*Original Article*

Exclusively endoscopic management of complicated pineal cysts in young children: Definitive treatment through single burr-hole technique

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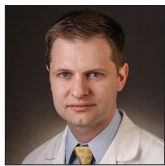
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ABSTRACT

Background: The management of complicated symptomatic pineal cysts in the pediatric population is challenging and variable. Surgical management may include treatment of hydrocephalus alone, or direct treatment of the cyst with or without direct hydrocephalus management. This is typically done through craniotomy-based microsurgical approaches to the pineal region or an endoscopic transventricular approach.

Methods: We present a stepwise minimally invasive technique to treat complicated pineal cysts in young children associated with an obstructive hydrocephalus in a single procedure through third ventriculostomy combined with an intraventricular marsupialization of the pineal cyst through a single burr-hole using stereotactic navigation.

Results: Two young patients with over 2 years of follow-up have done well without complication using this technique. Other literature reports for complex pineal cysts in pediatric patients are reviewed and this technique is not previously described for this population.

Conclusion: Endoscopic third ventriculostomy and cyst marsupialization using a single burr-hole and stereotactic navigation for symptomatic or enlarging pineal cysts in children allow for minimally invasive management, a rapid recovery, short hospital stay, and durable outcome owed to redundant CSF flow pathways.

Keywords: Complex pineal cyst, Endoscopic surgery, Endoscopic third ventriculostomy, Pineal apoplexy, Pineal cyst, Single burr-hole

INTRODUCTION

Pineal cysts are typically benign entities that are most commonly diagnosed in young women; the prevalence of pineal cysts is estimated at 2.4% (1.5% in males).[2] Some authors have postulated that pineal cysts may enlarge as a result of hormonal influences.[9,11,23] This may account for the sex differences seen in the age distribution of these cysts and can be a justification for serial observation of larger benign cysts through the post pubescent period. Pineal cyst etiology is not understood; the following theories have been proposed: (1) dysembryogenetic origin due to lack of obliteration of the cavum pineale,[5] (2) remnant of the embryological diverticulum that develops as an outgrowth from the floor of the third ventricle and forms the pineal gland (diverticulum pineale),[10,11] (3) degeneration of pinealocytes with cyst formation,[9,11] and

(4) ischemic necrosis and cyst formation.[12] Based on MR imaging studies, an estimated 4% of pineal cysts are >5 mm.[14] The vast majority of pineal region cysts are benign incidental findings and observational management strategies vary from no planned follow-up to serial imaging when detected in young children.

Complicated pineal cysts (defined here as symptomatic beyond isolated headache, causing mass effect, hydrocephalus, or papilledema) typically warrant surgical management, but likely owed to their rarity, no standardized management exists. There are limited reports in the pediatric literature regarding treatment of complicated pineal cysts. Published examples include observation only, endoscopic third ventriculocisternostomy (ETV) alone, external ventricular drain, ETV then craniotomy for cyst resection, ventriculoperitoneal shunt then craniotomy, and craniotomy only for cyst excision through supracerebellar-infratentorial approach [Table 1].[6,13,16,18,21,22] Due to anticipated hormonal changes and brain growth in pediatric patients, pineal cysts may be more likely to change than in adult patients.[2,15,23] Therefore, it is possible that this patient population may be more susceptible to late complications of mass effect if only the hydrocephalus is addressed. Furthermore, ETV failure is known to be higher in immature patients.[8]

Neuroendoscopic intraventricular techniques offer the advantage of treating or preventing any associated hydrocephalus by a third ventricular floor cisternostomy and can be done simultaneously with fenestration and marsupialization of the anterior cyst wall.[1,4,17,24,26] Given the benign histology of these lesions, this technique, which utilizes the principles of arachnoid cyst fenestration, offers definitive management despite incomplete cyst removal. With proliferation of frameless stereotactic technology and improvements in channeled flexible endoscopy, a single burr-hole technique can be achieved in pediatric patients and provide redundant protection against future complications by reintroducing bulk flow through the cerebral aqueduct and egress through an ETV in select cases.

