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Case Report

# Surgical Treatment of a Tarlov Cyst in a Pediatric Patient

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## Abstract

**Background:** Tarlov cysts, commonly known as perineurial cysts, are mostly asymptomatic benign lesions that are often accidentally discovered during imaging initiated for other reasons. The pathology is very rare in children, so it may be of scientific and clinical interest to present a pediatric case with a Tarlov cyst and discuss the best therapeutic options.

**Case Presentation:** This case report relates to a 5 year old patient who presented initially with nocturnal enuresis and later with the symptom of tiptoeing. magnetic resonance imaging (MRI) unmasked a large extradural cyst that extended from L4 to S3. The cyst was completely removed by a microsurgical laminotomy from L4 to L5, from which the otherwise healthy and normally developed patient still benefits today.

**Conclusions:** The surgery performed appears to be a preferable, effective, and safe therapy for pediatric patients with Tarlov cysts.

## BACKGROUND

Perineural cysts, also called Tarlov cysts, were first seen during an autopsy by Isadore Tarlov in 1938 [1]. Typically located sacral at the junction of a dorsal ganglion with its posterior nerve root between the endoneurium and perineurium [2], they contain both ganglion cells and nerve root fibers. They develop because of an imbalance between the influence and drainage of cerebrospinal fluid (CSF) from the subarachnoid space, but the exact etiology has not yet been entirely elucidated [3,4]. Overall, it can be assumed that Tarlov cysts are rare lesions, with a prevalence of 1-5% in patients with incidental findings of the lumbosacral spine region. Depending on the size, location and nerve root compression, neurological symptoms develop. Such symptomatic courses can only be seen in approximately 1% of patients [4]. Cysts measuring more than 1 cm are more symptomatic, with localized pain, through radiculopathies, sensory disorders and muscle weakness, up to a loss of bladder and bowel function [3-5]. Various therapies are available, including the extraction of CSF from the cyst, the injection of fibrin glue and the complete or partial removal of the cyst [4,5]. Currently there is a lack of any data on the incidence of Tarlov cysts in children and, consequently, uniform therapy standards or treatment recommendations. For this reason we would like to describe the successful treatment of the following pediatric case. Due to the mostly asymptomatic course, MRI is the gold standard in diagnostics [4,6]. The therapeutic interventions extend over interventional radiology with CT-guided aspiration or lumboperitoneal shunting. On the surgical side, microsurgical procedures like cyst fenestration, cyst neck ligature, incision or peeling off the cyst bed are preferred. Serious complications are recurrences, permanent neurological deficits, sepsis, negative pressure headaches, urinary incontinence and CSF leakage, the latter as in the present case [6.7]. Tarlov reports only low recurrence rates (<10%) after microsurgical treatment. However, there were different rates of symptomatic improvement, varying between 38 and 100% [1,8]. Systematic reviews for adult patients speak of success rates of between 70 and 100% for the combination of microsurgical excision and closure with fibrin glue [5]. Little reliable evidence is available regarding the contemporary literature on Tarlov cysts in children [9]. Only a few case reports are available, which mainly consider microsurgical laminotomy to be the method of choice [5,7,10]. In order to emphasize this, we also decided to undergo microsurgical laminotomy here and to report it towards clinical colleagues. Major limitation of the present case is surely the lack of a histopathological examination, but the surgical findings, the clinical outcome and the follow-up with no signs of recurrence suggest the presence of a benign epidural cyst.

## CASE PRESENTATION

In July 2013 a 5-year-old boy was presented within our pediatric neurosurgery consultation, who has been suffering

from nocturnal enuresis since birth and walking on tiptoe since he was three years old. Clinical examination showed no pain on tapping or pressure in the entire spine and free movement. Gait variations could be demonstrated on the same side, without any evidence of key muscle paresis or sensitive deficits. The reflex status was normal on both sides, with no clone or spasticity. In the performed MRI of the entire spinal axis a large lumbosacral Tarlov cyst from L4-S5 measuring 17mm times 350mm, compressing the dural sac and the sacral nerve root pairs, was unmasked (Figure 1D/E). The spinal canal at the level of the 5th limb and sacrum is widened and completely filled by the cyst (Figure 1A/B). No signs of bleeding or flow signals were seen, just as little as pathological accumulations after contrast medium application (Figure 1C).

