

## Case Report

# A case of spontaneous cerebrospinal fluid rhinorrhea: Accurate detection of the leak point by magnetic resonance cisternography

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## Abstract

**Background:** Spontaneous cerebrospinal fluid (CSF) rhinorrhea is a rare entity. The accurate preoperative localization of the leak point is essential for planning surgical treatment, but is sometimes difficult. To localize the leak point, magnetic resonance cisternography (MRC) is the method of choice, but its effectiveness remains unclear.

**Case Description:** A 34-year-old mildly obese female experienced spontaneous CSF rhinorrhea after an attack of bronchial asthma. High-resolution computed tomography (CT) failed to reveal the leak point, while MRC demonstrated an arachnoid herniation at the olfactory cleft. The patient underwent endoscopic endonasal repair of the CSF leak with success. There has been no recurrence of CSF rhinorrhea for 14 months after surgery followed by the administration of acetazolamide.

**Conclusion:** We report a rare case of spontaneous CSF rhinorrhea associated with benign intracranial hypertension, in which the leak point was successfully detected by MRC. The CSF leak was completely repaired by minimally invasive endoscopic endonasal surgery. MRC may be a reliable method for detecting CSF leak points.

**Key Words:** Benign intracranial hypertension, endoscopic endonasal surgery, leak point, magnetic resonance cisternography, spontaneous cerebrospinal fluid rhinorrhea

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## INTRODUCTION

The term spontaneous cerebrospinal fluid (CSF) rhinorrhea has been applied to describe the nasal discharge of CSF unrelated to trauma, surgery, malformation, or tumor.<sup>[12]</sup> It is related to elevated intracranial pressure (ICP) and it has been suggested that this high pressure might be the direct cause of

the leak.<sup>[3,16,18]</sup> Accurate preoperative localization of the CSF leak has become important since minimally invasive surgical techniques such as endoscopic endonasal surgery are now used for the treatment of CSF rhinorrhea. Unlike in the case of a standard transcranial repair, in endoscopic endonasal surgery, the leak point should be visualized during operation in order to achieve complete repair.

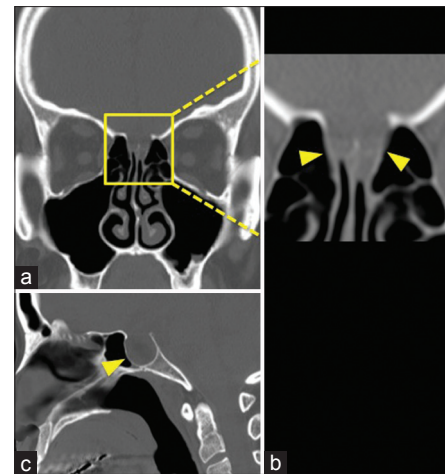
To localize the leak point, numerous techniques

have been used, including plain skull radiography, pluridirectional tomography, intraoperative injection of fluorescein dye, positive contrast studies, and radionuclide cisternography, however, these methods have some limitation to accurately detect leaks. In general, computed tomography (CT) cisternography has been considered the most reliable and accurate method of diagnosing CSF rhinorrhea, however, this technique is invasive and is contraindicated in patients with elevated ICP.<sup>[15]</sup> Since spontaneous CSF rhinorrhea usually accompanies elevated ICP, CT cisternography may be difficult to employ in this condition. In contrast, magnetic resonance cisternography (MRC) can be applied for any cases of CSF rhinorrhea. Although some previous researchers employed MRC to localize the leak point in CSF rhinorrhea, these reports consisted of heterogeneous patients, that is, traumatic, iatrogenic, or spontaneous CSF rhinorrhea,<sup>[2,8,12,13,15,16,18]</sup> and thus, evaluation of its usefulness for the treatment of spontaneous CSF rhinorrhea is difficult. To our knowledge, only three case reports state that preoperative MRC was able to identify a spontaneous CSF leak point when CT failed, however, in all of these cases, craniotomy was performed.<sup>[5,9,10]</sup> No reports, to our knowledge, directly showed the usefulness of MRC for identifying a leak point in spontaneous CSF rhinorrhea treated with endoscopic endonasal surgery.

