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CASE REPORT

Tarlov cyst—a rare lesion in children: case report

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Abstract

Perineural cysts, also known as Tarlov cysts, are benign lesions increasingly found in patients undergoing neuroimaging studies. These cysts can very rarely be identified in children and even then, they are not so likely to be responsible for some neurological deficit. It seems to be of scientific and clinical importance to present a pediatric case with Tarlov cyst. We report a case of a patient, a 7-year-old boy, previously treated for nocturnal enuresis (bedwetting), who later developed signs and symptoms of classic urinary incontinence. Magnetic resonance imaging (MRI) showed a relatively large extradural cyst at the level of S2. The cyst was approached by laminectomy of L5 to S2, excised, and completely removed from the belonging nerve root. The patient has established normal sphincter control without even a single episode of involuntary discharge of urine. A surgery is a powerful, safe, and efficacious option for treatment in pediatric patients with sacral Tarlov cysts.

Keywords Tarlov cyst · Sacral perineural cyst · Surgery · Pediatric

Introduction

Perineural or Tarlov cysts are named by the American neurosurgeon Isadore Tarlov who provided a methodical description of these cysts in 1938 as an incidental finding at an autopsy [1]. These cysts are typically located at the junction of the dorsal ganglion and the posterior nerve root and usually develop between the endoneurium and perineurium of the nerve root [2, 3]. Tarlov cysts characteristically contain nerve root fibers and ganglion cells in their walls or cavities and tend to be sacral [6]. Although the etiology is still unclear, microcommunication with the subarachnoid space at the dural sleeve of the nerve root may function as a valve, allowing CSF influx and restricting CSF efflux [4, 5].

Tarlov cysts are rare lesions. Paulsen reported their incidence as 4.6% in back pain patients ($n = 500$) but only 1% of them were symptomatic [7]. The incidence of these lesions in pediatric population is still unknown and there are not enough reports about this problem even in prestigious journals all over the world.

Cysts with diameters of 1 cm or larger are more likely to be symptomatic. The clinical appearance can mimic other spinal lesions: localized pain, radiculopathy, sensory disturbance, weakness of muscles, loss of reflexes, and bladder and bowel dysfunction [1, 6, 8, 9]. Various treatment methods were offered in the past, including the extraction of CSF from the cyst, fibrin glue injection, and the complete or partial removal of the cyst.

Case presentation

History

In June 2018, a 7-year-old boy was referred to our clinic with a history of involuntary discharge of urine since his birth. The mother stated that he had daily involuntary micturition in the last year. The patient has been investigated by several doctors for possible diagnosis of enuresis nocturna. He was also followed by a pediatric urologist who performed a voiding cystourethrogram (VCUG), but this procedure did not reveal the VU reflux or any suspected obstruction. The child occasionally complained of unspecified pain in the back area that spreads along the right leg.

Examination

Examination revealed a diminished ankle jerk, mild form of plantar flexion weakness, and hypoesthesia in S1 to S4 dermatomes on the right side.

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MRI of the lumbosacral spine undertaken before surgery showed a large cyst of 26 × 19 × 13 mm in size at the level S2 of the spinal canal (Fig. 1a–d).

MRI also showed that the cyst moved the distal dural sac on its right and pushed it completely to the left. We also noticed that there is a bony defect at the posterior aspect of the sacrum (S2–S4) indicating some form of spinal dysraphism–sacral spina bifida.

Operation

On June 12, 2018, we performed the surgery. The patient was placed in the prone position and we routinely monitored SSEPs and MEPs as well as the anal sphincter. An incision was made exposing from L5 to the midsacrum. An L5 laminectomy was carried out and we have noticed non-fusion malformation of the sacrum confirming a

