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Acute Hydrocephalus Following Otitis Media with Bilateral Transverse Sinus Stenosis: A Case Report and Management Approach

Canio Pietro Picciano*

Department of Biomorphology e. Scienze Neuromotorie (DIBINEM), Alma Mater Studiorum Università di Bologna, 40123 Bologna, Italy

Abstract

Background: We report a case of acute hydrocephalus in a 29-year-old woman following otitis media and bilateral stenosis of the transverse sinuses.

Observation: A 29-year-old woman presented to the Emergency Department of Parma Hospital with fever, headache, photophobia, nausea and altered sensorium. The blood tests, along with the concurrent fever, raised suspicion of an ongoing inflammatory process. Brain CT revealed acute hydrocephalus, requiring urgent external ventricular drainage. Given the suspicion of meningitis, a Cerebrospinal Fluid (CSF) sample was obtained in the operating room, yielding negative results both in terms of culture, cytology and biochemistry. Brain MRI conducted the following day showed progressive reduction of hydrocephalus. However, signs of intracranial hypertension persisted in the posterior fossa, with dilation of the fourth ventricle and bilateral stenosis of the transverse sinuses confirmed on cerebral angiography. Third ventriculocisternostomy was performed, followed by stenting of the dominant transverse sinus, resulting in resolution of the condition.

Keywords: Acute hydrocephalus • Cerebrospinal fluid • Bilateral stenosis

Introduction

Hydrocephalus is characterized by the progressive accumulation of Cerebrospinal Fluid (CSF) within the ventricular system. Acute hydrocephalus, in particular, presents as an emergency condition marked by a sudden increase in intracranial pressure, which can have significant implications for cerebral perfusion. Management typically involves the placement of an External Ventricular Drain (EVD) to mitigate the risk of irreversible brain damage associated with sustained elevated intracranial pressure.

The etiopathogenesis of acute hydrocephalus can be categorized into obstructive and non-obstructive (communicating) mechanisms. Obstructive hydrocephalus arises from a blockage in normal CSF circulation, resulting in CSF accumulation upstream of the obstruction, such as in cases involving colloid cysts of the third ventricle. Conversely, communicating hydrocephalus may stem from pathological increases in CSF production or diminished reabsorption capacity, as observed in conditions like leptomeningeal carcinomatosis. In this paper, we present a unique case of obstructive hydrocephalus in a 29-year-old woman, attributed to bilateral obstruction of the transverse sinuses secondary to otitis media ("otitic hydrocephalus").

Cerebrospinal fluid production and reabsorption

The rate of CSF formation in humans is estimated to be 0.3-0.4 ml/min, with a total volume of 90-150 ml in adults. CSF circulates through the ventricles, cisterns and subarachnoid space before being absorbed into venous blood at the level of the arachnoid villi [1-3]. Some CSF may also drain into cervical lymphatics *via* the perineural spaces of cranial nerves [4]. Traditionally, CSF flow has been described as a circulatory process, often referred to as the "third circulation" [5]. To maintain normal intracranial pressure, CSF production must be balanced by effective reabsorption to uphold intracranial homeostasis. Various theories have been proposed regarding CSF production and absorption. The classic theory implicates the choroid plexi as the primary sites of CSF production. However, recent findings challenge this notion, suggesting alternative mechanisms for water transport across epithelia.

According to the traditional view, the choroid plexus is considered the primary source of Cerebrospinal Fluid (CSF) production. These structures develop from the ependyma, which extends from the pia mater into the lateral, third and fourth ventricles. Comprising a single layer of epithelial cells resting on a basement membrane, connective tissue and fenestrated capillaries, the choroid plexus allows for the passage of compounds from the blood to the epithelial cells through tight junctions. While the classic theory has long held sway, recent research challenges its sole reliance on choroid plexus activity [1,2,6]. Studies involving AQP1 knockout mice have revealed intriguing insights. These experiments demonstrated a reduction of water permeability across the choroid plexus by 85%, whereas CSF secretion diminished by only 35%, suggesting alternative mechanisms for water transport through the epithelial layer. Despite this, the prevailing belief remains that the choroid plexus serves as the primary site of CSF production, albeit with potential contributions from extrachoroidal sites [7]. However, an alternative perspective posits that extrachoroidal sites may play a more significant role in CSF production, with the choroid plexus contributing as well. This viewpoint challenges the conventional understanding of CSF dynamics and underscores the need for further investigation into the mechanisms governing CSF production [7,8].

