

Delayed Cerebrospinal Fluid Cleft Formation Through the Midbrain and Thalamus Following Hemicraniectomy and Cranioplasty for Acute Subdural Hematoma

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BACKGROUND AND IMPORTANCE: Cerebrospinal fluid (CSF) cleft formation through brain parenchyma following nonpenetrating traumatic brain injury (TBI) is a rare phenomenon. Here we present a unique case of delayed CSF cleft formation months after initial injury.

CLINICAL PRESENTATION: A 41-yr-old male presented after a fall with a right convexity acute subdural hematoma and ipsilateral frontal contusion. He underwent emergent hemicraniectomy with subsequent autologous cranioplasty 2 mo later. At 10-mo follow-up his neurological status had improved. His magnetic resonance imaging (MRI) at that time demonstrated encephalomalacia at the site of his prior contusion and punctate right pontine traumatic shearing injury. The patient re-presented to clinic 13 mo after initial injury with 2 mo of progressively worsening dysarthria, left hand numbness, diplopia, and dysphagia. MRI revealed a new tubular-shaped CSF cleft extending from the fourth ventricle, through the right midbrain and thalamus that was not present on prior MRI. Computed tomography cisternogram confirmed communication with the ventricular system, and there was no clinical evidence for elevated CSF pressure. One month later, the patient's symptoms had not improved, and imaging revealed progression of the CSF cleft. Following placement of a ventriculoperitoneal shunt, progression of the cleft ceased. We postulate that this cleft was a late sequela of traumatic shearing injury. We discuss our efforts to diagnose the etiology of the cleft and the rationale for our management strategy.

CONCLUSION: To our knowledge, this represents the first reported delayed-onset CSF cleft through the midbrain and thalamus after closed TBI.

KEY WORDS: Cerebrospinal fluid, Cleft, Traumatic brain injury, Hemicraniectomy, Hydrocephalus, Ventriculoperitoneal shunt

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In rare traumatic brain injury (TBI) cases, clefts may form within brain parenchyma from shearing forces at gray-white matter junctions. This phenomenon, although rare, has been previously described, particularly

in children following severe nonaccidental trauma.^{1–5}

To our knowledge, no prior description exists for cerebrospinal fluid (CSF) cleft formation through the midbrain and thalamus long after traumatic injury. Here we present a patient with CSF cleft formation over 1 yr after initial TBI who presented a diagnostic and management challenge.

CLINICAL PRESENTATION

A 41-yr-old male presented to the emergency department after falling down a flight of stairs while intoxicated. On arrival he was GCS 8T, localizing in the upper extremities, and

ABBREVIATIONS: **CT**, computed tomography; **CSF**, cerebrospinal fluid; **DAI**, diffuse axonal injury; **DWI**, diffusion-weighted imaging; **EVD**, external ventricular drain; **GRE**, gradient echo; **ICP**, intracranial pressure; **LP**, lumbar puncture; **TBI**, traumatic brain injury; **MRI**, magnetic resonance imaging; **SDH**, subdural hematoma; **SWI**, susceptibility weighted imaging; **VPS**, ventriculoperitoneal shunt

had equal and reactive pupils. His initial head computed tomography (CT) revealed an acute right convexity subdural hematoma (SDH) with ipsilateral frontal hemorrhagic contusion and subarachnoid hemorrhage with resulting 9 mm midline shift (Figures 1A and 1B). The patient underwent external ventricular drain (EVD) placement revealing elevated intracranial pressure (ICP). He underwent emergent primary hemicraniectomy (Figure 1C) and subsequent autologous cranioplasty (Figure 1D) approximately 2 mo later.

At 6-mo follow-up, he had improved and was able to regain a commercial truck driver's license. Magnetic resonance imaging (MRI) of the brain obtained 10 mo after initial injury (Figure 2) showed encephalomalacia at the site of his prior contusion. Figure 2 displays gradient echo (GRE) and diffusion-weighted imaging (DWI) sequences obtained 10 mo postinjury revealing punctate gradient susceptibility in the right pons (Figures 2C and 2D) and midbrain (not shown) consistent with hemorrhagic diffuse axonal injury (DAI). Diffusion hyperintensity was seen along the right superior cerebellar peduncle consistent with traumatic shearing injury. The patient had no prior MRI before 10-mo follow-up; however, his CT imaging after injury at 2- and 4-mo follow-up showed no lesion in the pons or midbrain.

