

# Multiple Sacral Perineurial Cysts Presented Symptoms Triggered by Nonaneurysmal Perimesencephalic Subarachnoid Hemorrhage

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The origin and pathogenesis of perineurial cysts remain unclear. Here, we report a rare case of multiple sacral perineurial cysts presented symptoms triggered by nonaneurysmal perimesencephalic subarachnoid hemorrhage (SAH). A 48-year-old male presented with a severe headache of sudden onset. Brain computed tomography revealed a SAH in the basal cistern; however, no abnormal vascular lesion was detected. Four days after the onset of the SAH, he suffered from left buttock pain, followed by urinary dysfunction and paresthesia in the left S2 and S3 dermatomes. Magnetic resonance (MR) imaging demonstrated multiple cystic lesions at the left S1 and bilateral S2 sacral roots. The two cysts at the S2 level exhibited high signal intensity on both T<sub>1</sub>- and T<sub>2</sub>-weighted images, suggesting the content of the cysts included a hematoma. He was treated with intravenous injections of steroids, and the symptoms recovered completely within 2 months. Follow-up MR imaging revealed remarkable shrinkage of the cysts. Sacral perineurial cysts should be listed in the differential diagnosis if the patient suffers from low back pain or sacral radiculopathy after the onset of SAH.

**Keywords:** nonaneurysmal perimesencephalic subarachnoid hemorrhage, perineurial cyst, steroid, Tarlov cyst

## Introduction

Sacral perineurial cysts, which were first described by Tarlov in 1938<sup>1</sup>) as an incidental finding in the autopsies of 30 adults, are cystic lesions in the space between the peri and endoneurium of the posterior nerve root sheath.<sup>2,3</sup>) The prevalence of perineurial cysts has been estimated to be 1.5–4.6% based on serial magnetic resonance (MR) imaging studies.<sup>4,5</sup>) In clinical settings, the cysts are most often detected as an incidental finding of lumbosacral MR imaging and are asymptomatic throughout the patient's life.<sup>4</sup>) However, some cysts cause radicular pain, motor dysfunction, dysesthesia, and bowel, bladder, and sexual dysfunction.<sup>6–8</sup>) The origin and pathogenesis of

perineurial cysts remain unclear, and the optimal treatment for symptomatic cysts has not been established. We report a case of multiple sacral perineurial cysts that presented symptoms triggered by a nonaneurysmal perimesencephalic subarachnoid hemorrhage (SAH) (NAPH), which were conservatively treated with steroids and showed substantial shrinkage.

## Case Report

A 48-year-old male presented with a severe headache of sudden onset. Brain computed tomography (CT) demonstrated a SAH mainly located around the mesencephalon without a hematoma in the interhemispheric and lateral Sylvian fissures (Fig. 1A). Three-dimensional CT, MR, and conventional angiography were performed to determine the cause of the SAH; however, no abnormal vascular lesion was detected. Conservative treatment and careful observation were conducted, and the patient was gradually relieved of the headache. Four days after the onset of the SAH, he suffered from pain in the left buttock. The symptoms were aggravated by postural change, sitting and walking. Eight days after the onset of the SAH, urinary and bowel dysfunction occurred. He complained of paresthesia in the left S2 and S3 dermatomes, urinary retention and severe constipation. Lumbosacral MR imaging detected multiple cystic lesions at the sacral roots (Fig. 1B). The cyst at the S1 level had low signal intensity on T<sub>1</sub>-weighted images and high signal intensity on T<sub>2</sub>-weighted images, which were similar to those of cerebrospinal fluid (CSF). In contrast, the two cysts at the S2 level had high signal intensity on both T<sub>1</sub>- and T<sub>2</sub>-weighted images (Figs. 2A–2D). Additionally, the CT value of those cysts at S2 level was higher than that of normal CSF (Fig. 2E), suggesting the content of the cyst included a hematoma. No other abnormal radiological findings that could explain the symptoms were found.