Similarly, multiple theories exist regarding CSF absorption. The classic theory posits absorption *via* arachnoid granulations into the venous system, while studies in animal models indicate significant absorption through cervical lymphatics. Additionally, the dural venous plexus may play a role in

***Address for Correspondence:** Canio Pietro Picciano, Department of Biomorphology e. Scienze Neuromotorie (DIBINEM), Alma Mater Studiorum Università di Bologna, 40123 Bologna, Italy; E-mail: caniopietro.picciano@studio.unibo.it, caniopietro.picciano@gmail.it*

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CSF reabsorption, particularly in infants where arachnoid granulations are less developed [1,4,7]. The traditional theory of Cerebrospinal Fluid (CSF) absorption posits that absorption primarily occurs from the subarachnoid spaces into the venous bloodstream *via* the dural venous sinuses, facilitated by cranial arachnoid granulations. However, research conducted in rabbit and ovine models suggests that CSF absorption may also occur significantly through cervical lymphatics [9]. CSF not reabsorbed through arachnoid granulations may reach cervical lymphatics *via* two potential pathways. The first involves travel along the subarachnoid space of exiting cranial nerves, providing a direct route from the cisterns to extracranial lymphatics. The second pathway entails passage through the Virchow-Robin space surrounding arteries and veins penetrating brain parenchyma. This space's size can vary depending on pathological conditions. When CSF fails to be absorbed through the classical pathway, it may enter the Virchow-Robin space or be redirected to the Interstitial Fluid (ISF). The ISF exhibits bidirectional flow with the Virchow-Robin space and subarachnoid space, mediated by aquaporins, though ongoing research explores this mechanism further. CSF entering the ISF can ultimately be reabsorbed into the bloodstream, return to the Virchow-Robin space, or re-enter the subarachnoid space. From the Virchow-Robin space, CSF may return to the subarachnoid space or undergo reabsorption by cervical lymphatics, influenced by cardiac pulsations and pulmonary respiration [9,10].

Additionally, studies indicate the possibility of CSF reabsorption into the dural venous plexus, particularly in infants where arachnoid granulations are underdeveloped. While less extensive in adults, the dural venous plexus likely plays a role in absorption. Despite these insights, the precise mechanism of CSF uptake remains elusive [11]. In summary, while the mechanisms of CSF production and absorption continue to be elucidated, they are crucial for maintaining intracranial pressure and preventing conditions like obstructive hydrocephalus.

Case Presentation

A 29-year-old female, with an unremarkable medical history, presented to Parma Hospital's Emergency Department on December 16, 2023, complaining of a three-day history of headache, altered mental status and double vision. Her parents reported a preceding week of low-grade fever, left ear pain and vomiting. An otitis diagnosis was made by an otolaryngologist. Upon evaluation, the patient appeared lethargic but responsive to verbal stimuli, with intact motor function and no focal neurological deficits. Vital signs were mostly normal, except for a mild fever. Laboratory tests showed elevated white blood cell count and C-reactive protein levels. Empirical antibiotic therapy was initiated and cultures were obtained.

Imaging revealed hydrocephalic enlargement of ventricular cavities, particularly in the temporal horns of lateral ventricles, indicative of increased intracranial pressure. An urgent external ventricular drain placement was performed, resulting in a notable improvement in consciousness. (Figure 1) Subsequent analyses of cerebrospinal fluid, blood and urine cultures

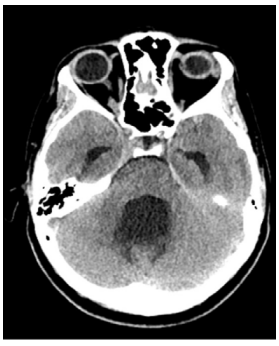

Figure 1. CT scan performed upon admission to the Emergency Department. Axial and sagittal slices. The images document the presence of acute hydrocephalus with signs of transependymal absorption. Note the compression of the basal cisterns, cisterna magna, and bulbospinal cistern, likely indicative of the obstructive nature of the hydrocephalus.

were negative. Biochemical analysis of CSF confirmed normal glucose and protein levels, ruling out meningitis. Cranial and cervical spine MRI showed reduced ventricular size and severe stenosis of transverse sinuses, suggesting obstructive hydrocephalus (Figure 2).