MATERIALS AND METHODS

Herein, we apply a stepwise surgical technique based on our experience with management of two recent cases of complicated pediatric pineal cysts, including a rare case of pineal apoplexy, successfully treated with endoscopic third ventriculostomy with combined endoscopic cyst fenestration to restore normal flow to the cerebral aqueduct and also achieve cyst wall biopsy [Figures 1-4]. In addition, we review the technical literature on management of complex pineal cysts with a novel focus on pediatric cases. A Supplementary Video 1 is provided as well.

Table 1: Summary of complicated pineal cysts in pediatric patients from the literature review.

Authors (Year)	Age/Sex	Symptom and Sign	Treatment	Apoplexy Reported	Outcome
Musolino *et al.*,1993[20]	16/F	HA, vomiting, visual deficit	Stereotactic aspiration	Yes	Asymptomatic
	12/F	HA, vomiting, diplopia	Ventriculoperitoneal shunt, stereotactic aspiration	Yes	Asymptomatic
Di Chirico *et al.*, 2001[7]	16/F	HA, papilledema	ETV only	Yes	Asymptomatic
Michielsen *et al.*, 2002[18]	4/F	HA, lethargy, vomiting	Stereotactic endoscopic subtotal excision	Yes	Asymptomatic
Mandera *et al.*, 2003[15]	15/F	HA, gaze palsy	Craniotomy, total excision	No	Improved
	12/F	HA, blurred vision	Craniotomy, total excision	No	No improvement
	10/F	HA, vertigo, VI palsy, nausea	Craniotomy, total excision	No	Improved, HA persisted
McNeely *et al.*, 2003[16]	12/F	HA, syncope	Drainage, ETV, craniotomy total excision	Yes	Asymptomatic
Nimmagadda *et al.*, 2006[21]	10D/F	Macrocephaly	Observation	Yes	Asymptomatic
Majeed and Enam, 2007[13]	10/F	HA, gaze palsy	Ventriculoperitoneal shunt, craniotomy, total excision	Yes	Asymptomatic
Sarikaya-Seiwert *et al.*, 2009[22]	16/F	HA, papilledema	Craniotomy, total excision	Yes	Asymptomatic
	16/F	HA, impaired concentration	Craniotomy, total excision	Yes	Asymptomatic
Davidson, 2020[6]	3/F	Lethargy, development delay, vertical gaze palsy	ETV and cyst fenestration	No	Asymptomatic
	14/F	HA, fatigue, nausea	ETV only		Improved

