Perineural cyst with intracystic hemorrhage following aneurysmal subarachnoid hemorrhage

A case report

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Abstract

Rationale: Most perineural cysts are asymptomatic and discovered incidentally at the time of imaging. Although enlargement of the perineural cyst (PC) through a ball-valve mechanism and resultant compression of the adjacent neural or bony structures are known to be a source of pain in PCs, the reason why asymptomatic PCs become symptomatic is unclear. The authors report a case of PC, which was presumed to become symptomatic after subarachnoidal hemorrhage without enlargement of the pre-existing PC.

Patient concerns: A 47-year-old woman complained of lumbosacral pain after neck clipping for a ruptured cerebral aneurysm.

Diagnoses: Magnetic resonance imaging (MRI) revealed a PC with intracystic hemorrhage at the S2 level. In comparison with the size of the PC on computed tomography performed 3 years ago, there was no change in the size. Electrodiagnostic studies performed 6 weeks after the onset of the pain showed subacute right S2 radiculopathy.

Interventions: With conservative treatment, her pain gradually diminished.

Outcomes: When the lumbosacral pain improved, follow-up MRI showed that a fluid-fluid level within the PC disappeared.

Lessons: Hemorrhage from the subarachnoid space, such as spontaneous aneurysmal SAH, into the pre-existing PC can cause an asymptomatic PC to become symptomatic without getting enlarged. Stretching of the nerve root due to hemorrhage or irritation of the nerve root due to an inflammatory reaction to blood products can make asymptomatic PCs symptomatic without enlargement of PCs.

Abbreviations: CT = computed tomography, MRI = magnetic resonance imaging, PCs = perineural cysts, SAH = subarachnoid hemorrhage, SI = signal intensity.

Keywords: intracystic hemorrhage, perineural cyst, subarachnoid hemorrhage, Tarlov cyst

1. Introduction

Perineural cysts (PCs), also known as Tarlov cysts are typically located within the nerve root sheath at the junction of the dorsal root ganglion and the posterior nerve root.[1,11] Most PCs occur in the sacral region and are discovered during diagnostic imaging of the lumbar spine (CT or MRI).[2,3] The prevalence of PCs has been reported to be approximately 1.5% to 4.6% in large series using MRI.[2,3] Although PCs presenting as coccydynia, pelvic pain, sacral radiculopathies, and sacral insufficiency fractures have been reported, most PCs remain asymptomatic.[3,4] The reported incidence of symptomatic PCs is 0% to 22% among the patients with PCs.[3,4] Although enlargement of the PC through a ball-valve mechanism and resultant compression of the adjacent neural or bony structures are known to be one of the sources of pain, the reason why some of the asymptomatic PCs become symptomatic is still unclear.[4,6]

We report a case of PC which we think became symptomatic as a result of aneurysmal subarachnoidal hemorrhage (SAH) without getting enlarged and discuss the possible mechanisms by which asymptomatic PCs may become symptomatic.

2. Case report

A 47-year-old woman presented with a complaint of severe headache of sudden onset. On neurological examination, she had signs of meningeal irritation. Brain computed tomography (CT) revealed Fisher grade 3 SAH. Three-dimensional brain CT angiography showed an anterior communicating artery aneurysm. Emergent craniotomy and neck clipping of the aneurysm were performed.

In the early postoperative period, she complained of pain mainly on the right side of the sacrum, buttock, and upper part of the posterior aspect of the right thigh. She complained of worsening pain when sitting down, walking, or coughing. She had a previous history of lumbago 3 years ago and it was relieved with conservative treatment. CT of the lumbosacral spine performed at that time revealed a bulging disc at the L4-L5
level and a PC at the S2 level (Fig. 1A). Thereafter, she did not have lumbago and there was no history of recent trauma. The nature and location of her pain were slightly different from the nature and location of pain she had experienced 3 years ago. During this episode, sacral and buttock pain was more severe than lumbago. She complained of severe tingling sensation and right-side predominant pain. The pain did not subside fully when lying down. She suffered from the pain despite using analgesics for 4 weeks.

Magnetic resonance imaging (MRI) of the lumbosacral spine performed at 1 month postoperatively showed an 18 × 13 × 25 mm-sized perineural cyst with high signal intensity at the S2 level, and a so-called “dark” and bulging disc at multiple levels of the lumbar spine. (C) Nonenhance T1-weighted sagittal magnetic resonance imaging shows mixed high and low signal intensity with a fluid-fluid level within the cyst, suggestive of intracystic hemorrhage. (D) Nonenhance T2-weighted axial magnetic resonance imaging shows that perineural cyst is located to the right side at S2 level. (E) Follow-up enhanced lumbosacral spine magnetic resonance imaging performed 3 months later shows no change in the size of the cyst. However, mixed signal intensity and a fluid-fluid level within the cyst disappear on the T1-weighted sagittal image.

The reason why an asymptomatic PC becomes symptomatic is still unknown. Regardless of the underlying cause, the mechanism of PC formation has been described as a ball-valve mechanism.\textsuperscript{4-6} With stenosis of the ostium of the nerve root sheath, the cyst neck serves as a valve.\textsuperscript{4-6} CSF is able to flow into the cyst with arterial pulsation and a patient’s postural changes, but outflow is restricted.\textsuperscript{4-6} When the cyst fills, pain occurs; when the cyst deflates, pain is relieved.\textsuperscript{4-6} The cysts can enlarge with time by the hydrostatic and pulsatile forces of CSF and they may become symptomatic by local compression of the adjacent
bone or by displacing the nerve root, if they become large.[4–6] However, in the present case, enlargement of the pre-existing PC was not evident. Although the size of the cyst measured on the CT and MRI could not be directly compared, comparison of the 2 methods did not show any obvious enlargement of the cyst. Because the size of the PC did not increase in comparison with the size of the PC on the lumbosacral spine CT performed 3 years before the onset of SAH and on the lumbosacral spine MRI performed when the patient complained of pain and after the resolution of pain.

