

## Case Report

# Orthostatic Headache from Spontaneous Spinal Cerebrospinal Fluid Leakage; Diagnosed by Heavily T2-Weighted Magnetic Resonance Myelography

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Orthostatic headache is derived from low cerebrospinal fluid (CSF) pressure as evidenced by cranial magnetic resonance myelography (MRM). This reports three cases of patients coming with orthostatic headache without previous obvious spine trauma. The first two cases had headache with radiating neck pain while the third case had headache with radiating pain to the eye sockets or occasional nausea. The third case was diagnosed from cranial MR imaging. The three cases were not done for CSF opening pressure measuring or criterion's method myelography, but had done T2-weighted MR. All of three cases had spinal epidural collections. The second case had meningeal diverticula. The present report found a possible site of leak in all cases. In the present report, T2-weighted MR myelography could avoid dural puncture. It was used as a non-radiation exposure investigating technique. This technique can be used as the first line of investigation prior to CTM, guiding a radiologist to seek the most likely site of leak during CTM study.

**Keywords:** Orthostatic headache, Spontaneous spinal cerebrospinal fluid leakage, Heavily T2-weighted magnetic resonance myelography

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Orthostatic headache is derived from assuming an upright position. The most likely pathophysiology is low cerebrospinal fluid (CSF) pressure<sup>(1)</sup>, which is the result of CSF leakage, either postdural puncture or spontaneous leakage. Without an obvious trauma, the diagnosis of spontaneous CSF leakage<sup>(2)</sup> needs the detail of headache presentation and investigation criteria. The authors reported three cases that were investigated between June and October 2010. These patients presented with orthostatic headache, slightly different in characteristic details. They underwent magnetic resonance myelography (MRM) as the initial investigation tool. It provided diagnosis of spontaneous spinal CSF leakage. This case report was approved by the ethical committee of Vichaiyut Hospital.

### Imaging technique and interpretation

MRM was performed with a superconducting magnetic resonance machine, 1.5 Tesla (Magnetom Avanto Version Syngo, Siemens, Erlangen, Germany). A spine matrix coil was utilized. A 3D imaging was acquired by using TSE sequence with restore pulse. Long echo time (TE > 600 ms) was used to obtain a heavily T2-weighted images. Axial and sagittal MRMs were acquired at all three levels (cervical, thoracic, and lumbar) or at the most suspected level of CSF leakage according to clinical judgment. The following parameters were performed; matrix size ranging 234 x 320 mm to 380 x 384 mm, slice thickness 1.5-2 mm on sagittal plane and 2-3 mm on axial plane, field of view ranging 280 x 208 mm to 370 x 370 mm on sagittal plane and ranging 120 x 160 mm to 195 x 240 mm on the axial plane. Suitable parameters were used differently for each spinal level of study. Accompanied T1-weighted images on both sagittal and axial planes were obtained for better soft tissue contrast details.

Imaging interpretation was done on a picture archiving and communication system (PACS). A radiologist assessed the MRM. The findings of abnormal extrathecal CSF were noted.

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## Cases Report

### Case 1

A 37-year-old Thai, office working woman came to the hospital with headache at the occipital area for four days. Her headache radiated along her neck and shoulders. Upright position made her symptom worse. Lying down for 30 minutes could relieve her headache but the symptom resumed within a few minutes after sitting or standing up. Sometimes, her symptom got worse, where it felt like it was pulling her head from behind and she needed to lie down. She used to practice yoga in the past. After stopping for several months, she started practicing again 10 days ago. Five days after resuming, she felt a minor pain in the middle of her back, moving downward to her waist. The next day she could not go to work and needed some rest.

Her physical exams were normal, except she had a point of mild tenderness between the lower thoracic and upper lumbar spines. Her blood exam, complete blood count, was normal. Inflammatory and immunologic profile; erythrocyte sedimentary rate of 25 mm/hour was in the upper end of normal limit and positive antinuclear antibody titers; homogeneous and speckled patterns 1:80, both of them were in the upper end of normal limit.