He was treated with a 10-day course of intravenous injection of steroids (betamethasone, tapering from 4 to 1 mg). Six days after the start of the medical treatment, his symptoms began to improve. He recovered completely from the symptoms in 2 months. Follow-up MR imaging at 2 months revealed remarkable shrinkage of the cysts. The two cysts at the S2 level, which had high signal intensity on both T<sub>1</sub>- and T<sub>2</sub>-weighted images with the initial MR imaging, changed to low signal intensity on both T<sub>1</sub>- and T<sub>2</sub>-weighted images (Figs. 3A–3D).

## Discussion

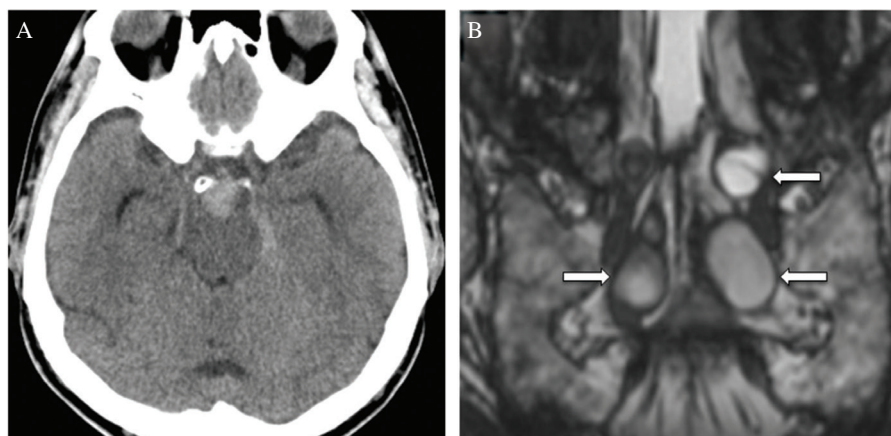
Since Tarlov<sup>1</sup>) reported the first case series of perineurial cysts in 1938, several authors have speculated the origin and

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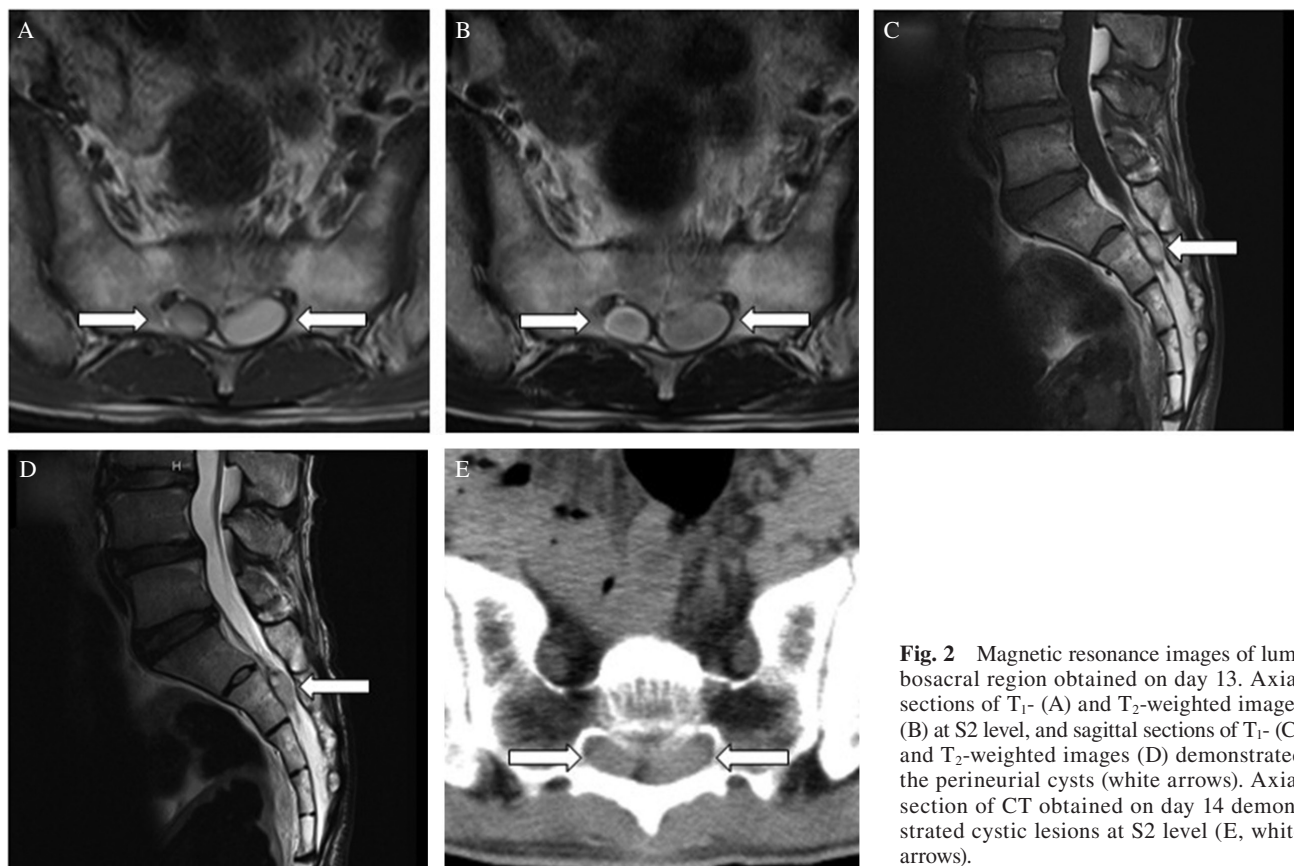
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**Fig. 1** Axial section of computed tomography (CT) obtained on day 0 demonstrated the subarachnoid hemorrhage around the midbrain (A). Coronal section of heavily T<sub>2</sub>-weighted image revealed multiple sacral perineurial cysts (B, white arrows).

