Child's Nervous System
https://doi.org/10.1007/s00381-021-05042-w

CASE REPORT

Porencephalic cyst after endoscopic third ventriculostomy and Ommaya reservoir placement: case report and review of the literature

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Received: 9 December 2020 / Accepted: 8 January 2021

Abstract

Background A 13-year-old female patient was diagnosed with a tectal glioma (TG), a subgroup of astrocytoma that can result in obstructive hydrocephalus secondary to aqueductal stenosis. Endoscopic third ventriculostomy (ETV) is used to treat this type of hydrocephalus with a good success rate. Our institution performs ETV and Ommaya reservoir (OR) placement in these cases. The OR allows measurement of intracranial pressure (ICP) and cerebrospinal fluid (CSF) access and a method for performing ventricular dye studies to evaluate third ventricular stoma (TVS) patency. In this case, a porencephalic cyst (PC) developed around the OR's ventricular catheter (OVC) two and a half months after surgery.

Conclusion The PC is thought to have developed in association with TVS stoma closure and resolved after ETV revision.

Keywords Endoscopic third ventriculostomy · Ommaya reservoir · Porencephalic cyst · Aqueductal stenosis · Hydrocephalus · Tectal glioma

Abbreviations

TG	Tectal glioma
ETV	Endoscopic third ventriculostomy
OR	Ommaya reservoir
OVC	Ommaya reservoir ventricular catheter
TVS	Third ventriculostomy
EVD	External ventricular drain
ICP	Intracranial pressure
CSF	Cerebrospinal fluid
PC	Porencephalic cyst
VS	Ventricular shunt
CT	Computed tomography
MRI	Magnetic resonance imaging

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Introduction

Porencephalic cysts (PC) are cerebrospinal fluid (CSF) filled cavities in the brain parenchyma often with ventricular communication [5, 14]. PCs can be found as a result of parenchymal injury secondary to vascular or genetic defects [6, 7, 26, 28], as well as trauma, radiation, and chemotherapy [16, 30]. Ventricular shunt (VS) malfunction has been associated with PC formation [13]. In some cases, ventricular non-compliance is thought to contribute to PC formation [21]. Treatment of a PC is tailored to the specific etiology and symptoms that are present in each individual patient.

We present a 13-year-old female who developed a right frontal PC after endoscopic third ventriculostomy (ETV) and Ommaya reservoir (OR) placement for tectal glioma (TG)–related hydrocephalus.

Case report

History, examination, and imaging

A 13-year-old female presented to the emergency room after routine ophthalmologist evaluation disclosing papilledema. The patient had headaches the preceding year approximately

Published online: 13 January 2021

twice a month. There was also an episode in which she may have passed out, and her school performance was worsening over the last few months. On exam, the patient was awake and alert, with no motor, sensory, or cranial nerve deficits. Magnetic resonance imaging of the brain (MRI) showed a TG with hydrocephalus and transependymal edema.

Operation

The patient went urgently to the operating room for treatment of hydrocephalus. Endoscopically, a blunt probe was used to perforate through the floor of the third ventricle and Liliequist membrane. A right frontal OR was placed through the same access tract. Lateral to this, an external ventricular drain (EVD) was inserted. The EVD was removed after intracranial pressure (ICP) was normal for 48 h. The patient was discharged home.

Follow-up

The patient presented for evaluation two and a half months post-operatively with a routine MRI (Fig. 1), demonstrating development of a right frontal PC and increased transependymal flow. An intraventricular computed tomography (CT) dye study revealed a non-patent third ventricular stoma (TVS) (Fig. 2). The patient was taken to the operating room where TVS closure was confirmed. She underwent ETV revision, placement of EVD, and removal of the OR. Immediate post-operative imaging revealed a patent TVS and a decrease of the PC. Sixteen-month follow-up imaging demonstrated decreased ventricular and PC size, with no transependymal flow (Fig. 3). She remains asymptomatic and doing well in school.

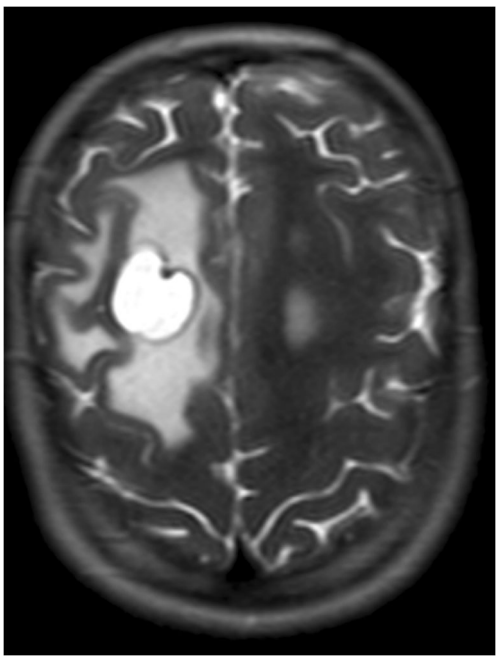

Fig. 1 Porencephalic cyst. Axial T2-weighted magnetic resonance imaging (MRI) of the brain showing a 2.6-cm right frontal PC. The right frontal Ommaya catheter can be seen associated with it. There is a large amount of edema in the white matter surrounding the PC

