Surgical Treatment of Sacral Perineural Cyst
—Case Report—

Hiroaki MATSUMOTO,1 Shigeo MATSUMOTO,1 Takanori MIKI,1 Yuki MIYAJI,1 Hiroaki MINAMI,1 Atsushi MASUDA,1 Shogo TOMINAGA,1 Yasuhisa YOSHIDA,1 Ikuya YAMAURA,1 Shigeatsu NATSUME,1 and Kozo YOSHIDA1

1Department of Neurosurgery, Eisyokai Yoshida Hospital, Kobe, Hyogo

Abstract

A 67-year-old man presented with persistent penis and scrotum pain due to S-2 and S-3 radiculopathy caused by a sacral perineural cyst. The cyst was treated with microsurgical partial cyst removal and cyst wall imbrication, together with closure of the point through which cerebrospinal fluid (CSF) flowed from the subarachnoid space into the cyst cavity. His pain resolved without recurrence of the cyst or complications. Symptomatic perineural cysts are quite rare. Surgical closure of the point through which CSF flows from the subarachnoid space into the cyst cavity is the most important intervention for symptomatic perineural cysts. If the source of CSF leakage cannot be detected, placement of a cyst-subarachnoid shunt should be considered in addition to partial cyst removal and cyst wall imbrication.

Key words: perineural cyst, spine surgery, Tarlov cyst, radicular pain, microsurgery

Introduction

Perineural cysts are lesions of the nerve roots most often found in the sacral region, which were first described in autopsy studies of the filum terminale.17) Perineural cysts arise at the junction of the dorsal ganglion and posterior nerve root, and develop between the endoneurium and perineurium, most commonly at the S2 or S3 level.17,18) The prevalence has been reported as approximately 1.5% to 4.6% in the general adult population.6,13) Although most perineural cysts are asymptomatic, approximately 1% cause symptoms related to local compression.13) Only symptomatic perineural cysts should be treated surgically. However, no consensus exists regarding the appropriate surgical procedures.2,3,16,18)

We describe a case of sacral perineural cyst treated successfully with microsurgical partial cyst removal and cyst wall imbrication, together with closure of the communication between the cyst and the arachnoid space with absorbable collagen sponge and fibrin glue.

Case Report

A 67-year-old man had been suffering from progressive penis and scrotum pain for 3 years. Although he had been treated at the urology department for a long time, his pain had not improved. He rated the pain as 7 of 10 on the visual analog scale (VAS). The pain was neither exacerbated by changes of the posture nor relieved by recumbence. Muscle strength and deep tendon reflexes were normal in both lower extremities. Sensations of light touch, pinprick, proprioceptive stimuli, and vibration were intact. Neither urinary nor fecal incontinence was noted. Magnetic resonance (MR) imaging revealed a 25 × 15-mm cystic lesion in the sacrum isointense with the cerebrospinal fluid (CSF) (Fig. 1A, B). The cyst was on the right side at S2 level and compressed the right S-2 and S-3 nerve roots toward the wall of the canal (Fig. 1C–E). Computed tomography (CT) myelography demonstrated delayed filling of the cyst, indicating communication between the cyst and the arachnoid space (Fig. 1F, G).

Our diagnosis was penis and scrotum pain due to S-2 and S-3 nerve root radiculopathy caused by a sacral perineural cyst. Microsurgical treatment was performed. The cyst was exposed via an S2 laminectomy. The cyst wall appeared tough and tense (Fig. 2A). To avoid nerve and venous plexus injury, dissection of the cyst from the surrounding structures was not performed. After fenestration of the cyst wall, clear CSF-like fluid was found, which was evacuated with suction, and the cyst wall was then partially removed. The right S-2 nerve root ran caudally along the inner surface of the cyst wall (Fig. 2B). There was a dural tear in the vicinity of the S-2 nerve root sleeve. Pulsatile CSF flow passed out through the dural tear, but the reverse phenomenon did not occur. To close the communication between the subarachnoid space and the cyst, the dural tear was closed with absorbable collagen sponge and fibrin glue (Fig. 2C). After the repair, no further inflow of CSF was observed. Imbrication of the cyst wall was performed using 6–0 nylon (Fig. 2D).