Cranial magnetic resonance imaging (MRI) demonstrated normal brain parenchyma, normal brainstem location, and no evidence of subdural fluid collection. Gadolinium-enhanced MRI showed no abnormal enhancement, no finding of pituitary hyperemia. Magnetic resonance venography revealed normal appearance of dural venous sinuses. There was CSF tracking in posterior epidural space from about T2-3 to T11 levels and along bilateral nerve roots at T4-level. The possible point of leak located at the posterior aspect of T6 level, of which a small gap of discontinued dural sleeve was seen, (Fig. 1). A spontaneous spinal CSF leakage was diagnosed.

After counseling for epidural blood patching (EBP), she preferred bed rest with the abdominal binder and oral analgesics given as the supportive treatments and her symptom had gradually recovered after one month.

### Case 2

A 36-year-old Thai, silo manager woman, came to the hospital with headache at the occipital area for a few months, the headache radiated to her neck. Her symptom started in late morning during working, sitting or standing. The symptom would be absent,

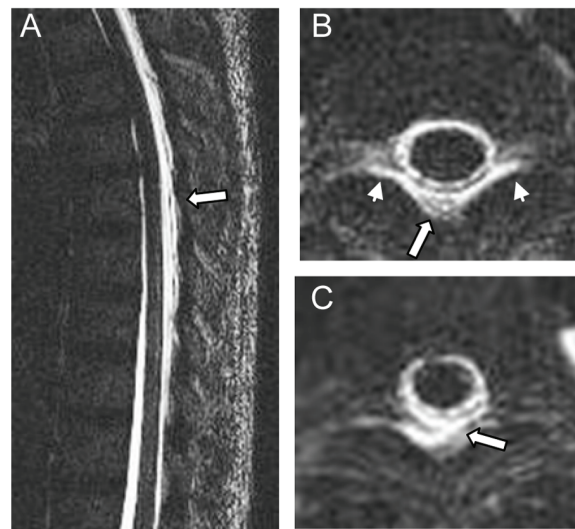
mostly only just after waking up, and then the intensity had been gradually increasing up to the maximum level in the afternoon, which she needed to lie down to relieve her headache. At the maximum intensity, she felt like pulling her head from the rear, pain score of 9/10, without nauseating. After a period of lying down during the day, she could work in the evening and the headache did not occur when she slept during the night.

She often has sciatica pain on the left leg, which radiates from the lower back to the dorsum of her left foot since two years ago after bending her back to carry some heavy bags of sugar.

Her physical exams were normal, except slight weakness of extensor hallucis longus of the left foot. Her immunologic profile; antinuclear antibody titer was positive 1:80 for speckled pattern which was in the upper end of normal limit.

Cranial MRI showed no intracranial abnormality. Gadolinium-enhanced MRI demonstrated neither abnormal thickened-enhancing meninges nor subdural collection. Thoraco-lumbar spine MRI with and without contrast study revealed broad-based meningeal diverticula through right T12-L1 neural

### Case 1



**Fig. 1** Heavily T2WI (TSE T2-weighted images with long echo time), sagittal (A) and axial sections (B and C), showed presence of fluid collection in the posterior epidural space (arrows in A and B), with evidence of fluid tracking along bilateral nerve root sleeves at T4-level (B, short arrows). A small gap of discontinued dural sleeve was seen posteriorly to T6-level, possible site of leak (C, arrow)

foramen and a smaller one at right L1-2 neural foramina opening (Fig. 2). Posterior and right-sided epidural fluid collections were seen from mid T12 to L1-2 disc level (Fig. 3). A spontaneous spinal CSF leakage was diagnosed.

After counseling for EBP, she preferred bed rest with the abdominal binder as a supportive treatment and the symptom had gradually recovered after one month.