Due to the considerable space occupation, microsurgical removal using a laminotomy for L4/5 was agreed with the parents. We performed surgery on the first days of August 2013. Therefore the child was laid in a modified prone position and L5 was marked with methylene blue injection using a image converter. The arches were notched lengthways on both sides of the lamina. After relieving the dural sac the swollen cyst was windowed and also relieved, which was followed by the attachment of both holes to each other with silk thread. Layer-appropriate wound closure using silk thread and sealing matrix was performed as well as wound irrigation. No drainage was required and a primary wound closure supplemented by fibrin glue was made.

Postoperatively, the patient was maintained in a flat position to minimize stress on sacral nerves. We started with physiotherapy assisted mobilization at the first postoperative day and kept the child in hospital for five days for monitoring and pain medication. Due to an oozing wound and the incipient headache of the patient, we performed a wound revision, removal of the sutures and adhesive bonding of a CSF-fistula (Figure 2), four weeks later. The subsequent course and mobilization were normal, without neurological deficits or pain and dry wound conditions. Clinical observation was carried out quarterly in the first year, every six months in the second and finally annually until the neurosurgical final examination in summer 2017. Walking on tiptoes improved noticeably and disappeared after 10 weeks postoperatively. The nocturnal enuresis improved a little more slowly, lasted into elementary school and ended 1.5 years after the intervention. Regularly performed MRIs showed a completely regressed cyst with unfolded dural sac and orthotopic nerve fibers (Figure 3A/B). The connection to our social pediatric center of the cooperating children's hospital continued.

## CONCLUSIONS

Microsurgical removal of cysts including fibrin glue closure appears to be a safe and successful technique for symptomatic sacral perineural (Tarlov) cysts in pediatric populations as well. This conclusion was derived from the observed neurological improvement with no recurrence within the 5-year follow-up

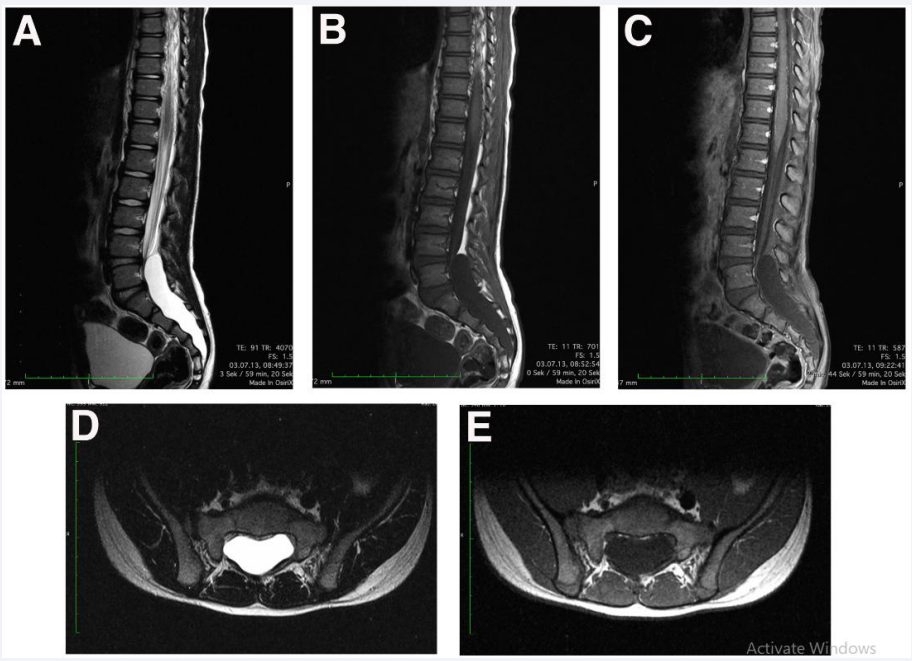


**Figure 1** Preoperative MRI imaging A : T2W sequence, sagittal view showing a large hyperintensive cystic structure projecting onto the L4 / 5 transition in the spinal canal. B : T1W sequence, sagittal view showing the same lesion with hypointense content. C : T1W sequence, sagittal view, with contrast media application, no pathological accumulations. D : T2W sequence, axial view, shows the cyst that occupies the middle part of the spinal canal and exerts considerable compression of the dural sac. E : T1W sequence, axial view, correspondingly shows a hypointense lesion.

