Spinal endoscopy combined with selective CT myelography for dural closure of the spinal dural defect with superficial siderosis: technical note

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The authors describe a new procedure to detect the tiny dural hole in patients with superficial siderosis (SS) and CSF leakage using a coronary angioscope system for spinal endoscopy and selective CT myelography using a spinal drainage tube. Under fluoroscopy, surgeons inserted the coronary angioscope into the spinal subarachnoid space, similar to the procedure of spinal drainage, and slowly advanced it to the cervical spine. The angioscope clearly showed the small dural hole and injured arachnoid membrane. One week later, the spinal drainage tube was inserted, and the tip of the drainage tube was located just below the level of the dural defect found by the spinal endoscopic examination. This selective CT myelography clarifies the location of the dural defect. During surgery, the small dural hole could be easily located, and it was securely sutured. It is sometimes difficult to detect the actual location of the small dural hole even with thin-slice MRI or dynamic CT myelography in patients with SS. The use of a coronary angioscope for the spinal endoscopy combined with selective CT myelography may provide an effective examination to assess dural closure of the spinal dural defect with SS in cases without obvious dural defects on conventional imaging.

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KEY WORDS superficial siderosis; spinal endoscopy; coronary angioscope; CT myelography; dural closure; dural defect; surgical technique

SUPERFICIAL siderosis (SS) of the CNS is a rare and intractable disease with hemosiderin deposition in the subpial layers of the brain and spinal cord caused by chronic or repeated subarachnoid hemorrhage (SAH).^{6,12} Patients with SS typically show the slow progression of cerebellar ataxia, sensorineural hearing loss, and dementia.^{5,9,12,17,20} MRI usually shows hemosiderin deposition at the surface of the CNS, especially in the cerebellum, brainstem, and spinal cord.^{11,13,22,24}

Some patients with SS show the presence of CNS tumors or arteriovenous malformations, or a history of head trauma or CNS surgery;^{5,17,20,29} however, some studies have reported spinal dural defects with longitudinal extradural fluid collection in the spinal canal on spinal MRI or CT myelography.^{2,4,5,7,8,12,14–16,22} Recently, these characteristic features have received a great deal of at-

tention as causes of SS,^{2,12,13,15} and dural closure has been performed for SS patients with a spinal dural defect.^{2,4,5,7,8,12,14–16,22}

Kumar et al., who first reported the repair of the dural defect for patients with SS, reported the usefulness of dynamic CT myelography to identify the site of the dural defect.¹⁴ Egawa et al. reported the usefulness of constructive interference in steady state (CISS) MRI, showing dural defects in the thoracic spinal canal in 2 patients with SS.⁵ If the site of communication between the epidural and subarachnoid spaces is very small, dynamic CT myelography and CISS MRI may have a limited capacity to show the tiny dural hole.

In this article, we describe a new modality to detect the dural defect in 2 patients with SS using a coronary angioscope system in the spinal canal and selective CT

ABBREVIATIONS CISS = constructive interference in steady state; COSMIC = coherent oscillatory state acquisition for the manipulation of an image contrast; SAH = subarachnoid hemorrhage; SS = superficial siderosis.

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FIG. 1. **A:** Photograph showing the test setup for spinal endoscopic examination with a coronary angioscope system (FT-203F angioscope and VISIBLE fiber). **B:** Photograph showing a patient in the lateral position in a hybrid operating room. A TV monitor can show both images of fluoroscopy and spinal endoscopy. **C:** Photograph showing an angioscope fiber being introduced into the lumbar subarachnoid space through the outer needle of the lumbar spinal drain. **D:** Photograph showing a spinal endoscope view demonstrating the dura matter (arrow), vessel (arrowhead), and spinal cord (double arrows). Figure is available in color online only.

myelography with a spinal drainage tube. Intraoperative findings agreed with the preoperative findings based on these examinations, and we could suture the tiny dural holes during open surgery.

