

Case Report

Idiopathic Intracranial Hypertension With Transcribriform Plate Intranasal Meningocele Without Cerebrospinal Fluid Rhinorrhea: A Case Report

Khalid Mohamed Abdalla*, Awajimijan Nathaniel Mbaba, and Hamza Mustapha Ahmed

Department of Radiology, King Abdullah Hospital, Bisha, Saudi Arabia **ORCID:**

Khalid Mohamed Abdalla: http://orcid.org/0009-0004-1707-8522 Awajimijan Nathaniel Mbaba: https://orcid.org/0000-0001-6167-869X

Abstract

Idiopathic intracranial hypertension (IIH) is a rare disorder of unclear pathogenesis occurring commonly in obese women of childbearing age and characterized by increased intracranial pressure in the absence of intracranial space-occupying lesion or cerebrospinal fluid outflow obstruction. We report a rare case of a middle-aged obese female with intractable headache referred to the department of radiology for neuroimaging on account of suspected dural sinus thrombosis and brain Magnetic Resonance Imaging (MRI) which revealed signs of IIH with asymptomatic transcribriform plate intranasal meningocele with a view to underscore the quiddity of IIH in middle-aged obese females and also emphasize the need for radiologists to diligently review previous patient's images where available for possible finding that could contribute to a suspected diagnosis.

Keywords: headache, idiopathic intracranial hypertension, intranasal meningocele

Corresponding Author: Khalid Mohamed Abdalla; email: dr.khalidmabdalla@gmail.com

Received 11 November 2022
Accepted 15 July 2023
Published 29 December 2023

Production and Hosting by Knowledge E

Khalid Mohamed Abdalla et al. This article is distributed under the terms of the Creative Commons Attribution License,

which permits unrestricted use and redistribution provided that the original author and source are credited.

Editor-in-Chief:
Prof. Nazik Elmalaika Obaid
Seid Ahmed Husain, MD, M.Sc,
MHPE, PhD.

1. Introduction

Idiopathic intracranial hypertension (IIH), also known as benign intracranial hypertension and psudotumor cerebri, is a disorder of poorly understood pathogenesis [1]. This entity was first described by Quincke in 1893, and in 1937 Dandy further expounded it as a syndrome characterized by elevated intracranial pressure in the absence of intracranial space-occupying lesion or cerebrospinal fluid (CSF) outflow obstruction[2]. The incidence of IIH has been put at approximately 1–3 per 100,000 adults [3, 4] and is said to be on the rise with increase in the rate of obesity [2, 5]. Its manifestation is usually with headache of variable phenotype, vomiting, pulse-synchronous tinnitus, diplopia, and eye pain in association with elevated intracranial pressure(ICP) [6], predominantly

□ OPEN ACCESS

affecting middle-aged obese females [7]. IIH is a rare neurological disorder in children and presentation is not different from that of adults [8].

Spontaneous meningocele with associated cerebrospinal fluid (CSF) leak have been observed in patient with IIH. Although the pathophysiology is still debated, it is believed to evolve from progressive erosion of the thin bone of the skull base by persistent increased intracranial pressure[9]. Skull base meningocele is herniation of the meninges through points of weakness or defects in the bone of the skull base. Meningocele are commonly asymptomatic but depending on location and persistent increased ICP can rupture and produce leak. Similar complications include herniation of brain tissue only (encephalocele) or herniation of brain tissue and overlying meninges (meningoencephalocele) [10]. Perez et al. [11], reported a case of IIH in an overweight female who developed rhinorrhea, and brain imaging revealed a right cribriform plate encephalocele which was responsible for the CSF leak from a ruptured encephalocele due to formation of a defect in the cribriform plate following elevated ICP for three years.

Neuroimaging, computed tomography (CT), and magnetic resonance imaging (MRI) of the brain and skull base are required for the diagnosis and detection of associated complications, such as skull base defect which predisposes to formation of meningocele. In patient with suspected IIH, CT and MRI are also necessary to exclude signs of elevated ICP which may be due to other causes such as brain tumor, dural sinus thrombosis, and hydrocephalus. We report a rare case of a middle-aged female with intractable headache referred to the department of radiology for neuroimaging on account of suspected dural sinus thrombosis but was later diagnosed as IIH with asymptomatic transcribriform plate intranasal meningocele with a view to underscore the quiddity of IIH in females with weight in the obese range as well as emphasize the need for radiologists to diligently review previous patient's images where available for possible finding that could contribute to a suspected diagnosis.

2. Case Presentation

A 42-year old obese female was referred to the department of radiology for neuroimaging following intractable headache. The referring doctor had a concern about possible dural sinus thrombosis. There was no history of rhinorrhea, trauma, or previous cranial surgery.

