



Case Report

Severe constipation due to sacral perineural cysts in a pediatrics patient: A case report

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ABSTRACT

Background: Symptomatic perineural cysts are rare in pediatric patients. Severe bowel dysfunction caused by the perineural cysts at the sacral level is particularly rare in children. Moreover, the long-term outcome of surgery for the perineural cysts in pediatric patients is uncertain. Here, we describe a rare case of perineural cysts originating in the S3 roots in a pediatric patient that manifested as severe constipation.

Case Description: The case was a 13-year-old girl who presented with intractable vomiting and constipation. She also had low back and buttock pain, urinary incontinence, and periproctal sensory disturbance. Radiological studies revealed bilateral perineural cysts originating from the S3 nerve roots, which were considered to be the cause of her symptoms. Microsurgical decompression of the cysts relieved her intractable bowel dysfunction. There has been no recurrence in the 5 years since surgery.

Conclusion: This case suggests that microsurgery for severe bowel dysfunction due to symptomatic perineural cysts could have a satisfactory long-term outcome in pediatric patients.

Keywords: Bowel dysfunction, Pediatric patient, Perineural cyst, Tarlov cyst

INTRODUCTION

Perineural cysts, also known as Tarlov cysts, develop between the endoneurium and perineurium of a nerve root.^[6] Sacral perineural cysts are usually asymptomatic, but may be detected incidentally by magnetic resonance imaging (MRI).^[6] However, the incidence in children is still unknown.^[7,9] These lesions may cause a variety of symptoms, including low back pain, sciatic pain, and bowel and bladder dysfunction.^[6] Although meticulous care is needed to clearly illustrate the relationship between the symptoms and radiological findings,^[6] several reports suggest that surgery may be effective for these lesions.^[2,3,8,11] However, only a few reports in pediatric patients have been published.^[4,5,7,12] Moreover, there is uncertainty about the long-term outcome of surgery for these lesions in children. Here, we describe a very rare pediatric case of perineural cysts, originating in the S3 roots that manifested as severe bowel dysfunction in which a good outcome was achieved by microsurgical decompression.

CASE DESCRIPTION

A 13-year-old girl presented with an intractable vomiting and constipation that did not respond to medical treatment. She was referred to our department when MRI revealed cysts

at the sacral level. She was found to have low back and buttock pain, urinary incontinence, and periproctal sensory disturbance.

The MRI showed intestinal and bladder distension, suggesting severe bowel and bladder dysfunction [Figure 1] with perineural cysts, originating in both the S3 nerve roots at the S2 vertebral level [Figure 2]. The intensity of the cysts was the same as that of cerebrospinal fluid [Figure 2]. Myelography revealed an immediate influx of contrast medium into the cysts, indicating influx of cerebrospinal fluid [Figure 3]. Computed tomography (CT) revealed erosion of the sacral lamina and scalloping of the S2 vertebra. The cysts were considered to be the cause of her symptoms. The decision was made to shrink the cysts by microsurgery.

A bilateral partial laminectomy was performed at S1-2. The cysts, dural tube, and nerve roots were detected [Figure 4a]. The cyst wall was incised and the S3 nerve root was detected inside the cyst [Figure 4b]. The flaccid cyst wall was suitably cut with caution, to prevent S3 nerve root injury. Moreover, then, the autologous fat tissue and fibrin glue were used

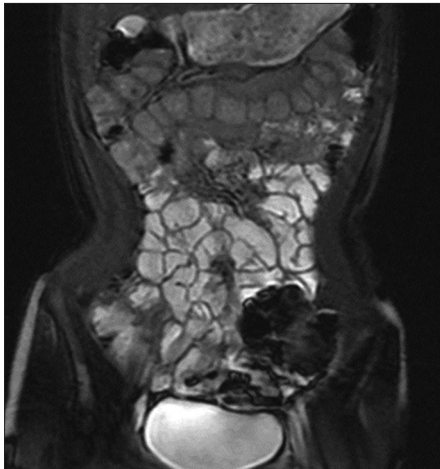


Figure 1: Magnetic resonance image showing intestinal and bladder distension.

