cysts: a study of 10 cases with review of the literature, *J. Neurosurg.* 95 (1 Suppl) (2001) 25–32.
- [5] X. Zheng, S. Li, H. Sheng, B. Feng, N. Zhang, C. Xie, Balloon-Assisted Fistula Sealing Procedure for Symptomatic Tarlov Cysts, *World Neurosurg.* 88 (2016) 70–75.
- [6] K.T. Cho, K. Nam, Perineural cyst with intracystic hemorrhage following aneurysmal subarachnoid hemorrhage: A case report, *Medicine (Baltimore)* 98 (8) (2019), e14184.
- [7] M.R. Mijalcic, B. Djurovic, I. Cvrkota, M. Jokovic, V. Bascarevic, M. Micovic, Tarlov cyst-a rare lesion in children: case report, *Childs Nerv. Syst.* 35 (4) (2019) 701–705.
- [8] Z. Aljuboory, A. Yaseen, J. Simpson, M. Boakey, Surgical excision of a symptomatic thoracic nerve root perineural cyst resulting in complete resolution of symptoms: a case report, *Cureus* 9 (6) (2017), e1343.
- [9] K.D. Than, S.U. Rahman, P.E. McKeever, A.C. Wang, F. La Marca, P. Park, Symptomatic calcified perineural cyst after use of bone morphogenetic protein in transforaminal lumbar interbody fusion: a case report, *Spine J* 13 (8) (2013) e31–e35.
- [10] H. Matsumoto, S. Matsumoto, T. Miki, Y. Miyaji, H. Minami, A. Masuda, S. Tominaga, Y. Yoshida, I. Yamaura, S. Natsume, K. Yoshida, Surgical treatment of sacral perineural cyst—case report, *Neurol. Med. Chir. (Tokyo)* 51 (12) (2011) 867–871.
- [11] A. Neulen, S.R. Kantelhardt, S.M. Pilgram-Pastor, I. Metz, V. Rohde, A. Giese, Microsurgical fenestration of perineural cysts to the thecal sac at the level of the distal dural sleeve, *Acta Neurochir (Wien)* 153(7) (2011) 1427–34; discussion 1434.
- [12] D. Guo, K. Shu, R. Chen, C. Ke, Y. Zhu, T. Lei, Microsurgical treatment of symptomatic sacral perineural cysts, *Neurosurgery* 60(6) (2007) 1059–65; discussion 1065–6.
- [13] I. Fiss, M. Danne, C. Hartmann, M. Brock, R. Stendel, Rapidly progressive paraplegia due to an extradural lumbar meningocele mimicking a cyst. Case report, *J. Neurosurg. Spine* 7 (1) (2007) 75–79.