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Endoscopic treatment of intracranial cysts in infants: personal experience and review of literature

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Abstract

Background A wide variety of intracranial cysts is known to occur in infants. If symptomatic, they require treatment; the ideal surgical treatment and indications of surgery are yet a matter of discussion. Traditional treatment is either by cystoperitoneal shunting, or microsurgical fenestration. Endoscopic treatment is an alternative procedure that avoids the invasiveness of open craniotomy and the complications caused by shunting.

Methods This article reviews the endoscopic treatment of intracranial cysts in infants. The author presents personal experience by reviewing the results of endoscopic treatment in different subgroups among his series of pediatric patients extending over 20 years.

Results Different types of intracranial cysts in infants were discussed and the role of endoscopy in the management of these patients was reviewed. The author also presented the results of endoscopic treatment of a personal series including 87 infants with intracranial cysts operated by the endoscopic procedure.

Conclusions It has been recommended to use the endoscopic procedure in the treatment of intracranial cysts in infants, because it is effective, simple, minimally invasive, and associated with low morbidity and mortality rates. However, an important prerequisite is the presence of an area of contiguity with the subarachnoid cisterns and/or the ventricular system.

Keywords Arachnoid cysts · Endoscopic fenestration · Loculated hydrocephalus

Abbreviations

AOS	Aqueduct of Sylvius
CPS	Custoperitoneal shunting
CSF	Cerebrospinal fluid
CT scan	Computed tomography scan
IPACs	Intraparenchymal arachnoid cysts
IVECs	Intraventricular ependymal cysts
MCFACs	Middle cranial fossa arachnoid cysts
QACs	Quadrigeminal arachnoid cysts
SACs	Suprasellar arachnoid cysts
VC	Ventriculocystostomy
VCC	Ventriculocystocisternostomy

Introduction

Variable intracranial cysts can occur in children, and their differentiation is important in order to choose the proper treatment option [1]. Intracranial cysts are classified into two groups; the first group includes cysts derived from the central nervous system such as arachnoid cysts, ependymal cysts, loculated hydrocephalus, choroid plexus cysts, and porencephalic cysts. The second group includes cysts resulting from the intrusion of non-nervous tissue into the neuroaxis, usually in the midline, such as colloid cysts and Rathke's cleft cysts [2].

Conventional treatment is either by cystoperitoneal shunting (CPS) or microsurgical fenestration (with or without partial cyst excision). Endoscopic treatment is an alternative procedure that can be used; it avoids the invasiveness of open craniotomy and the complications caused by shunting. Intracranial cysts in infants are known to be more challenging for neurosurgeons than cysts occurring in older pediatric age groups. Data about surgical outcome of endoscopic treatment of intracranial cysts in adults and older children have been well reported; however, in infants, such data is

Important note The material included in this study had been previously published in different subgroups among separate publications.

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still sparse. There is paucity of information available in the literature about endoscopic treatment of intracranial cysts in infants.

This article reviews the endoscopic treatment of intracranial cysts in infants, whether intra-arachnoid, intraventricular, or intraparenchymal. Cystic lesions or neoplasms and parasitic diseases are excluded from the discussion. Other types of intracranial cysts will be also excluded, either because there are prevalent in adults (e.g., colloid cysts), they are not candidates for endoscopic treatment (e.g., porencephalic cysts), or they are usually symptomless and considered to be an incidental finding (e.g., choroid plexus cysts, Rathke's cleft cysts, cavum septum pellucidum). Pathogenesis, symptomatology, radiological features, and details of surgical techniques are beyond the scope of this discussion. The author will also present personal experience by reviewing the results of outcome of endoscopic treatment of intracranial cysts in infants stressing on some important technical considerations [3–10].

Discussion

Endoscopic treatment in infants

The clinical presentation of intracranial cysts in infants may offer considerable insight into the relationship between these lesions and underlying aberrancies in CSF flow. Many authors had postulated a correlation with hydrocephalus; however, the precise relationship between both entities remains to be fully elucidated [11].

