

Spontaneous Cerebrospinal fluid Rhinorrhea: An enigma

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Abstract:

Aims: Evaluation of an association between Spontaneous Cerebrospinal Rhinorrhea and obesity

Settings and Design: Study was conducted in the Departments of ENT and Neurosurgery, ASTER MIMS Hospital, Kozhikode from 2010 to 2013. The study design was a prospective observational study.

Methods and Material: Adult patients (age >18 years) admitted for the management for spontaneous CSF rhinorrhea from 2010 to 2013 were studied. A total of 11 adult patients presented were studied. Body Mass Index, blood cholesterol levels were documented at presentation. Imaging confirmation of site of the leak was done by CT Cisternography.

Of the eleven patients, three underwent endoscopic surgical repair, whereas eight patients refused surgical intervention. Follow up regarding recurrence of CSF rhinorrhea, and symptoms of idiopathic intracranial hypertension (IIH) was obtained.

Statistical analysis used: No analytical tools used

Results: Majority of patients (seven) had a high body mass index (25-29.99) The lipid profile was deranged in most patients. Patients did not present with symptoms of IIH at any point of diagnosis or follow-up.

Conclusions: The study reports a possible correlation between Spontaneous CSF rhinorrhea and Idiopathic Intracranial Hypertension. A relook into the necessity of operative intervention in all cases of spontaneous CSF rhinorrhea is also needed.

Key-words: Spontaneous Cerebrospinal fluid Rhinorrhea; BMI

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I. Introduction

Spontaneous Cerebrospinal fluid (CSF) rhinorrhea is an enigmatic disease. The accurate diagnosis and treatment of the disease is a formidable challenge for any physician, and a high index of suspicion is necessary to detect these cases. Meningitis^[1], a possible sequela of the disease, can result in significant morbidity and even death. An association between intracranial hypertension and CSF rhinorrhea has been noted in the literature. However, the vast majority of patients with raised intracranial hypertension do not present with CSF rhinorrhea at any point of the primary disease process. An exception to this rule is the association noted between idiopathic intracranial hypertension and spontaneous CSF rhinorrhea^[2]. Both of these diseases are known to have subclinical presentations. An increase in Body Mass Index has been documented to be associated with Idiopathic Intracranial Hypertension. The present study was done to find out whether an association existed between spontaneous CSF rhinorrhea and obesity.^[3] The incidence of Spontaneous CSF rhinorrhea is rare, with most reported series consisting of only a few numbers of cases. Occasional articles point out the co-existence of Idiopathic Intracranial Hypertension. There are also reports of relief of IIH symptoms when CSF rhinorrhea occurs.^[4] The behaviour of IIH has been varied with patients of varying symptoms, and symptom duration reported. We present our findings from a long term follow-up of patients who suffered from spontaneous CSF rhinorrhea.

II. Material and Methods

Adult patients (age >18 years) admitted for the management for spontaneous CSF rhinorrhea were studied. Over a period ranging from 2010 to 2013, a total of 11 adult patients presented with spontaneous unilateral CSF rhinorrhea. There was no history suggestive of meningitis at any point in all of the 11 patients. None had a history of trauma or neurosurgical interventions. Six patients had right-sided nasal discharge, and

five had left-sided nasal discharge at presentation. Of these, eight patients were female and three male with ages ranging from 33 to 65 years with a mean age of 46.7 years. Their nasal discharge in all patients was collected and sent for biochemical analysis. The biochemical findings were suggestive of CSF with high sugar and protein values. Four patients had a history of hypertension, and two had a history of diabetes mellitus, another had a history of hypothyroidism. None of the patients had features of intracranial hypertension or visual loss. There was no evidence of papilledema in any of the patients. Body Mass Index was documented in all patients at presentation. Patients also underwent evaluation with blood cholesterol levels. Detailed imaging was done in all patients to document anatomical defects causing the rhinorrhea. CT Cisternography done in all patients showed defects in the anterior cranial fossa (ACF) base corresponding to the side of rhinorrhea in all cases. CT evaluation of 9 patients showed a defect in the cribriform plate corresponding to the side of the leak. Two patients showed a defect in the roof of the posterior and middle ethmoid. MRI cisternography of one patient showed a small herniation of right medial frontal lobe across Cribriform plate defect to the nasal cavity. Three patients who also underwent Magnetic Resonance Imaging (MRI) showed empty sella as an additional finding. The fasting lipid profile also showed a high level of triglyceride, total cholesterol with lower levels of HDL in most of the patients. Four had a total cholesterol level less than 200 and seven had levels more than 200. Ten of the patients had a triglyceride level of less than 150, and only one had levels more than 150. Ten patients had high-density lipoprotein values more than 35, and only one had values less than 35. Low-density lipoprotein values were less than 129 for six patients and five had values more than 129. However, none of the values could reach statistical significance. Majority of patients (seven) had a high body mass index (25-29.99) One had very high value (>30). Two had a BMI of less than 19. One had values between 19 and 24.99

