Chronic encapsulated intraventricular hematoma in a pediatric patient: case report

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Chronic encapsulated intraventricular hematoma (CEIVH) is a rare, intraventricular, nonneoplastic mass lesion that can become symptomatic from mass effect or obstructive hydrocephalus. Only 5 cases have been reported in the literature, and only one of these occurred in a pediatric patient and dates back to the pre-modern neuroimaging and pre-microsurgical era of neurosurgery. Imaging features can mimic those of many more common intraventricular lesions, such as choroid plexus tumors or cavernous malformations. In all reported symptomatic cases, resection was safely performed and led to a cure and symptom resolution. Here, the authors present a case of CEIVH in a pediatric patient, describe the operative techniques of resection, review the available literature, and discuss current understanding of the pathophysiology, making this the most comprehensive report on this disease entity to date. The case is a 14-year-old boy who presented with headaches and emesis. Computed tomography showed a hyperdense mass in the trigone of the right lateral ventricle. Magnetic resonance imaging showed a contrast-enhancing well-circumscribed mass. Right temporal craniotomy utilizing a posterior middle temporal gyrus transcortical approach was performed, and gross-total resection was achieved. Pathology revealed a CEIVH. The boy's postoperative course was uncomplicated, and he was discharged 2 days after surgery.

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CHRONIC encapsulated intracerebral hematoma (CEIH) is a rare, solid, nonneoplastic, well-circumscribed intracranial mass that can present with a variety of signs and symptoms depending on the location of the lesion. First defined by Hirsh et al., CEIH can mimic neoplasms or vascular malformations in its presentation and radiological findings and is often distinguishable only on pathological examination of resected tissue.¹⁰ Approximately 60 cases of CEIH have been reported in the literature, and by far the majority of them are intraparenchymal in location.²²

To our knowledge, only 5 cases of chronic encapsulated intraventricular hematoma (CEIVH) have been reported in 4 papers.^{2,6,13,23} Of these, only 1 case has occurred in a child; however, its report dates back to 1955.² The etiology of these lesions is not well defined. Here, we present the

case of a chronic encapsulated hematoma of the trigone of the right lateral ventricle in a patient who presented with headaches. To our knowledge, this is the most comprehensive report on this lesion, supplemented with intraoperative photographs and video, as well as a review of the literature. Furthermore, it is the first report of a CEIVH mimicking a neoplasm in a pediatric patient in the modern era of neurosurgery.

Case Report

Clinical Presentation

A 14-year-old, previously healthy male presented with 2 days of right-sided headache, blurred vision, nausea, and vomiting. He had a normal neurological exam. No papilledema was noted. The patient was having intermittent

ABBREVIATIONS CEIH = chronic encapsulated intracerebral hematoma; CEIVH = chronic encapsulated intraventricular hematoma.

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FIG. 1. Preoperative CT scans demonstrating an isodense mass within the trigone with peripheral hyperdensity (arrow, A) and early entrapment of the right temporal horn (B). Figure is available in color online only.

high fevers ($> 39^{\circ}\text{C}$) while in the hospital prior to surgery. Full infectious workup was negative, and the significance of the fevers was unclear. There was no history of trauma preceding his presentation. He had no personal or family history of any coagulopathy or bleeding diathesis. His laboratory workup, which included a complete blood count, basic metabolic panel, and standard coagulation tests, was unremarkable. Given the exclusion of any personal identifying information pertaining to this case, specific permission from this patient was neither required nor sought. No institutional review board approval was indicated to produce this case report and literature review.

Imaging Examination

Head CT without contrast revealed an isodense mass in the trigone of the right lateral ventricle with surrounding hyperdensity (Fig. 1A). There was enlargement of the right temporal horn indicating early entrapment and suggesting that the patient's headaches were indeed related to the presence of this mass (Fig. 1B).

Gadolinium-enhanced MRI revealed a $2.9 \times 2.2 \times 2.4$ -cm mass that was T1 hypointense, T2 isointense, avidly contrast enhancing, and non-diffusion restricting (Fig. 2). Magnetic resonance angiography did not show any obvious vascular malformation. Given these imaging characteristics, we believed that this probably represented a choroid plexus tumor, favoring a papilloma or, less likely, an intraventricular cavernous malformation.