### Case 3

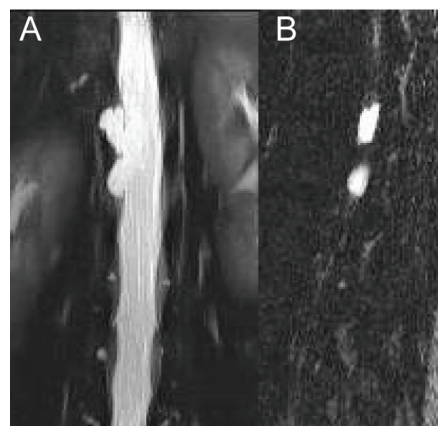
A 41-year-old Thai man, business owner, came to the hospital with gradually progressive headache for three months. His headache started at the occipital area and radiated to the eye socket of both sides. He occasionally had nausea when the headache became more severe. He could sit upright for a short period then the headache started. He then needed to lie down. Later, his headache worsened when lying down. He often needed to shake his head to make him feel comfortable.

He had hypotrophy of the right arm since birth. He had taken an uncertain amount of ginkgo and four capsules of fish oil every day over the last three months due to the test result of hyperlipidemia.

His physical exams found a high blood pressure of 140/90 mmHg. Hypotrophy and hyperreflexia of his right upper extremity reflected intrauterine defect. His blood exam, complete blood count, and prothrombin time were normal. Inflammatory profiles; erythrocyte sedimentary rate of 20 mm/hour and high sensitivity C-reactive protein of 1.3 mg/L were within the normal range. Immunologic profile; positive antinuclear antibody titer of nucleolar pattern 1:80 was in the upper end of normal limit.

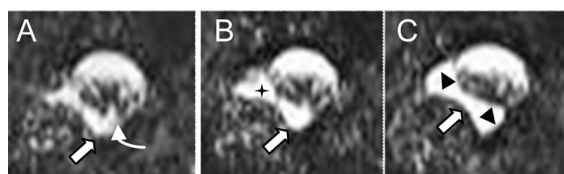
Cranial MRI with and without contrast revealed bilateral subdural collections and left-sided subdural hematoma. Evidence of downward displacement of the brainstem, diffusely thickened enhancing pachymeninges, and pituitary hyperemia were seen (Fig. 4). Whole spinal MRI showed a long segment of posterior epidural collection from C3 to T10 levels, anterior epidural collection from C5-6 to T3 levels and left-sided anterior epidural collection from T5 to T7 levels. There was a 3-mm-wide barely-perceptible dural sleeve line at right-sided T3 level, which was a possible site of leak (Fig. 5). Degenerative change of C5-6 level, causing narrowing of left C5-6 neural foramen. A spontaneous spinal CSF leakage was diagnosed together with intracranial evidences of SIH.

### Case 2



**Fig. 2** Heavily T2WI, coronal (A) and sagittal (B) sections demonstrated two broad-based meningeal diverticula through right T12-L1 and right L1-2 neural foramina

### Case 2



**Fig. 3** Heavily T2WI, axial planes, showed posterior and right-sided epidural fluid collections from mid T12 to L1-2 disc levels (arrows in A, B and C). Diverticular-like projection was noted through right T12-L1 neural foramen (star in B). Intact low-intense thin dural sleeve was seen, separating central thecal sac and the abnormal epidural fluid (arrow heads in C) except at a small portion posteriorly to T12-level (curved arrow in A), which was a possible site of leak

After counseling for EBP, he preferred bed rest with the abdominal binder. He was done for craniotomy and subdural collections removal at the university hospital. His symptom had gradually recovered after continuing bed rest for one month.

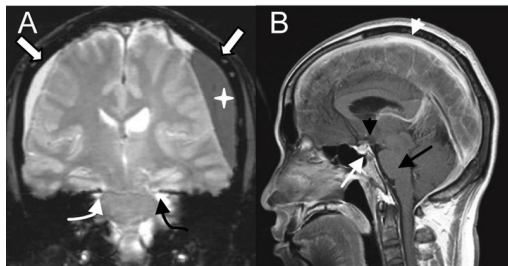
### Discussion

Orthostatic headache is the usual leading symptom of intracranial hypotension. Dural puncture or other penetrating spinal trauma is excluded in spontaneous spinal CSF leakage.