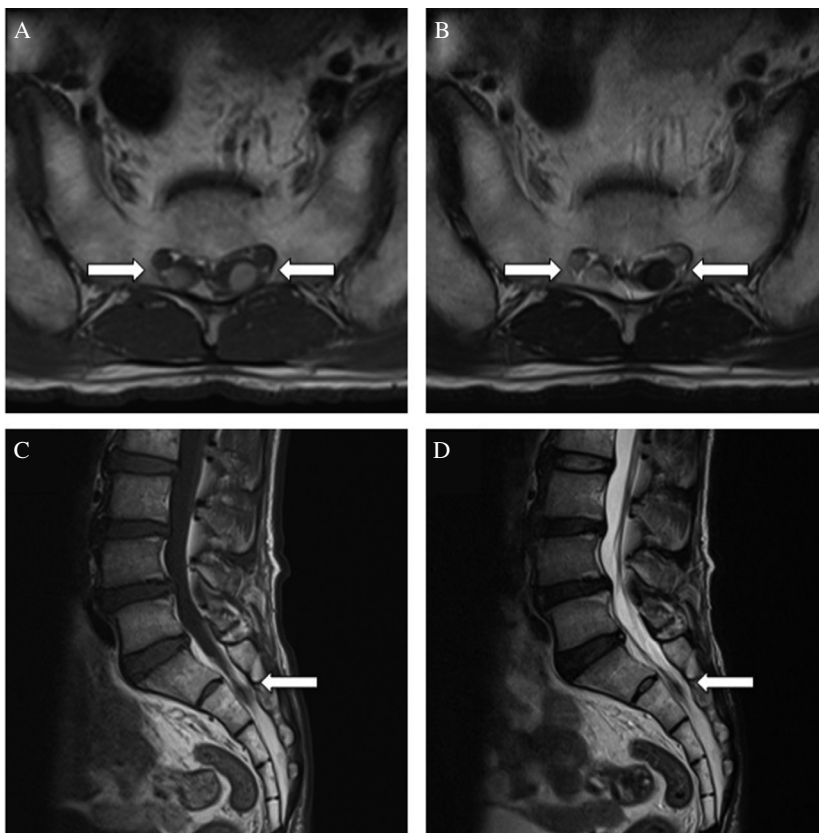


**Fig. 2** Magnetic resonance images of lumbosacral region obtained on day 13. Axial sections of T<sub>1</sub>- (A) and T<sub>2</sub>-weighted images (B) at S2 level, and sagittal sections of T<sub>1</sub>- (C) and T<sub>2</sub>-weighted images (D) demonstrated the perineurial cysts (white arrows). Axial section of CT obtained on day 14 demonstrated cystic lesions at S2 level (E, white arrows).

