Clinical Case Report

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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 subarachnoid 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] Most PC's occur in the sacral region and are discovered during diagnostic imaging of the lumbar spine (CT or MRI).[1] 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 subarachnoid 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

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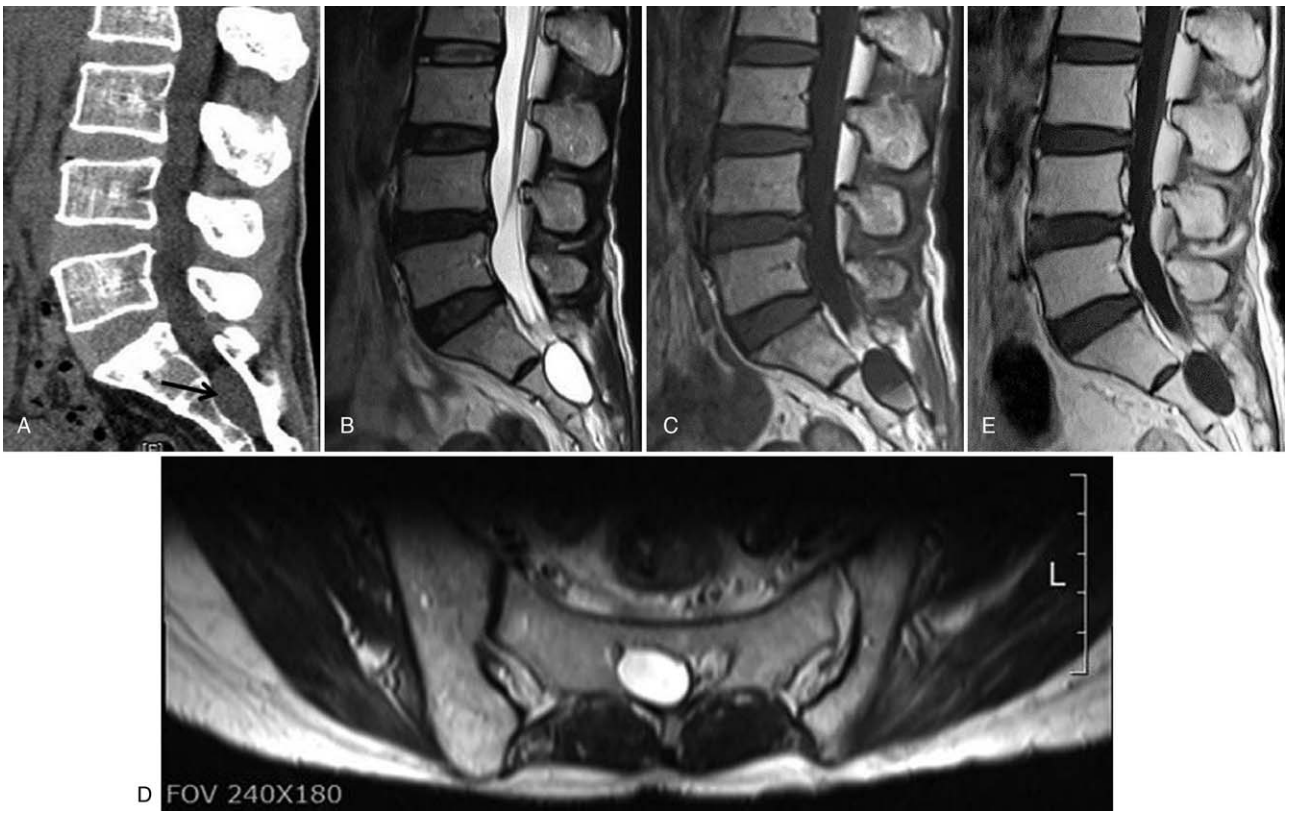

Figure 1. (A) Computed tomography of the lumbosacral spine performed 3 years before the onset of subarachnoid hemorrhage shows a perineural cyst at the S2 level (arrow). (B) T2-weighted sagittal magnetic resonance imaging of the lumbosacral spine shows an 18×13×25 mm-sized perineural cyst with high signal 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.

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 PC with high signal intensity (SI) at the S2 level affecting the right S2 root and a so-called dark' and bulging disc at multiple levels of the lumbosacral spine on the T2-weighted sagittal image (Fig. 1B). On the nonenhance T1-weighted sagittal image, mixed high and low SI with a fluid–fluid level within the cyst, suggestive of intracystic hemorrhage, was revealed (Fig. 1C). There is a perineural cyst that is located to the right aspect at S2 level (Fig. 1D). The size of the cyst measured on MRI showed no apparent enlargement of the cyst compared with the size on the CT performed 3 years ago.

It seemed that the size of the cyst had not increased as compared with the size of the cyst on the CT performed 3 years ago. An electrodiagnostic study was performed 6 weeks after the onset of the pain, and it showed subacute right S2 radiculopathy. There were abnormal findings such as delayed onset latency of the right bulbocavernosus response, spontaneous denervation activity, and fibrillations in the right side of the anal sphincter muscle, soleus muscle and foot intrinsic muscles (abductor digiti minimi, abductor halluces). The fibrillation potentials had increased amplitude of 300uV, which suggested that 6 weeks to 3 months had elapsed after denervation.[7]

With conservative treatment, her pain gradually diminished. Follow-up MRI performed 3 months later showed no change in the size of the PC. However, mixed SI and a fluid–fluid level within the PC disappeared on the enhanced T1-weighted sagittal image (Fig. 1E). As this is a case report, ethical approval was not required. Informed patient consent was obtained for the publication of this report.

3. Discussion

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.[4–6] With stenosis of the ostium of the nerve root sheath, the cyst neck serves as a valve.[4–6] CSF is able to flow into the cyst with arterial pulsation and a patient's postural changes, but outflow is restricted.[4–6] When the cyst fills, pain occurs; when the cyst deflates, pain is relieved.[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 lumbar spine CT performed 3 years before the onset of SAH and on the lumbar 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 intracystic 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 intracystic 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 subacute right S2 radiculopathy. The amplitude of the fibrillation potential can be used to predict the lesion timing.[7] 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 asymptomatic PC to become symptomatic without getting enlarged. Transient stretching of the nerve root by hemorrhage or irritation of the nerve root by an inflammatory reaction to blood products 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 intracystic hemorrhage following SAH, because intracystic hemorrhage is absorbed and the symptoms improve.

Author contributions

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