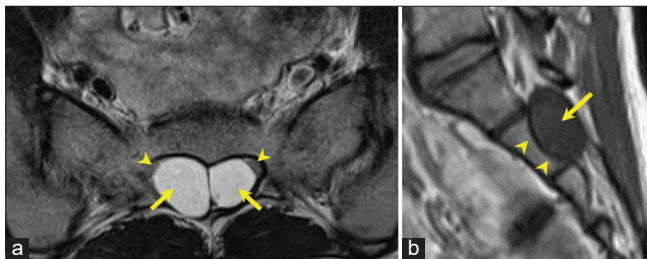


Figure 2: Findings on preoperative magnetic resonance images. (a) T2-weighted image showing high-intensity cysts (arrows), compressing S2 roots (arrowheads) on both sides. (b) T1-weighted image showing a low-intensity cyst (arrow) with scalloping of the S2 vertebra (arrowheads).

to seal the proximal end of the S3 nerve root in the cyst [Figure 4c]. The cyst wall was sutured continuously along the S3 nerve root [Figure 4d], and the epidural space was packed with fat tissue and fibrin glue. Finally, the cysts were shrunk on both sides and the epidural space was released from the compression caused by the cysts.

After the surgery, her severe bowel dysfunction was relieved. Although she complained of radicular pain in the left thigh after the surgery, the pain was relieved later. Moreover, there has been no recurrence until 5 years after her surgery [Figure 5]. The histological diagnosis was consistent with that of perineural cyst.

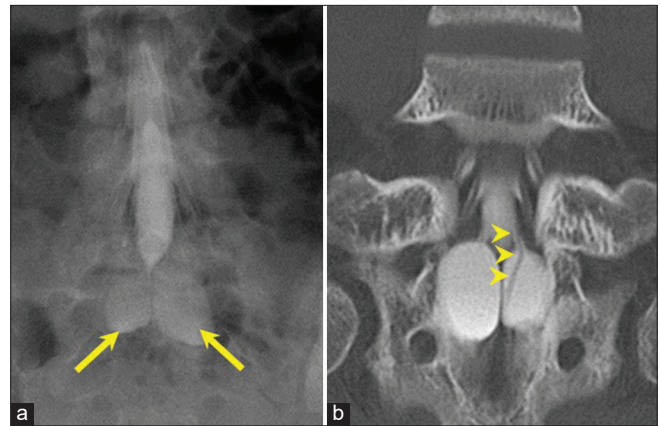


Figure 3: Preoperative myelogram and computed tomography myelogram. (a) Myelogram showing bilateral sacral cysts (arrows) with immediate influx of contrast medium. (b) Computed tomography myelogram obtained 3 h after injection shows contrast medium in the cysts that originated from the S3 roots (arrowheads).

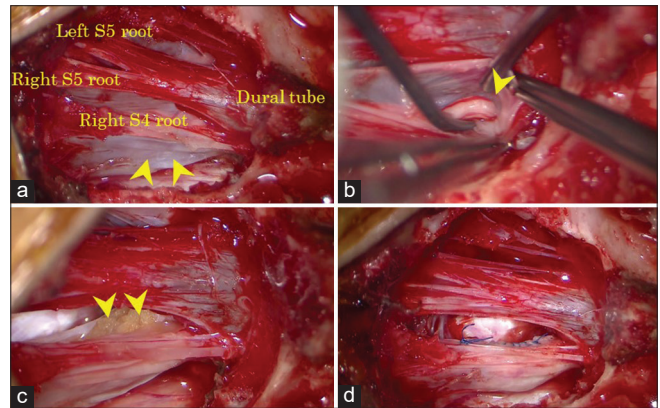


Figure 4: Intraoperative photographs. (a) Photograph showing the perineural cyst (arrowheads), dural tube, and nerve roots after bilateral S1-2 partial laminectomy. (b) The right S3 root is seen within the cyst after the cyst wall incision. The proximal end of the S3 root in the cyst (arrowhead). (c) Sealing of the proximal end of the S3 root in the cyst using autologous fat tissue (arrowheads) and fibrin glue. (d) Cyst wall was sutured.

