

# From Allergic Rhinitis to Spontaneous Cerebrospinal Fluid Leak: A rare Case Report of Sphenoid Sinus Defect

Alireza Mohebbi and Mohammad Aghajanpour\*

Received: 08 February 2018 / Received in revised form: 17 March 2018, Accepted: 18 March 2018, Published online: 19 March 2018  
© Biochemical Technology Society 2014-2018  
© Sevas Educational Society 2008

## Abstract

Cerebrospinal fluid leak in the patients without a history of trauma, surgery, malformation, tumor, or radiotherapy has been known as spontaneous cerebrospinal fluid leak. The present report introduced a patient that was hospitalized due to a three-month rhinorrhea of cerebrospinal fluid. The patient was an elderly 65-year-old woman with obesity who had been referred to the clinic since three months ago because of intermittent nasal rhinorrhea. The patient did not have any history of trauma in the head or hypertension. The patient was initially treated due to allergic rhinitis diagnosis, however, no improvement was observed. The patient then was referred to the neurological clinic for headache, and antibiotic treatment was started for her due to sinusitis diagnosis, but as the headache was not resolved, she received various treatments in the later stages of referrals due to other diagnoses. During the referral, the patient was also complaining of headache besides rhinorrhea. The examinations at the time of referral were normal. The patient did not mention the history of any certain diseases, and did not consume any special medications before the onset of symptoms. A beta-2 transferrin test was requested for the patient, which was positive. The patient also underwent CT scan of paranasal sinuses, according to it, in the coronal and sagittal views, a defect was observed in the base of skull and in the sphenoid sinus region. The sella turcica was full of air, and there was an evidence of pneumocephalus. The patient eventually underwent surgery. Currently, according to the follow-ups, the patient had a good general health and her headache and rhinorrhea were resolved. Finally, it should be mentioned that the treatment of cerebrospinal fluid leak was associated with significant surgical complications. The importance of this matter has been much more because of specific anatomical structure as well as various variations of the sphenoid sinus.

---

### Alireza Mohebbi

Rhinology Director, Associate Professor, ENT and Head and Neck Research center Hazrat Rasoul Hospital, The Five Senses Institute, Iran University of Medical Sciences, Tehran, Iran.

### Mohammad Aghajanpour\*

Otorhinolaryngologist, Ghods Hospital, Arak, Iran.

\*Email: Aghajanpour560@yahoo.com

**Keywords:** Spontaneous Cerebrospinal Fluid Rhinorrhea, Sphenoid Sinus, Surgery.

## Introduction

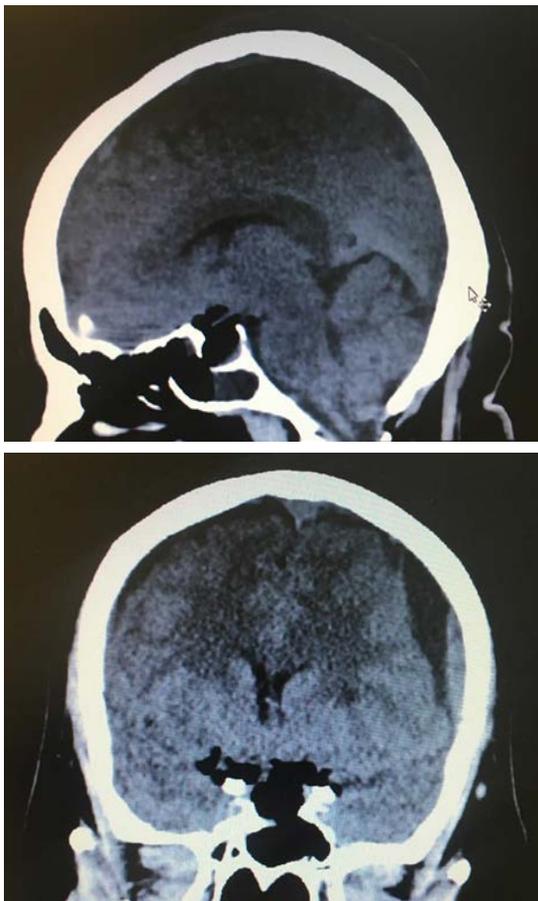
Cerebrospinal fluid leak in the patients without a history of trauma, surgery, malformation, tumor, or radiotherapy has been known as spontaneous cerebrospinal fluid leak (Ommaya et al., 1968; Schuknecht et al., 2008). This disorder is rare and its complications (e.g. meningitis, brain abscesses or pneumocephalus) can be fatal (Chaaban et al., 2014). Cerebrospinal fluid leak is the result of an abnormal connection between the space inferior to the arachnoid mater and a pneumatized area at the base of skull, which includes the sinonasal tract. This connection or fistule should involve the end of arachnoid mater and dura mater, the bone of the base of skull and the underlying mucus (Zapalac, Marple and Schwade, 2002). Galen was the first person who introduced a patient with the intermittent secretion of the cerebrospinal fluid from the nose in about 200 BC; it was initially thought to be a physiological issue, but in 1989, another person called Thomson introduced 21 patients with spontaneous rhinorrhea of cerebrospinal fluid and referred to this as an abnormal phenomenon (Ommaya et al., 1968; Schuknecht et al., 2008; St Clair, 1899).