The patient re-presented to clinic 13 mo postinjury with new onset of progressively worsening slurred speech, left hand clumsiness and numbness, double vision, and trouble swallowing liquids. On exam he was alert and oriented fully. Pupils were equal and reactive bilaterally. Nystagmus was observed on right lateral gaze as well as diplopia on convergence testing. He had subtle left-sided facial paralysis sparing the forehead. Speech was dysarthric. Numbness to light touch in the left hand, wrist, and forearm was observed. He had full strength throughout and was not hyperreflexic. Ataxia was observed in the left upper extremity. A formal ophthalmology consult was obtained; findings were notable for full visual fields, impaired convergence, and right esotropia. Funduscopic examination was negative for papilledema.

MRI sequences obtained 13 mo postinjury (Figures 3A and 3B) revealed a tubular-shaped CSF-isointense lesion in the right midbrain and thalamus that communicated with the superior aspect of the fourth ventricle. CT cisternogram (Figures 3C and 3D) showed communication between the CSF cleft and the ventricular system. Opening pressure on his lumbar puncture (LP) was 13 cm CSF. T2-weighted MRI sequence of the midbrain 13 mo postinjury at re-presentation (Figure 3E) revealed CSF cleft formation in the posterolateral pons at the sites of previous axonal injury on gradient and DWI 3 mo earlier. Susceptibility weighted imaging (SWI) sequence imaging at 13 mo postinjury of the midbrain and thalamus demonstrated foci of susceptibility in the right midbrain and bilateral thalamus (Figure 3F). The fourth ventricular origin of the cleft is best appreciated on coronal T2 imaging (Figure 3G). The cerebral aqueduct was patent on CT cisternogram (Figure 3H).

Although there was no evidence for hydrocephalus, we recommended placement of a ventriculoperitoneal shunt (VPS) in hopes of reducing the risk of cleft progression. The patient

initially declined, favoring a course of observation. He returned for follow-up 2 mo later (15 mo postinjury) with imaging (Figure 4A) revealing cleft progression. Angiogram did not reveal any vascular abnormality or an enlarged Virchow Robin space (Figure 4B). Given the progressive CSF cleft enlargement and unchanged symptoms, the patient consented to VPS. At writing, the VPS has demonstrated success in preventing progression of the cleft for 3 mo following placement (Figure 4C).

DISCUSSION

Prior Literature

To our knowledge, only 1 prior report of delayed post-traumatic CSF cleft exists. Anderson et al reported a case of a 28-yr-old man struck by a cement truck.⁵ He underwent craniotomy for SDH evacuation and had residual deficits from his injury including left upper extremity clumsiness. Eight months after initial injury, he began experiencing dysarthria, dysphagia, left facial weakness, left ataxia, and diplopia. Head CT revealed a previously unseen midbrain CSF cleft with ventricular communication on CT myelogram. The patient was treated with a VPS resulting in cessation of cleft progression; however, this had no effect on his neurological outcome.

Prior reports of traumatic CSF clefts involve nonaccidental trauma to infants with grey-white matter interface or acute subcortical white matter tears that lead to CSF cleft formation.^{1,2} Such cases involve contusional tears discovered immediately after the initial traumatic insult, which was not observed for our patient.

Wan et al⁶ reported a case series of 25 patients undergoing decompressive hemicraniectomy who developed outward bulging of the bone flap (high-pressure complications) or "sinking skin flap syndrome" (low pressure complications). Of 25 patients, 2 developed ipsilateral cystic encephalomalacia communicating with the lateral ventricle treated using a lumbar drain. Wan et al⁶ did not comment on CSF pressure or drainage parameters for the patients in their series undergoing lumbar drainage. Cystic encephalomalacia with ventricular communication developed prior to cranioplasty in both patients presented by Wan et al⁶ in contrast to our patient.

