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A Case of Symptomatic Multiple Tarlov Cysts Treated with Microsurgical Wrapping Technique —Efficacy and Limitation of Surgical Procedure—

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Abstract

Tarlov cysts (TCs) rarely cause clinical symptoms, such as leg pain, buttock pain, and bladder/bowel dysfunction. Surgery is considered when these symptoms persist despite medical treatments. Among several surgical procedures, microsurgical wrapping (MSW) is a relatively novel, simple technique with few complications, including cerebrospinal fluid leakage. Herein, we report a case of multiple TCs treated with MSW and present the mechanism of symptoms generated by TC and the procedure's limitations. A 58-year-old man complained of severe right leg and buttock pain for 3 months and was admitted to our hospital. His symptoms aggravated with sitting and standing and improved with the prone position. Spinal magnetic resonance imaging (MRI) demonstrated multiple sacral cysts containing intense cerebrospinal fluid. The cysts connect to the right S3 and S4 nerve roots. He was treated conservatively with medications; however, his symptoms were not improved. Therefore, MSW was performed for TCs connected to the S3 and S4 roots. The postoperative course was uneventful, and cerebrospinal fluid leakage did not occur. MRI performed 1 year after the operation demonstrated no recurrence of the TCs, and his leg pain was completely relieved; however, the buttock pain remained. MSW for TCs is effective for symptoms of adjacent nerve root compression; however, repairing the damaged nerve root in TCs is sometimes difficult. This may be a limitation of present surgical interventions because these symptoms may be difficult to treat even with other interventions.

Keywords: buttock pain, microsurgical wrapping, perineural cyst, sacrum, Tarlov cyst

Introduction

Tarlov cysts (TCs) are extradural meningeal cysts containing cerebrospinal fluid (CSF) arising between the endoneurium and perineurium of the nerve root sheath.[1)] The incidence of TCs is approximately 1%-4.6%, and they most commonly occur in the sacrum.[2,3)] Nearly all sacral TCs are asymptomatic, and those detected incidentally do not require any treatment. However, they rarely cause symptoms, such as leg pain, buttock pain, leg weakness, bladder and bowel dysfunction, dyspareunia or sexual dysfunction, and positional headache.[4)] Surgical intervention is considered when these symptoms persist despite medical treatments. Many surgical procedures for symptomatic TCs were reported. Among them, microsurgical wrapping (MSW) for TCs is a relatively novel simple procedure with few complications.[5)] Herein, we report a case of multiple sacral TCs treated with MSW and present the mechanism of symptoms generated by TC and the limitations of surgical procedures, including MSW.

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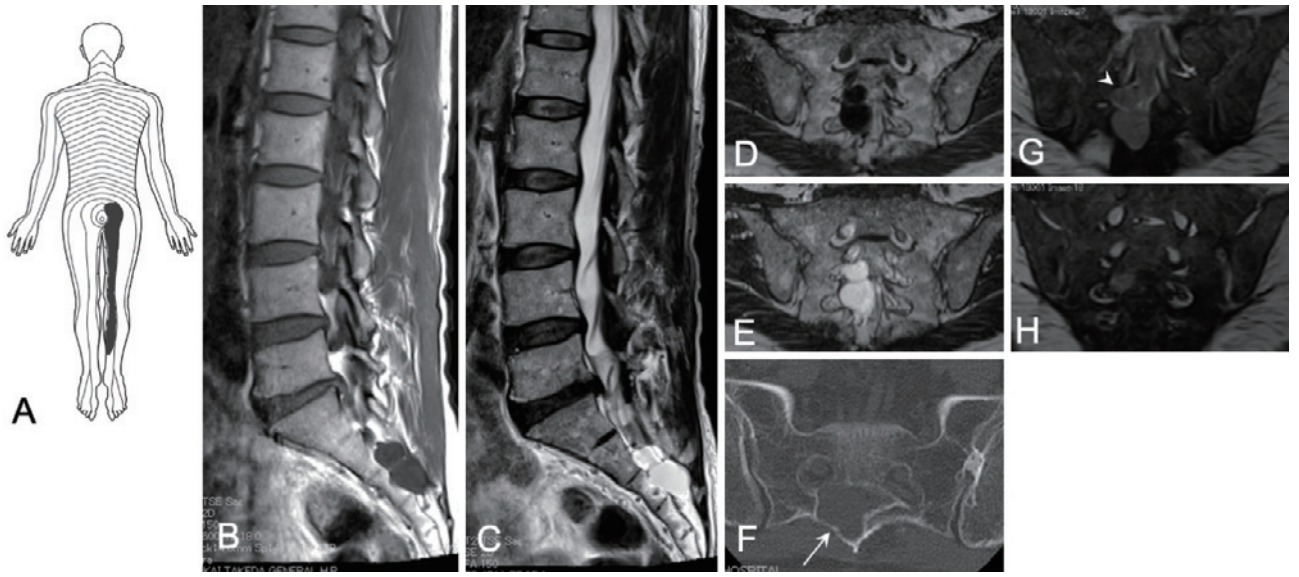