ETV: Endoscopic third ventriculocisternostomy
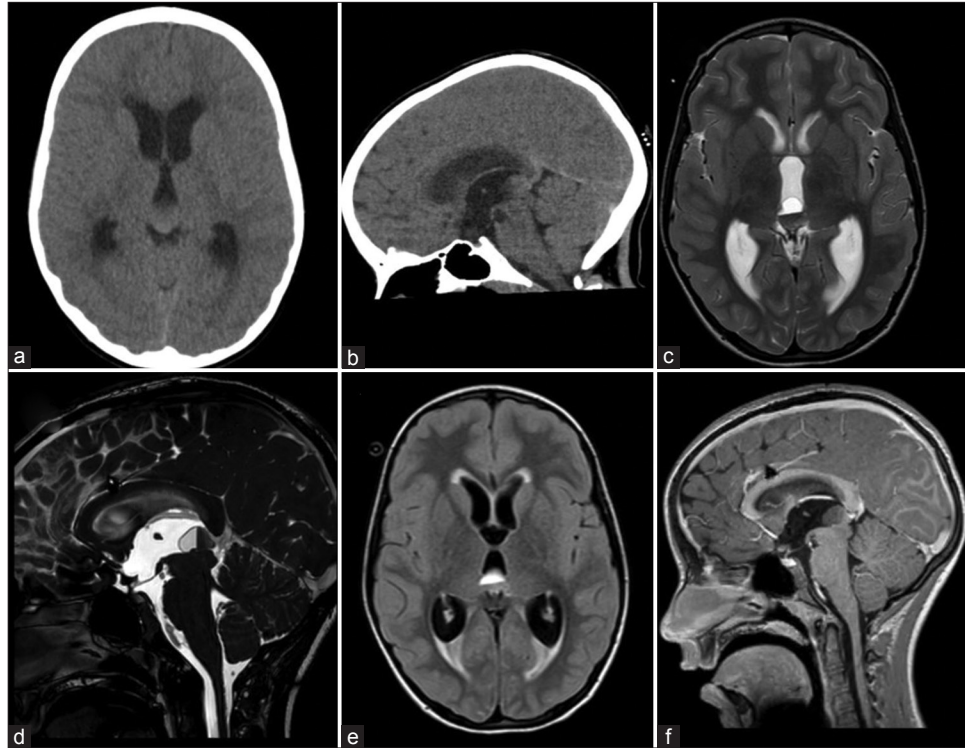
Figure 1: Case 1 preoperative radiographs. (a) Axial, (b) and sagittal noncontrasted CT head demonstrating lateral and third ventriculomegaly and a mixed density pineal region mass. (c) Axial T2, (d) sagittal T2, (e) axial FLAIR, and (f) sagittal T1 with contrast MRI of the brain revealing a nonenhancing 1.5 cm pineal region cyst with a dependent region of T1 hyperintense, T2 hypointense, with magnetic susceptibility, and consistent with hemorrhage. The ventral portion displays T1 hypointense, T2 hyperintense fluid collection, and consistent with blood components. The cerebral aqueduct is obstructed.
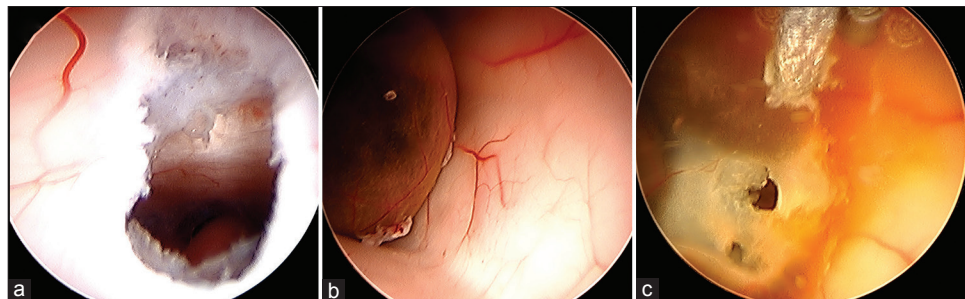
Figure 2: Case 1 operative endoscopic photographs. (a) Endoscopic view of third ventricle floor after ventriculostomy. The basilar artery is visible at the bottom of the field (posterior aspect of ventriculostomy), (b) Endoscopic view of the posterior third ventricle revealing a dark cystic mass that obstructs the cerebral aqueduct, (c) Coagulation of the cyst with copious debris, early cyst fenestration with cyst collapsing posteriorly, and revealing hemosiderin staining.

RESULTS

Case 1

Presentation

A 7-year-old girl with medical history of mild hereditary spherocytosis presented to the emergency department complaining of 2 days history of progressive headache associated with nausea, vomiting, confusion, and photophobia. Her initial head computed tomography (CT)

[Figures 1a and b] revealed moderately dilated third and lateral ventricles and a 1.5 cm pineal region mass.

Shortly thereafter, a right frontal ventriculostomy catheter was placed and kept open to drain at 20 cm H_2O. Her mental status dramatically improved post procedurally. Both AFP and beta-HCG were undetectable in CSF, and CSF cytology was normal.

After temporary CSF diversion, we completed magnetic resonance imaging (MRI) of the entire neural axis. This

revealed a mixed density 1.5 cm pineal region cyst, with dependent hemorrhagic components, and obstruction of the cerebral aqueduct [Figures 1c-f].