Proposed Causative Mechanisms

Several potential contributing etiologies of this patient's presentation were considered (Table). These included cystic encephalomalacia, ischemic infarct, hydrocephalus, enlarged Virchow-Robin space (perivascular space), syringobulbia, and traumatic parenchymal tear. Mechanistically, we propose that the patient's brainstem was subjected to shearing injury during trauma, potentially being accelerated-decelerated against the tentorium. The GRE and DWI imaging sequences 10 mo postinjury revealed punctate gradient susceptibility and diffusion restriction consistent with shearing injury in the right superior pons and midbrain, including the right superior cerebellar peduncle.

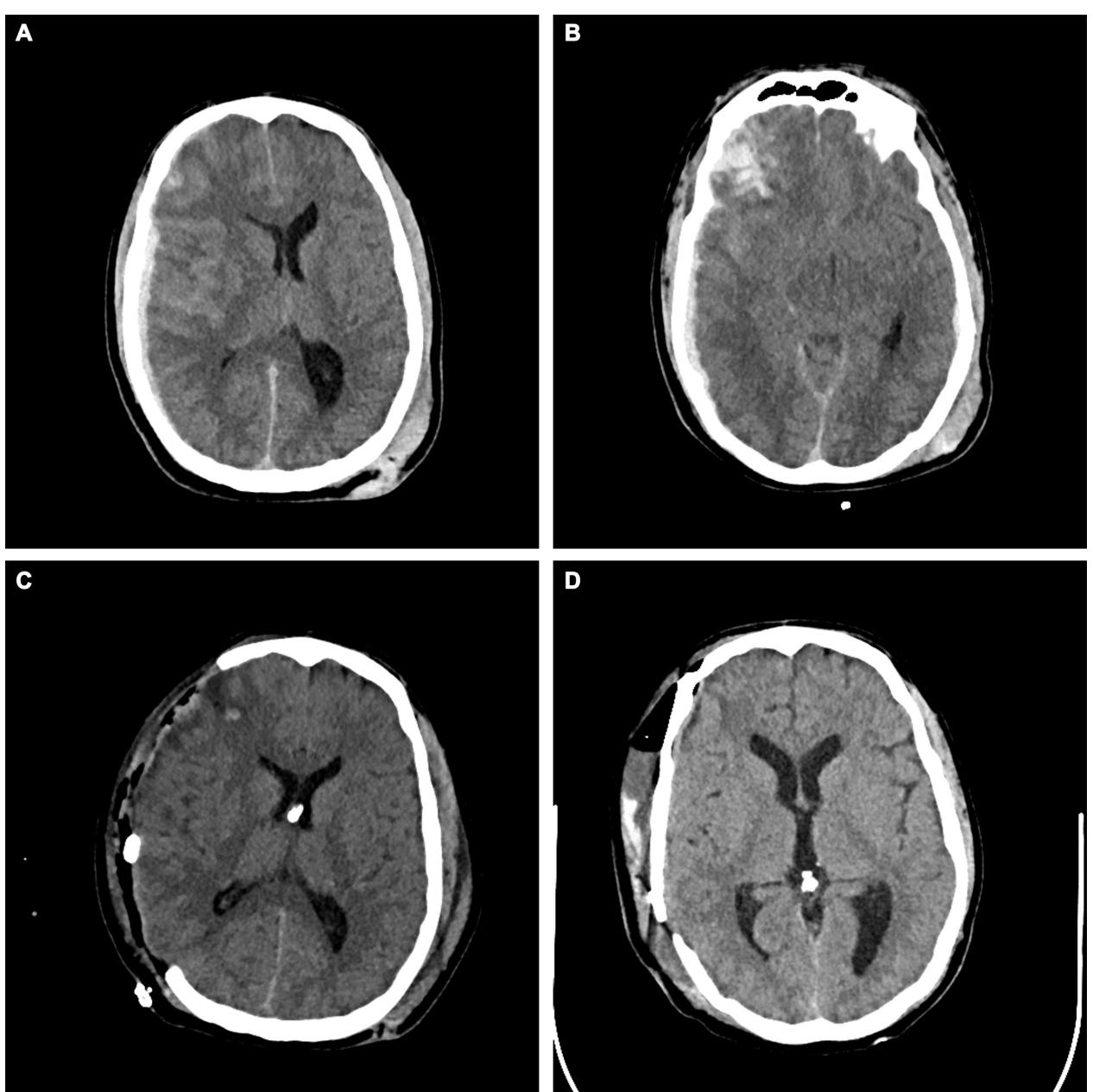


FIGURE 1. Images obtained at initial injury and post-cranioplasty. **A**, Noncontrast head CT at initial presentation showing right-sided convexity acute SDH with 9 mm of midline shift and **B**, right frontal hemorrhagic contusion. **C**, Postoperative head CT obtained after right-sided hemicraniectomy and external ventricular drain placement 1 day after initial injury. **D**, Postoperative head CT obtained after primary cranioplasty 2 mo following initial presentation. Mild ventriculomegaly was observed compared to prior CT imaging; however, the patient did not have clinical symptoms of hydrocephalus.

Furthermore, foci of susceptibility in the right thalamus were seen on SWI sequences consistent with DAI. The cleft expanded laterally and superiorly from the areas of brainstem injury. The brainstem could also have been subjected to injury from ICP increases prior to decompressive hemicraniectomy and EVD

placement. Brain arterial/tissue pulsatility has been implicated in causing communicating hydrocephalus even in cases of normal ICP.^{7,8} We propose that brain pulsations may have contributed to development and progression of his CSF cleft. We elected to proceed with palliative VPS given the progressive enlargement