Fig. 1 Preoperative images and neurological findings.
Painful area (shaded black in panel A). Magnetic resonance imaging (MRI) demonstrated multiple cystic lesions at the S2-S3 levels. These cysts contain fluid with a similar intensity to cerebrospinal fluid. (B) T1-weighted sagittal image. (C) T2-weighted sagittal image. (D) T1-weighted coronal image. (E) T2-weighted coronal image. Axial computed images demonstrated bony erosion on the right side of the sacral lamina (arrow) (F). Coronal MRI demonstrated that each cyst appeared to connect the S3 and S4 roots, respectively, and the S2 root (arrowhead) was compressed by the cysts (G, H).

Case Report

A 58-year-old man was admitted to our institution with complaints of right leg and buttock pain for several months. When aggravated, he became irritable and could not endure sitting and standing because of pain, which was resolved with a prone position. He took nonsteroidal analgesic drugs (1300 mg/day of Acetaminophen) and gabapentin (1200 mg /day) for 2 months; however, the symptoms gradually worsened. The painful area is described in Fig. 1A. He did not complain of bowel/bladder dysfunction. Magnetic resonance imaging (MRI) (Fig. 1B-E) demonstrated multiple cystic lesions at the S3-S4 levels. These cysts contain fluid with the same intensity as the CSF. Axial computed tomography (Fig. 1F) demonstrated bony erosion on the right side of the sacral lamina, indicating slow-growing lesions. On coronal MRI, each cyst appeared to connect the S3 and S4 roots, and the cyst compressed the S2 root (Fig. 1G, H). According to the imaging findings, the patient was diagnosed with multiple sacral TCs (sacral perineural cysts), and MSW was planned.

Surgical procedures

The patient was placed in a prone position under general anesthesia, and a midline lumbosacral linear skin incision was made. The right sacral laminas were exposed microscopically, and a bony defect was confirmed in the S4 lamina (Fig. 2A). The spinal dural sac and multiple cyst walls were exposed widely after a right hemi-laminectomy of S1-S4 using a high-speed drill and ultrasound aspirator (Fig. 2B). After puncturing the cysts with a 26-gauge needle for the internal decompression of the cysts (Fig. 2C), the cyst wall was meticulously dissected from the other cyst wall and epidural venous plexus (Fig. 2D). Following the surgical procedure reported by Sugawara et al.,[5] an expanded polytetrafluoroethylene (ePTFE) membrane (GORE-TEX, W. L. Gore & Associates, Inc., DE, USA) was passed under the shrinking cyst (Fig. 2E), and the punctured hole was sealed with polyglycol acid membrane and fibrin glue (Fig. 2F). The ePTFE membrane was closed with multiple titanium clips (Fig. 2G). The same procedures were performed on the other cyst (Fig. 2H). During wrapping, both cysts did not re-expand with early CSF filling after the single puncture and aspiration, indicating that CSF flow volume into both cysts was originally minimal in the present case. Finally, we confirmed that the CSF leakage did not occur with the Valsalva maneuver, and the bony defect of the sacral lamina was reconstructed with a fibrin glue-soaked absorbable gelatin sponge.