MRI of the brain without contrast was done using standard protocols. The scan revealed bilateral dilatation of the perioptic CSF space, partial empty sella, and bilateral stenosis of the lateral part of the transverse sinuses as well as prominent gyrus rectus on

the side of the meningocele. A right-sided transcribriform plate intranasal meningocele measuring 12.1×7.2 mm was also detected (Figures 1–3).

On review of the patient's previous brain CT images, which were done a week earlier based on the complaint, a defect was revealed in the right aspect of the cribriform plate, which serves as the lead point for the intranasal meningocele (Figure 4). These findings are consistent with IIH with transcribriform plate intranasal meningocele. Following this diagnosis, patient was placed on appropriate management including weight loss and she responded well to the treatment regimen. Nonetheless, patient was referred to a higher center for further treatment and possible closure of the cribriform plate defect.



Figure 1: Axial T2W MRI showing bilateral dilated perioptic CSF space (arrow).

3. Discussion

IIH is a rare disorder of unclear pathogenesis occurring commonly in obese women of childbearing age and characterized by increased intracranial hypertension in the absence of intracranial space-occupying lesion or cerebrospinal fluid outflow obstruction[1, 12]. Several mechanism have been proposed to explain the pathophysiology of IIH and popular among them include an increase in production of CSF, a higher



Figure 2: Sagittal T2W MRI showing partial empty sella (arrow).

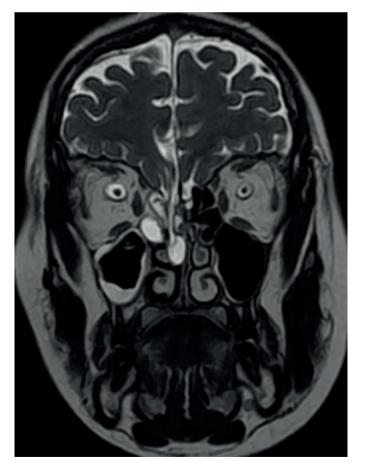


Figure 3: Coronal T2W MRI showing right transcribriform plate intranasal meningocele.

levels of cerebral blood flow resulting in increased fluid content, and a restriction of

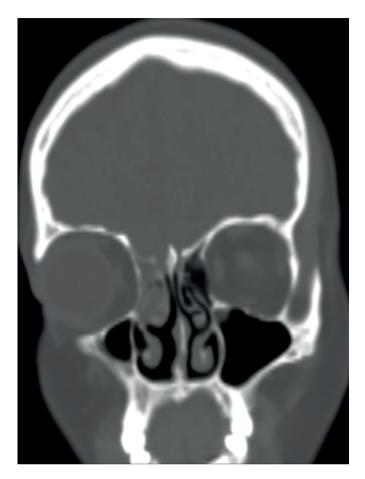


Figure 4: Coronal CT bone window image showing a defect in the right cribriform plate (arrow).

venous drainage [13]. IIH has also been associated with spontaneous CSF leak through formation of meningocele in the skull base with subsequent rupture due to chronically elevated intracranial pressure[9].

Obese female of childbearing age are at increased risk of having IHH with incidence of up to 20 times that of the general population [14]. Our case is a 42-year old female with body mass index in the obese range that was being investigated for dural sinus thrombosis. Similarly, Perez et al. [10] reported two cases of obese middle-aged female diagnosed with IIH, one of them was initially mistaken for sinusitis. Bidot et al. [14] and Illing et al. [16] in their studies noted that 94% and 81%, respectively, of the study population diagnosed with IIH were middle-aged obese women. Aaron et al. [17] on the other hand also reported obesity in 94% of their study population.

Most patients with IHH present with headache requiring investigations with both neuroimaging and laboratory. Headache is a common presenting complaint in IIH. Our patient presented with headache of which dural sinus thrombosis was suspected clinically. Fiebai and Chukwuka [18] and Ghimire et al. [1], similarly, reported headache as the cardinal symptom in their patients. In Wawhal et al.'s [19] case, the patient presented

with global headache necessitating ophthalmologic consultation but could not get relief from ophthalmologic treatment regimen and further review lead to the diagnosis of IIH.

