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 T1- and T2-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 19381 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.2,3 The prevalence of perineurial cysts has been estimated to be 1.5–4.6% based on serial magnetic resonance (MR) imaging studies.4,5 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.4 However, some cysts cause radicular pain, motor dysfunction, dysesthesia, and bowel, bladder, and sexual dysfunction.6–8 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 T1-weighted images and high signal intensity on T2-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 T1- and T2-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 T1- and T2-weighted images with the initial MR imaging, changed to low signal intensity on both T1- and T2-weighted images (Figs. 3A–3D).

Discussion

Since Tarlov1 reported the first case series of perineurial cysts in 1938, several authors have speculated the origin and
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.\textsuperscript{1,6,9} Sacral perineurial cysts arise in the sacrococcygeal nerve roots, most commonly at the second and third sacral nerve root levels.\textsuperscript{4} 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.\textsuperscript{4} 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.\textsuperscript{4,7}

To the best of our knowledge, there is only one previous report of symptomatic perineurial cysts related to the aneurysmal SAH,\textsuperscript{10} 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.\textsuperscript{10} 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
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Perineural cysts, including lumbar-peritoneal shunt, cyst-subarachnoidal shunt, decompressive laminectomy, percutaneous cyst aspiration, CT-guided injection of fibrin glue, and a direct microsurgical approach have been described.\(^{12,13}\) Although the efficacy of conservative treatments has not been studied sufficiently, Mitra et al.\(^{14}\) 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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