Subsequently, various researchers introduced patients with spontaneous rhinorrhea of cerebrospinal fluid in the form of case reports. The present report has also introduced a patient that was hospitalized due to a three-month rhinorrhea of cerebrospinal fluid.

### *Patient introduction*

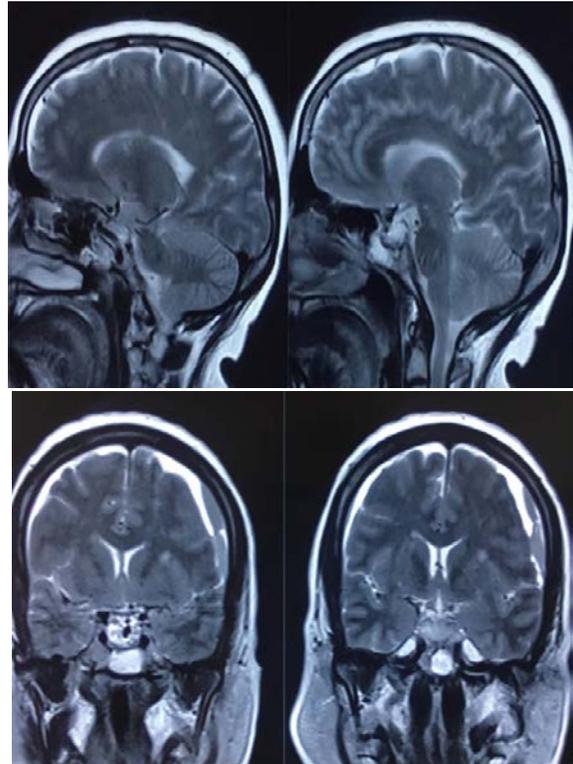
The patient was an elderly 65-year-old woman with obesity (a weight of 90 kg and a height of 163 cm) who had been referred to an Ear, Nose, and Throat (ENT) clinic since three months ago because of intermittent nasal rhinorrhea. The patient did not have any history of trauma to the head or hypertension, but her blood pressure was 140/90 mmHg at the time of referral. The onset of symptoms was a flu-like syndrome, and consequently the patient experienced bilateral intermittent rhinorrhea. The patient was initially treated with corticosteroid (fluticasone nasal spray) and

antihistamine (cetirizine) due to an allergic rhinitis diagnosis, however, no improvement was observed. The patient then was referred to the neurological clinic for headache, and antibiotic treatment was started for her due to sinusitis diagnosis, but as the headache was not resolved, she received various treatments in the later stages of referrals due to other diagnoses. During the referral, the patient was also complaining of headache besides rhinorrhea. The examinations at the time of referral included body temperature (37.1 ° C), heart beats (84 beats per minute), respiratory rate (20 breaths per minute), as well as a complete neurological assessment; and the results of all examinations were normal. The patient did not mention the history of any certain diseases, and did not consume any special medications before the onset of symptoms. During the referral, and when the patient was placed in the leaning forward position, a drop by drop clear unilateral secretion was observed. A beta-2 transferrin test was requested for the patient, which was positive. The patient also underwent CT scan of paranasal sinuses, according to it, in the coronal and sagittal views, the defect was observed in the base of skull and in the sphenoid sinus region (Fig. 1). The sella turcica was full of air, and there was an evidence of pneumocephalus.



**Figure 1.** Defect in the base of skull and sphenoid sinus in the patient's CT scan.

The patient also underwent MRI, according to which the sella turcica was full of air, and bilateral subdural effusions were evident (Fig. 2).



**Figure 2.** Patient's MRI indicating air accumulation in sella turcica and bilateral subdural effusion.

The patient eventually underwent surgery, and the largeness of sella turcica was observable during the surgery. The cerebrospinal fluid leak was high flow and in the posterior inferior part of the sphenoid, and also in the middle line. To repair this defect, the fat and harvested fascia from the anterior lateral surface of the thigh of the patient, the mucosal flap of the nasal septum was utilized with modified rescue technique; moreover, gel foam and biosynthetic glue were used. After repairing the location of cerebrospinal fluid leak (due to subdural effusion), the patient was subjected to midline shifts, and thus was operated by a neurosurgeon. Currently, according to the follow-ups, the patient had a good general health, and her headache and rhinorrhea were resolved.