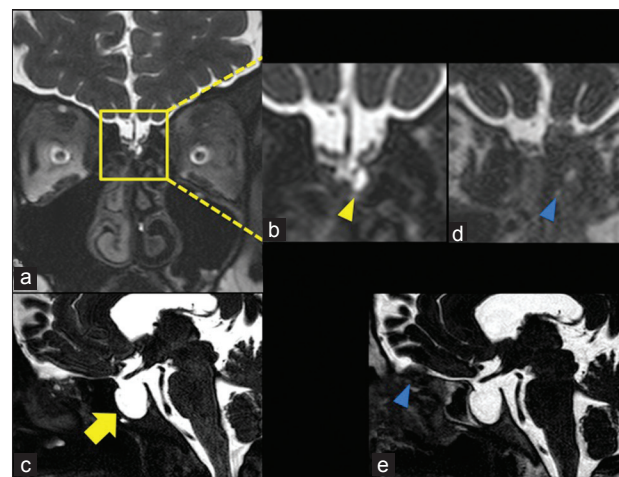
We report here on a patient with spontaneous CSF rhinorrhea associated with benign intracranial hypertension (BIH) and originating at the cribriform plate, in which the leak point was successfully detected by MRC, resulting in complete repair via minimally invasive endoscopic endonasal surgery.

## CASE REPORT

A 34-year-old female suddenly experienced marked rhinorrhea from the left nostril, with no history of trauma. She had been having intermittent headache. She had been coughing due to bronchial asthma for one month and was taking medication including steroid hormone inhalation. The diagnosis of CSF rhinorrhea was made based on clinical findings and a glucose-oxidase test. The patient's physical examination revealed mild obesity (BMI 34.1 kg/m<sup>2</sup>), but no signs of meningitis, visual disturbance, or olfactory dysfunction. High-resolution CT revealed enlargement of the sella turcica and broad thinning of the bony wall of both the sphenoid sinus and ethmoid sinus, including the cribriform plate [Figure 1]. Some of this thinning might have been attributed to bone defect, however, no apparent bony defect was found. Coronal T2-weighted 3D drive equilibrium magnetic resonance imaging (MRI; T2-3D DRIVE), a kind of MRC, revealed an extracranial extension of CSF space as a small pouch protruding into the left olfactory cleft [Figure 2a-c]. Arachnoid herniation was indicated by soft tissue



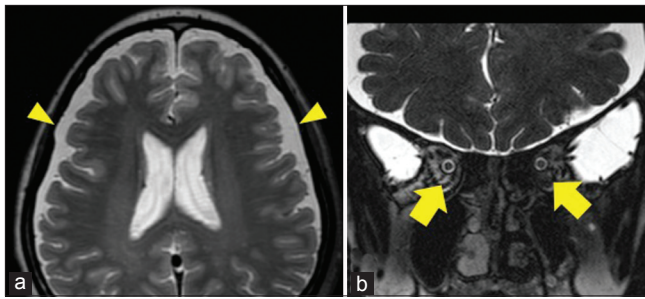
**Figure 1:** High-resolution computed tomography (CT) reveals enlargement of the sella turcica (c) and the broadly attenuated bony wall of both the sphenoid sinus (c, arrowhead) and ethmoid sinus (a,b, arrowheads). No apparent bony defect can be found.