Meningocele may be congenital, iatrogenic, or spontaneous. The anatomical locations of the meningoceles vary. Spontaneous meningoceles are common in the skull base with the ethmoid and lateral wall of the sphenoid sinus being the most common locations [20]. In a study conducted by Bialer et al. [21], all meningoceles were located in the temporal bone, either petrous apex or Meckel's cave. Spontaneous intranasal meningocele, through the cribriform plate with rhinorrhea, have been reported in patients with IIH [11], however, meningocele without rhinorrhea is seldom reported. Our patient denied any history of rhinorrhea except headache. Intranasal meningocele without rhinorrhea was incidentally discovered in our patient. Perez et al. [11] illustrated the case of a middle-age obese woman with transcribriform plate intranasal meningocele with rhinorrhea who developed bacterial meningitis due to delay in diagnosis. In a related study, Randonjic et al. [10] reported a case of bilateral meningocele in adult male involving both transethmoidal and lateral recess intrasphenoidal lesions.

Symptomatically, IIH is characterized by headaches of variable severity, vomiting, and visual disturbances ranging from blurred vision to visual field loss. Some patients may present with complications such as meningocele with associated CSF leaks which may manifest in the form of rhinorrhea or otorrhea and recurrent bacterial meningitis. The role of neuroimaging has been reiterated in the diagnosis of IIH and its complications. Imaging findings of IIH include papilledema manifesting on imaging as flattening of the posterior globe and intraocular protrusion of the optic nerve head, vertical tortuosity of the optic nerve with widening of the perioptic nerve sheath, empty sella which may be partial and transverse venous sinus stenosis [8].

4. Conclusion

IIH is common among middle-aged obese female and could be associated with spontaneous meningocele, which may rupture and predispose to complications such as meningitis. Young women presenting with intractable headaches and sometimes rhinorrhea should be investigated with neuroimaging to exclude among others IIH. Early diagnosis of a disease is panacea for the avoidance of terrible complications.

Clinicians should prioritize early neuroimaging in the diagnostic arsenal of young women with headache and radiologists should ensure systematic review of both new and old images where available to establish diagnosis and complications which may not manifest clinically.

Acknowledgements

None.

5. Competing Interests

None declared.

Ethical Considerations

Not applicable (It was waived due to the retrospective nature of the study).

Availability of Data and Material

The data used to support the findings of this study are available from the corresponding author upon reasonable request.

Funding

None.

References

- [1] Ghimire, A., Acharya, A. R., Karn, A., & Kumar Jha, M. (2021). Idiopathic intracranial hypertension: A case report. *JNMA; Journal of the Nepal Medical Association*, 59(234), 197–199. https://doi.org/10.31729/jnma.5176
- [2] Sunderland, G. J., Jenkinson, M. D., Conroy, E. J., Gamble, C., & Mallucci, C. L. (2021). Neurosurgical CSF diversion in idiopathic intracranial hypertension: A narrative review. *Life*, 11, 393. https://doi.org/10.3390/life11050393
- [3] Wall, M., Kupersmith, M. J., Kieburtz, K. D., Corbett, J. J., Feldon, S. E., Friedman, D. I., Katz, D. M., Keltner, J. L., Schron, E. B., McDermott, M. P., & the NORDIC Idiopathic Intracranial Hypertension Study Group. (2014). The idiopathic intracranial hypertension treatment trial: Clinical profile at baseline. *JAMA Neurology*, 71(6), 693–701. https://doi.org/10.1001/jamaneurol.2014.133
- [4] Kilgore, K. P., Lee, M. S., Leavitt, J. A., Mokri, B., Hodge, D. O., Frank, R. D., & Chen, J. J. (2017). Re-evaluating the incidence of idiopathic intracranial