Histological examination revealed that the cyst walls mostly consisted of dense, fibrocollagenous bundles with
Fig. 1 Preoperative investigations. A, B: Sagittal T1-weighted (A) and T2-weighted (B) magnetic resonance images demonstrating a 25 × 15-mm cystic lesion at the S2 level, and the cyst contents isointense with the cerebrospinal fluid. C: Axial T1-weighted magnetic resonance image demonstrating the cyst compressing the right S-2 nerve (arrow) toward the wall of the canal. D, E: Coronal T1-weighted magnetic resonance images demonstrating the cyst (asterisk) compressing both the right S-2 nerve root (D: arrow) and the right S-3 nerve root (E: arrow). F, G: Computed tomography myelograms at the S2 level immediately (E) and 3 hours (F) after intrathecal injection showing delayed filling of contrast medium.

sparse cellularity, with no nervous tissue identified (Fig. 3). The histological diagnosis was perineural cyst. Postoperatively, he rated his pain at 3 of 10 on the VAS immediately after the operation. He was discharged without complications or pain (0 of 10 on the VAS) on postoperative Day 10. Postoperative MR imaging revealed collapse of the cyst and the decompressed right S-2 and S-3 nerve roots (Fig. 4). He experienced neither pain nor recurrence of the cyst at the 12-month follow-up examination.

Discussion

Asymptomatic perineural cysts or Tarlov cysts are generally found coincidentally when lumbar MR imaging or CT is performed for other reasons, whereas symptomatic perineural cysts are quite rare.6,13 Depending on the location, size, and relationship to the nerve roots, perineural cysts may cause local or radicular pain, sensory disturbance, motor deficits, neurogenic claudication, bowel and bladder dysfunction, or sexual impotence.1-19 Specific radicular pain may be the result of distortion, compression, or stretching of the nerve root by the space-occupying features of cysts. Symptoms can be exacerbated by changes in posture, coughing, Valsalva maneuvers, standing, lifting, and climbing stairs, all of which increase CSF pressure. The symptoms can be relieved by recumbence. These phenomena can be explained by the ball-valve mechanism.3,5,11,12 In our case, although the pain was neither exacerbated by movements which increase the CSF pressure nor relieved by recumbence, we diagnosed S-2 and S-3 nerve root radiculopathy caused by a sacral perineural cyst, because MR imaging had revealed the perineural cyst at the S2 level affecting the right S-2 and S-3 nerve roots without lumbar or sacral lesions.

Consensus has been achieved that asymptomatic perineural cysts should be followed up. However, whether patients with symptomatic perineural cysts should be treated surgically remains under discussion.2,3,16,18 In fact, the majority of symptomatic perineural cysts have been treated surgically, but conservative treatment with steroid or gabapentin has been reported.8,9 Successful surgical treatment of perineural cysts depends on appropriate patient selection.11,16,18 Several predictors of an effective operative outcome have been proposed, including large cysts greater than 15 mm in diameter, delayed filling of the cyst on CT myelography, and radicular pain or bowel/bladder dysfunction.11,16,18 The curable symptoms are coccydynia, dyspareunia, sacral pain, and perianal pain.16 On the other hand, sciatica and vague lumbago are residual symptoms. In our case, preoperative investigations revealed the cyst was 25 × 15 mm in size, and delayed filling of the cyst was observed on CT myelography. Moreover, the cyst caused S-2 and S-3 nerve root radiculopathy. Therefore, a good operative outcome was expected, and surgical treatment was performed.

Several surgical procedures for perineural cysts have been reported,1-7,10-16,18,19 but no consensus exists of the most appropriate. The procedures are of three types: procedures to lower CSF hydrostatic and pulsatile pressures, such as external CSF drainage1 or lumbar peritoneal shunt1; procedures to decompress cysts, such as simple decompressive laminectomy,15 CT-guided percutaneous aspiration,7,13 or cyst-subarachnoid shunt5,10; and direct surgical procedures such as bipolar cautery to shrink cysts,14 total cyst removal,2,12 partial cyst removal with neck ligation,4,18 or cyst wall imbrication.3,6,11,16,19 These surgical options are summarized in Table 1. Each procedure has some advantages and disadvantages. Recently, simple decompressive laminectomy and CT-guided percutaneous aspiration have not been recommended, because of the relatively high rates of complication and lower rates of success.2,3,18