pathogenesis of the cyst. Hypotheses include inflammation within the nerve root sheath followed by inoculation of CSF, arachnoidal proliferation around the nerve roots, posttraumatic hemorrhage into the subarachnoid space, which causes breakage of venous drainage in the peri- and endoneurium of the nerve roots, and congenital origin.<sup>1,6,9)</sup> Sacral perineurial cysts arise in the sacrococcygeal nerve roots, most commonly at the second and third sacral nerve root levels.<sup>4)</sup> Although the specific mechanisms that make the cysts symptomatic have not been elucidated, the most important factor appears to be stenosis of the ostium of the nerve root sheath.<sup>4)</sup> CSF can flow into the cysts through the ostium by

the hydrostatic pressure and arterial pulsatile forces, but cannot exit from the cysts because of a ball-valve effect.<sup>4,7)</sup>

To the best of our knowledge, there is only one previous report of symptomatic perineurial cysts related to the aneurysmal SAH,<sup>10)</sup> and our case is the first report of those triggered by NAPH. In the previous report, the patient complained of worsening pain in the lumbosacral, buttock, and the posterior aspect of the right thigh, following neck clipping of ruptured aneurysm.<sup>10)</sup> The pain was intermittent at onset and exacerbated by, sitting, walking, and coughing. The patient relieved from pain after CT-guided aspiration of the cyst, and 1 cc of old blood was aspirated from the cysts, which proved



**Fig. 3** Axial sections of T<sub>1</sub>- (A) and T<sub>2</sub>-weighted images (B), and sagittal sections of T<sub>1</sub>- (C) and T<sub>2</sub>-weighted images (D) obtained 2 months after the onset of the subarachnoid hemorrhage revealed shrinkage of the cysts (white arrows).

the accumulation of the SAH into the cysts.<sup>10)</sup> In our case, the perineurial cysts presented symptoms 4 days after the onset of the SAH. Assuming that it takes a few days for the accumulation of the SAH into the sacral cysts to become symptomatic, this time lag appears to be reasonable. The elevation of the CSF pressure following a SAH might prompt the hematoma in the subarachnoid space to flow into the pre-existing cysts and contributes to the expansion of the cysts. Symptomatic perineurial cysts should be listed in the differential diagnosis if a patient suffers from low back pain or sacral radiculopathy after the onset of a SAH.

Magnetic resonance imaging has been recommended as the initial imaging modality when perineurial cysts are suspected, because of its high resolution of soft tissue density, multiplanar imaging capabilities, and the absence of artifact induced by the spine.<sup>4,7,11)</sup> The typical radiological findings of perineurial cysts on MR imaging demonstrate CSF-like characteristics, that is, low signal intensity on T<sub>1</sub>-weighted images and high signal intensity on T<sub>2</sub>-weighted images.<sup>4)</sup> The two cysts at the S2 level in our patient had high signal intensity on both T<sub>1</sub>- and T<sub>2</sub>-weighted images, which indicated the content of the cysts included the SAH. The shrinkage of the cysts and degeneration of the content of the cysts were concomitant with relief of the symptoms. Mechanical compression and irritation of the sacral nerve roots by the expanded cysts containing the SAH appeared to be the cause of symptoms.

The management of symptomatic perineurial cysts remains controversial. Various surgical options for symptomatic

perineurial cysts, including lumbar-peritoneal shunt, cyst-subarachnoid shunt, decompressive laminectomy, percutaneous cyst aspiration, CT-guided injection of fibrin glue, and a direct microsurgical approach have been described.<sup>12,13)</sup> Although the efficacy of conservative treatments has not been studied sufficiently, Mitra et al.<sup>14)</sup> described two cases of symptomatic perineurial cysts treated successfully with oral and epidural steroid therapy. In our case, the intravenous administration of steroids was effective for improvement of the symptoms. The mechanism behind the effectiveness of steroids for symptomatic cysts remains unclear and needs further validation. Nevertheless, our case suggests intravenous administration of steroids could be an initial treatment option for symptomatic perineurial cysts after a SAH.

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#### Conflicts of Interest Disclosure

All authors completed a self-declaration of the conflicts of interest (COI) to the Japan Neurosurgical Society and declare no potential COI regarding this manuscript.

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