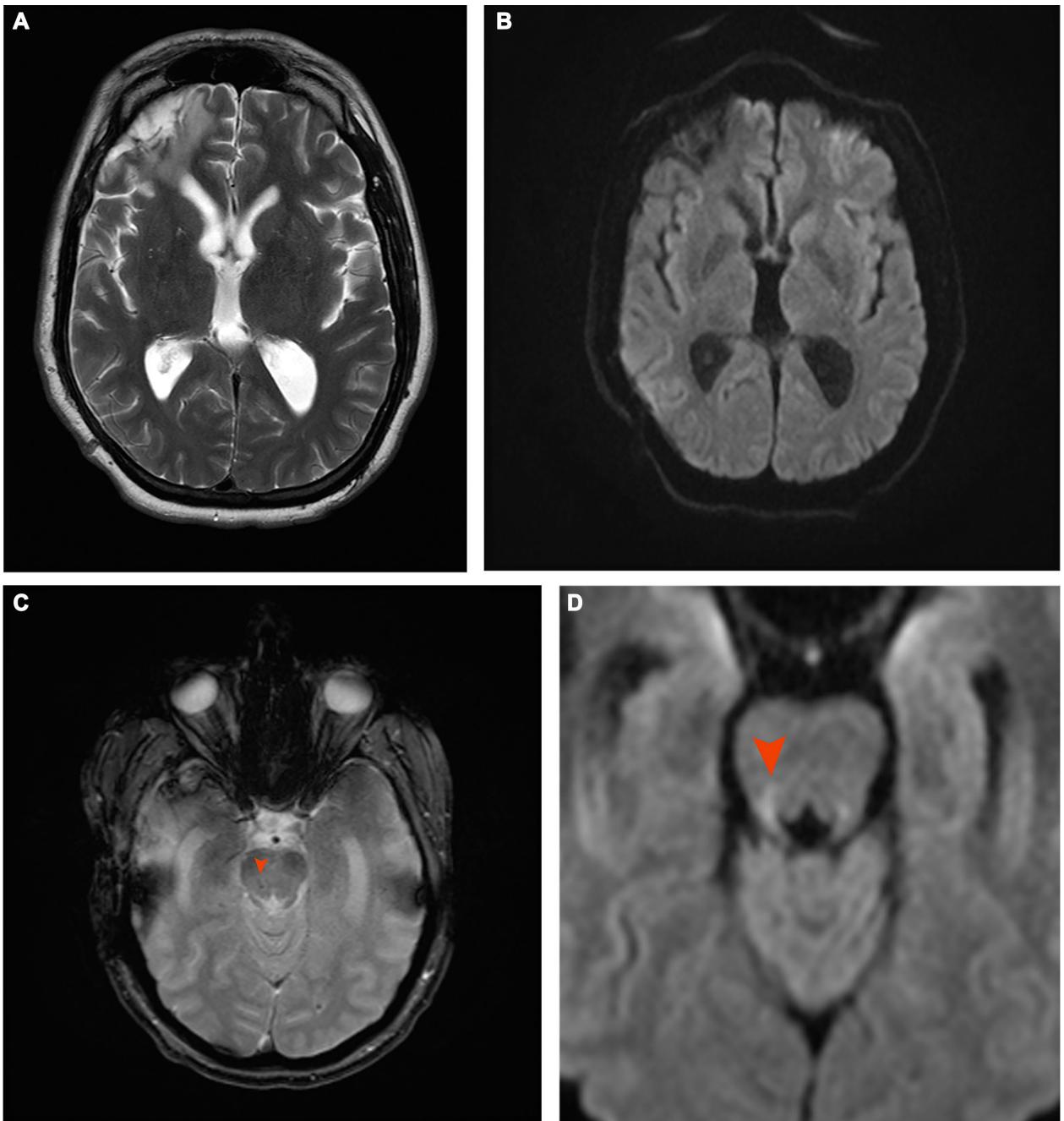


FIGURE 2. Images obtained at 10 mo postinjury prior to symptom onset. **A**, Axial T2 weighted and **B**, DWI MRI sequences showing no grossly visible CSF cleft or signal changes in the area of future cleft formation (compare to Figure 3A). **C**, Axial GRE and **D**, DWI sequences showing punctate gradient susceptibility and diffusion restriction in the right pons consistent with hemorrhagic shearing injury (arrows).

of the CSF cleft despite the absence of ICP elevation. Indeed, the patient's opening pressure on LP was normal at the time of abnormal imaging findings and no papilledema was observed. The exact mechanism and pathophysiology remain unclear.

CONCLUSION

Here we present a case of CSF cleft formation over 1 yr after initial TBI. We propose that the CSF cleft resulted from traumatic