The goals of surgical treatment are to relieve neural compression and prevent recurrence and CSF leakage. The former is achieved by decompressing the cysts, and the latter by repair of defects or closure of communications between the cyst and arachnoid space. Microsurgical partial cyst removal and cyst wall imbrication with
Fig. 2  Intraoperative photographs (upper row) and corresponding illustrations (lower row) showing the surgical steps and findings. 
A: After S2 laminectomy, the cyst (arrowheads) was exposed.  B: After fenestration of the cyst wall and evacuation of the fluid, the right S-2 nerve root ran caudally along inner the surface of the cyst wall (asterisk). Pulsatile cerebrospinal fluid flow was observed through the right S-2 nerve root sleeve (arrow).  C: The dural tear was closed with absorbable hemostat and fibrin glue.  D: The cyst wall was imbricated with 6–0 nylon. Arrows indicate the closure line.

Fig. 3  Photomicrographs showing the cyst wall consisting of dense, fibrocollagenous bundles with sparse cellularity, with no identified nervous tissue. Hematoxylin and eosin stain, original magnification A: × 40, B: × 200.

defect repair are frequently recommended.2,3,6,11,16,19) This procedure may be the most reasonable, since the goals of treatment can be achieved without venous and nerve injury. Total cyst removal or partial cyst removal with neck ligation may also be useful. However, nerve injury is possible during dissection of the entire cyst or neck ligation, because the sacral nerve root runs from the cyst neck along the medial cyst wall.2,3) Electrophysiological monitoring may help minimize injury to sacral nerve roots during procedures,11) but excision or ligation of cysts together with the involved nerve roots should be avoided, since such injury often results in neurological deficits, even if with monitoring confirmation that no major component of the nerve root enters the cyst.2,3) Bipolar cautery to shrink cysts may also be hazardous because of the possibility of nerve injury. On the other hand, cyst-subarachnoid shunting may be an effective option, since continuous CSF excretion can reduce the volume of cysts and prevent regrowth. The possibilities of dysfunction and infection should always be considered when performing this procedure.4–3)

In our case, the strategy of surgical treatment was both to decompress the cyst and to close the communication between the cyst and the arachnoid space completely, since CT myelography demonstrated delayed filling of the cyst. We consider that detection and closure of the point where CSF flows from the subarachnoid space into the cyst cavity is most important. Therefore, we chose to perform microsurgical treatment first; if we had not been able to locate the source of leakage of CSF, a cyst-subarachnoid shunt would have been required in addition to partial cyst removal and cyst wall imbrication. However, we were able to detect and close the point of CSF leakage, so partial
cyst removal and cyst wall imbrication were performed without shunt placement.

A total of 32 cases of sacral perineural cysts treated with partial cyst removal and cyst wall imbrication have been reported, including the present case (Table 2).²,⁷,¹¹,¹⁶,¹⁰ Twenty-seven of the 32 patients experienced complete or substantial resolution of their symptoms. Five patients had no significant improvement. Four patients suffered postoperative complications such as bladder dysfunction, CSF leak, prostatitis, and cerebellar bleeding. Only one patient had recurrence of the cyst and underwent reopera-

tion during the follow-up period ranging from 1 month to 73 months. Our patient experienced immediate resolution of pain without complication or recurrence of the cyst at 12-month follow up, but recurrence is possible, because a part of the cyst remained.

We recommend microsurgical partial cyst removal and cyst wall imbrication together with closure of the point through which CSF flows from the subarachnoid space into the cyst cavity for symptomatic perineural cysts. If the source of CSF leakage cannot be detected, placement of a cyst-subarachnoid shunt should be considered in addition to partial cyst removal and cyst wall imbrication.