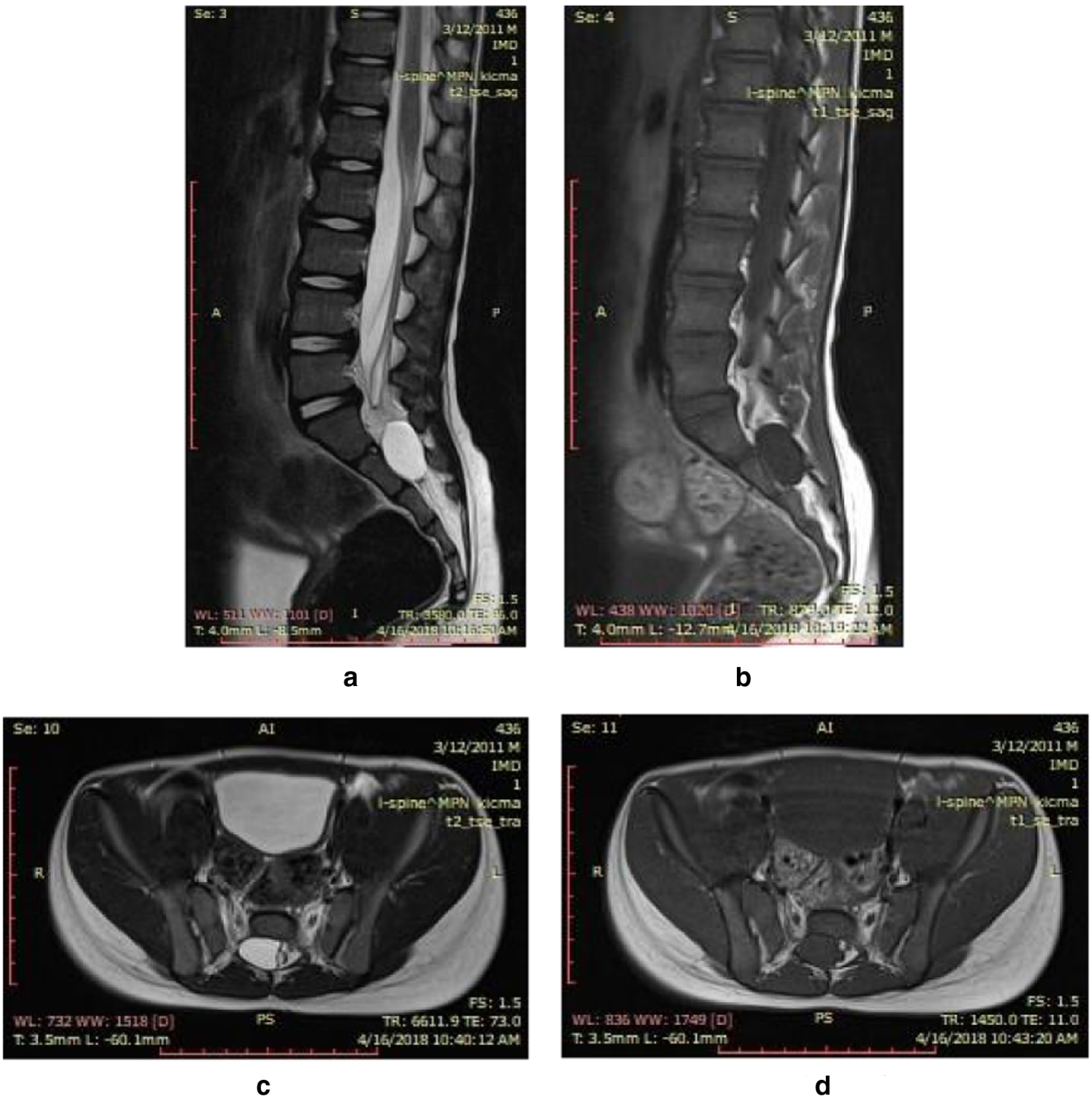

Fig. 1 **a** Preoperative MR, T2W sequence, sagittal view, showing a large hyperintense cystic structure, located in the spinal canal, at the S2 level. **b** Preoperative MR, T1W sequence, sagittal view, revealing the same cyst with hypointense content. **c** Preoperative MR, T2W sequence, axial view, showing the cyst occupying the central and right portion of the spinal canal and pushing the dural sac to the left. **d** Preoperative MR, T1W sequence, axial view

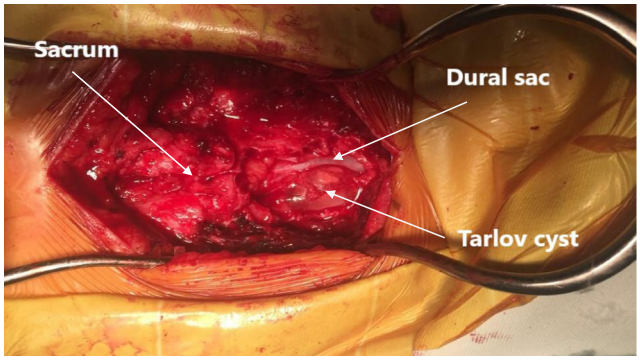

Fig. 2 Intraoperative image showing the exposed dural sac with Tarlov cyst on its right, attached to the S2 nerve root

preoperative MR finding. We have removed the dorsal wall of the sacrum up to the level of S2–S3 and identified Tarlov cyst just attached to the S2 nerve root (Fig. 2).