Postoperative course

The postoperative course was uneventful, the symptoms suggested CSF leakage, and posture headache did not occur. MRI performed 1 week after the operation did not reveal CSF leakage. The right leg pain was relieved early after the operation; however, the buttock pain partially improved but persisted at discharge 10 days after the operation. The symptom persisted 13 months after the operation (Fig. 3A). At this time, the MRI demonstrated ePTFE artificial sheathes, and the CSF-containing cysts did not re-

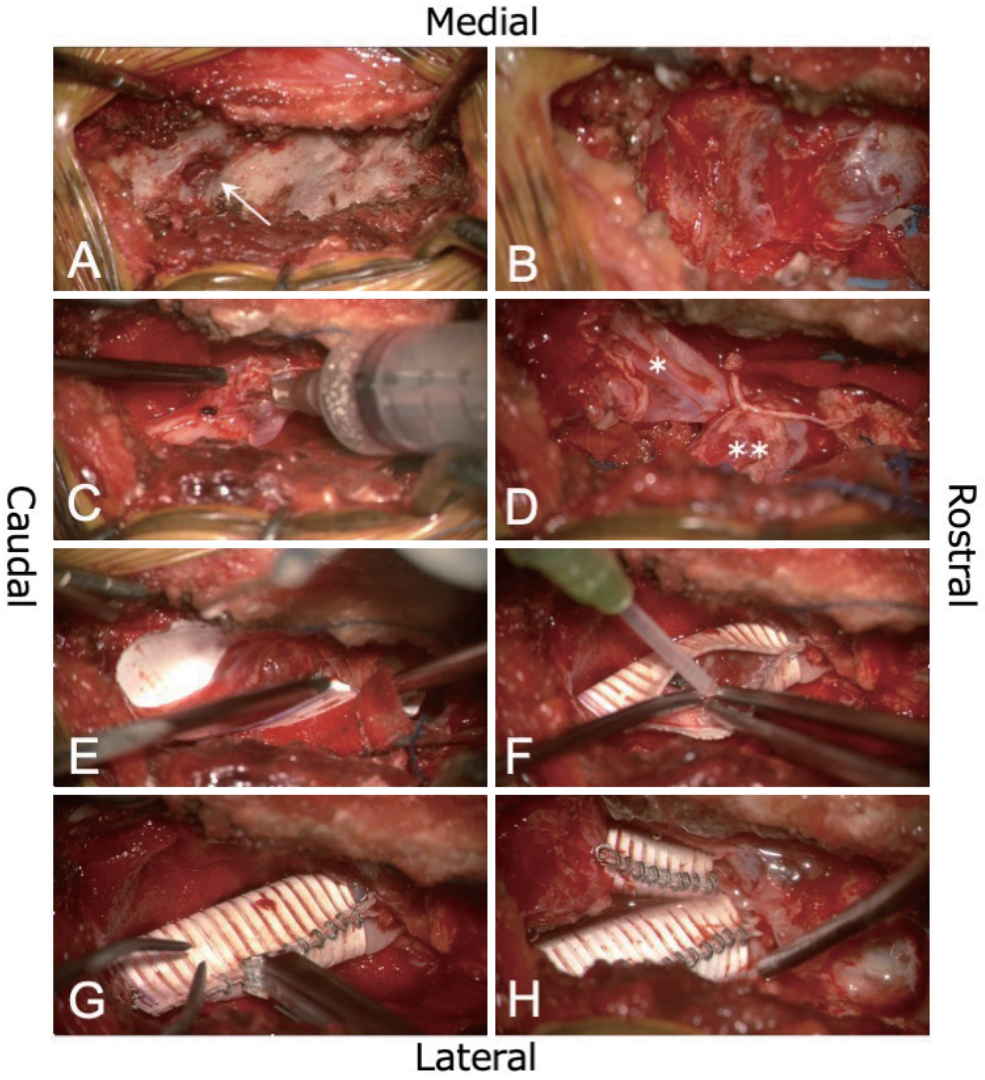

Fig. 2 Intraoperative photographs of microsurgical wrapping in the present case.
The right sacral laminas were exposed, and bony defects (arrow) were found in the S4 lamina (A). The spinal dural sac and multiple cyst walls were exposed after the hemi-laminectomy of S1-S4 (B). After puncturing the cysts with a 26-gauge needle for internal decompression of the cysts (C), a cyst wall (single asterisk) was dissected from another cyst wall (double asterisks) and epidural venous plexus (D). An expanded polytetrafluoroethylene (ePTFE) membrane was passed under the shrinkage cyst (E), and the puncture hole was sealed with a polyglycol acid membrane and fibrin glue (F). The ePTFE membrane was closed with multiple titanium clips (G). The same procedure as E-G was performed in another cyst (H).

cur (Fig. 3B-D).

Discussion

