Adhesive arachnoiditis after percutaneous fibrin glue treatment of a sacral meningeal cyst

Case report

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The authors present the case of a 64-year-old woman who was referred for severe sacral pain. She reported that her pain had been longstanding, and had greatly increased after percutaneous fibrin glue placement therapy for a sacral meningeal cyst 2 months earlier at a different hospital. An MRI scan obtained immediately after fibrin glue placement at that hospital suggested that fibrin glue had migrated superiorly into the subarachnoid space from the sacral cyst to the level of L-4. On admission to the authors’ institution, physical examination demonstrated no abnormal findings except for perianal hypesthesia. An MRI study obtained at admission demonstrated a cystic lesion in the peridural space from the level of S-2 to S-4. Inhomogeneous intensity was identified in this region on T2-weighted images. Because the cauda equina and nerve roots appeared to be compressed by the lesion, total cyst excision was performed. The cyst cavity was filled with fluid that resembled CSF, plus gelatinous material. Histopathological examination revealed that the cyst wall was composed of hyaline connective tissue with some calcification. No nervous tissue or ganglion cells were found in the tissue. The gelatinous material was acellular, and appeared to be degenerated fibrin glue. Sacral pain persisted to some extent after surgery. The authors presumed that migrated fibrin glue caused the development of adhesive arachnoiditis.

The risk of adhesive arachnoiditis should be considered when this therapy is planned. Communication between a cyst and the subarachnoid space should be confirmed to be sufficiently narrow to prevent the migration of injected fibrin glue.

| Key Words | • sacral meningeal cyst • fibrin tissue adhesive • arachnoiditis |

Since Patel et al. introduced CT-guided percutaneous fibrin glue placement to treat a sacral meningeal cyst in 1997, multiple additional cases of this procedure, with different variations, have been described. Reports of complications after this therapy are rare. We present a case in which a sacral meningeal cyst persisted after fibrin glue placement, with the glue migrating into the subarachnoid space, exacerbating the patient’s sacral pain and inducing adhesive arachnoiditis. To our knowledge, this has not previously been reported in the literature.

Case Report

History. This 64-year-old woman was seen in our outpatient clinic reporting severe pain in the sacral region of 2 months’ duration. Her history was significant for the onset of mild sacral pain 1 year previously. The MRI studies performed at a different hospital had shown a sacral meningeal cyst (Fig. 1). Two months before being seen in our clinic, after multiple cyst aspirations provided no relief, she underwent CT-guided percutaneous fibrin glue placement. For this procedure, 8 ml of fluid in the cyst was aspirated and then an equal volume of fibrin glue was placed with a 2-needle technique. Following the CT-guided procedure, she noted a marked increase in sacral pain, requiring increasing doses of opioid analgesics. An MRI scan performed immediately after the treatment had revealed a cyst in the peridural space extending from the level of S-2 to S-4, with heterogeneous material in the subarachnoid space extending from the level of L-4 to the superior margin of the cyst. Below the level of L-5 the cauda equina roots were distributed peripherally, consistent with adhesive arachnoiditis (Fig. 2).

Examination. When seen in our clinic, the patient reported taking 160 mg of codeine phosphate per day. She described the pain as “feeling as if the spine had exploded,” and evaluated its intensity as 10 of 10 on estimation by a visual analog scale. The Japanese Orthopaedic Association score of back pain was 9 of 29, indicating severe pain. Physical examination revealed that she could neither stand nor sit owing to severe sacral pain. Strength in her lower extremities was 5 of 5. Sensation to pinprick and light touch was intact throughout all lower extremity dermatomal distributions, except in the perianal region. Patellar and Achilles deep tendon reflexes were increased symmetrically.

Neuroimaging Studies. An MRI study performed on admission to our hospital (Fig. 3) demonstrated a persistent sacral cyst with inhomogeneous contents visible on T2-weighted images. The heterogeneous subarachnoid material seen on the previous scan had resolved. The cau-
da equina and nerve roots seemed to be compressed by the cyst. The peripheral distribution of nerve roots seen inferior to L-5 on the postprocedural examination was unchanged (Fig. 3D). A CT myelography study showed no early-phase accumulation and slight late-phase contrast accumulation in the cyst. We considered her sacral pain to be caused by a combination of persistent compression by the cyst and adhesive arachnoiditis.

**Operation.** Total cyst excision and decompression were performed. After laminectomy from S-2 to S-4, the whole cyst was brought under direct vision. The dura mater and nerve roots were severely compressed to the left side of the canal by the cyst, and adhered closely to the cyst wall. We attempted dissection of the cyst from the dura mater, but could not preserve its integrity because of the adherence. Although the cyst wall was perforated during dissection, it did not collapse, because the wall was thick and fibrotic. The cyst’s contents consisted of a combination of what appeared to be CSF and gelatinous material (Fig. 4).

**Pathological Findings.** The resected cyst was submitted for histopathological examination. Light microscopic findings demonstrated that the cyst wall was composed of hyaline connective tissue with areas of calcification (Fig. 5). The gelatinous material was acellular, and appeared to be residual, partially clotted fibrin glue. There was no malignant change. No nerve cells or ganglion cells were found in the sample, which was also negative for S100 protein.

**Postoperative Course.** The patient experienced partial relief of pain after the operation, and was able to be weaned away from oral codeine phosphate. However, 1 year after surgery she still feels some sacral pain, and perianal numbness and abnormal lower-extremity deep tendon reflexes persist.