Methods

Patients

Two patients with SS were diagnosed based on typical brain MRI findings. Spinal MRI clearly showed longitudinally extensive CSF at the epidural ventral surface, but thin-slice axial MRI could not clearly detect any dural defect.

Spinal Endoscopy

Spinal endoscopic examination was performed with a coronary angioscope system (FT-203F angioscope and VISIBLE fiber, Fiber Tech Co.; Fig. 1A). This fiber catheter contains 6000 optic fibers as imaging and illuminating fibers within its diameter. The outside diameter and visual angle are 0.75 mm and about 60°, respectively. The angioscope fiber is very flexible, and the effective length of the fiber is 1.62 m, which is sufficient to be advanced to the upper cervical level.

The examination procedure was approved by the IRB of Fukui University Hospital and performed in a hybrid

operating room. With the patient in a lateral position under local anesthesia (Fig. 1B), the angioscope fiber is introduced into the lumbar subarachnoid space through the outer needle of the lumbar spinal drain (Fig. 1C). Surgeons slowly advance the angioscope fiber upward while confirming the position of the fiber tip, which is visible under fluoroscopy. The outer needle should be removed to prevent CSF hypovolemia due to CSF leakage through the needle. The fiber tip should be advanced along the ventral part of the spinal cord because the epidural fluid collection through the dural defect is usually at the ventral part of the spinal cord. If the surgeon feels resistance on attempting to advance the fiber, it should never be advanced forcibly. The surgeon should pull the angioscope fiber down and search for another space where the fiber can be advanced smoothly. The angioscope fiber can show the arachnoid membrane, dura mater, spinal cord, spinal vessels, and nerve roots (Fig. 1D). The tiny dural defect can be detected as a small dural hole or an abnormal pulsation of the arachnoid membrane. When surgeons detect the dural defect with the angioscope camera, they should confirm the spinal level of the defect under fluoroscopy. Neuromonitoring was not used during this manipulation because surgeons could communicate with patients under local anesthesia, and directly asked the patients about sensorimotor disturbance.

FIG. 2. Case 1. **A:** Brain T2*-weighted axial MR image showing the low-level deposition of hemosiderin predominantly in the brainstem and cerebellum. **B and C:** Preoperative T2-weighted sagittal MR images showing longitudinal epidural fluid collection ventral to the spinal cord from C-2 to T-12 (arrows). **D:** Thin-slice axial MR image with COSMIC sequence at the level of the dural defect (T7–8) could not clearly show the small dural hole.

Selective CT Myelography

The intracranial pressure in patients with SS is sometimes low.^{5,7,13,15,21,28} CSF leakage after spinal endoscopy may cause further intracranial hypotension; therefore, we performed CT myelography 1 week after the spinal endoscopy.

With the patient in a lateral position, the spinal drainage tube is inserted into the lumbar subarachnoid space. Under fluoroscopic guidance, the tip of the drainage tube is located 1 level below the dural defect, confirmed by endoscopic examination. In the CT room, the patient adopts a prone position with a supporting cushion under the ilium on the CT bed so the contrast medium can flow in a rostral direction. CT must be performed just after injection of the contrast medium with 2 ml from the spinal drainage tube. Contrast medium remains within the intact dura mater on the caudal side of the dural defect; however, at the level of the dural defect, the contrast medium fills not only the intradural space but also the ventral epidural space through the dural hole due to its high specific gravity in a prone position. Then, the contrast medium immediately diffuses in both the intra- and extradural spaces. In this way, surgeons can verify the position of the dural defect. Selective CT myelography performed a few minutes after the injection of contrast medium cannot precisely detect the dural hole because the contrast medium longitudinally diffuses not only in the intradural space, but also in the epidural space of the spinal canal, rostrally and caudally.