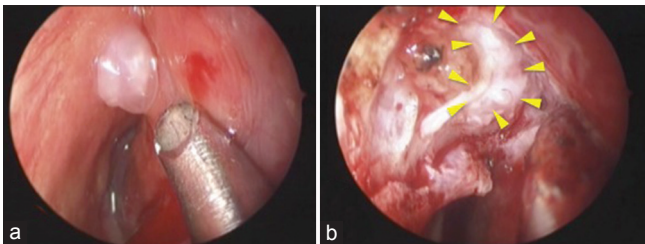
**Figure 2:** Magnetic resonance cisternography (MRC) reveals an extracranial extension of cerebrospinal fluid (CSF) space as a small pouch protruding into the left olfactory cleft (a,b). Arachnoid herniation is indicated by soft tissue isointense strands of tissue intermingled with CSF signal intensity probably through the osteodural defect at the olfactory cleft (b, arrowhead). Note that the empty sella is depicted (c, arrow). MRC taken one week after surgery reveals the arachnoid herniation diminishing with a solid mass (abdominal fat) (d,e, blue arrowhead)

isointense strands of tissue intermingled with CSF signal intensity probably through the osteodural defect at the olfactory cleft. In addition, enlargement of the subarachnoid space and dilated optic nerve sheaths and empty sella were depicted [Figures 2c and 3], suggesting a chronic state of increased ICP. There was no evidence of cerebral venous thrombosis, hyperviscosity syndrome, or chronic heart failure. Thus, the patient was diagnosed with spontaneous CSF rhinorrhea due to suspected BIH.<sup>[1,18]</sup>

The patient underwent endoscopic endonasal repair of the CSF leak via the left nostril. During the surgery, arachnoid



**Figure 3: Preoperative magnetic resonance imaging depicts enlargement of the subarachnoid space (a, arrowheads) and dilated optic nerve sheaths (b, arrow). This most likely indicates a chronic state of increased intracranial pressure**



**Figure 4: Intraoperative photography shows the arachnoid herniation at the olfactory cleft (a). After removing the mucosa surrounding the bony defect, the arrow heads indicate the edge of the osteodural defect (b)**

herniation was observed at the olfactory cleft, precisely as demonstrated by the preoperative MRC [Figure 4a]. After detecting the leak point, the mucosa surrounding the bony defect was thoroughly removed in a range of several square millimeters [Figure 4b]. The herniated arachnoid pouch was then ablated using bipolar cautery. The dura was gently elevated above the bony skull base defect, then abdominal fat tissue, which was slightly larger than the osteodural defect, was inserted in an underlay fashion in the epidural space. Finally, the osteodural defect was covered by a pedicled local mucoseptal flap in an overlay fashion.

After the surgery, lumbar drain management was carried out for one week. ICP monitoring revealed that the patient's mean ICP was 22-23 cmH<sub>2</sub>O. Furthermore, a sudden impulse of elevated ICP over 36 cmH<sub>2</sub>O (B wave) was found (BIH). Thus, acetazolamide was administered to decrease CSF production. At the time of writing, 14 months after surgery, there has been no recurrence of CSF rhinorrhea [Figure 2d and e].

## DISCUSSION

Over the past two decades, open transcranial approaches for the repair of CSF rhinorrhea have been replaced by a minimally invasive endoscopic endonasal approach. Improved cosmetics, olfaction preservation, and reduced morbidity are the main reasons for this preference for endoscopic repairs.<sup>[8]</sup> The success rate for traditional

transcranial repair is generally considered to be approximately 70-80%.<sup>[6]</sup> In contrast, a systematic review shows successful leak closure in 90.6% of endoscopic CSF leak repairs after the first attempt, increasing to 96.6% with the second attempt.<sup>[17]</sup> The overall complication rate is very low at only 0.03%.<sup>[17]</sup>

In the present patient, preoperative MRC precisely identified the location of the CSF leak, which made it possible to perform minimally invasive endoscopic surgery, avoiding a craniotomy and damage the olfactory nerve. If we had chosen conventional transcranial repair, the leak point would have been covered with a pericranial flap over the olfactory nerve and would therefore not be seen directly. The endoscopic endonasal approach made it possible to directly visualize the skull base defect and facilitated direct repair of the leak point.