## Discussion and Conclusion

As previously mentioned, cerebrospinal fluid leak in the patients without a history of trauma or surgery has been known as spontaneous or non-traumatic cerebrospinal fluid leak (Ommaya et al., 1968; Schuknecht et al., 2008). In about 6% to 23% of all cases, this disorder included cerebrospinal fluid leak (Schuknecht et al., 2008; Crozier et al., 2004; Ahmad et al., 2008). All types of cerebrospinal fluid leaks have had a common pathophysiology: disorder in arachnoid mater, dura mater and bone structure, as

well as cerebrospinal fluid pressure that increases continuously or intermittently, and leads to the leak of cerebrospinal fluid through the damaged structure (Ahmad et al., 2008; Ntsambi-Eba, Fomekong and Raftopoulos, 2014). There have been many indications mentioning that the major cause of most cases of spontaneous cerebrospinal fluid leak has been the elevated intracranial pressure for reasons such as idiopathic intracranial hypertension (also known as pseudotumor cerebri) (Chaaban et al., 2014; Ntsambi-Eba, Fomekong and Raftopoulos, 2014; Wang, Vandergrift and Schlosser, 2011). The patient introduced in this report was also an elderly and obese woman who was multipara, and despite she did not have a history of hypertension, her blood pressure was 140/90 mmHg at the time of referral.

Patients with a rhinorrhea caused by a cerebrospinal fluid leak who do not have a history of surgery or trauma are usually obese, with hypertension, multiparous women in the four or five decades of life, and the ratio of female to male is 2 to 1 (Kortbus, Roland Jr and Lebowitz, 2003). Clinical manifestations of this disorder are usually rhinorrhea and in some cases otorrhea (Ntsambi-Eba, Fomekong and Raftopoulos, 2014). Patients often refer with tension headaches and tinnitus (Wang, Vandergrift and Schlosser, 2011). The patient introduced in this report also referred while complaining about a three-month intermittent rhinorrhea and headache.

Medical imaging is preferred to biochemical tests for diagnosing the cause of rhinorrhea of cerebrospinal fluid, which is important in locating the cerebrospinal fluid leak and also in selecting the surgical procedure. Imaging methods in patients with cerebrospinal fluid rhinorrhea include CT scan of sinuses, CT cisternography, MRI scan of sinuses, and MRI water imaging (Reddy and Baugnon, 2017). According to studies, CT scan is able to accurately detect the location of bone defects leading to cerebrospinal fluid leak (Tumturk et al., 2016). CT cisternography has better features as compared with other types of CT scans, and is able to directly detect the morphology of the location of the leak, its size and location, as well as the amount of cerebrospinal fluid that is secreted from the location of leak; the flaw of this diagnostic method is that it cannot clearly show bone structure. Therefore, based on the results of various studies, it is suggested that CT cisternography to be used along with CT scan in order to improve diagnostic accuracy. Spiral CT scan and imaging accompanied with 3D simulation can provide a three dimensional view of the structure of the base of skull, as well as the place of existing bone defect and its surrounding structures to facilitate the process of surgery (Connor, 2010). If the above diagnostic methods fail, radionuclide cisternography can be helpfully used (Schaller, 2005). The introduced patient also underwent CT scan of paranasal sinuses and MRI and these imaging methods were helpful in locating the cerebrospinal fluid leak, and no other diagnostic methods were needed.

Among the various types of disorders leading to cerebrospinal fluid leak, patients suffering from cerebrospinal fluid leak due to sphenoid disorder expose doctors to specific challenges; this is due to the specific anatomical structure as well as various variations of the sphenoid sinus (Muscatello et al., 2010). In the introduced patient, the cerebrospinal fluid leak was high flow and

located in the posterior inferior part of the sphenoid and also in the middle line,

The treatment of cerebrospinal fluid leak is associated with significant surgical complications. In patients who do not recover during 3 to 4 weeks in spite of maintenance therapy, surgical repair is performed. Published reports on the therapeutic procedures of these patients suggested that patients who do not recover during 7 to 14 weeks after receiving treatment should undergo surgical repair as soon as possible. The first endoscopic repair of the location of cerebrospinal fluid leak was performed in 1981 by a physician called Wigand (1981). Many authors believed that the endoscopic approach is a good approach to treat rhinorrhea of cerebrospinal fluid, because this therapeutic approach is followed by low complications (Tumturk et al., 2016). In recent years, an endoscopic extracranial approach has been developed rapidly to repair rhinorrhea of cerebrospinal fluid, which would possibly take the place of traditional repair methods in the coming years (Ibrahim, Okasha and Elwany, 2016). In the present study, the patient suffered from subdural effusion after the surgery, and thus underwent another surgical procedure. After that, the patient had a good general condition.