**Discussion**

Tarlov described sacral meningeal cysts as “perineurial cysts” in 1938, but later labeled them as “meningeal cysts” to distinguish them from perineurial cysts. These terms are often confused in the literature. A strictly perineurial cyst arises between the perineurium and endoneurium, and its lining contains nerve fibers and/or ganglion cells. The present case fits the definition of a sacral meningeal cyst and not of a perineurial cyst, because of the lack of nervous tissue in its lining.

Although asymptomatic sacral meningeal cysts are not considered to require treatment, the treatment of symptomatic sacral meningeal cysts remains controversial. Several recently reported nonsurgical therapies include oral or epidural steroid therapy, lumbar CSF drainage, CT-guided percutaneous aspiration, and fibrin glue placement after aspiration. Surgical therapies include simple decompressive laminectomy and recapping laminectomy with cyst fenestration, or with either total cyst excision (which sometimes sacrifices the involved sacroccygeal nerve roots) or partial resection of the cyst and wall imbrication with or without neck ligation. Other procedures include microsurgical excision and cyst wall imbrication together with repair of the defect with muscle.

Simple percutaneous aspiration is of limited value because of the postulated ball-valve effect and cyst reinflation. Paulsen et al. reported instant pain relief lasting from 3 weeks to 6 months with this method, but that repeated aspiration was required to maintain pain control.

Patel et al. described fibrin glue therapy in 1997 as a more definitive treatment for patients with cyst recurrence after initial aspiration. After CT-guided percutane-
ous aspiration is continued until no more fluid can be aspirated, an equal volume of fibrin glue is injected into the cyst cavity. The injected fibrin glue is expected to remain in the cyst and occlude the communication between the cyst and the subarachnoid space, obliterating any ball-valve one-way occlusion and preventing the reaccumulation of CSF. Used appropriately, fibrin glue clearly provides good tissue adhesion, which will occlude the communication. Once the communication has been occluded, the fibrin glue is expected to dissolve or be gradually resorbed by fibrinolysis, which may result in subsequent fibrosis. Zhang et al. reported 31 cases treated by this method, and almost 80% of the patients had sufficient improvement in symptoms to allow a return to regular work. These authors also found that there was no significant distinction between groups with or without initial aspiration of the cyst. Murphy and colleagues reported more than 100 cases treated using a 2-needle technique, and attained 65% improvement in symptoms, which is equivalent to surgical treatment.

Some complications have been reported. Our review of the literature found 5 cases of aseptic meningitis. Patel et al. reported that 3 of 4 patients suffered headache, dizziness, and fever within a few days of the procedure. However, Murphy et al. reported no occurrence of postprocedure aseptic meningitis in more than 100 cases. Overall, percutaneous fibrin glue treatment is considered to have an acceptable risk compared with the risks of surgery.

To our knowledge, adhesive arachnoiditis after fibrin glue treatment has not been previously reported. However, in the event of migration into the subarachnoid space, fibrin glue could theoretically cause inflammation of the arachnoid mater and adherence to the cauda equina. Subsequently, the cauda equina may coalesce with itself and/or with the arachnoid mater, and be vulnerable to ischemia. In our case, an MRI study performed immediately after fibrin glue placement showed that this material had migrated upward into the subarachnoid space from the sacral cyst to the level of L-4, resulting in adherence of the cauda equina roots to the surrounding arachnoid (Fig. 2).

The patient’s sacral pain progressed at the same time, presumably in part due to adhesive arachnoiditis. This is a case of failure and poor outcome of the fibrin glue therapeutic approach for a sacral meningeal cyst. Although no severe complication with this procedure has been reported before, there is some concern that it would be the tip of the iceberg.

Additionally, the injected fibrin glue, which should have been resorbed by fibrinolysis, was noted several months later in the cyst, which had not collapsed, most likely due to persistent communication with the CSF, and which accordingly continued to compress the thecal sac. Although we performed cyst excision to decompress the sac, its therapeutic value was limited due to persistent adhesive arachnoiditis.

At a minimum, clinicians who intend to use this procedure have to ensure that the communication between the cyst and the subarachnoid space is narrow enough to prevent migration of the fibrin glue into the subarachnoid space. Murphy and colleagues compared T2-weighted MRI studies of a cyst to those of the CSF around the cauda equina and reported that if the signal in the cyst is brighter than the signal in the CSF around the cauda equina, then the cyst has a narrow neck. In our case, the signal in the cyst was indeed higher than that of the subarachnoid space, and myelography studies performed...
before fibrin glue injection showed that the cyst did not immediately fill with contrast medium. Although the precise cause of glue migration remains to be elucidated, once adhesive arachnoiditis develops it is difficult and sometimes impossible to treat. This risk needs to be considered when planning fibrin glue replacement therapy of a sacral meningeal cyst. Furthermore, it may be no exaggeration to say that there is little place for this therapy due to the risk.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: Hayashi, Hattori. Acquisition of data: Hayashi. Analysis and interpretation of data: Hayashi. Drafting the article: Hayashi. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Hayashi. Administrative/technical/material support: Nagano, Hattori. Study supervision: Nagano.

References


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Fig. 4. Intraoperative photographs of the cyst. Upper: Whole view after laminectomy from S-2 to S-4. Lower: Inside view after suctioning of the fluid, which contained gelatinous material (arrow).

Fig. 5. Photomicrograph of the resected cyst showing the cyst wall, which was composed of hyaline connective tissue with some calcification. No nerve or ganglion cells were found. H & E, original magnification ×15.