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A Case of an S2 Tarlov Cyst Causing S1 Radiculopathy

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Abstract

Symptomatic Tarlov cysts are rare. We present a case of an S2 Tarlov cyst causing severe S1 radiculopathy. A 37-year-old male presented with intractable pain over the right S1 dermatomal distribution. MRI revealed a right S2 Tarlov cyst that slightly compressed the adjacent S1 nerve root. His pain could not be controlled with medication. He underwent right-sided laminectomy at the S1 and S2 levels. The right S1 nerve root was found to be compressed by the right S2 Tarlov cyst. The right S2 Tarlov cyst wall was carefully incised and re-sutured to reduce the cyst size. This procedure relieved the compression of the S1 nerve root by the cyst.

Postoperatively, the lower limb pain disappeared. One month later, his lower limb pain recurred, but it was manageable with medication, and there was no further worsening. The patient has been stable with reduced size of the cyst for two years. This case highlights that Tarlov cysts can cause pain in dermatomal distributions different from their involved nerve roots.

Categories: Orthopedics, Neurosurgery
Keywords: microsurgery, perineural cysts, radiculopathy, spine, tarlov cysts

Introduction

Tarlov cysts are cystic lesions caused by the accumulation of cerebrospinal fluid between the perineurium and the endoneurium [1]. Symptomatic Tarlov cysts are rare [2]. However, Tarlov cysts can cause lower limb pain, bowel dysfunction, and sexual dysfunction [2]. Some reports suggest that pain in the lower extremities is caused by the enlargement of Tarlov cysts or increased intra-cystic pressure [3,4]. Nevertheless, the detailed underlying mechanism remains unclear [4]. Furthermore, these hypotheses cannot explain why symptoms occur in nerve roots different from the root involved with the Tarlov cyst. In this report, we present a case of S2 Tarlov cyst showing intractable lower limb pain over the S1 dermatome. During surgery for the S2 Tarlov cyst in this case, the adjacent S1 nerve root was found to be compressed by the S2 cyst. The possible mechanisms and symptoms are discussed.

Case Presentation

A 37-year-old male presented to our outpatient clinic with the chief complaint of pain in his right lower limb, which was resistant to one year of medications, including loxoprofen, acetaminophen, tramadol, and eperisone. He exhibited weakness of the right gastrocnemius muscle (manual muscle testing (MMT) 5/5, but he had difficulty standing on his tiptoes), along with right S1 dermatomal paresthesia. Magnetic resonance imaging (MRI) revealed a 34-mm Tarlov cyst involving the right S2 nerve root (Figures *1A*, *1D*). There were no other obvious lesions that could cause right S1 radiculopathy, such as disc herniation at the L5/S1 level or stenosis of the right S1 sacral foramen. The patient lived far from our hospital, and due to the patient's time limitation, we were unable to perform somatosensory evoked potential (SEP)/electrodiagnosis (EDX) or transforaminal selective nerve root blocks (SNRB). However, based on the combination of S1 dermatomal paresthesia, weakness of the gastrocnemius muscle, and MRI findings, S1 radiculopathy caused by the S2 Tarlov cyst was the most likely cause of the patient's symptoms.

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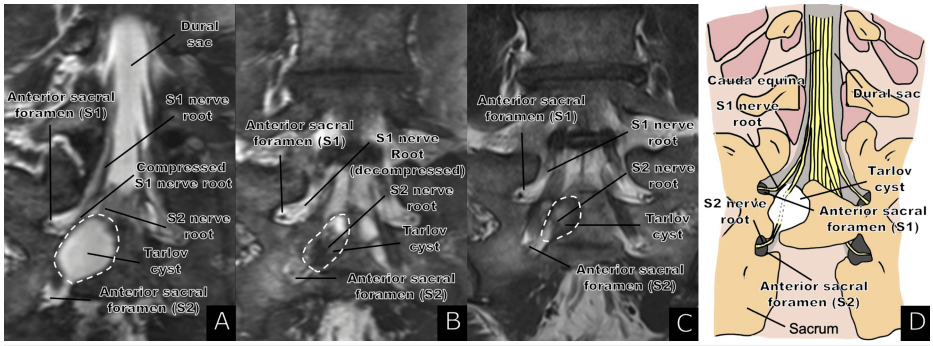


FIGURE 1: Pre and postoperative magnetic resonance imaging (MRI) findings

A: Preoperative T2-weighted coronal images revealed a 34-mm-diameter Tarlov cyst containing the right S2 nerve root and compressing the right S1 nerve root.

B: The Tarlov cyst had shrunk (27-mm-diameter) on the T2-weighted coronal images taken 3 days after surgery.