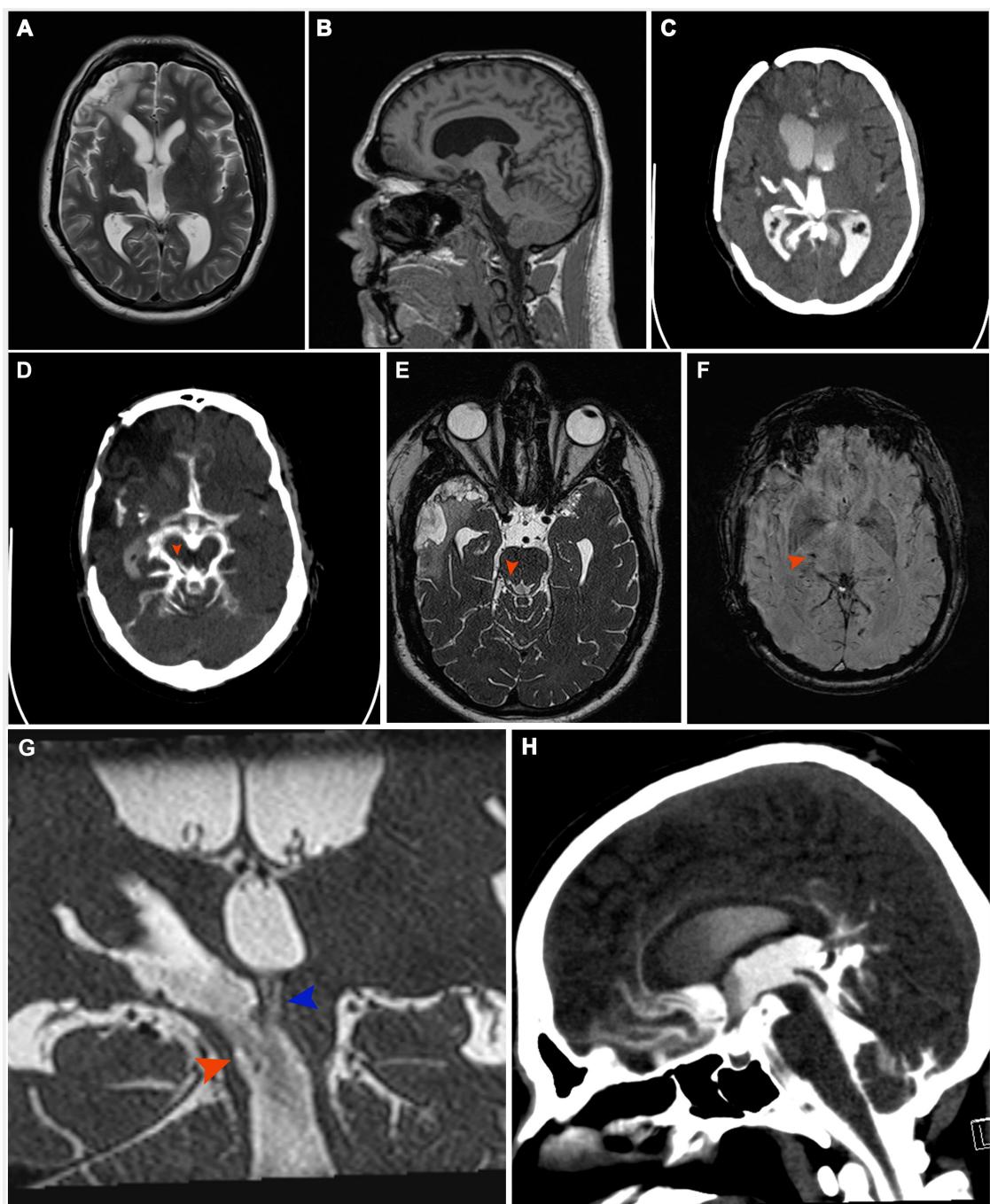


FIGURE 3. Images obtained at symptom onset 13 mo postinjury. **A**, Axial T2-weighted MRI and **B**, sagittal T1-weighted MRI demonstrating a tubular shaped right-sided CSF cleft forming through the midbrain and thalamus. **C**, Axial CT cisternogram confirming CSF cleft communication with the ventricular system. **D**, Axial CT cisternogram revealing the cleft extension from the cranial and lateral aspect of the fourth ventricle (arrow). **E**, T2-weighted MRI sequence of the midbrain showing CSF-isointense cleft formation extending from the lateral wall of the fourth ventricle in posterolateral pons (arrow). **F**, SWI sequence of the thalamus showing foci of susceptibility in the right thalamus (arrow). Foci of susceptibility were also observed in the midbrain and contralateral thalamus (not shown). **G**, Coronal T2-weighted MRI sequence revealing the fourth ventricular origin of the cleft as it extends cranially and laterally from the superior aspect of the fourth ventricle (orange arrow). Note the patency of the cerebral aqueduct without evidence of stricture (blue arrow). **H**, Sagittal CT cisternogram revealing a patent cerebral aqueduct with appropriate contrast opacity throughout, indicating that the ventriculomegaly observed on imaging and normal measured opening pressure on LP was not the result of aqueductal stenosis.