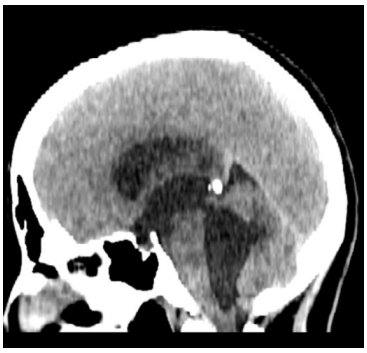

Figure 2. Brain MRI with and without contrast performed following the placement of an external ventricular drain. T2-w Sagittal view and MR angiography sequence. The resolution of the acute hydrocephalus is documented. Despite the near-complete resolution of supratentorial hydrocephalus, abnormal enlargement of the fourth ventricle persists, with signs of hypertension in the posterior cranial fossa (flattening of the cerebellar folia, brainstem compression against the clivus). MR angiography shows the appearance of bilateral stenosis of the transverse sinuses, likely causing increased pressure in the posterior cranial fossa (reduced resorption and increased venous pressure).

Further CSF analysis post-ventricular drain placement yielded negative results.

On December 21st, an attempt was made to wean the patient off the external ventricular drain with subsequent closure. On December 22nd, a neurological evaluation advised further diagnostic investigation, including immunofixation for the detection of oligoclonal bands and serum protein electrophoresis (both negative). A follow-up chest X-ray showed no signs of ongoing consolidation, an abdominal ultrasound revealed no significant findings and COVID-19, Treponema pallidum and HIV tests were negative. A cranial CT scan on December 22nd, following closure of the external ventricular drain, showed minimal enlargement of the ventricles. On December 24th, neurological deterioration manifested with lethargy, slowing and poor reactivity, prompting the reopening of the external ventricular drain and a repeat cranial CT scan, which revealed further enlargement of the ventricles. In light of the clinical picture, on December 24th, the patient underwent Third Ventriculo Cisternostomy (TVCS) and repositioning of the external ventricular drain, which remained closed, to protect against potential neurological deterioration. On December 28th, a cranial CT scan showed a decrease in ventricular size, prompting the removal of the external ventricular drain.

On January 2nd, cerebral angiography was performed with measurement of intravenous pressures. The angiographic control revealed a reduction in caliber of the right transverse sinus (dominant), consistent with findings from the brain MRI on December 18th. Venous sinus manometry was performed, measuring pressures at the level of the superior sagittal sinus, torcular sinus, right transverse sinus, right sigmoid sinus and right jugular vein. A gradient of 15 mmHg between the superior sagittal sinus and the right jugular vein was documented (Figure 3). Therefore, antiplatelet therapy was initiated in preparation for stenting of the transverse sinus. On the follow-up brain MRI on January 5th, with cerebrospinal fluid flow study, the expected outcomes of the third ventriculocisternostomy with patent and functioning stoma were observed. There was initial reduction in hydrocephalic dilatation of the fourth ventricle and the aqueduct of Sylvius. The ventricular cavities remained substantially unchanged in size compared to the previous study, with resolution of periventricular signal alteration related to altered transependymal absorption. There was also partial enlargement of the bilateral transverse sinuses compared to the previous MRI.