C: The Tarlov cyst remained shrunk (22-mm-diameter) even on the T2-weighted coronal images taken two years after surgery.

D: An original illustration representing the anatomical structures corresponding to the MRI images, created by the authors using Clip Studio Paint (Version 2.0; Celsys, Tokyo, Japan).

Surgery was attempted based on the assumption that a Tarlov cyst involving the right S2 nerve root, slightly compressing the S1 nerve root, was the cause of the lower limb pain. A large nerve root cyst was exposed through a right-sided laminectomy at the S1 and S2 levels (Figures *2A*-*2D*). The large S2 cyst wall was incised, and the underlying arachnoid membrane was opened (Figures *2E*-*2H*). This procedure confirmed an S2 nerve root on the inner surface of the cyst wall (Figures *2G*, *2H*). After careful separation of the fanning nerve root from the arachnoid membrane, the cyst wall was partially resected and sutured to reconstruct the right S2 nerve root (Figures *2I*, *2J*). Subsequent to the retraction of the S2 Tarlov cyst, another nerve root was visualized, which was confirmed to be the right S1 nerve root by intraoperative motor evoked potential (Figures *2K*-*2M*). We used a micro-dissector to confirm that adequate decompression of the S1 nerve root had been achieved. To prevent postoperative cerebrospinal fluid leakage, fibrin glue was applied after the dura mater was sutured.

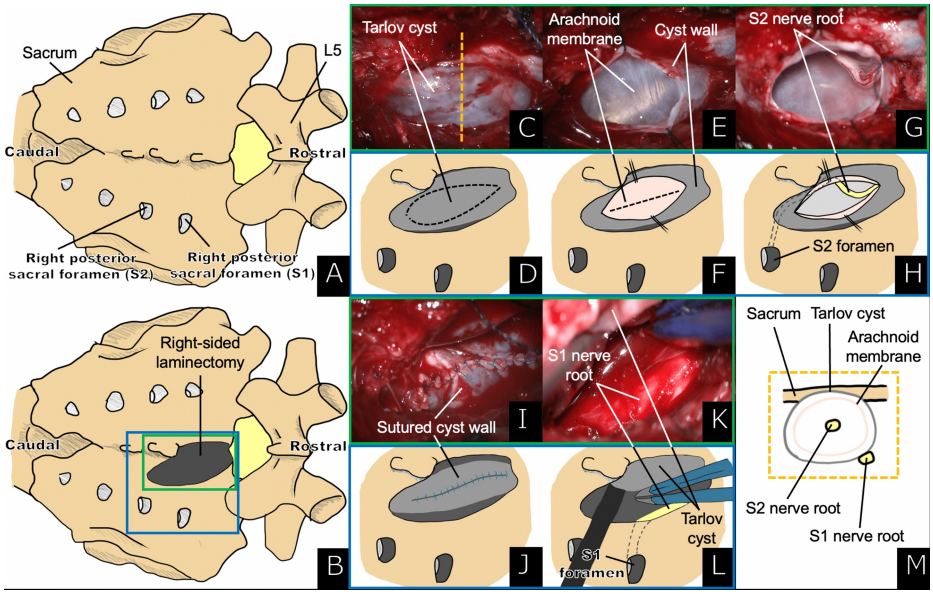


FIGURE 2: Intraoperative findings of the surgery for the Tarlov cyst. In each figure, the rostral is to the right side.

A: An illustration showing the dorsal surfaces of the sacrum and L5 vertebral body. The locations of the posterior sacral foramina of S1 and S2 are indicated.

B: A right-sided laminectomy was performed from S1 to S2. In the surgical images below, the area outlined in green is shown, while in the corresponding illustrations, the area outlined in blue is shown. To facilitate anatomical understanding, the illustrations include the right posterior sacral foramen of S1 and S2.

C, D: A right-sided laminectomy revealed the posterior surface of the Tarlov cyst. The Tarlov cyst within the area indicated by the dashed line was resected.

E, F: After opening the Tarlov cyst's wall, the arachnoid membrane containing the cerebrospinal fluid and S2 nerve root in the cyst appeared. The arachnoid membrane was incised along the dashed line.

G, H: A partial resection of the arachnoid membrane in the Tarlov cyst was performed. The right S2 nerve root was identified in the cyst and carefully separated from the cyst wall. The dashed line in the illustration indicates the course of the S2 nerve root, which is not visible in the surgical images.

I, J: The cyst wall was sutured to form smaller-sized S2 nerve root. When reconstructing the cyst wall, we sutured while confirming the position of the S2 nerve root to ensure it was protected by the arachnoid membrane and not damaged.