Patients complained of headache as “head pulling sensation from behind” and “headache radiated



### Case 3

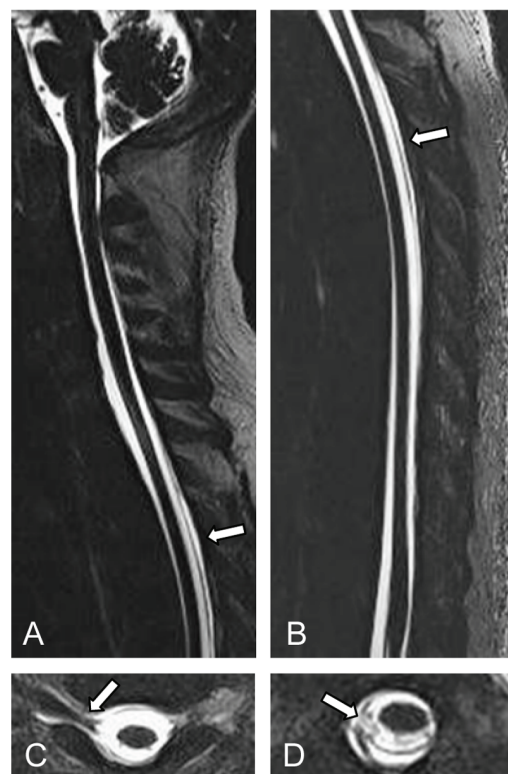


**Fig. 4** Coronal GRE T2-weighted image (A) demonstrated bilateral subdural fluid collection (arrows) and left-sided subdural hematoma (star), the blood product was seen as low intense part within left-sided collection. There was effacement of the perimesencephalic cistern, bilaterally, due to downward displacement of the midbrain (curved arrows). Sagittal SE T1-weighted image with gadolinium (B) demonstrated diffuse thickened enhancement of the pachymeninges (white arrow head), mild prominent size and prominent enhancement of the pituitary gland was visualized (white arrow). Sagging midbrain with pons against posterior clivus (black arrow) and optic chiasm (black arrow head) attaching against the clival tip showed declining course

to neck and shoulders". Those resembled symptom of neck stiffness<sup>(2,3)</sup>. In the first two cases, the symptoms were alleviated when lying down and using a pillow as abdominal binder. However, the third reported case got worse sometimes when lying down. In this case, cranial MRI revealed a subdural hematoma, which lying down did not compensate the symptom of intracranial hypotension. Actually, it would be increasing intracranial pressure. This was differentiated from the condition in previous report<sup>(4)</sup> of not getting better when lying down in spontaneous intracranial hypotension (SIH) complicated by venous thrombosis.

The initial onset of headache occurred gradually after trivial traumatic event<sup>(5)</sup> in the first two cases, practicing yoga (case 1) and pulling heavy things with back bending posture (case 2). However, the third case had taken the maximum doses of fish oil. However, shaking his head might be implicated in neck trauma. While different from spontaneous CSF leakage in connective tissue disorder cases<sup>(6-8)</sup> and spinal bony structural defect case<sup>(9)</sup>, those had repeated trivial traumatic episodes related to pre-existing dural weakness or spinal bony pathology and needed complicated specific treatments. Among the consecutive

### Case 3



**Fig. 5** Sagittal heavily T2WI, cervicothoracic level (A), thoracolumbar level (B) demonstrated a long segment of posterior epidural collection (arrows in A and B). Axial heavily T2WI at C7-T1 level (C) showed evidence of right C8 root piercing dura through the right-sided epidural CSF collection (arrow). There was also leakage of CSF along right C8 nerve root sleeve. Axial heavily T2WI at T3 level (D) showed a tiny dural defect (3-mm gap) at right anterolateral aspect, possible to be the site of leak (arrow)

cases of SIH and spontaneous spinal CSF leakage from previous report<sup>(10)</sup>, three cases (2%) were taking anticoagulant (warfarin) and had chronic subdural hematoma. Anticoagulant has been suspected to be related to non-traumatic chronic subdural hematoma. In addition, the relationship of clinical scenario among three factors, SIH with subdural hematoma, spontaneous spinal CSF leakage, and current use of anticoagulant are suspicious. Fish oil (case 3) could be a predisposing factor for spontaneous CSF leakage because of the fish oil antithrombotic effects<sup>(11)</sup>. This should be closely observed.