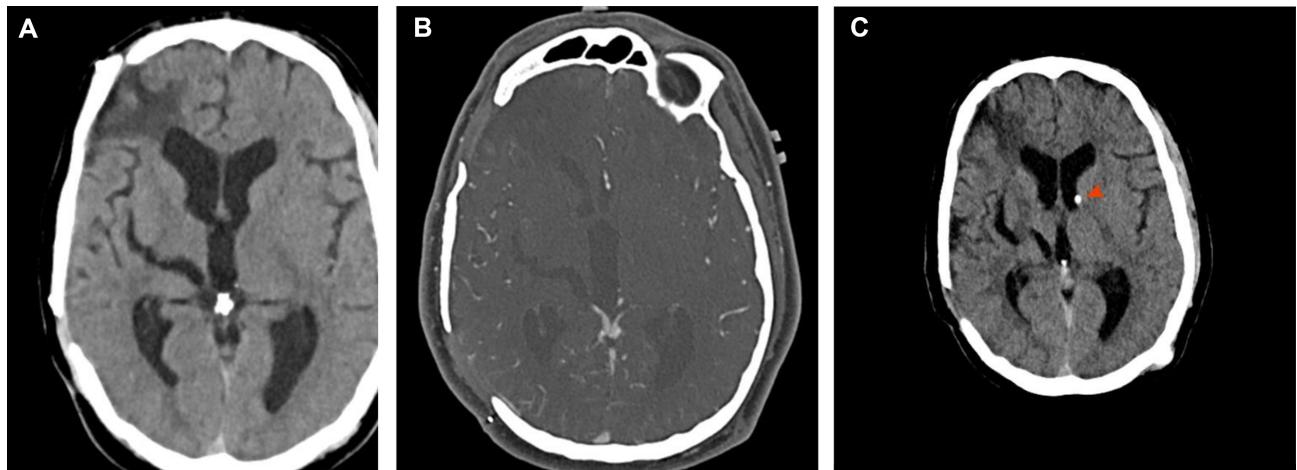


FIGURE 4. **A**, Noncontrast head CT obtained 15 mo after initial injury revealing progressive enlargement of the prior CSF cleft during conservative management. **B**, CT angiogram obtained 15 mo after initial injury showing no traversing blood vessels across or through the cleft space which might otherwise suggest that the lesion is an enlarged Virchow-Robin space. **C**, Noncontrast head CT obtained 3 mo after left occipital ventriculoperitoneal shunt placement (18 mo after initial injury; VPS catheter tip depicted by arrow) showing interval decrease in ventricular size and stable size of the CSF cleft.

TABLE. Differential Diagnoses Considered as Potential Etiologies of CSF Cleft Formation

Differential diagnosis	Supporting evidence	Contradictory evidence
Cystic encephalomalacia	History of ipsilateral traumatic brain injury, delayed onset signal abnormalities of midbrain	No imaging evidence of encephalomalacia directly at site of CSF cleft formation prior to or at time of cleft onset
Traumatic CSF cleft/parenchymal tear (secondary to closed or penetrating TBI)	History of traumatic brain injury, ipsilateral CSF cleft lesion, evidence of ipsilateral shearing injury in right midbrain and pons on MRI at 10 mo follow-up	Delayed onset of CSF cleft imaging findings (not observed until 13 mo after injury, symptom onset 11 mo after injury), no history of penetrating head trauma
Virchow Robin space (perivascular space)	CSF-isointense cleft near basal ganglia	CT angiogram without evidence of traversing vessels
Hydrocephalus	Ventriculomegaly on follow-up imaging, communication of the CSF cleft with ventricular spaces (representing ex vacuo change instead of hydrocephalus), increased ICP recordings after EVD placement during initial TBI episode	Normal CSF opening pressure on lumbar puncture (13 cm CSF), no papilledema on funduscopic exam
Syringobulbia/syringomesencephaly	Cystic fluid-filled space on imaging with bulbar symptoms (dysphagia, dysarthria)	NA

CT, computed tomography; CSF, cerebrospinal fluid; EVD, external ventricular drain; ICP, intracranial pressure; TBI, traumatic brain injury

damage to brainstem structures that progressed to cavitation and progressive cleft formation over time perhaps as a result of arterial pulsations. The patient was successfully treated with a VPS aimed at arresting further cleft progression.

Disclosures

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

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COMMENT

This is a fascinating adult case of traumatic brainstem and thalamic parenchymal cleft most likely resulting from shearing injury at the level of the fourth ventricle. The cleft extended into the midbrain and thalamus

possibly because of CSF pulsations in the fourth ventricle. Progression of the cleft, which was nearly a year after acute right-sided SDH, which needed decompressive hemicraniectomy, resulted in brainstem, cerebellar, and thalamic symptoms. Enlargement of the cleft eventually arrested following insertion of a shunting device. Two major points in this case report attract attention. 1, In a patient with a devastating subdural hematoma which needed decompressive hemicraniectomy and responded well, any new symptoms should not be taken lightly and screening for a parenchymal cleft must be considered. 2, The cleft was not associated with hydrocephalus, however its enlargement arrested with a ventriculoperitoneal shunt.

The authors of this case report should be commended for their genuine attempt to follow the patient and their literature-based review to explain the working pathogenetic mechanisms.

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