Given the patient's history of hereditary spherocytosis, she underwent a thorough workup led by our hematologic colleagues. Given a lack of anemia and asplenia, with only a mild reticulocytosis, and normal coagulation profile, her spherocytosis was deemed very mild and unlikely to be the cause of her pineal cyst hemorrhage.
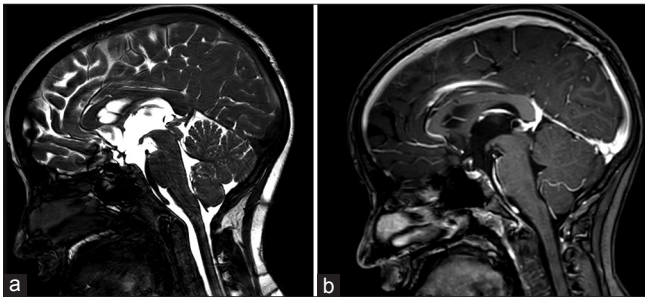

Figure 3: Case 1 postoperative radiographs (a) 15 months postoperative sagittal T2 MRI revealing a patent third ventriculostomy, a decompressed mixed-density pineal cyst, and a patent cerebral aqueduct, (b) T1 sagittal with contrast MRI brain revealing normal ventricular size and smaller cyst size.

Treatment

We considered various treatment options including craniotomy for pineal mass resection (especially with concern for possible hemorrhagic neoplastic tissue), observation of the cyst with ventriculoperitoneal shunt to manage her hydrocephalus, endoscopic third ventriculostomy, endoscopic cyst fenestration, or a combination thereof. We ultimately felt that the lesion was anatomically accessible through the Foramen of Monro for a frameless stereotactic endoscopic combined ventriculostomy and cyst fenestration with partial resection through a single entry point, a technique previously described in detail by Berhouma *et al.* in adults.[4] The patient tolerated the procedure without complication.

Outcome

Postoperative day 1 MRI revealed a patent third ventriculostomy, a decompressed mixed-density pineal cyst, a patent cerebral aqueduct, and normalized ventricular system. The ventriculostomy catheter was removed on postoperative day 1. She discharged home on postoperative day 3. The gross and microscopic pathology analysis found cystic pineal gland with numerous hemosiderin-laden macrophages consistent with pineal cyst lining without any elements to suggest neoplasm.
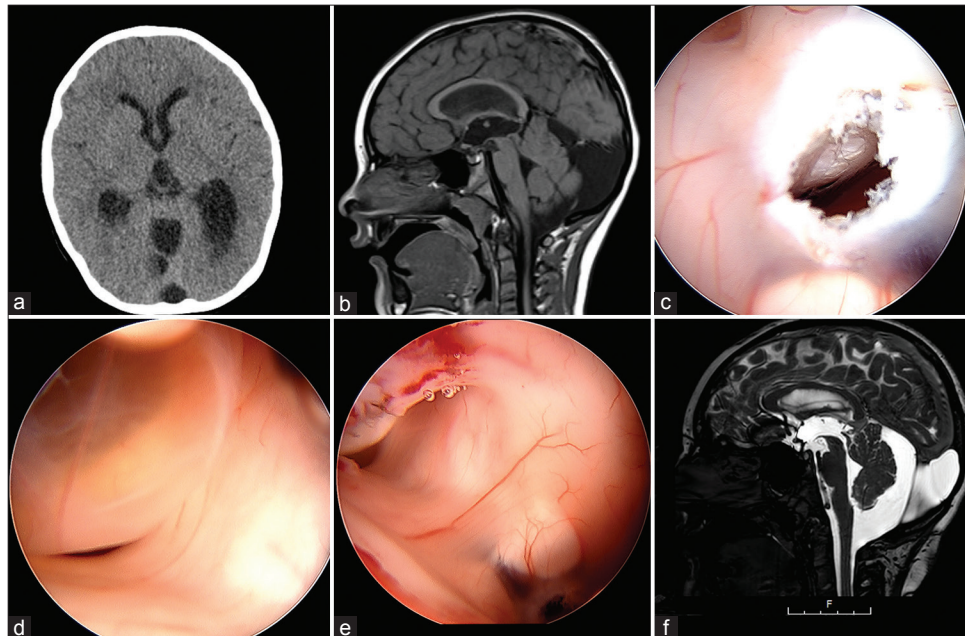
Figure 4: Case 2 (a) Axial noncontrast CT head showing the pineal region cyst is not discernible from the adjacent third ventricle CSF, ventriculomegaly, (b) Sagittal T1-weighted MRI image showing the pineal cyst and the superior portion of the tectum inferiorly displaced and partially occluding the superior cerebral aqueduct, (c) Endoscopic view of third ventricle floor after ventriculostomy. The basilar artery is visible, (d) Posterior third ventricle, revealing the pineal cyst that obstructs the cerebral aqueduct, (e) Fenestrated pineal cyst and the patent cerebral aqueduct, (f) 15 month postoperative sagittal T2-weighted image showing patent third ventriculostomy opening, decompressed pineal cyst, and patent cerebral aqueduct.