Controversy still exists regarding whether infants have a higher risk of treatment failure after neuroendoscopic procedures for the treatment of hydrocephalus and intracranial cysts. An increase in intracranial pressure causing sutural diastases rather than flow of cerebrospinal fluid (CSF) via the fenestration might be a reason of failure. Another explanation might be the plasticity of the developing brain, which may lead to occlusion of the fenestration and opening of the cranial sutures [12]. It could be also explained by immaturity of the subarachnoid CSF dynamics in infants, as well as the deficiency of absorptive capacity of the subarachnoid space due to scarring of the arachnoid following intraventricular hemorrhage or meningitis [3].

Many authors reported favorable outcome with endoscopic procedures in infants with intracranial cysts [12, 13]. However, the number of patients was small, and if associated with hydrocephalus, postoperative shunt dependency still constitutes a main problem in the treatment of these patients. In a study including 15 infants who underwent neuroendoscopic surgery for the treatment of CSF space loculation and hydrocephalus, the incidence of shunt dependency was 46.7% [12].

Among 87 infants included in our series (48 boys and 39 girls), with mean age of 11.9 months, the endoscopic procedure was associated with postoperative clinical improvement in 71 patients (81.6%), and radiological improvement in 72 patients (82.8%). Postoperative complications occurred in 17 patients (19.5%). Among the long-term follow-up duration (44.8 months), recurrence was encountered in 21 patients (24.1%). The incidence of shunt dependency was 32.2% (Table 1).

Technical considerations

Endoscopic treatment of intracranial cysts in infants is of clinical importance and is considered to be technically more challenging than in older children. It is of particular importance to highlight specific nuances related to the management of these cysts at such young age. Although the indications of surgery are still debatable, it is well-known that infants

Table 1 Results of outcome and morbidity in a series of 87 infants with intracranial cysts operated by endoscopy

Type of Cyst	MCFACs	SACs	QACs	IPACs	IVECs	Uniloculated hydrocephalus	Multiloculated hydrocephalus	Total
Material	18	14	12	1	2	19	21	87
Age range (mean) mos	8–24 (15)	6–24 (13.6)	3–21 (13)	21	8–23 (15.5)	5–24 (13.6)	1–24 (7.9)	1–24 (11.9)
Boys/girls	12/6	8/6	7/5	0/1	1/1	11/8	9/12	48/39
Clinical improvement	16 (88.9%)	11 (78.6%)	11 (91.7%)	0 (0%)	2 (100%)	14 (73.7%)	17(81%)	71 (81.6%)
Radiological improvement	17 (94%)	12 (85.7%)	10 (83.3%)	0 (0%)	2(100%)	14 (73.7%)	17 (81%)	72 (82.8%)
Morbidity	4 (22%)	1 (7.1%)	3 (25%)	0 (0%)	0 (0%)	5 (26.3%)	4 (16.7%)	17 (19.5%)
Shunt dependency	0	0	2 (16.7%)	0 (0%)	0 (0%)	7 (36.8%)	19 (90.5%)	28 (32.2%)
Follow-up (mos)	69	53.9	43.6	22	60	52.4	30.4	44.8
Recurrence	3 (16.7%)	0(0%)	3 (25%)	0 (0%)	0 (0%)	9 (47.3%)	6 (28.6%)	21 (24.1%)

IPACs intraparenchymal arachnoid cysts, *IVECs* intraventricular ependymal cysts, *MCFACs* middle cranial fossa arachnoid cysts, *mos* months, *QACs* quadrigeminal arachnoid cysts, *SACs* suprasellar arachnoid cysts

are prone to cyst enlargement, hence the need of surgical intervention as early as possible. There is a general trend to encourage surgical interference in such age group (24 months old and younger), if the cyst increases in size or exerts brain compression even if there are no symptoms, because, at such age, the possibility of brain expansion is still available [14].