In the present case, the MSW performed for TCs with buttock or leg pain was originally reported by Sugawara et al.[5] They hypothesized that the sacral symptoms were caused by adjacent nerve root stimulation due to pulsation or hydrostatic pressure exerted by TCs. They developed MSW for symptom relief and decompression of the adjacent nerve roots. They reported seven MSW-treated cases and achieved pain relief in five patients. Aside from the high pain remission rate, this procedure is notably simple with low complication rate, particularly for postoperative CSF leakage, because the cyst can be decompressed only by puncture aspiration with a fine needle and sealed with an artificial membrane (e.g., polyglycol acid membrane) and a small amount of fibrin glue.

Several surgical procedures for symptomatic TCs were reported. These procedures divided manipulation for the ball-valve mechanism, one of the mechanisms for cyst formation and growth, decreasing the mass effect of the cyst. The former includes packing with fat, muscle pedicle flap, or fibrin glue[6,7] and clipping the neck of the TCs;[8] the latter includes fenestration and imbrication of the cyst wall.[9] Recently, endoscopic fenestration of the cyst was also re-

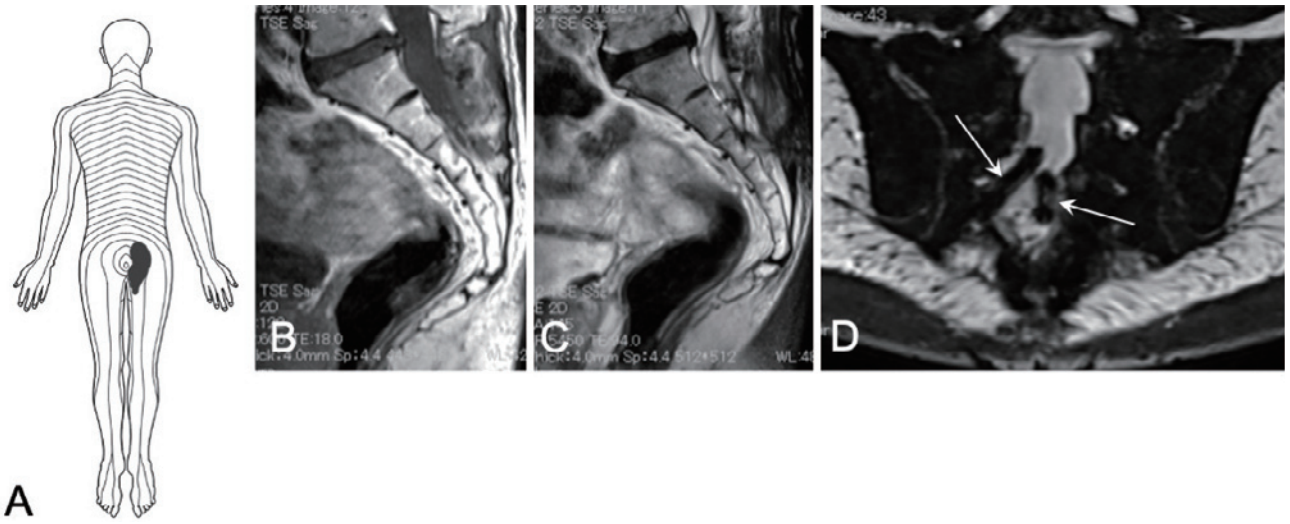

Fig. 3 Magnetic resonance imaging13 months after the operation and its neurological findings.
(A) The pain remained even after 13 months from the operation (shaded black). Sagittal images revealed a significant reduction in the cyst size. (B) T1-weighted sagittal image. (C) T2-weighted sagittal image. (D) Coronal T2-weighted image demonstrating artificial seethe of the expanded polytetrafluoroethylene membranes (arrows) wrapping the cysts.

ported.[10] Because all these procedures require a wide opening of the cyst wall, postoperative CSF leakage is inevitable and is expected to be high for MSW.