The neurological status progressively improved and the patient regained autonomy. Consequently, she was discharged home on January 6th with an appointment for subsequent admission to undergo the transverse sinus stenting procedure on the right (dominant) side. On January 22nd, the patient was readmitted for the angiographic procedure. Upon return, the patient was

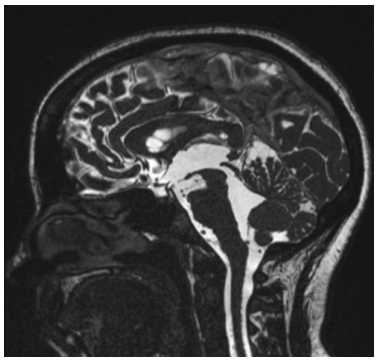

Figure 3. Pre and post-stenting angiography of the dominant transverse sinus. Pre-stenting sinus manometry documented a gradient of 15 mmHg between the superior sagittal sinus and the right jugular vein. Post-stenting control showed the gradient had decreased to 1 mmHg.

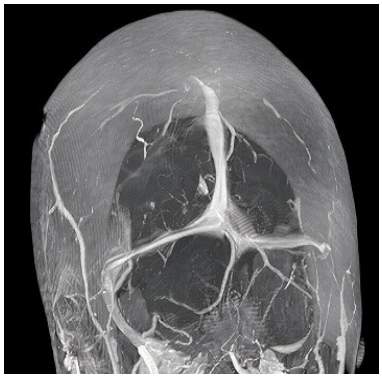

Figure 4. The two-month follow-up brain MRI showed proper functioning of the stoma and complete resolution of hydrocephalus.

awake and fully oriented, with a GCS of 15\15, but diplopia persisted due to abducens nerve palsy on the right side. The follow-up cranial CT scan upon admission showed further slight reduction in the ventricular system size. On January 23rd, she underwent cerebral angiography for stenting of the dominant transverse sinus. Pre-stenting venous sinus manometry showed a gradient of 4 mmHg, indicating resolution of the previously present stenosis. A Carotid Wallstent 7 × 50 mm was placed in the stenotic right transverse sinus. Venous drainage control prior to stent release showed no obstructions. Post-stenting pressure checks showed a physiological gradient of 1 mmHg. The postoperative CT scan showed no complications.

At the clinical follow-up two months post-procedure, the patient exhibited clinical well-being, with no neurological deficits except for persistent right abducens nerve palsy. Control angiography documented normal pressure gradient between the superior sagittal sinus and the right jugular vein. The two-month follow-up brain MRI showed proper functioning of the stoma and complete resolution of hydrocephalus (Figure 4).

Results and Discussion

In this paper, we present a rare case of obstructive hydrocephalus of unknown etiology. From the evaluation of the case, we have hypothesized two pathogenetic mechanisms: on one hand, bilateral transverse sinus stenosis evidently led to a reduction in Cerebrospinal Fluid (CSF) reabsorption, while on the other hand, increased venous pressure within the posterior cranial fossa caused an obstruction to normal CSF outflow [9,10]. Supporting the first hypothesis are known studies of CSF dynamics and the close relationship between the subarachnoid space, interstitial matrix and blood flow. An increase in pressure within the intracranial venous compartment evidently leads to a reduction in CSF reabsorption capacity. However, this does not explain the obstructive hypothesis underlying the development of acute hydrocephalus in this patient [7]. Focusing on the morphology of the patient's posterior fossa and calculating its overall volume using the five-sided polygon and the additional Chamberlain's line suggested by Raybaud and Jall, it emerges that the conformation of the Posterior Cranial Fossa (PCF) in this patient may be a predisposing factor to the obstruction of CSF outflow [12-14].

Given that the morphology of the patient's posterior fossa is "smaller" compared to a normotype, a variation in venous pressures within the structures involved in venous outflow in this location may have led to an increase in pressure within the compartment, evidently causing obstruction of the outflow from the fourth ventricle, the cisterna magna and the bulbo-spinal cistern.

Conclusion

We have described a rare case of obstructive hydrocephalus of unknown etiology. From the assessment of the case, we hypothesized that in a patient with anatomical predisposition (a "small" posterior cranial fossa), a subacute obstruction of venous outflow could lead to an increase in pressure within the subtentorial compartment, resulting in an obstruction of CSF outflow and the development of acute hydrocephalus. The resolution of the venous outflow obstruction consequently leads to a return to cerebrospinal fluid homeostasis and resolution of hydrocephalus.

Acknowledgement

None.

Conflict of Interest

None.

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