Table 1: Literature review.

Journal	Year	First author	Age, sex	Main symptoms	Treatment	Follow-up (months)	Outcome
<i>Eur Spine J</i>	2016	Elsawaf et al.	7 years, M	Nocturnal enuresis	Surgical excision	27	Improving
<i>Eur Spine J</i>	2016	Elsawaf et al.	7 years, F	Nocturnal enuresis	Surgical excision	77	Improving
<i>World Neurosurg</i>	2019	Dayyani and Zabihyan	8 months, F	Abdominal distension Instability in sitting position (Atypical giant perineural cyst)	Surgical excision	6	Improving (Cyst peritoneal shunt for recurrence)
<i>Childs Nerv Syst</i>	2019	Mijalcic et al.	7 years, M	Urinary incontinence	Surgical excision	2	Improving
<i>Childs Nerv Syst</i>	2021	Yoshioka et al.	7 years, F	Urinary and fecal incontinence	Rotation flap technique	24	Improving

Eur Spine J: European Spine Journal, World Neurosurg: World Neurosurgery, Child's Nerv Syst: Child's Nervous System, M: Male, F: Female

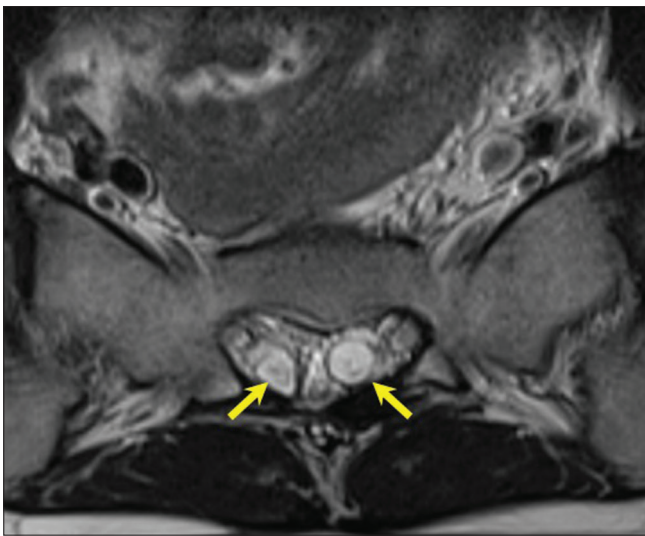


Figure 5: Magnetic resonance image showing no recurrence in the 5 years since surgery. Arrows indicate shrunken perineural cysts.

DISCUSSION

We have encountered an exceedingly rare case of perineural cysts originating from the S3 nerve roots of a pediatric patient that manifested as severe bowel dysfunction and was treated successfully by microsurgical decompression. A search of the literature revealed only a few reports on surgical outcome of pediatric perineural cysts [Table 1].^[4,5,7,12] In adults, although sacral perineural cysts are usually asymptomatic, these lesions cause a variety of symptoms, including low back pain, sacrococcygeal pain, perineal pain, sciatic pain, leg weakness, neurogenic claudication, and bowel, bladder, and sexual dysfunction.^[6] Urinary disturbance was the main symptom mentioned in the previous reports involving pediatric patients.^[5,7] In our case, the main symptoms were vomiting and constipation, suggesting very severe bowel disturbance. Sacral MRI should be performed in pediatric patients who

present with severe bowel disturbance of unknown etiology to exclude these lesions.