Illustrative Cases

Case 1

A 50-year-old man was referred to our hospital with a 10-year history of slow, progressive worsening motor disturbance of the bilateral upper limbs. A neurological examination showed a normal mental state, intact cranial nerves, ataxia of the extremities, mild motor weakness in his upper limbs dominant on the left, and hyperactive tendon reflexes in all limbs. Although his sensory function was almost normal, he complained of numbness in his upper limbs. He also complained of gait difficulty, and his gait was ataxic. He had a history of surgery for bilateral chronic subdural hematoma 15 years ago. He continued to have mild headache even after the surgery.

Brain T2*-weighted MRI (Fig. 2A) showed the low-level deposition of hemosiderin in the superficial brain, predominantly in the brainstem, cerebellum, and bilateral sylvian cisterns. Spinal T2-weighted MRI (Fig. 2B and C) demonstrated longitudinal epidural fluid collection ventral to the spinal cord from C-2 to T-12; however, thin-slice axial MRI with CISS and coherent oscillatory state acquisition for the manipulation of an image contrast (COSMIC) sequence (Fig. 2D), as well as dynamic CT myelography, did not clearly detect any dural defect in the spinal canal. CSF analysis revealed xanthochromia with a low opening pressure of 20 mm H₂O.

We performed spinal endoscopy using the coronary angioscope system as stated above, which revealed a tiny

FIG. 3. Case 1. **A:** The spinal endoscope showing the tiny dural hole (arrow) and spinal cord (arrowhead) at the T7–8 level. **B and C:** Selective CT myelography with the tip of the drainage tube at the T8–9 level with the patient prone. At the T8–9 level (B), the tip of the drainage tube (arrow) and contrast medium can be detected just at the intradural spinal canal. At the T7–8 level (C), contrast medium fills not only the intradural space but also the ventral epidural space (arrow) through the dural hole due to its high specific gravity. **D:** An intraoperative photograph showing the tiny dural hole at the T7–8 level (arrow). **E and F:** Postoperative T2-weighted sagittal MRI showing the disappearance of the epidural fluid collection. Figure is available in color online only.

dural hole at the T7–8 level (Fig. 3A). After a week, we located the tip of the drainage tube at the T8–9 level and performed selective CT myelography. The tip of the drainage tube and contrast medium could be detected at the intradural spinal canal (Fig. 3B); however, at the T7–8 level, contrast medium fills not only the intradural space but also the ventral epidural space through the dural hole due to its high specific gravity (Fig. 3C), which verified the dural defect at the T7–8 level.

After T6–8 laminectomy and opening of the dura mater, we detected the tiny hole in the anterior median dura mater at the T7–8 level (Fig. 3D), which was repaired with three 8–0 nylon sutures. Postoperative spinal MRI showed the disappearance of epidural fluid collection ventral to the spinal cord (Fig. 3E and F). The patient's headache completely disappeared after the operation. Other neurological symptoms were stable without progression during the 17-month follow-up.

Case 2

A 59-year-old man was referred to our hospital with a 3-month history of progressive worsening of motor disturbance of the right upper and lower limbs. A neurological examination showed a normal mental state, slight bilateral hearing disturbance, mild motor weakness of the right upper and lower limbs, and hyperactive tendon reflexes in all

limbs. Although his sensory function was almost normal, he complained of intermittent numbness in his right upper and lower limbs. His gait was almost normal, and cerebellar ataxia was not observed. He had a history of surgery for bilateral chronic subdural hematoma 20 years ago. He continued to have chronic headache.

Brain T2*-weighted MRI (Fig. 4A) showed the characteristic features of SS. Spinal T2-weighted MRI (Fig. 4B and C) demonstrated longitudinal epidural fluid collection ventral to the spinal cord from C-2 to T-12; however, thin-slice axial MRI with CISS and the COSMIC sequence (Fig. 4D), and dynamic CT myelography, did not detect the dural defect. CSF analysis revealed xanthochromia with a low opening pressure of 0 mm H₂O.