Table 1  Summary of surgical procedures for perineural cysts

<table>
<thead>
<tr>
<th>Procedure</th>
<th>Advantage</th>
<th>Disadvantage</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Procedures to lower the hydrostatic and pulsatile pressures</td>
<td></td>
<td></td>
</tr>
<tr>
<td>1–1. External CSF drainage</td>
<td>ease</td>
<td>high recurrence rate</td>
</tr>
<tr>
<td>1–2. Lumboperitoneal shunt</td>
<td>ease</td>
<td>possibilities of dysfunction and infection</td>
</tr>
<tr>
<td>2. Procedures to decompress the cysts</td>
<td></td>
<td></td>
</tr>
<tr>
<td>2–1. Simple decompressive laminectomy</td>
<td>ease</td>
<td>low success rate</td>
</tr>
<tr>
<td>2–2. CT-guided percutaneous aspiration</td>
<td>ease</td>
<td>high recurrence rate and complications such as meningitis, nerve injury, and hemorrhage into the cyst</td>
</tr>
<tr>
<td>2–3. Cyst-subarachnoid shunt</td>
<td>ease</td>
<td>possibilities of dysfunction and infection</td>
</tr>
<tr>
<td>3. Direct surgical procedures</td>
<td></td>
<td></td>
</tr>
<tr>
<td>3–1. Bipolar cautery to shrink the cysts</td>
<td>complete cure</td>
<td>possibilities of nerve injury and CSF leakage</td>
</tr>
<tr>
<td>3–2. Total cyst resection</td>
<td>complete cure</td>
<td>possibilities of nerve injury</td>
</tr>
<tr>
<td>3–3. Partial cyst removal and neck ligation</td>
<td>complete cure</td>
<td>possibilities of CSF leakage</td>
</tr>
<tr>
<td>3–4. Partial cyst removal and cyst wall imbrication</td>
<td>complete cure</td>
<td>possibilities of nerve injury</td>
</tr>
</tbody>
</table>

CSF: cerebrospinal fluid, CT: computed tomography.

References

Table 2  Summary of reported cases of sacral perineural cysts treated with partial cyst removal and cyst wall imbrication

<table>
<thead>
<tr>
<th>Author (Year)</th>
<th>No. of patients</th>
<th>Age (yrs)</th>
<th>Sex (M/F)</th>
<th>Repair materials</th>
<th>Improved</th>
<th>Complications</th>
<th>Recurrence</th>
<th>Follow up (mos)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yucesoy et al. (1999)</td>
<td>1</td>
<td>48</td>
<td>0/1</td>
<td>FG and cementation</td>
<td>1/1</td>
<td>no</td>
<td>no</td>
<td>3</td>
</tr>
<tr>
<td>Mummaneni et al. (2000)</td>
<td>8</td>
<td>34–72</td>
<td>2/6</td>
<td>FG and fat or muscle</td>
<td>7/8</td>
<td>no</td>
<td>no</td>
<td>1–73</td>
</tr>
<tr>
<td>Lee et al. (2004)</td>
<td>2</td>
<td>24–46</td>
<td>0/2</td>
<td>none</td>
<td>2/2</td>
<td>no</td>
<td>no</td>
<td>6–12</td>
</tr>
<tr>
<td>Tanaka et al. (2006)</td>
<td>9</td>
<td>43–72</td>
<td>6/3</td>
<td>none</td>
<td>7/9</td>
<td>2/9</td>
<td>no</td>
<td>6–24</td>
</tr>
<tr>
<td>Guo et al. (2007)</td>
<td>11</td>
<td>28–44</td>
<td>0/5</td>
<td>FG, absorbable gelatin sponge, and muscle</td>
<td>9/11</td>
<td>2/9</td>
<td>1/11</td>
<td>28–44</td>
</tr>
<tr>
<td>Present case</td>
<td>1</td>
<td>67</td>
<td>1/0</td>
<td>FG and absorbable collagen sponge</td>
<td>1/1</td>
<td>no</td>
<td>no</td>
<td>12</td>
</tr>
</tbody>
</table>

FG: fibrin glue.

2008

Address reprint requests to: Hiroaki Matsumoto, MD, PhD, Department of Neurosurgery, Eisyokai Yoshida Hospital, 9–2–6 Daikai–dori, Hyogo–ku, Kobe 652–0803, Japan. e-mail: hiroaki-matsu@umin.ac.jp