Accurate localization of the CSF leak is essential for planning surgical treatment.<sup>[1,14]</sup> For more than a decade, contrast-enhanced CT cisternography has been considered the standard of reference for the diagnosis of a CSF leak. CT cisternography has a higher success rate than other techniques in locating CSF leaks, reaching 92% in active leaks and 40% in inactive leaks.<sup>[15]</sup> However, CT cisternography is invasive, time-consuming, and uncomfortable for the patient, and carries a slight risk for such complications as headache and infection. In addition, it is contraindicated with active meningitis or elevated ICP, latter was present in our patient.

Focusing the topic to *spontaneous* CSF rhinorrhea, CT is not sufficient compared with that following trauma especially in localizing CSF leak point, based on the following reasons. First, patients with a spontaneous CSF leak naturally lack skull fracture, which is usually present in traumatic patients and can be detected by high-resolution CT (accuracy 92%, sensitivity 92%, specificity 100%).<sup>[11,15]</sup> Second, the spontaneous group is associated with the highest rate (50-100%) of encephalocele formation, and there are often rather large meningoencephaloceles herniating through relatively small bony defects.<sup>[11]</sup> Initially, CT can provide only indirect evidence of CSF leaks such as bone defects, pneumocephalus, or a fluid level in the paranasal sinus.<sup>[4]</sup> Small bony defects with encephalocele/meningoencephaloceles may be mistaken on CT to be mere secretions of the paranasal sinuses or small bony defects perhaps not actively leaking CSF.<sup>[11]</sup> Third, high-resolution CT occasionally demonstrates thinning or dehiscence of the skull base, mimicking a small bone defect at a glance, but they are usually not the bone defect (false positive finding).<sup>[1,15]</sup> In contrast, MRC has some merits over CT: MRC (i) complements high-resolution CT in demonstrating the extracranial extension of the CSF column (accuracy 89%, sensitivity 87%, specificity 100%),<sup>[11,15]</sup> and MRC more accurately evaluates the leak point than CT, (ii) when there are multiple fractures of the skull base, or (iii) when there is



soft tissue in the paranasal sinuses; that is, when hematoma or fibrous tissue in the paranasal sinuses may preclude optimum evaluation of the correct sites of the CSF leak.<sup>[7,15]</sup> Thus, MRC should be added when CT cannot detect the precise leak point in spontaneous CSF rhinorrhea.

In contrast, the efficacy of MRC in localizing spontaneous CSF rhinorrhea compared with other types of CSF rhinorrhea has not yet been fully evaluated. Only one previous study showed the efficacy of MRC: Schuknecht *et al.* demonstrated spontaneous intermittent CSF rhinorrhea in 27 patients; MRC was correct in 93.3% of cases, CT cisternography in 62.5%, and CT in 50%.<sup>[12]</sup> In an earlier report on 45 patients with posttraumatic and spontaneous CSF rhinorrhea, the accuracy of MRC was 86% (CT 93%) but the authors (Shetty *et al.*) state that none of the patients with spontaneous CSF rhinorrhea had increased ICP,<sup>[15]</sup> and thus it is doubtful whether the patients really had spontaneous CSF rhinorrhea. Some previous researchers described patients with spontaneous CSF rhinorrhea,<sup>[13,16,18]</sup> however, they focused mainly on their clinical course and made no mention of radiological diagnostic modalities. Mirza *et al.* mention 97 CSF rhinorrhea patients including 29 with spontaneous rhinorrhea, almost all of whom were diagnosed with CT, though 4 needed MRC,<sup>[8]</sup> in which the authors did not mention the comparison between MRC and CT.

In conclusion, the present clinical course suggested that MRC was effective for revealing the precise CSF leak point in a patient with spontaneous CSF rhinorrhea and thereby making it possible to perform less invasive endoscopic endonasal surgery. We believe that significant number of patients may be unreported, in whom MRC can provide more information to identify the leak point than CT, and endoscopic surgery was available. Further studies are needed to determine the usefulness of MRC in localizing the leak point of spontaneous CSF rhinorrhea.

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