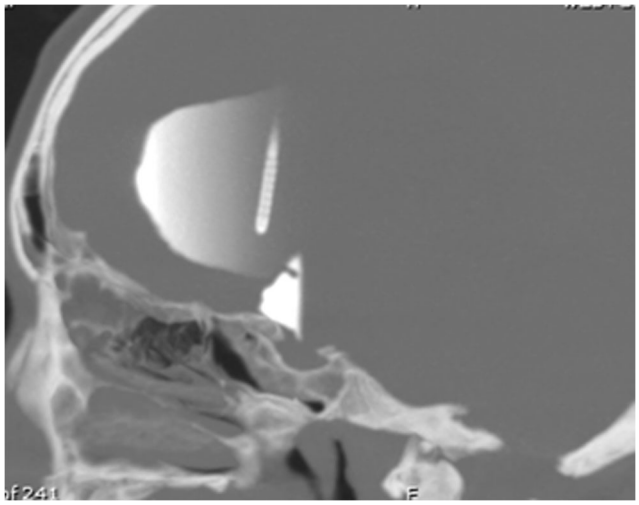

Fig. 2 Failed ETV. Sagittal computed tomography scan of the brain acquired after injection of 10 cc of Omnipaque 180 into the OR. No evidence of contrast opacification is seen beyond the third ventricle or in the basal cisterns

Discussion

PCs are CSF filled cavities in the brain parenchyma, usually contiguous with the ventricular system, with surrounding edematous gliotic and spongiotic white matter [5, 14]. The imaging modality of choice to evaluate these lesions is MRI [10]. Features include a non-diffusion restricting parenchymal

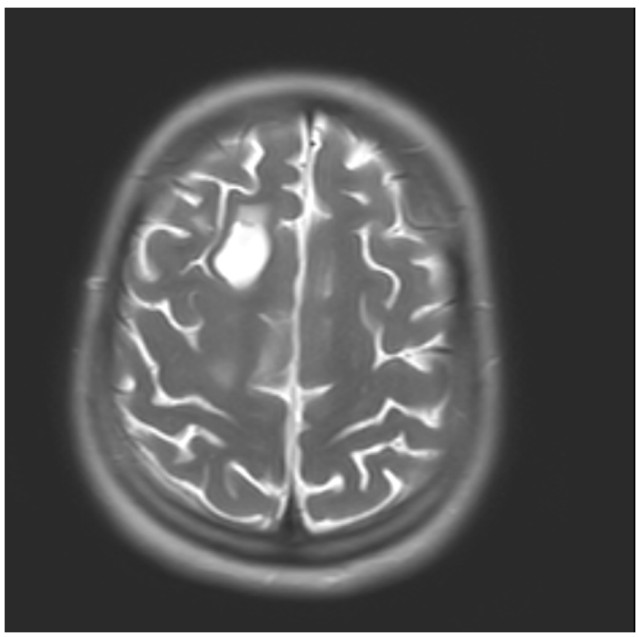

Fig. 3 16-month follow-up imaging. Axial T2-weighted MRI of the brain demonstrating decreased PC and edema at 16 months

cyst following CSF signal lined with white matter [28]. PCs can be found in neonates with an incidence of 3.5 per 100,000 live births, as a result of parenchymal injury secondary to vascular or genetic defects [6, 7, 26, 28]. Other conditions associated with acquired or idiopathic PC including trauma, radiation, and chemotherapy [16, 30]. Patients have a range of sequelae including seizures, developmental delay, language impairment, psychosis, and motor deficit [14, 15, 28].

VS malfunction has been infrequently associated with formation of PCs. A series of 208 children with CSF shunting reports 0.4% of shunt revisions were associated with a PC [13]. In patients with PC secondary to VS malfunction, symptoms of increased ICP can be seen [3, 8, 16, 18, 19, 21, 22]. In some cases, the ventricular size has been found to be stable, suggesting a degree of ventricular non-compliance thought to contribute to PC formation [21]. Treatment of a PC is tailored to the specific etiology and symptoms that are present in each individual patient.

A theoretical mechanism of PC formation involves CSF escaping from the ventricular puncture site and subsequently tracking to the parenchyma surrounding the ventricular catheter [10]. This CSF forms the cyst, particularly in a setting of increased ventricular pressure and well-formed intraventricular gliosis, resulting in decreased ventricular compliance [3, 8, 21]. Therefore, the PC and the associated edema is a phenomenon similar to periventricular subependymal interstitial CSF edema in patients with hydrocephalus [1, 21].