Considering the international classification of headache disorders<sup>(3)</sup>, some of which attributed to

spontaneous low CSF pressure, all three reported cases' presenting symptom met diagnostic criterion of gradual onset of headache after assuming the upright position. The first two cases had radiating symptoms to the neck and the third case had referring pain to periorbital areas and occasional nausea. The third case had presence of subdural collections, thickened enhancing meninges, and sagging of midbrain, which had met cranial MRI criterion (Table 1).

From previous studies, cranial MRI<sup>(12)</sup> could reveal the indirect evidences of intracranial hypotension, which are contributed by spinal CSF leakage, sagging of brain, subdural fluid collection, enhancement of the pachymeninges, pituitary hyperemia, and engorgement of venous structures. Subdural hematomas (case 3) with some degrees of mass effect have been described<sup>(13-16)</sup>. These may be caused by tearing of bridging veins or rupture of the dilated thin-walled blood vessels in the subdural zones.

The first two cases had normal cranial MRI and had been investigated for the evidence of spinal CSF leakage by MRM, which did not meet the criterion's detail. The criterion<sup>(3)</sup> included three modalities for demonstration of spinal CSF leakage, conventional myelography, and computed tomography myelography (CTM) or cisternography). All three reported cases in the present study (Table 1) demonstrated spinal CSF leakage by MRM, which was

proposed as newly developed criteria<sup>(2)</sup>. MRM is a reliable and well-established imaging finding. MRM also has been discussed.

CTM has been a choice of investigation to locate the exact site of CSF leak<sup>(2)</sup>. The procedure needs lumbar puncture for intrathecal injection of the iodinated contrast material and exposure to radiation. Delayed scan may be required in slow or intermittent leak and ultra early computed tomography scan with dynamic technique might be required in the rapid high flow leak<sup>(2,17)</sup>. Adversely, additional scans increase radiation exposure.

Recent studies have given more attention to spinal MR imaging, particularly heavily T2-weighted image, mainly because it is a non-invasive procedure that requires neither radiation nor iodinated contrast material injection. The previous studies found it promising for detecting and following-up of spinal CSF leakage<sup>(18,19)</sup>. Multiple spinal manifestations and indirect evidences of SIH have been described, including dilated epidural or intradural veins, dural enhancement, subdural fluid collection, meningeal diverticula, extrathecal CSF collection, and presence of retrospinal fluid collection at C1-C2 levels<sup>(20-25)</sup>. The degree of CSF leakage may vary from a small amount of CSF tracking along a single or multiple nerve roots, to extensive bilateral collections of CSF within the paraspinal soft tissue. Heavily T2-weighted image provides an excellent resolution between soft tissue

**Table 1.** Diagnostic criteria for intracranial hypotension, low CSF pressure and spontaneous spinal CSF leak correlate to this study's cases

Reference	Criteria	Case 1	Case 2	Case 3
3	Headache worsens within 15 minutes after sitting or standing	Occipital area	Occipital area	Occipital area and eye sockets
	With described accompanying symptoms	Neck stiffness	Neck stiffness	Nausea
	Evidence from cranial MR imaging	Not found	Not found	Present (Fig. 4)
	CSF opening pressure measuring	Not done	Not done	Not done
	Conventional myelography, CT myelography or cisternography	Not done	Not done	Not done
2	Demonstration of a spinal CSF leak (This cases report used MR myelography)	Epidural fluid collection (Fig. 1)	Epidural fluid collection (Fig. 3)	Epidural fluid collection (Fig. 5)
	Meningeal diverticulum	Not found	Two sites (Fig. 2A and 2B)	Not found
This cases report	The additional finding from MR myelography; site of CSF leak	Possible site of leak (Fig. 1C)	Possible site of leak (Fig. 3A)	Possible site of leak (Fig. 5D)

CSF = cerebrospinal fluid; MR = magnetic resonance; CT = computed tomography

and fluid, thus the sequence is sensitive in depiction of abnormal extrathecal CSF collection. Single or multiple meningeal diverticula may be demonstrated.