MRI at 3 months follow-up revealed normalized pineal appearance with stable normal ventricular size and persistent patency of the cerebral aqueduct that was persistent at 42 months [Figure 3].

She remains asymptomatic, continues to do well after surgery, and demonstrates normal development more than 3 years out from surgery.

CASE 2

Presentation

A 5-year-old male with severe developmental delay, attention deficit hyperactivity disorder, and autism presented through outpatient neurosurgical clinic, becoming increasingly difficult to control with behavioral outbursts. A CT and subsequent MRI were ordered, and he was found to have ventriculomegaly compared [Figure 4a-b] to a prior MRI 3 years earlier done for workup of developmental delay. This appeared to be related to aqueductal obstruction from an enlarging pineal region cyst that did not appear to be associated with a tumor or hemorrhage.

Treatment

Given the patient's young age and therefore greater potential for additional cystic expansion, CSF diversion alone might not address issues with pineal region mass-effect. We, therefore, considered strategies that would both eliminate cystic mass effect and ensure long-term normalization of CSF circulation without implants and again elected for the combined ETV and endoscopic cyst fenestration technique through a single frontal burr-hole entry point [Figures 4c-e].

Outcome

The patient was discharged home on postoperative day 3. Three month and 1 year postoperative MRIs revealed a patent third ventriculostomy, a decompressed pineal cyst, a patent cerebral aqueduct, and normalized ventricular system [Figure 4f]. This patient experienced significant improvement in behavioral issues, and while developmental delays persist, a marked rise in developmental performance including language development was noted shortly after surgery and has persisted over 2 years out from surgery.

Indications

The endoscopic transventricular approach to pineal cysts represent an excellent option in pediatric patients with hydrocephalus, as it allows both third ventriculostomy and marsupialization of the pineal cyst into the cavity of the third ventricle. Histological sampling is also feasible with minimal risks. Navigational planning software is helpful in determining the feasibility of a single burr-hole approach based on the individual patient's anatomy. Considerations for use of flexible endoscopy are reasonable here as well.

Surgical technique

At the time of these procedures, we were not aware of the manuscript by Berhouma *et al.* that describe this approach in adult patients, but our technique was indeed similar for these pediatric patients.[4]

Our patients were placed supine on the operating room table. The cervical spine was placed in slight flexion and the head is secured in a Mayfield three pin head holder with pediatric pins. A soft scapular bump was used to avoid excessive flexion. Navigation is especially helpful for locating a single frontal entry point that can provide a feasible working trajectory to both the floor of the third ventricle and the anterior membrane of the obstructive cyst. It is important to consider that as soon as the cyst wall is punctured, the cyst will begin to deflate, and the anterior wall may retrude posteriorly. The ideal entry point was marked on the scalp and should be anterior to the coronal suture. The area was prepped and draped and a small linear incision was made. Consideration can be made for a burr-hole slightly larger than one would for a typical ETV; we used a 9 mm burr-hole to allow for anterior-posterior angulation to reach the two different target points during this procedure. A cruciate durotomy was performed and a gyral surface was identified and the pia coagulated.