K, L: After reconstruction of the S2 Tarlov cyst, the right S1 nerve root became fully visible. The right S1 nerve root was adherent to the ventral aspect of the spinal canal along with the surrounding tissues. The adhesions were carefully dissected and the S1 nerve root was released.

M: Axial view of a Tarlov cyst, corresponding to the orange dashed line in 2C. The S2 nerve root was contained within the arachnoid membrane inside the cyst. The Tarlov cyst was compressing the S1 nerve root.

A, B, D, F, H, J, L, M: Are original illustrations created by the authors using Clip Studio Paint (V 2.0; Celsys, Tokyo, Japan).

The postoperative course was uneventful, and the lower limb pain disappeared. There was no cerebrospinal fluid leakage, pseudomeningocele, or new neurological deficit. One month later, his lower limb pain recurred, but it was kept under control for two years with mirogabalin medication. Postoperative MRI revealed a shrunken S2 cyst, which was maintained for as long as two years after surgery (Figures *1B*, *1C*). Since heavily T2-weighted sequences were prioritized to obtain a detailed understanding of the structures surrounding the nerve roots, contrast-enhanced MRI was not performed. Due to patient-related constraints, such as place of residence, we were unable to perform EDX after surgery or following a recurrence of symptoms.

Discussion

The prevalence of Tarlov cysts is reported to be 4.18% [5]. Most Tarlov cysts are asymptomatic and require no treatment [2]. Surgical intervention is occasionally performed for symptomatic Tarlov cysts [1-4,6]. The

detailed mechanism underlying the development of symptoms in Tarlov cysts remains unclear [4]. Several reports suggest that the enlargement of the cysts or increased pressure causes lower limb pain [3,4,7].

The present case developed right S1 nerve root symptoms while the large Tarlov cyst was present at the S2 nerve root. Such discrepancy between the level of the lesion and dermatomal distribution of pain often confuses clinicians. Imaging studies showed a large cyst of the right S2 nerve root, slightly compressing the S1 nerve root in the caudal direction. Although it could not be performed in this case due to patient-related constraints, SEP/EDX and transforaminal SNRB may be useful for identifying the affected nerve root. Furthermore, if puncture of the Tarlov cyst is technically possible, percutaneous drainage may be performed as a diagnostic and therapeutic option [1,2]. In this case, however, the Tarlov cyst was surrounded by thick bone, making puncture impossible. Intraoperative findings revealed that the S1 nerve root was compressed by the S2 cyst and adhering to the surrounding tissues. The lower extremity pain was alleviated by decompressing the S2 cyst and releasing the S1 nerve root. Recent reports suggest that releasing the nerve root is effective in symptom improvement [8,9]. The recurrence of mild lower extremity pain one month after surgery may reflect postoperative adhesions or inflammatory changes.

Surgery is considered for Tarlov cysts that are resistant to medical treatment [2]. However, there is no established surgical procedure for Tarlov cysts [1,2]. It has been reported that, among 10 patients who underwent Tarlov cyst resection following laminectomy, all 7 patients with cysts measuring 1.5 cm or larger experienced symptom improvement [10]. Thus, symptomatic Tarlov cysts larger than 1.5 cm in diameter are considered to be an indication for surgery [1,10]. Although percutaneous procedures such as CT-guided drainage or fibrin glue injection are sometimes performed, there are concerns about symptom recurrence [1,2]. Furthermore, a recent systematic review has shown that the cyst recurrence rate is significantly lower with microsurgical procedures than with percutaneous procedures (8% vs 20%) [11]. Therefore, microsurgical procedures such as cyst suturing and cystectomy, as performed in this case, will continue to be one of the valuable options for treating drug-resistant Tarlov cysts. However, the 1.5 cm cutoff is based on a limited number of case series, and the decision to perform surgery should be made based on each patient's symptom severity, clinical-radiological concordance, exclusion of alternative diagnoses, response to conservative treatment, and the potential risks of intervention.

Our case report clearly demonstrated that Tarlov cysts can cause pain in dermatomal distributions different from the involved nerve root. Clinicians should be aware of these clinical features of Tarlov cysts when they manage lower extremity pain.

Conclusions

This case highlights that Tarlov cysts can cause pain in dermatomal distributions different from their involved nerve roots. Surgical treatment is considered for drug-resistant Tarlov cysts. Microsurgical procedures, such as cyst suturing and cystectomy, are valuable options for Tarlov cysts.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Takuma Takano, Genki Uemori, Nobuyuki Mitsui, Izumi Koyanagi, Manabu Kinoshita

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Drafting of the manuscript: Takuma Takano

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Supervision: Manabu Kinoshita

Disclosures

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