A literature review of PC in pediatric shunt-dependent cases with ventricular catheters yielded 12 cases (Table 1) [3, 8, 18, 19, 21, 22, 25]. The patients were aged 20 days to 10 years old. All cases, except for one that had valve adjustment, underwent VS revision [18]. We also found a series of cases that were ultimately not shunt-dependent and treated with removal of their ventricular catheters, including one Ommaya reservoir ventricular catheter (OVC) [2, 16, 23]. These were the exception and considered to have clinically compensated hydrocephalus. All cases had clinical improvement and interval decrease of PC.

ETV in patients with TG avoids permanent VS, which is associated with malfunction and subsequent revisions [4, 9, 20, 24, 27]. The success rate for treatment of this lesion as evaluated by the ETV success score is high [11]. The OR serves as a CSF diversion method should there be acute hydrocephalus secondary to TVS closure. The literature describes emergent access to the OR during such a crisis [12, 17, 29]. Given the possible benefit and relative lack of complications seen, ORs are commonly placed at the time of ETVs in our institution [17].

Table 1 Pediatric cases of PC associated with ventricular catheters

PMID (source)	Age	Clinical presentation	Hydrocephalus etiology	Treatment	Outcome
10912207 (Iqbal et al. [8])	10 years	2 months H/A, N/V, R arm weakness	s/p myelo-repair, VPS at birth	VPS revision	Clinical improvement, decreased cyst size
10912207 (Iqbal et al.[8])	10 years	5 days of headache, nausea, mild right paresis	Post-traumatic hydro	VPS revision	Clinical improvement, decreased cyst size
24610692 (Santín-Amo et al. [22])	7.5 years	Intermittent headache	Unclear	VPS revision	Clinical improvement, decreased cyst size
22053237 (Rim et al.[19])	1 month	Follow-up imaging	Neonatal hydro	VPS revision	Decreased cyst
29380111 (Park et al. [18])	6 years	3 days of malaise, lethargy	Obstructive hydrocephalus secondary to brainstem glioma	Shunt valve adjustment	Decreased cyst size
8027798 (Sakamoto et al. [21])	7 years	Subcutaneous fluid collection	Myelomeningocele	VPS revision	Decreased cyst size
8027798 (Sakamoto et al. [21])	7 years	Vomiting, subcutaneous fluid collection	Achondroplasia	VPS revision	Decreased cyst size
8027798 (Sakamoto et al. [21])	6 years	Drowsiness, vomiting	Occipital encephalocele	VPS revision	Decreased cyst size
7131072 (Chiba et al. [3])	20 days	Disconnected catheter	Meningitis	VSP revision	Decreased cyst size
7131072 (Chiba et al. [3])	2 years	Nonspecific increased ICP signs	Congenital	VPS revision	Decreased cyst size
1794120 (Sugimoto et al. [25])	5 months	Vomiting	Meningomyelocele	VPS revision	Resolution
1794120 (Sugimoto et al.[25])	3 years	Vomiting	Meningitis	VPS revision	Resolution
1794120 (Sugimoto et al.[25])	2 years	Vomiting	Congenital, Meningitis	VPS revision	Resolution

To our knowledge, this is the only PC described in association with failure of an ETV. There are two proposed mechanisms for formation of the PC in this case. In the first, the PC developed after TVS closure, preventing CSF diversion into the pre-pontine cistern. Instead, the CSF accumulated in the weaker peri-OVC parenchyma after tracking through the ventricular puncture site. An alternative explanation for PC development is that the OVC tract provided an alternative path for CSF diversion with less resistance compared to that seen in the TVS. As such, this decreased the pressure gradient and flow across the TVS, resulting in its ultimate closure.

Conclusion

Non-communicating hydrocephalus secondary to TG is frequently treated with ETV and OR. Routine follow-up imaging in our case revealed the development of a PC associated with the OVC. The patient was asymptomatic. Workup revealed a closed TVS. The OVC tract provided an alternative pathway for CSF as the TVS closed. The finding of PC after ETV and OR placement should prompt investigation into patency of TVS.

Code availability Not applicable.

Authors' contributions Drs. Dominguez and Tobias contributed to the study conception and design. The manuscript drafts were written by Drs. Dominguez, Smit, Kim, Eric Feldstein, and Boyi Li. The manuscript was critically revised by Dr. Tobias. Drs. Tobias, Kim, and Dominguez contributed to the care of the patient. All authors reviewed the submitted version of the manuscript and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Data availability Data sharing is not applicable to this article as no datasets were generated or analyzed for this manuscript.

Compliance with ethical standards

Conflict of interest The authors have no conflicts of interest relevant to this article to disclose.

Ethics approval As this is a case report of a single patient, our institutional review board has confirmed that no ethical approval is required.

Consent for publication Written informed consent was obtained from the parent of the patient, who consented to the submission of the case report to the journal.

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