If navigation is used already for trajectory planning, we recommend the relatively simple additional step of securing a navigation guide to the endoscope so that the tip of the endoscope can be tracked. This can be of utility if visualization is temporarily compromised at any point and helpful for precise tracking through the cortex and white matter to stay aligned with the optimal trajectory into the lateral ventricle. We first proceed with the ETV procedure. Since the entry point is slightly more anterior than for a typical ETV to optimize access to the posterior third ventricle, an angled scope may be preferable. We used a 30 degree rigid endoscope for both ETV and cyst fenestration in these cases. When ETV is satisfactorily completed, the angled scope is turned toward the posterior aspect of the ventricle and slid under the massa intermedia. While it was not necessary in either of the cases described here, partial sectioning of the massa intermedia with sharp midline dissection can provide a less obstructed view if necessary. If the cyst is not easily reachable or there is concern for tension on the Foramen of Monroe, consider use of flexible endoscopy if available or if a secondary more anteriorly located burr-hole might provide better access.

We opened the anterior border of these cysts with a combination of blunt and sharp electrocautery probes. We

also applied cautery to the inferior aspect of the cyst wall, thus shrinking it posteriorly and superiorly away from the cerebral aqueduct [Figures 2 and 4]. Extrusion of dark brown fluid consistent with aging blood products was seen and grasping forceps were used to biopsy several regions of the cyst wall in case 1. We did this until the cerebral aqueduct was visibly patent and the cyst was well fenestrated in such a way that the remaining shrunken flaccid membrane would be unlikely to even periodically obstruct the aqueduct [Figure 4].

The combination of a young, thin, and pediatric skull with a slightly larger (9 mm) burr-hole can increase the risk of pseudomeningocele formation. In addition to an on-lay dural allograft, we placed a titanium (case 1) and resorbable (case 2) burr-hole cover.

Technical considerations

1. The entry point in the calvarium is slightly anterior to the one used for classical third ventriculostomy to allow access to the posterior third ventricle without excessive tension on the Foramen of Monro.
2. Stereotactic navigation is useful in cases, where single burr-hole is intended, especially for planning the most appropriate entry site and confirming that tension on the Foramen of Monro and fornix can be avoided. If line of site access does not appear feasible on stereotactic planning, flexible endoscopy may be an option.
3. Performing the third ventriculostomy before fenestrating the pineal cyst is preferable to avoid any bloody pollution of the CSF that can settle easily in the region of the tuber cinereum and limit endoscopic visibility.
4. A Fogarty balloon can be used to enlarge the ETV and/or the cyst fenestration site, especially if the cyst retrudes and can be difficult to reach with other instruments.
5. Coagulation of the cyst wall is done with low intensity to avoid adhesion to the cyst wall and subsequent bleeding.

Potential complications

Intraventricular hemorrhage may require temporary external ventricular drainage, meningitis, cranial nerve palsy, seizure, forniceal injury, diabetes insipidus, recurrence, additional hemorrhage, or regrowth of the cyst.

DISCUSSION

There is a surprisingly small body of the literature guiding management of complicated pineal cysts in young children. Existing literature that describe management of these lesions utilize various methods from observation, craniotomy for resection or fenestration, and endoscopic techniques [Table 1]. [7,13,16,18,20-22] Even patient selection can be complicated. While almost all patients reviewed here do have headache, they all had additional signs or symptoms as well. Given the high prevalence of headache and the typically idiopathic etiology of that symptom, we would not consider the combination of a noncompressive, nonapoplectic, and stable pineal cyst complicated in the setting of headache as an isolated symptom. There are arguments made for sometimes treating non complicated cysts with surgery, but such cysts would likely be less technically feasible with the approach described here and less likely to require the dual therapy that we have described.