Another possible mechanism is that hemorrhage or hematoma within the PC can cause stretching or irritation of the nerve roots.[8,9] Lam[8] reported transient radicular pain induced by SAH after spinal anesthesia and suggested that the hematoma displaced and irritated the lumbosacral nerve roots. Desai et al.[9] postulated the inflammatory role of heme and suggested that hemolysis can incite an inflammatory response which can irritate the neural structures and leptomeninges. In the present case, it was speculated that intracisternal hemorrhage occurred as a result of the SAH. Intracranial SAH during aneurysmal rupture can migrate to the spinal subarachnoid space because the anterior spinal and posterior spinal cisterns communicate through the foramen magnum with the posterior fossa cisterns.[10] Then, spinal SAH can migrate into the pre-existing PC with arterial pulsation and a patient’s postural changes.

In the present case, it was difficult to clarify whether the pain originated from intracisternal hemorrhage or from degenerative changes in the lumbosacral spine solely based on the nature, location, and aggravating or relieving factors of pain. Although the nature and location of the pain was slightly different from the nature and location of the pain which the patient had experienced 3 years ago, patients with degenerative changes could also have pain on the lumbosacral spine, buttock, and posterior thighs and their symptoms may persist while lying down. At such a time, an electrodiagnostic study can help to diagnose and to determine the duration of radiculopathy.[7] In the present case, the electrodiagnostic study suggested subacuate right S2 radiculopathy. The amplitude of the fibrillation potential can be used to predict the lesion timing.[17] The mean fibrillation amplitude during the 1.5–3 months following nerve injury approximated 300 uV.[7] In our case, the fibrillation amplitude was 300uV, which suggested that 6 weeks to 3 months had elapsed after denervation. Based on all these findings observed in the present case, we carefully speculated that the source of pain was hemorrhage within the PC rather than degenerative changes in the lumbosacral spine. Transient stretching of the nerve root by hemorrhage or irritation of the nerve root by an inflammatory reaction to blood products may be the source of the pain in the absence of enlargement of the pre-existing PC.

Hemorrhage from the subarachnoid space, such as spontaneous aneurysmal SAH, into the pre-existing PC can cause an inflammatory reaction to blood products that can cause radiculopathy in the absence of enlargement of the pre-existing PC. An electrodiagnostic study in these diagnostic procedures and can help to diagnose and to determine the duration of radiculopathy. Many methods have been described for treatment of symptomatic lesions, with variable results. From percutaneous CT-guided needle aspiration and fibrin glue injection to surgical treatment involving complete cyst removal and excision of the affected posterior root and ganglion is tried.[11] However, there is little consensus in the literature regarding the best treatment of symptomatic perineural cysts.[12] Conservative treatment should be given priority in symptomatic perineural cyst with intracisternal hemorrhage following SAH, because intracisternal hemorrhage is absorbed and the symptoms improve.

Author contributions

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References

[1] Voyadzis JM, Bhargava P, Henderson FC. Tarlov cysts: a study of 10 cases with review of the literature. J Neurosurg 2001;95(1 Suppl):25–32.
[2] Guo D, Shu K, Chen R, et al. Microsurgical treatment of symptomatic sacral perineural cysts. Neurosurgery 2007;60:1039–65.
[3] Paulsen RD, Call GA. Murtagh FR: Prevalence and percutaneous drainage of cysts of the sacral nerve root sheath (Tarlov cysts). AJNR 1994;15:293–7.
[4] Langdown AJ, Grundy JR, Birch NC. The clinical relevance of Tarlov cysts. J Spinal Disord Tech 2005;18:29–33.
[5] Lucantoni C, Than KD, Wang AC, et al. Tarlov cysts: a controversial lesion of the sacral spine. Neurosurg Focus 2011;31:E14.
[6] Marino D, Carluccio MA, Di Donato L, et al. Tarlov cysts: clinical evaluation of an Italian cohort of patients. Neurol Sci 2013;34:1679–82.
[7] Dumitr uD, Zwarts M, Dumir uD, Amato A. Needle electromyography. Electrodiagnostic Medicine 2nd edn. Philadelphia: Hanley & Belins; 2002.
[8] Lam DH. Subarachnoid haematoma after spinal anaesthesia mimicking transient radicular irritation: a case report and review. Anaesthesia 2008;63:423–7.
[9] Desai MJ, Dave AP, Martin MB. Delayed radicular pain following two large volume epidural blood patches for post-lumbar puncture headache: a case report. Pain Physician 2010;13:257–62.
[10] Di Rienzo A, Iacoangeli M, Alvaro L, et al. Subarachnoid haematoma of the craniovertebral junction and upper cervical spine after traumatic cerebral contusion: case report. Neurol Med Chir (Tokyo) 2013;53:620–4.
[11] Nkwerem SP, Ito K, Ichinose S, et al. Resection and imbrication of symptomatic sacral Tarlov cysts: A case report and review of the literature. Surg Neurol Int 2018;9:180.
[12] Elsawaf A, Awd TE, Fesal SS. Surgical excision of symptomatic sacral perineural Tarlov cyst: case series and review of the literature. Eur Spine J 2016;25:3385–92.