The cyst was aspirated removing approx. 2 ml of CSF in order to get more space for preparation and identification of the anatomical landmarks. During direct stimulation of S2 nerve root using bipolar forceps (stimulation intensity max. 2 mA, single pulse; response 20 μV), we got potentials for the right tibialis anterior muscle and external anal sphincter which was monitored by bilateral insertion of needles.

We have excised the superficial part of the cyst wall and sent it as a thin tissue fragment for further histopathological verification. We were able to excise the complete cyst wall up to the point where there is a continuation as the perineurium, and then we got a small amount of CSF coming from the proximal part of the nerve root. It was possible to see nerve fascicles of the right S2 nerve root and to preserve them from damage during cyst wall removal. In order to prevent a CSF leak, we have put a piece of fatty tissue and fixed it using a fibrin glue. We then closed the fascia and skin in a running fashion.

Postoperative course

Postoperatively, the patient was maintained on flat bed rest to minimize stress on the nerve root sheath repair. We kept the child flat for 24 h postoperatively and then allowed him to be upright. There was not even a single episode of urinary incontinence during postoperative recovery period. The child was discharged on the 6th postoperative day. Histopathology finding reveals the layers of flattened cells, actually connective tissue cells originating from the perineurium that built the wall of the Tarlov cyst, surrounded by a basement membrane and collagen fibers (Fig. 3).

We have done a follow-up MR lumbosacral spine 2 months after the surgery which confirmed that Tarlov cyst was completely removed (Fig. 4a–c). At that time, 2 months after the surgery, the patient's mother mentioned that he had only one episode of involuntary voiding during jumping, playing, and effort, but this event never occurred again.

Discussion

Perineural cysts are formed in the perineural space among the endoneurium derived from the pia mater and the perineurium shaped by the arachnoid mater. MRI is now the technique of choice in finding perineural cysts.

The surgical options are as follows: (1) diversion of the CSF flow (CT-guided percutaneous aspiration, lumboperitoneal shunt); and (2) direct microsurgical methods (cyst fenestration, cyst neck ligation, cyst resection, and combinations of the above). Each option comes with the possibility of serious complications such as cyst recurrence, neurological deficits, infection and inflammation, spinal headache, urinary disturbances, and CSF leakage.

Despite low rates of cyst recurrence (range 0–10%), different rates of symptomatic improvement have been reported in association with microsurgical treatment, varying from 38 to 100% [10, 11]. Nevertheless, there is an extremely small number of publications in which one can find some valuable data regarding Tarlov cysts in children and that was the primary motive for us to report this case.

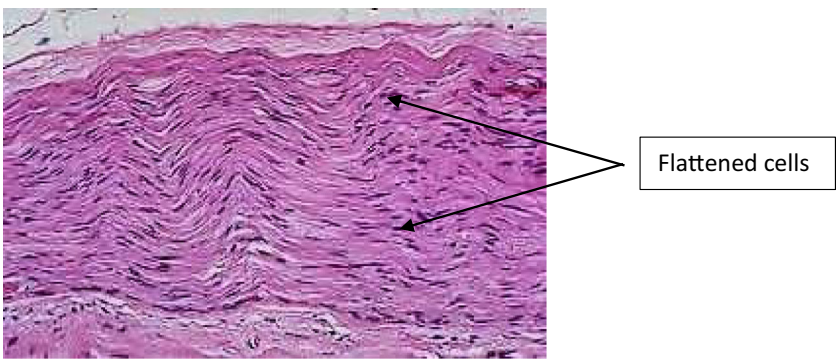

Fig. 3 Histopathology sample seen under the microscope revealing the layers of flattened cells that actually belong to perineurium and build the wall of the Tarlov cyst, surrounded by a basement membrane and collagen fibers