The abnormalities in the present study were not different from the abnormal extrathecal CSF collection found in previous reports<sup>(2,25)</sup>. Long segments of epidural CSF collections with CSF tracking along multi-leveled bilateral nerve root sleeves were observed in two cases (case 1 & 3). Short segments of epidural CSF collections were visualized in the other patient (case 2), who's epidural fluid minimally extending into adjacent two neural foramina, giving the appearance of meningeal diverticula. In the second case, a thin dural sleeve could be identified, separating between central thecal sac and the abnormal epidural fluid. This helped to confirm extrathecal location of the fluid collection (Fig. 3C).

A meningeal diverticulum may or may not be associated with CSF leakage. Nerve root sleeve dilatation or meningeal diverticula may represent only primary pathology of dural weakness<sup>(26)</sup>. The size of abnormality, although important and helpful, may not always or necessarily favor the larger lesion over the smaller one as the potential site of the leakage<sup>(27)</sup>. Evidence of abnormal extradural fluid at the site of diverticula on MRI or contrast leakage on CTM has to be seen to document the leakage.

MRM showed evidence of CSF leakage in static rather than dynamic information<sup>(25)</sup>. This was because such leak could be concealed in the background of readily present hyperintense CSF collection, which was not absorbed and tracked up or down below the exact level of dural defect or true site of leak.

Although MRM could not demonstrate the exact site of leak, it could be easily performed as a first line of investigation in the patients who clinically suspected SIH, prior to any invasive procedures. Moreover, treatment of SIH usually begins in a conservative way, even when CTM shows the leak. When the initial MRM shows extrathecal CSF collection, conservative treatment may be first given to the patient. If it fails, the patient has to go for targeted EBP or surgery, then the exact site of CSF leak has to be identified, and then more invasive procedure such as CTM<sup>(22)</sup> would be performed.

When short segments (1-2 vertebral levels) of abnormal CSF collection were found, a certain region of leak would be able to be localized, such as in the second case, targeted EBP might be further performed without other invasive investigations<sup>(21)</sup>. However, when long segments of abnormal CSF

collection were identified, untargeted EBP may be a choice of treatment, the injected blood at the lumbar level may spread cranially over several segments<sup>(28-30)</sup>, although rate of response is not high<sup>(25)</sup>. CTM would still be reconsidered when MRM failed to demonstrate any CSF leakage or when the actual location of CSF leak is needed to be identified either prior to surgery or targeted EBP. Myelography with intrathecal administration of gadolinium followed by MRI<sup>(31)</sup> would be an excellent alternative.

Heavily T2-weighted MRM<sup>(32-34)</sup> was used in reported cases in the present study, which could provide sufficient evidences for diagnosis. Neither lumbar puncture for measuring of opening CSF pressure nor intrathecal administration of gadolinium was done. Beneficially, the reported cases did not take the risk of post-dural puncture headache<sup>(3)</sup>.

After discussion of specific treatment, the reported cases preferred bed rest, denied untargeted EBP and were concerned of the risk of spinal cord or radicular compression from targeted EBP<sup>(35)</sup>. Fortunately, their symptoms gradually resolved after prolonged bed rest for one month. However, orthostatic headache often disappears over time if left untreated then chronic sequelae could be observed<sup>(15,36,37)</sup>.

From previous reports, following-up imaging of meningeal enhancement, it results in dilatation of the meningeal vasculature secondary to a reduced CSF volume<sup>(38,39)</sup>, this had resolution after treatment, when headache had markedly improved<sup>(8)</sup>.