The repair of the location of the cerebrospinal fluid leak is often done using autologous materials such as cartilage, bone, mucus of nasal septum, fascia, abdominal fat or cartilage of the ear. Wigand was the first to use the free tissue transplant to treat the rhinorrhea of cerebrospinal fluid using endoscopic method (Gjuric et al., 1996). In this patient, the fat and harvested fascia from the anterior lateral surface of the thigh of the patient, the mucosal flap of the nasal septum, as well as gel foam and biosynthetic glue were used.

## References

- Ahmad FU, Sharma BS, Garg A, Chandra PS. Primary spontaneous CSF rhinorrhea through the clivus: possible etiopathology. *J Clin Neurosci*. 2008; 15 (11): 1304–8.
- Chaaban MR, Illing E, Riley KO, Woodworth BA. Spontaneous cerebrospinal fluid leak repair: a five-year prospective evaluation. *Laryngoscope*. 2014; 124(1): 70–5.
- Connor SE. Imaging of skull-base cephaloceles and cerebrospinal fluid leaks. *Clin Radiol* 2010; 65 (10): 832–41.
- Crozier F, Goubert JL, Gobert P, Bohé D, Martin N, Roumieu G, Arwidson I. About a case of spontaneous breccia of the posterior wall of the sphenoidal sinus. *Annals of Otolaryngology and Cervico-facial Surgery*. 2004; 121 (5): 306–8.
- Gjuric M, Goede U, Keimer H, et al. Endonasal endoscopic closure of cerebrospinal fluid fistulas at the anterior cranial base. *Ann Otol Rhinol Laryngol* 1996; 105 (8): 620–3.
- Ibrahim AA, Okasha M, Elwany S. Endoscopic endonasal multilayer repair of traumatic CSF rhinorrhea. *Eur Arch Otorhinolaryngol* 2016; 273 (4): 921–6.
- Kortbus MJ, Roland Jr JT, Lebowitz RA. Sphenoid sinus cerebrospinal fluid leak: Diagnosis and management.

- Oper Tech Otolaryngol Head Neck Surg. 2003; 14 (3): 207–11.
- Muscattello L, Lenzi R, Dallan I, Seccia V, Marchetti M, Sellari-Franceschini S. Endoscopic transnasal management of cerebrospinal fluid leaks of the sphenoid sinus. *J Cranio-Maxillofac Surg.* 2010; 38 (5): 396–402.
- Ntsambi-Eba G, Fomekong E, Raftopoulos C. Spontaneous submucosal sphenoidal fistula discovered intraoperatively. A case report. *Neurochirurgie.* 2014; 60 (5): 262–4.
- Ommaya AK, Di Chiro G, Baldwin M, Pennybacker JB. Non traumatic cerebrospinal fluid rhinorrhoea. *J Neurol Neurosurg Psychiatry* 1968; 31 (3): 214-25.
- Reddy M, Baugnon K. Imaging of Cerebrospinal Fluid Rhinorrhea and Otorrhea. *Radiol Clin North Am* 2017; 55 (1): 167–87.
- Schaller B. Subcranial approach in the surgical treatment of anterior skull base trauma. *Acta Neurochir (Wien)* 2005; 147 (4): 355–66.
- Schuknecht B, Simmen D, Briner HR, Holzmann D. Nontraumatic skull base defects with spontaneous CSF rhinorrhea and arachnoid herniation: imaging findings and correlation with endoscopic sinus surgery in 27 patients. *Am J Neuroradiol* 2008; 29 (3): 542-9.
- St Clair T. *The cerebrospinal fluid: its spontaneous escape from the nose.* London: Cassel & Co; 1899.
- Tumturk A, Ulutabanca H, Gokoglu A, et al. The results of the surgical treatment of spontaneous rhinorrhea via craniotomy and the contribution of CT cisternography to the detection of exact leakage side of CSF. *Turk Neurosurg* 2016; 26 (5): 699–703.
- Wang EW, Vandergrift WA, Schlosser RJ. Spontaneous CSF leaks. *Otolaryngol Clin N Am.* 2011; 44 (4): 845–56.
- Wigand ME. Transnasal ethmoidectomy under endoscopic control. *Rhinology* 1981;19:7-15.
- Zapalac JS, Marple BF, Schwade ND (2002) Skull base cerebrospinal fluid fistulas: comprehensive diagnostic algorithm. *Otolaryngol Head Neck Surg* 126 (6):669–676.