Operation

Given the diagnostic uncertainty and early entrapment of the temporal horn, resection was recommended to the patient and his family. He was taken for a temporal craniotomy for image-guided resection of the mass. A posterior middle temporal gyrus transcortical approach was employed to gain access to the posterior part of the temporal horn, which in turn provided access to the atrium. Upon entering the ventricle, we encountered normal-appearing choroid plexus of the atrium, which was partially overlying, but separate from, the mass. The lesion appeared as a well-demarcated bluish-gray mass with a thick capsule, which was well vascularized (Fig. 3A). It had feeding and draining vessels from a relatively normal-appearing cho-

FIG. 2. Preoperative MR image (A) showing a T2 isointense lesion within the right trigone with relative dilatation of the trigone. Preoperative contrast-enhanced T1-weighted MR image (B) showing heterogeneous enhancement.

roid plexus. There was a robust vascular pedicle attached to the lesion (Fig. 3B). This was likely the choroidal artery feeding the glomus of the choroid plexus, but it may have been supplying the lesion as well. Initially, the capsule was coagulated and opened sharply, revealing a chronic liquefied dark brown hematoma (Video 1).

VIDEO 1. Intraoperative video of resection of a CEIVH. Copyright Jeremy Wetzel. Published with permission. [Click here to view.](#)

The mass was circumferentially dissected, and it separated easily from the ependymal surface, which was hemosiderin stained. The vascular pedicles were coagulated and cut, and the mass was removed en bloc.

Postoperative Course

The patient was discharged on postoperative day 3. He suffered no neurological deficit. Postoperative MRI revealed gross-total resection (Fig. 4). Interestingly, he had at least two temperatures recorded as $> 39^{\circ}\text{C}$ each day prior to surgery and not a single temperature $> 38^{\circ}\text{C}$ postoperatively. Again, the significance of the fevers is unknown, but they were believed to be incidental and unrelated to the lesion.

Pathological Findings

Gross specimen examination revealed a soft, tan-brown fibrous mass. On H & E staining, there were regions with organized hematoma with fibroblasts (Fig. 5A). There was a choroid plexus stalk containing large blood vessels, meningotheelial clusters, and a blood-filled cavity lined with hemosiderin-stained fibroblasts and chronic inflammation. Some of the large vessel walls had extensive hematoidin deposition. Psammoma bodies were seen (Fig. 5B). Cholesterol clefts appeared in areas representative of xanthogranulomatous change (Fig. 5C). No source of hemorrhage was seen. No neoplastic tissue was identified, nor were any malformed capillary caverns indicative of a cavernous malformation.

Discussion

Here, we present the first case of CEIVH in a pediatric

FIG. 3. Intraoperative microscope photographs. **A:** Intraoperative view after opening ependyma and entering the ventricle. Asterisk indicates normal ependyma. Arrow points to the lesion with its blue-gray pseudocapsule with neovascularization. **B:** Intraoperative view into the trigone after resection of the mass. Asterisk indicates hemosiderin-stained ependyma. Arrow points to the divided vascular pedicle, likely representing the choroidal artery.

patient in the modern neuroimaging era.²⁶ Only 5 other cases of CEIVH have been reported in the literature (Table 1). While there are approximately 60 cases of CEIH in the literature, an intraventricular lesion of this sort is a rare phenomenon. Below we discuss the definition, etiology, presentation, radiological features, operative considerations, and pathology of these lesions.

Intracerebral and intraventricular hematomas are common diagnoses seen by the neurologist and neurosurgeon, generally occurring in the setting of long-standing untreated hypertension or rupture of a vascular malformation.¹¹ Immediately after hemorrhage occurs, platelets in contact with tissue factor begin to form a clot. Ultimately, immune cells and fibroblasts migrate into the clot and initiate an inflammatory response.⁵ Granulation tissue forms around the hemorrhage periphery, which may contribute to the peripheral hyperdensity seen on CT.^{5,29}