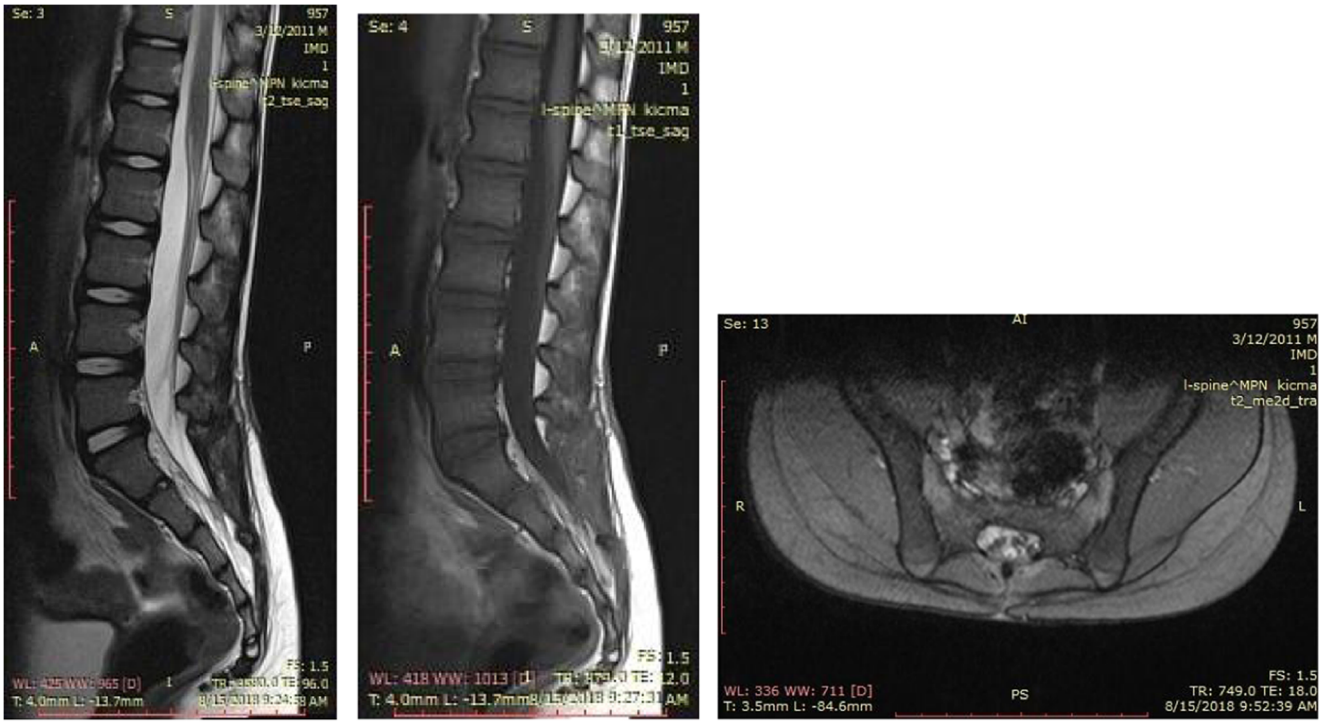

Fig. 4 **a** Postoperative MR, T2W sequence, sagittal view, confirming the total excision of cyst. **b** Postoperative MR, T1W sequence, sagittal view. **c** Postoperative MR, T2W sequence, axial view, showing early postoperative changes

Conclusion

A small percentage of Tarlov cysts may be symptomatic, and an even smaller percentage of all these symptomatic lesions can be found in pediatric population.

We can assume that the surgical treatment of Tarlov cysts is optional if symptomatic cases are selected suitably. The best management option must be determined by the size (surgery is usually indicated for the cysts larger than 1.5 cm in diameter) and location of the cyst. In our opinion, the indications for surgery in pediatric patients should be more flexible than in adults, knowing that symptoms in children disappear more quickly and usually completely withdraw.

Compliance with ethical standards

Conflict of interest On behalf of all authors, the corresponding author states that there is no conflict of interest. None of the authors have any competing interests in the manuscript.

Declarations All authors agree that this manuscript could be published.

Ethics, consent, and permissions The patient and his parents gave us the approval report for publishing this manuscript. Informed consent was obtained from all individual participants included in the study. The ethics committee of the Clinical Center of Serbia approved publication of this manuscript.

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