As Sugawara et al. hypothesized, one of the causes of symptomatic TCs is adjacent nerve root compression by enlarged cysts.[5] The other mechanism of the symptoms is stretching the nerve root running on the inner wall of the TCs, i.e., the injury of the TC-originated nerve root itself. In the present case, no complications, including CSF leakage, occurred postoperatively, and the preoperative leg pain was completely relieved and did not aggravate with sitting or standing. In symptomatic TC cases, as the CSF volume is shifted caudally when changing from lying to sitting, the pain increases when sitting or standing and is relieved when lying down, especially in the prone position.[11] Symptom aggravation via postural change and Valsalva maneuver are the characteristics of TCs-related symptoms, which contribute to differential diagnosis[11] or decide surgical indication.[2,12] In the present case, postural aggravation of the symptoms also disappeared, dramatically improving his quality of life after the MSW. However, buttock pain only partially improved and persisted even after 13 months of follow-up.

The schematic illustration of two speculations in the mechanism of symptoms onset and the clinical course after MSW is described in Fig. 4: i) the symptoms of severely injured adjacent nerve root (probably S2) compression by TCs were not completely resolved after MSW; meanwhile, those with mildly injured or intact TC-originated nerve roots (probably S3 and/or S4 root) were completely resolved or unchanged after MSW (the same speculation with Sugawara et al.[5]) (Fig. 4A); ii) the symptoms of the adjacent nerve roots (probably S2 and S3 roots) compressed by TCs were resolved after MSW but that of the severely injured TC-originated nerve root (probably S3 and/or S4 roots) was partially resolved. The symptoms of nerve root compression easily improved because the adjacent nerve root was compressed by the cyst indirectly through the dural root sleeves (Fig. 4B).

We meticulously dissected the cyst walls intraoperatively; however, surgical manipulations might have stretched and affected the nerve roots. We decompressed the cysts with a single puncture and dissected between each cyst wall and the adjacent nerve root sleeve. Thus, staged decompression of the cysts and dissection may have been the best technique to avoid stretching the inner nerve roots.

However, in any case, the degree of postoperative improvement of symptoms caused by the affected nerve root depends on the severity of injury by compressing with the cyst (the speculation in above i)) or stretching with the cyst (the speculation in above ii)). The larger the cyst, the more compressed and severely stretched the nerve root. Thus, as in the present case, we hypothesized that the symptoms caused by the large cyst, which induced severe nerve root injury, might be challenging to improve postoperatively anyway. This may be one of the limitations of MSW for TCs. However, this may also be a limitation of surgical procedures for TCs because all the reported surgical procedures for TCs cannot directly reconstruct the injured nerve root. This may be important and should be indicated in the preoperative informed consent. Thus, we believe that MSW has some advantages over other surgical procedures because of its simplicity and low complication rates, such as for CSF leakage.