The majority of these clots go through a process whereby they become liquefied and resorbed over 6–8 months.^{9,24} Rarely, intracerebral hematomas will become encapsulated in a process that has been theorized to be similar to the formation of a chronic subdural hematoma.^{19,24,25} It is hypothesized that in the subacute phase of a slow hemorrhage, fibroblasts enter the intracerebral parenchyma and begin to form a capsule around the hemorrhage area.^{1,10} As the capsule wall forms, it becomes vascularized through chemical signaling shown to include vascular endothelial growth factor (VEGF).^{21,28} The friable vasculature of the hematoma capsule wall can re-hemorrhage, further adding to the volume of the hematoma core.^{4,15–17,24} It is important to note that the fibroblastic capsule is critical to the definition of CEIH and CEIVH, and in the absence of this capsule a lesion is not considered as such.²⁰

It has been postulated that one-half of all CEIHs first form due to a vascular malformation or microaneurysm.⁸ The typical culprit is a cavernous malformation because

of its indolent, slow hemorrhage pattern, but small and cryptic high-flow malformations have also been shown to create a CEIH.^{3,14} The overwhelming majority of these vascular malformations are angiographically occult, but diagnosis is made after histological analysis demonstrates thin-walled sinusoids with a single level of endothelium in a cavernoma or thick and thin-walled vessels of various calibers in an arteriovenous malformation.^{3,14} A CEIH is a known complication after incomplete obliteration of vascular malformations in stereotactic radiosurgery.^{7,21,28} Other postulated causes of CEIH have included vascular amyloidosis and subclinical hemorrhage after trauma.^{18,30} The use of antiplatelets and anticoagulation may have a role in the progression of CEIH and CEIVH.⁶ Hypertension may be associated with the formation of CEIH.^{5,16} No etiology is found for many cases of CEIH.²⁷ It is a largely benign growth until it causes significant neurological deficit or mass effect. Unique to the CEIVH is the development of

FIG. 4. **A:** Postoperative MR image showing total resection of the lesion. **B:** Postoperative MR image showing the surgical corridor to the temporal horn and atrium.

FIG. 5. Photomicrographs demonstrating an organized hematoma with infiltrating fibroblasts (A), psammoma bodies (B), and xanthogranulomatous change with cholesterol clefts (C). H & E, original magnification $\times 4$ (A and C) and $\times 10$ (B).

obstructive hydrocephalus, which may result in a smaller lesion becoming symptomatic sooner than it would in the parenchyma of the brain.

The presentation of CEIH and CEIVH largely varies depending on each lesion's location, size, and rate of growth. Patients present with seizures, hemiparesis, ataxia, and other focal neurological deficits.^{6,8,16,24} Moreover, common presentations include signs of increased intracranial pressure, such as headaches, papilledema, nausea, and vomiting.^{5,13} Our patient presented with headaches related to early entrapment of the temporal horn of the lateral ventricle, which resolved after complete resection. A CEIVH is probably more rare because a resolving liquefied hematoma is more readily absorbed through the ventricular system and rarely consolidates to a nidus large enough to undergo encapsulation. The age of patients presenting with CEIH or CEIVH is variable, though cases in pediatric patients seem to be quite rare. The reason for this is unknown, but perhaps the insidious growth of these hematomas causes them to take many years to become symptomatic.

Both CEIH and CEIVH can pose challenges from a radiological perspective because there are no characteristics on standard modern images that clearly distinguish them from neoplastic or vascular lesions. Prior to the modern imaging techniques of CT and MRI, skull radiography and ventriculography demonstrated only a vague hyperdensity in the brain parenchyma or ventricular system.^{2,20} Computed tomography may show a circumscribed intracranial lesion with mass effect.¹³ Magnetic resonance imaging will demonstrate a well-circumscribed, variably enhancing mass with T2 signal heterogeneity and perilesional edema.²¹ Given the high rates of vascular malformations in cases of CEIH and CEIVH, we do recommend some form of vascular imaging, such as MR angiography. Conventional angiography is unnecessary in most cases. An abscess can usually be distinguished on the basis of restricted diffusion, which CEIH does not normally display. Given the diagnostic uncertainty based on imaging alone, biopsy or resection will likely be indicated at some point. However, given the overall benign appearance of these lesions, it would not be unreasonable to offer observation