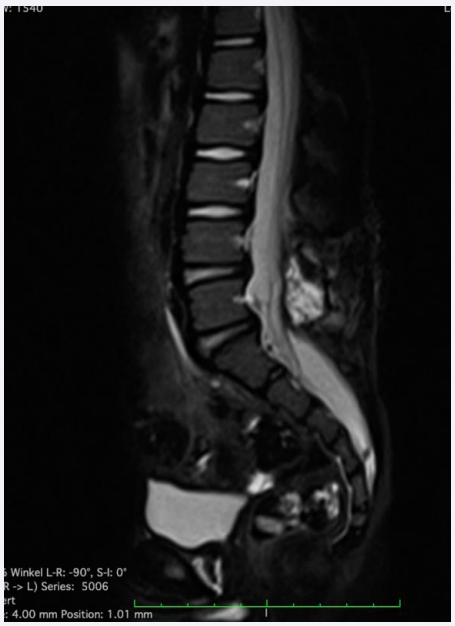


**Figure 2** Postoperative MRI imaging, T2W sequence, sagittal view showing the regressing cyst, at the level of the laminotomy a hyperintense structure with suspicion of Liquor leakage and granuloma can be seen, surrounding hypointense soft tissue swelling

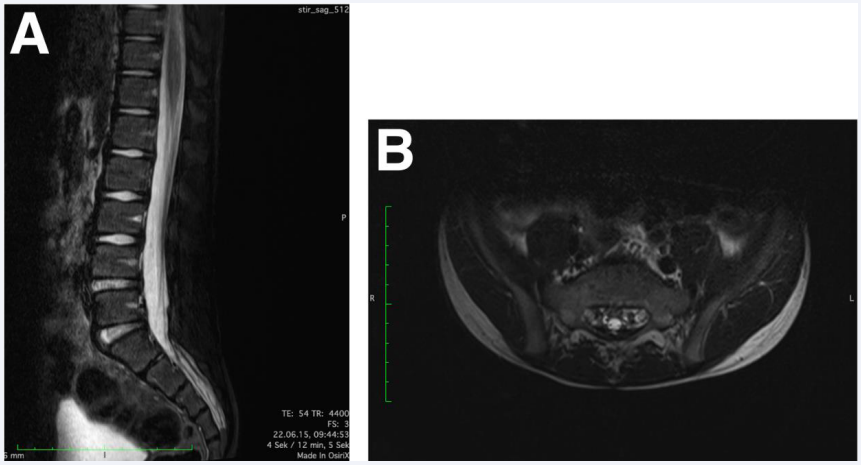


**Figure 3** Postoperative MRI imaging, A : T2W sequence, sagittal view, complete regression of the cyst, no signs of leakage or granuloma. B : T2W sequence, axial view, properly expanded dural sac with orthotopic nerve fibers

after surgery. Careful patient selection, especially in pediatric patients, remains of vital importance. In the case of large Tarlov cysts (>15mm a.p. diameter) in children, there is increasing evidence for prompt surgical treatment.

## Ethics Approval and Consent to Participate

Informed consent was obtained from all individual participants included in the study. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Ethical approval was not required for this study in accordance with national guidelines.

## Consent for Publication

The patient and his parents gave us the approval for publishing this manuscript including the MRI dataset.

## Availability of Data and Material

All data generated or analysed during this study are included in this article. Further enquiries can be directed to the corresponding author.

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## Authors' Contributions

Conceptualization, M.O.; methodology, M.O. and E.-D. R.-G.; validation, M.O., E.-D. R.-G. and V.B.; formal analysis, M.O. and E.-D. R.-G.; investigation, M.O. and E.-D. R.-G.; resources, M.O., E.-D. R.-G. and V.B.; data curation, M.O. and E.-D. R.-G.; writing - original draft preparation, M.O. and E.-D. R.-G.; writing - review and editing, M.O., E.-D. R.-G. and V.B.; visualization, M.O. and E.-D. R.-G.; supervision, E.-D. R.-G. and V.B.; project administration, E.-D. R.-G. and V.B. All authors have read and agreed to the published version of the manuscript.

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