Among several procedures, MSW is an effective and safe

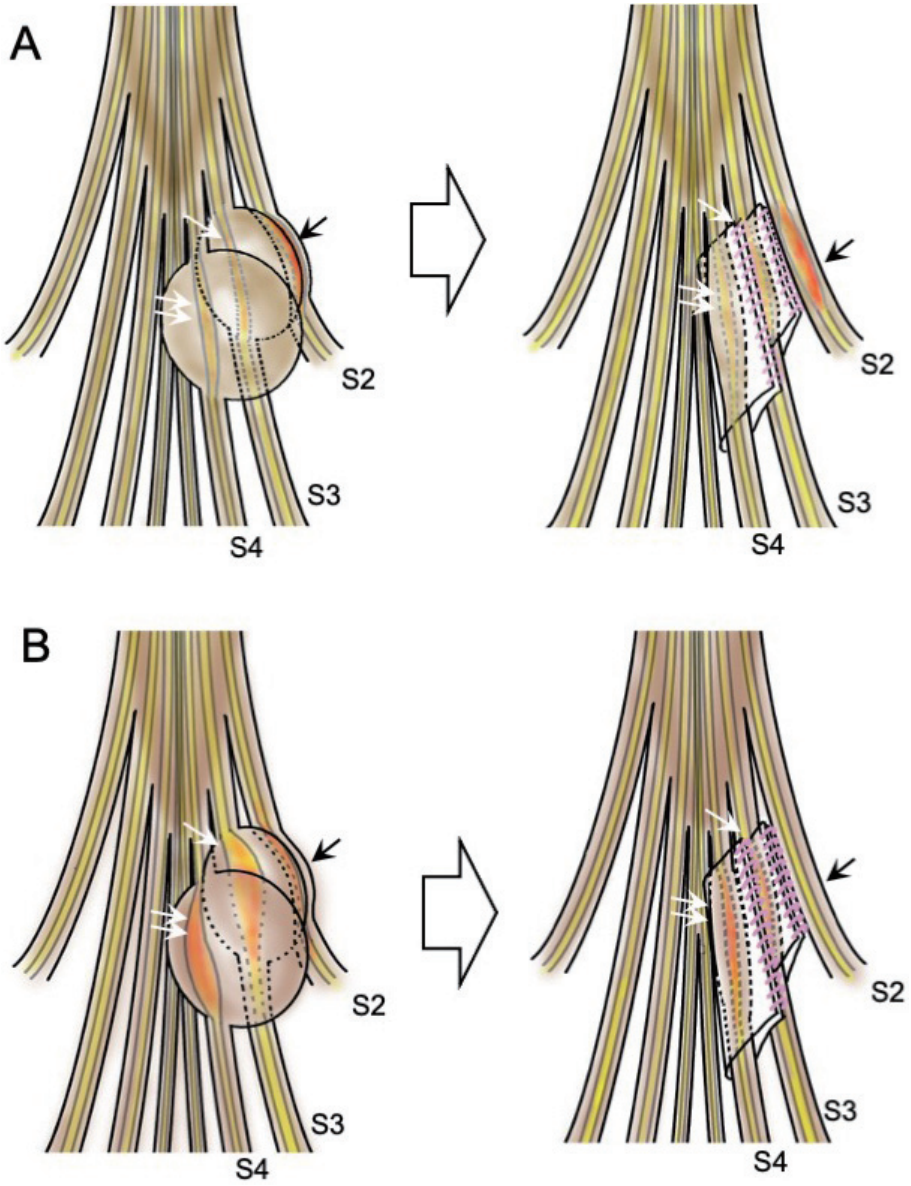

Fig. 4 Schematic illustration of two speculations in the mechanism of symptoms onset (left column) and the clinical course after the microsurgical wrapping (MSW) (right column).
A: Symptoms of severely injured adjacent nerve root (probably S2 [black arrow] in the present case) compression by Tarlov cysts (TCs) were not completely resolved after MSW; meanwhile, those with mildly injured or intact TC-originated nerve roots (probably S3 [white arrow] and/or S4 [white double arrows] root in the present case) were completely resolved or unchanged.
B: The symptoms of the adjacent nerve root (probably S2 [black arrow] and S3 roots [white arrow] in the present case) compression by TCs were resolved after MSW; however, those with severely injured TC-originated nerve roots (probably S3 [white arrow] and/or S4 [white double arrows] root in the present case) were not completely resolved.

surgical procedure for symptomatic TCs. However, we should recognize the general limitations of the surgical procedures for TCs. When considering surgery for symptomatic TCs, surgeons should inform patients preoperatively of some residual symptoms postoperatively if they complain of symptoms caused by the large TCs.

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Informed Consent

Informed consent was obtained from the patient involved in the study.

Conflicts of Interest Disclosure

All authors declare no conflicts of interest associated with this manuscript. The manuscript has not been published previously and is not under consideration for publication elsewhere.

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