Microsurgical total excision through an supracerebellar-infratentorial approach has been described as a curative method.[3,11,16,19,22,25,27] There have also been reports on successful stereotactic interventions. In these cases, only the contents of the cysts were aspirated, and one of these cases had a ventriculoperitoneal shunt placed 3 months before the aspiration.[20]

A report of four patients (one pediatric) with endoscopically treated pineal apoplexy and associated occlusive hydrocephalus due to aqueductal occlusion, without intervention for hydrocephalus, showed good results. In these cases, the cyst walls were removed as much as possible using endoscopy. The postoperative images showed persistent cyst remnants, but flow through the cerebral aqueduct was normal.[18] This same study compared endoscopically-treated patients with an open surgical arm (two patients operated through supracerebellar-infratentorial approach) and revealed that the endoscopic group had a shorter duration of surgery (30 min for endoscopic group, 4–5.5 h for open group) and mean hospitalization (3 days for endoscopic group and 19 days for open group).[18]

Ventriculocisternostomy is an additional option for occlusive hydrocephalus secondary to a pineal cyst.[6,16] In an illustrative case, a 12-year-old girl with pineal apoplexy and hydrocephalus was treated with endoscopic third ventriculostomy. Despite lasting cure of her hydrocephalus, follow-up imaging revealed an increase in the size of the unmanaged pineal cyst. The cyst was subsequently resected through a craniotomy.[16] Thus, despite management of hydrocephalus, the cyst may continue to grow without direct intervention. It should also be noted that long-standing aqueductal compression may result in persistent stenosis refractory to resection of the cyst.[22] Conversely, in another report of a complex pineal cyst, isolated endoscopic third ventriculostomy was done for CSF diversion and the cyst spontaneously decreased on follow-up imaging.[7]

In this study, we present a single burr-hole technique for ETV and cyst marsupialization using stereotactic navigation for the management of complicated pineal cysts in young children. This technique offers redundancy for hydrocephalus treatment in the growing child, permits nonaggressive management of the vascular cyst wall to prevent issues with mass effect and restore aqueductal patency, allows for tissue biopsy, minimizes the likelihood of future shunt dependence, and utilizes a skillset familiar to pediatric neurosurgeons.

CONCLUSION

Pineal cysts are typically benign incidental findings; however, complicated cysts that cause mass effect, hydrocephalus, and papilledema can be found in pediatric patients and warrant neurosurgical intervention. Pineal apoplexy with subsequent obstructive hydrocephalus is the rarest and most dangerous form with resultant acute neurological deterioration which can lead to sudden death. Most reported cases of surgically treated complicated pediatric pineal cysts discovered in this review involved apoplectic etiologies. To date, the pediatric literature on management of these cysts has been variable, but the majority of authors advocate for either total cyst excision through craniotomy or a combination of cyst surgery and hydrocephalus management. The transventricular endoscopic approach with stereotactic navigation allows simultaneous third ventriculostomy and cyst marsupialization to restore patency of the cerebral aqueduct and provides an opportunity to histologically sample the cyst wall and coagulate cyst edges to prevent further hemorrhage and minimizes the likelihood for future surgery. The patient selection to include complex cysts that project into the third ventricle and stereotactic planning to assess single burr-hole access feasibility is key.

Declaration of patient consent

Patients' consent not required as patients' identities were not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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MULTIMEDIA
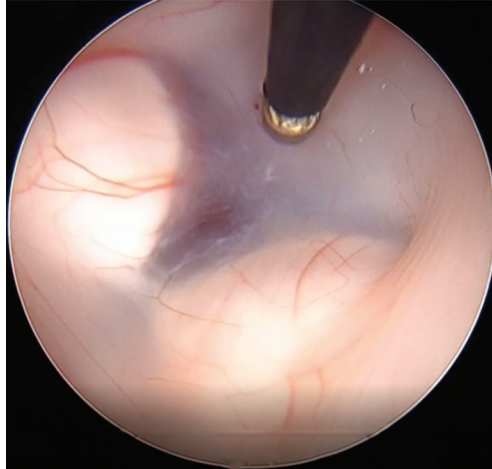
Supplementary Video 1: Endoscopic third ventriculostomy and pineal cyst marsupialization.

Annotations

00.32 – Using the endoscope, entering through Foramen of Monro, using the electrocautery, Fogarty balloon, and grasping forceps for the ETV.

00.42 – Exposure of the pineal cyst, using blunt and sharp electrocautery for cyst fenestration.