- hypertension in an era of increasing obesity. *Ophthalmology, 124*, 697–700. https://doi.org/10.1016/j.ophtha.2017.01.006
- [5] Mollan, S. P., Aguiar, M., Evison, F., Frew, E., & Sinclair, A. J. (2019). The expanding burden of idiopathic intracranial hypertension. *Eye*, *33*, 478–485. https://doi.org/10.1038/s41433-018-0238-5
- [6] Galgano, M. A., & Deshaies, E. M. (2013). An update on the management of pseudotumor cerebri. *Clinical Neurology and Neurosurgery*, 115, 252–259. https://doi.org/10.1016/j.clineuro.2012.11.018
- [7] Smith, S. V., & Friedman, D. I. (2017). The idiopathic intracranial hypertension treatment trial: A review of the outcomes. *Headache*, *57*(8), 1303–1310. https://doi.org/10.1111/head.13144
- [8] Albakr, A., Hamad, M. H., Alwadei, A. H., Bashiri, F. A., Hassan, H. H., Idris, H., Hassan, S., Muayqil, T., Altweijri, I., & Salih, M. A. (2016). Idiopathic intracranial hypertension in children: Diagnostic and management approach. *Sudanese Journal of Paediatrics*, *16*(2), 67–76.
- [9] Aggarwal, V., Nair, P., Shivhare, P., Jayadevan, E. R., Felix, V., Abraham, M., & Nair, S. (2017). A case of evolving bilateral sphenoidal meningoencephaloceles: Case report and review of the literature. World Neurosurgery, 100, 708.e11–708.e17. https://doi.org/10.1016/j.wneu.2017.02.037
- [10] Radonjic, A., Kassab, A. M., Moldovan, I. D., Kilty, S., & Alkherayf, F. (2019). Idiopathic intracranial hypertension presenting as bilateral spontaneous lateral intrasphenoidal and transethmoidal meningoceles: A case report and review of the literature. *Journal of Medical Case Reports*, *13*(1), 62. https://doi.org/10.1186/s13256-018-1959-6
- [11] Pérez, M. A., Bialer, O. Y., Bruce, B. B., Newman, N. J., & Biousse, V. (2013, December). Primary spontaneous cerebrospinal fluid leaks and idiopathic intracranial hypertension. *Journal of Neuro-Ophthalmology*, 33(4), 330–337. https://doi.org/10.1097/WNO.0b013e318299c292
- [12] Madriz Peralta, G., & Cestari, D. M. (2018, November). An update of idiopathic intracranial hypertension. *Current Opinion in Ophthalmology, 29*(6), 495–502. https://doi.org/10.1097/ICU.00000000000018
- [13] Mollan, S. P., Ali, F., Hassan-Smith, G., Botfield, H., Friedman, D. I., & Sinclair, A. J. (2016). Evolving evidence in adult idiopathic intracranial hypertension: Pathophysiology and management. *Journal of Neurology, Neurosurgery, and Psychiatry*, 87, 982–992. https://doi.org/10.1136/jnnp-2015-311302

- [14] Sinclair, A. J., Kuruvath, S., Sen, D., Nightingale, P. G., Burdon, M. A., & Flint, G. (2011). Is cerebrospinal fluid shunting in idiopathic intracranial hypertension worthwhile? A 10-year review. *Cephalalgia*, *31*, 1627–1633. https://doi.org/10.1177/0333102411423305
- [15] Bidot, S., Levy, J. M., Saindane, A. M., Narayana, K. M., Dattilo, M., DelGaudio, J. M., Mattox, D. E., Oyesiku, N. M., Peragallo, J. H., Solares, C. A., Vivas, E. X., Wise, S. K., Newman, N. J., & Biousse, V. (2021). Spontaneous skull base cerebrospinal fluid leaks and their relationship to idiopathic intracranial hypertension. *American Journal of Rhinology & Allergy*, 35(1), 36–43. https://doi.org/10.1177/1945892420932490
- [16] Illing, E., Schlosser, R. J., Palmer, J. N., Curé, J., Fox, N., & Woodworth, B. A. (2014). Spontaneous sphenoid lateral recess cerebrospinal fluid leaks arise from intracranial hypertension, not Sternberg's canal. *International Forum of Allergy & Rhinology, 4*(3), 246–250. https://doi.org/10.1002/alr.21262
- [17] Aaron, G., Doyle, J., Vaphiades, M. S., Riley, K. O., & Woodworth, B. A. (2014). Increased intracranial pressure in spontaneous CSF leak patients is not associated with papilledema. *Otolaryngology Head and Neck Surgery, 151*(6), 1061–1066. https://doi.org/10.1177/0194599814551122
- [18] Fiebai, B., & Chukwuka, I. O. (2011). Presumed idiopathic intracranial hypertension: A case report and literature review. [Erratum in: *Nigerian Journal of Medicine*. 2011 Jul-Sep;20] [3] [:390. Chukwuka, I F ,corrected to Chukwuka, I O]. *Nigerian Journal of Medicine*, 20(2), 289–291.
- [19] Wawhal, M., Mogal, V., Patil, P., Kakde, R., Sonawane, M., & Gole, D. P. (2014). Idiopathic intracranial hypertension: A case report and brief review of literature. *IOSR Journal of Dental and Medical Sciences*, 13(12), 55–57. https://doi.org/10.9790/0853-131265557
- [20] Yang, Z., Wang, B., Wang, C., & Liu, P. (2011). Primary spontaneous cerebrospinal fluid rhinorrhea: A symptom of idiopathic intracranial hypertension? *Journal of Neurosurgery*, 115, 165–170. https://doi.org/10.3171/2011.3.JNS101447
- [21] Bialer, O. Y., Rueda, M. P., Bruce, B. B., Newman, N. J., Biousse, V., & Saindane, A. M. (2014). Meningoceles in idiopathic intracranial hypertension. *AJR. American Journal of Roentgenology*, 202(3), 608–613. https://doi.org/10.2214/AJR.13.10874