TABLE 1. Summary of cases of CEIVH in the literature

Case No.	Authors & Year	Age (yrs)/ Sex	Presentation	Imaging Findings & Lesion Location	Treatment/Outcome	Pathological Findings
1	Avol & Vogel, 1955	48/M	Headaches, nausea	Ventriculogram: hyperdensity in rt frontal horn of lat ventricle	Rt frontal craniotomy/ full recovery w/o deficits	Well-circumscribed organized hematoma
2	Avol & Vogel, 1955	14/M	Headaches, nausea, & vomiting	Ventriculogram: large hyperdensity in lt lat ventricle	Lt parietal craniotomy/ full recovery w/o deficits	Well-organized & fibrotic blood clot associated w/ vascular malformation of choroid plexus
3	Ooba et al., 2003	68/F	Gait disturbance, hemiparesis, on warfarin	MRI: dumbbell-shaped mass in body of rt lat ventricle, T1 isointense, T2 mixed intensity, heterogeneous contrast enhancement	Rt frontal craniotomy/ full recovery w/ resolution of gait disturbance	Organized hematoma & granulomatous tissue
4	Kechagias et al., 2010	30/M	Fever, photophobia, diplopia, cervical pain	MRI: circular mass from roof of third ventricle, T1 & T2 hyperintense w/ ring enhancement	Rt frontal craniotomy/ full recovery w/o deficits	Hematoma encapsulated by accumulation of fibroblasts, granulation tissue w/ neovascularization
5	Dey et al., 2011	47/F	Numbness, incoordination of lt hand	MRI: bilat multilobulated masses in atria of lat ventricles, T1 & T2 hyperintense w/ heterogeneous contrast enhancement	Rt parietal craniotomy/full recovery w/ resolution of sensation in lt hand	Firm capsule w/ hematoma of different ages, cholesterol crystals, calcification & thin-walled capillaries c/w cavernous malformation

c/w = consistent with.

with serial imaging in an asymptomatic patient and to intervene only in the setting of rapid growth or symptom onset. We believed that resection was indicated in our patient given the headaches and early entrapment of the temporal horn. Resection has been shown to be safe and curative. Of 57 total cases of CEIH reported in the literature, 55 were surgically removed with good results. Approximately three-quarters of the patients had no deficits following surgery, and 14% were reported to have a persistent neurological deficit, but it is unclear how many of these patients had a preoperative deficit.²⁶ Table 1 shows that among the CEIVH cases reported, all patients had successful surgical removal of the mass without postoperative deficits, as in our present case.

Choroid plexus papilloma and intraventricular cavernoma were the diagnostic considerations in our case given the patient's presentation, age, and neuroimaging findings. Preoperative MR angiography revealed a large right anterior choroidal artery but no underlying vascular malformation.

It is generally thought that these lesions do not regress on their own; we found only one case of spontaneous resorption of a CEIH.⁸ Surgery should not be sought in asymptomatic patients; however, if symptoms can be clearly tied to the lesion, surgery should be considered. Our operative video demonstrates the ease of removing a CEIVH given the nice tissue plane between the capsule and surrounding brain.

Histological examination is necessary for diagnosis of this entity. Grossly, the mass is usually well circumscribed and tan in color.¹⁰ There is a capsule with granulation tissue and fibroblast proliferation. Occasionally, there can be findings consistent with a cavernoma or arteriovenous malformation inside the encapsulated mass. Roda et al. theorized that a CEIH initially caused by a vascular malformation could lose gross or histological evidence of the malformation secondary to degradation of the malformation in the hematoma core over time.²⁵ There is always deteriorating hemorrhage volume with various stages of blood breakdown in the capsule.⁴ In our patient, there was an organized hematoma with infiltrating fibroblasts (Fig. 5A). There were psammoma bodies (Fig. 5B), which can also be seen in normal choroid plexus histology.¹² Xantho-granulomatous changes are possible within the hematoma core (Fig. 5C).

Conclusions

Both CEIH and CEIVH are benign intracranial masses that are extremely rare in pediatric patients and have a presentation that depends on lesion location and neuroimaging findings, which are nonspecific. Resection is recommended in symptomatic patients.

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Disclosures

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Author Contributions

Conception and design: Wetzel. Analysis and interpretation of data: Wetzel, Wrubel. Drafting the article: all authors. Critically revising the article: all authors. Reviewed submitted version of manuscript: Wetzel.

Supplemental Information

Videos

Video 1. <https://vimeo.com/254901372>.

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