All patients were counselled on the risk of meningitis and were advised surgical repair. However, eight patients were managed conservatively as the patients were not willing for surgical intervention. These patients were treated with oral Acetazolamide. Three patients underwent endoscopic repair of the defect. Follow up regarding CSF rhinorrhea and treatment for headache or vision loss could be obtained in all of the patients by telephonic consultation. No patient had a recurrence of CSF rhinorrhea. There were no reported symptoms suggestive of Idiopathic Intracranial Hypertension during the intervening period.

III. Discussion

CSF rhinorrhea is caused due to defects in the skull base resulting in abnormal communication of the subarachnoid space with the paranasal sinuses.^{[5][6]} This communication or fistula must involve a breach of the arachnoid and dura mater, bone of skull base, and the underlying mucosa. Spontaneous CSF otorrhea was first described in 1897 by a French physician in a 10-year old girl who had had intermittent otorrhea for eighteen months and a fistulous opening in the roof of the inner third of her ear canal. Omayya's classification^[7] is the most widely used, which divides the causes into congenital and acquired; which is further divided into traumatic^[8] and nontraumatic. The traumatic cases may be either acute (<48 hours) or delayed (within three months). The non-traumatic group is divided into high and normal intracranial pressure groups. 80% of all cases of CSF rhinorrhoea occur secondary to head trauma; 16% are postoperative, secondary to operations in nasal/ paranasal cavities and skull base.^[9] About 3-4% are considered non-traumatic/ spontaneous. Spontaneous CSF rhinorrhea presents with or without nasal discharge associated with attacks of meningitis. Early recognition of spontaneous CSF rhinorrhea is critical to institute treatment. A high index of suspicion is required while evaluating patients suffering from repeated attacks of meningitis. The defects within the skull bone include defects within the ethmoid, sphenoid sinus.^[10], sella^[11], and clivus rarely.^[12] Dandy first described an intracranial repair of a CSF leak in 1926, and Dohlman in 1948 was the first to describe the extracranial repair, which he did via a naso-orbital incision for an anterior cranial fossa CSF leak. Hirsch and later Vrabec described trans-nasal approaches^[13], and Wigand in 1981 was the first to describe the use of endoscopes in the repair of small leaks incurred during ethmoidectomy.^[14] The last twenty years have seen a refinement in the techniques and materials used for endoscopic repair of CSF leaks^[15] and encephaloceles.^[16]

No significant laterality was noted in the patients reported in the study. Endoscopic repair done in three patients was successful with no recurrence noted on follow up. Most of the female patients were obese, with a body mass index >25 kg/m². We reviewed the postulated etiologic factors of this uncommon condition and studied the pathophysiologic basis for obese patients to have CSF rhinorrhea. This study suggests a relationship between obese women and the risk of developing primary spontaneous CSF rhinorrhea. Two additional interesting facts also have been noted in this study. First is the presence of empty sella, high body mass index in these patients is an interesting finding. Both these features of an empty sella and high BMI are factors associated with Idiopathic Intracranial hypertension. A statistical correlate of the same could not be ascertained given the observational nature of the study and the number of patients studied. Though no other aetiological factors of IIH could be noted in our series, the association of these two independent factors with spontaneous CSF rhinorrhea is of importance. A detailed evaluation of the association would need a long term study with more significant patient numbers. The second factor is the spontaneous resolution seen in the majority of the patients. The

patients who refused surgery had minimal symptoms at presentation. The grounds of refusal of surgery in all these patients was because of the cost of surgical intervention. However, during the follow-up period, these patients have reported resolution of rhinorrhea following treatment with Acetazolamide. Also, none of these patients suffered from meningitis at any point during presentation nor follow-up.

IV. Conclusion

The study reports a possible correlation between Spontaneous CSF rhinorrhea and Idiopathic Intracranial Hypertension. The association between a high BMI and spontaneous CSF rhinorrhea and empty sella would need a detailed prospective study. Similar to cardiovascular diseases which are associated with obesity, the association between Spontaneous CSF rhinorrhea and obesity also needs to be highlighted. A detailed study also would be necessary to define the role of surgical intervention in patients presenting with minimal symptoms. The option of medical management though associated with risks would be a favourable treatment option in selected cases.

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