The spinal endoscopy revealed the dural defect with abnormal pulsation of the arachnoid membrane at the C-7 level (Fig. 5A). After a week, we performed selective CT myelography with the tip of the drainage tube at the T-1 level with the patient prone. At the T-1 level, the tip of the drainage tube and contrast medium could be detected just at the intradural spinal canal (Fig. 5B); however, at the C-7 level, contrast medium leaked into the epidural space through the dural defect (Fig. 5C), which verified the dural defect at the C-7 level.

During the C6–T1 laminectomy, we detected the small hole in the anterior median dura mater (Fig. 5D), which

FIG. 4. Case 2. **A:** Brain T2*-weighted axial MRI showing the low-level deposition of hemosiderin in the brainstem, cerebellum, and surface of the temporal lobes. **B and C:** Preoperative T2-weighted sagittal MRI showing longitudinal epidural fluid collection ventral to the spinal cord from C-2 to T-12 (arrows). **D:** Thin-slice axial MRI with COSMIC sequence at the level of the dural defect (C-7 level) could not clearly show the small dural hole.

was repaired with two 8-0 nylon sutures. Postoperative spinal MRI showed the disappearance of the epidural fluid collection (Fig. 5E and F). His headache completely disappeared after the operation. The intermittent numbness in the patient's right upper and lower limbs disappeared, and motor weakness of the right upper and lower limbs gradually improved during the 9-month follow-up.

Discussion

Since Kumar et al. first reported surgical treatment for SS patients with a dural defect,¹² patients with SS treated by dural closure using sutures or patches with muscle, fat, or fibrin glue have been reported.^{2,4,5,7,8,12,14–16,22} Previous reports stated that surgical treatment with dural closure could not markedly resolve the neurological symptoms associated with SS, except for symptoms associated with CSF hypovolemia.^{2,4,5,7,8,12,14–16,22} A long-term neuronal injury due to neurotoxic hemosiderin deposition and free radical damage is believed to cause irreversible neurological deterioration.¹⁰

The origin of chronic or repeated SAH in SS with a spinal dural defect is unknown. Kumar et al. suggested that friable vessels in the dural defect may be a possible source of the chronic bleeding.¹² Cheng et al. proposed a hypothesis in which an increased epidural pressure may micro-traumatize the fragile internal venous plexus, leading to

recurrent microbleeding.⁴ Based on these hypotheses, dural closure may stop chronic or repeated SAH through the dural defect, and this may prevent the progressing symptoms of SS. Patients with SS and a dural defect occasionally show intracranial hypotension.^{5,7,13,15,21,28} We think that dural closure for SS with a spinal dural defect is a reliable procedure. Before surgery, surgeons should verify the position of the dural defect to prevent the need for an unnecessarily large laminectomy to find it.

Kumar et al. identified the site of communication between the epidural cavity and arachnoid space using dynamic CT myelography, which revealed the spinal dural defect of SS.¹⁴ In our 2 cases we initially used this method, but contrast medium immediately diffused up and down the epidural cavity through the small dural hole. We could not accurately identify the dural defect in either case. Well-timed dynamic CT myelography may be difficult in some cases. Usually, T2-weighted imaging can reveal the ventral epidural fluid collection in the spinal canal; however, it may be difficult to detect the small dural hole by axial T2-weighted imaging. We could not find the tiny dural defect even with high-resolution CISS MRI.⁵ In addition, we used the COSMIC sequence, which is suitable to visualize the intervertebral foramen for spinal root disorders;²³ however, we could not verify the dural defect with conventional methods.

FIG. 5. Case 2. **A:** The spinal endoscope showing the tiny dural hole (arrow) and injury of the arachnoid membrane (arrowhead) at the C-7 level. A spinal cord with capillary vessels can be seen (double arrows). **B and C:** Selective CT myelography with the tip of the drainage tube at the T-1 level with the patient prone. At the T-1 level (B), the tip of the drainage tube (arrow) and contrast medium can be detected just at the intradural spinal canal. At the C-7 level (C), contrast medium leaked into the epidural space (arrow) through the dural defect. **D:** An intraoperative photograph showing the tiny dural hole at the C-7 level (arrow). **E and F:** Postoperative T2-weighted sagittal MRI showing the disappearance of epidural fluid collection. Figure is available in color online only.