## Conclusion

Orthostatic headache without a history of dural puncture should be investigated for evidence of SIH. Following the clinical criterion of headache is helpful in making the decision. Cranial MRI provides evidence of low CSF pressure. There are several techniques to detect spinal CSF leakage. The suitable one could be selected. Heavily T2-weighted MRM helped to confirm the diagnosis of spontaneous spinal CSF leakage among these reported cases. MRM might localize a certain region of the CSF leak. Furthermore, MRM could avoid some procedures such as spinal puncture or intrathecal CSF monitoring. In addition, this is a non-radiation exposure method, used beneficially during the reproductive ages. This technique could be used as the first line of investigation. MRM can be performed prior to CTM, guiding a radiologist to the most possible level of CSF leak during CTM study.

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### Potential conflicts of interest

None.

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**โรคปวดศีรษะเมื่อเปลี่ยนเป็นทำนองหรืออื่น เกิดจากน้ำหล่อไขสันหลังรั่วแบบเกิดขึ้นเอง ที่ระดับสันหลัง;  
วินิจฉัยโดยการตรวจไขสันหลังด้วยสนามแม่เหล็กไฟฟ้า *Heavily T2-weighted***

ชัยรัช เทียนวิบูลย์, ภัทรภรณ์ เซาวนะปัญญา

โรคปวดศีรษะเมื่อเปลี่ยนเป็นทำนองหรืออื่น วินิจฉัยได้จากประวัติ การตรวจหาความดันน้ำหล่อไขสันหลังต่ำ และหลักฐานจากการตรวจศีรษะด้วยสนามแม่เหล็กไฟฟ้า (MR) เอกซเรย์ไขสันหลังตามวิธีในข้อกำหนด (เอกซเรย์ไขสันหลังด้วยวิธีมาตรฐาน เอกซเรย์คอมพิวเตอร์ (CT) ไขสันหลัง หรือ เอกซเรย์ช่องน้ำไขสันหลัง) การศึกษาฉบับนี้รายงานผู้ป่วยที่มาด้วยอาการปวดศีรษะเมื่อเปลี่ยนเป็นทำนองหรืออื่น โดยไม่มีประวัติอุบัติเหตุชัดเจนที่สันหลังมาก่อน สองรายแรกมีปวดศีรษะร่วมกับอาการปวดร้าวไปต้นคอ รายที่สามปวดศีรษะร่วมกับอาการปวดร้าวไปกระบอกตาสองข้างหรือบางครั้งคลื่นไส้ รายที่สามพบหลักฐานการวินิจฉัยจากการตรวจศีรษะด้วยสนามแม่เหล็กไฟฟ้า ทั้งสามรายไม่ได้ตรวจวัดความดันเปิดน้ำหล่อไขสันหลัง หรือ เอกซเรย์ไขสันหลังตามวิธีในข้อกำหนด แต่ตรวจด้วยสนามแม่เหล็กไฟฟ้า *heavily T2-weighted* ทั้งสามรายพบการสะสมของน้ำเหนือเยื่อหุ้มไขสันหลัง รายที่สองพบถุงกระเปาะของเยื่อหุ้มไขสันหลัง การศึกษานี้ยังพบตำแหน่งที่น่าจะมีการรั่วของน้ำหล่อไขสันหลังในผู้ป่วยทั้งหมดในการศึกษานี้การตรวจไขสันหลังด้วยสนามแม่เหล็กไฟฟ้า *heavily T2-weighted* สามารถหลีกเลี่ยงการเจาะเยื่อหุ้มไขสันหลังและเป็นวิธีที่ไม่ได้รับรังสีจากการตรวจ วิธีนี้อาจใช้เป็นการตรวจขั้นแรก ก่อนที่จะตรวจเอกซเรย์คอมพิวเตอร์ไขสันหลัง แนะนำแนวทางแก่รังสีแพทย์หาตำแหน่งรั่วที่เป็นไปได้มากที่สุด ขณะตรวจเอกซเรย์คอมพิวเตอร์ไขสันหลัง

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