Although various treatment strategies have been described, there is no consensus on the treatment of perineural cysts.^[10] In addition, because these lesions are usually asymptomatic, meticulous care is needed to clearly uncover the relationship between the symptoms and radiological findings and to determine the indication of intervention.^[6] Minimally invasive nonsurgical procedures, including aspiration and percutaneous injection of fibrin glue, have been reported.^[10] Although there was no difference in symptom recurrence between surgical and percutaneous interventions, the incidence of cyst recurrence was reported to be lower after surgical interventions than after percutaneous interventions.^[10] Surgical methods are reportedly effective in adults,^[2,3,8,11] with younger age and a shorter duration of preoperative symptoms being associated with good outcome.^[1] Surgery is also effective for associated bladder dysfunction.^[3] However, most of the previous reports on surgical treatment of these lesions were in adults, and the long-term outcome of the surgery in pediatric patients remains unclear. A few reports indicate that surgery is an effective treatment for urinary dysfunction caused by this lesion in pediatric patients^[5,7,12] and one report mentioned a good long-term outcome in a pediatric patient with bowel symptom^[12] [Table 1]. This study suggests that early surgical treatment for not only urinary but also bowel dysfunction due to this lesion could also have a satisfactory long-term outcome in pediatric patients.

CONCLUSION

This case suggests that microsurgery for severe bowel dysfunction due to symptomatic perineural cysts could have a satisfactory long-term outcome in pediatric patients.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Burke JF, Thawani JP, Berger I, Nayak NR, Stephen JH, Farkas T, *et al.* Microsurgical treatment of sacral perineural (Tarlov) cysts: Case series and review of the literature. *J Neurosurg Spine* 2016;24:700-7.
- Cantore G, Bistazzoni S, Esposito V, Tola S, Lenzi J, Passacantilli E, *et al.* Sacral Tarlov cyst: Surgical treatment by clipping. *World Neurosurg* 2013;79:381-9.
- Caspar W, Papavero L, Nabhan A, Loew C, Ahlhelm F. Microsurgical excision of symptomatic sacral perineurial cysts: A study of 15 cases. *Surg Neurol* 2003;59:101-5.
- Dayyani M, Zabihyan S. Giant Tarlov cyst of infancy. *World Neurosurg* 2019;123:348-50.
- Elsawaf A, Awad TE, Fesal SS. Surgical excision of symptomatic sacral perineurial Tarlov cyst: Case series and review of the literature. *Eur Spine J* 2016;25:3385-92.
- Lucantoni C, Than KD, Wang AC, Valdivia-Valdivia JM, Maher CO, La Marca F, *et al.* Tarlov cysts: A controversial lesion of the sacral spine. *Neurosurg Focus* 2011;31:E14.
- Mijalcic MR, Djurovic B, Cvrkota I, Jokovic M, Bascarevic V, Micovic M. Tarlov cyst-a rare lesion in children: Case report. *Childs Nerv Syst* 2019;35:701-5.
- Mummaneni PV, Pitts LH, McCormack BM, Corroo JM, Weinstein PR. Microsurgical treatment of symptomatic sacral Tarlov cysts. *Neurosurgery* 2000;47:74-8.
- Ramadorai UE, Hire JM, DeVine JG. Magnetic resonance imaging of the cervical, thoracic, and lumbar spine in children: spinal incidental findings in pediatric patients. *Glob Spine J* 2014;4:223-8.
- Sharma M, SirDeshpande P, Ugiliweneza B, Dietz N, Boakye M. A systematic comparative outcome analysis of surgical versus percutaneous techniques in the management of symptomatic sacral perineural (Tarlov) cysts: a meta-analysis. *J Neurosurg Spine* 2019;30:623-34.
- Weigel R, Polemikos M, Uksul N, Krauss JK. Tarlov cysts: Long-term follow-up after microsurgical inverted plication and sacroplasty. *Eur Spine J* 2016;25:3403-10.
- Yoshioka F, Shimokawa S, Masuoka J, Inoue K, Ogata A, Abe T. Elimination of the check-valve mechanism of the sacral Tarlov cyst using a rotation flap technique in a pediatric patient: Technical note. *Childs Nerv Syst* 2021;37:1741-5.

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