We used a coronary angioscope system for the spinal endoscopy to examine the dural defect inside the spinal canal, with approval by the IRB of Fukui University Hospital. To the best of our knowledge, this is the first report on the detection of small dural defects using the coronary angioscope system. The coronary angioscope fiber is usually used for examination of the inner coronary artery using the guiding catheter by saline solution perfusion.^{1,18,25,26} In the spinal canal, an angioscope fiber can be used without saline solution perfusion, and operators can handle it like a spinal drainage tube and obtain clear images of the intradural spinal canal. The color images from spinal endoscopy were very helpful but were of low resolution, which may be a limitation of the endoscopic technology. This technology has no potential for therapeutic applications.

The drawback of using the coronary angioscope for spinal endoscopic examination is the poor manipulation of the fiber tip during the procedure. Although the tip of the angioscope fiber is flexible, operators cannot change the direction of the angioscope tip. Operators should perform spinal endoscopic examination while confirming the location of the fiber tip under fluoroscopy. It is important to confirm that the tip advances in the ventral subarachnoid space of the spinal cord, and check the level of the angioscope tip. Although the angioscope fiber is very flexible with the blunt tip, important complications during the spinal endoscopy are spinal cord injury, SAH, and subdural hematoma. Intrathecal baclofen therapy, which requires a technique similar to spinal endoscopy, can be

safely performed without cord injury.²⁷ We identified 2 cases of intraoperative complications inducing slight SAH. One might occur during the intrathecal insertion of the catheter,³ and the other might arise from the migration of a broken fragment of the catheter.¹⁹ To prevent injury of intraspinal vessels, the spinal cord, and spinal roots, operators should never advance the angioscope fiber upward when the tip cannot be easily advanced. Operators should take care to avoid damaging the angioscope fiber with the outer needle of spinal drain. In our cases, only 2 neurosurgeons with sufficient experience in the catheter technique of cerebral angiography, spinal drainage, and intrathecal baclofen therapy could operate the scope without the help of cardiac surgeons. Inexperienced neurosurgeons should not operate the scope. There is the potential for new-onset CSF leak due to the 2 lumbar punctures for the spinal endoscope and selective CT myelography. Intracranial hypotension should be considered after these examinations.

Selective CT myelography cannot be performed at multiple levels in the spinal canal at once, and this is an auxiliary examination after spinal endoscopy. If thin-slice axial MRI strongly suggests a dural defect, spinal endoscopy or selective CT myelography can be omitted. Our technique should be considered only in cases with an unknown dural hole even by conventional examinations. Although this procedure may not bring about marked neurological improvement, we believe that the detection and repair of the spinal dural hole can prevent the further progression of neurological deficits. Patients with severe disability or pro-

gressive deficit should be selected with appropriate patient counseling.

Conclusions

We have presented a new procedure to detect the tiny dural holes in SS patients with CSF leakage using a coronary angioscope system for the spinal endoscopy and selective CT myelography using the spinal drainage tube. These procedures may prevent a large and unnecessary laminectomy to detect a tiny dural hole, and are useful for surgeons to accurately detect the location of dural defects that cannot be detected by conventional imaging.

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Disclosures

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

Conception and design: Arishima, Kikuta. Acquisition of data: Arishima, Higashino, Awara. Analysis and interpretation of data: Arishima, Higashino, Awara. Drafting the article: Arishima. Critically revising the article: Arishima, Kikuta. Reviewed submitted version of manuscript: Arishima, Higashino, Kikuta. Approved the final version of the manuscript on behalf of all authors: Arishima. Administrative/technical/material support: all authors. Study supervision: Arishima, Kikuta.

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