For endoscopic treatment in infants, it is important to be extremely cautious while doing the burr hole because skull bone is very thinned out in young infants who are few months old. One should also take greater care on how the dura matter is opened and closed; sometimes, it is very thinned out, and dealing with it should be done in a gentle proper way in order to avoid any inadvertent injury. The dura is opened in a cruciate way before introducing the endoscopic sheath, and edges are approximated at end of procedure, and better to be closed, if possible, by few stitches.

Every attempt should be taken in order to avoid intraoperative bleeding during endoscopy; children at such age are especially vulnerable to any amount of blood loss even if minor. Intraoperative identification of anatomical landmarks and proper selection of the target point of fenestration remains the most important factor that minimizes the incidence of any vascular injury with consequent intraoperative bleeding.

Arachnoid cysts

Intracranial arachnoid cysts are congenital CSF collections that occur between arachnoid layers [15]. These lesions are developmental in nature; however, trauma could play a role in cyst formation or enlargement [16, 17]. They are typically supratentorial, and approximately 50–66% are present in the middle cranial fossa [18]. Clinically 60–80% of these cysts are thought to be symptomatic [19, 20].

There is debate about indications of surgery among different authors; however, there is a general consensus to interfere surgically only if symptoms exist. It is our policy to operate only on symptomatic patients who have cyst-specific symptoms [21].

Although pure endoscopic procedure has been generally justified in the treatment of intracranial arachnoid cysts, notably, suprasellar arachnoid cysts (SACs), quadrigeminal arachnoid cysts (QACs), and intraparenchymal arachnoid cysts (IPACs), there remains considerable controversy regarding its role in the treatment of middle cranial fossa arachnoid cysts (MCFACs) [22, 23].

Middle cranial fossa arachnoid cysts

Endoscopically, MCFACs can be approached either through a transcoronal, transtemporal, or superior orbital approach. Treatment is based on making fenestration on medial cyst wall communicating it to subarachnoid spaces surrounding the brainstem (endoscopic cystocisternostomy). Target point of fenestration is chosen either between optic nerve and carotid artery (optico-carotid angle), between carotid artery and oculomotor nerve, or below the oculomotor nerve. This depends on the neurosurgeon's assessment during surgery, which one of these 3 areas has enough room for the endoscope to perform safe fenestration [5].

The process of fenestration is considered to be technically challenging as the space available for fenestration is small and surrounded by important neurovascular structures. The juxtaposition of critical neural and vascular structures to the medial cyst wall, the limited space available for fenestration, and the reliance on a single endoscopic instrument, all result in a procedure that can be intimidating. The use of neuronavigation has been advocated by many authors; since orientation within large MCFACs can be difficult, neuronavigation increases the precision and safety of the procedure [23]. Recently, navigated neuroendoscopy has been used together with intraoperative magnetic resonance cysternography [24].

A 7-month child with bilateral temporobasal arachnoid cyst operated by cystoventriculostomy with reservoir placement at site of burr hole showed significant postoperative clinical and radiological improvements [13]. In our cohort of patients with MCFACs, among 18 infants operated by pure endoscopy, postoperative clinical and radiological improvements were encountered in 88.9% and 94%, respectively [5].

Suprasellar arachnoid cysts

Due to the presence of hydrocephalus, there is a general consensus with endoscopic procedure as the primary option in the treatment of SACs [25, 26]. It has the advantage of not being invasive like open craniotomy, and it avoids the insertion of foreign bodies which carries the risk of malfunction [16, 27].

Suprasellar arachnoid cysts and colloid cysts are appropriate for endoscopy because they exist in anterior part of the third ventricle and because of the hydrocephalic changes they produce [4, 28]. Although endoscopic fenestration is considered to be the procedure of choice in the management of SACs, the choice of using either ventriculocystostomy (VC) or ventriculocysto-cisternostomy (VCC) is still a matter of debate. Our results proved that both procedures are equally effective clinically and radiologically; however, VC was associated with a higher incidence of recurrence (27.3% versus 0%, $P = 0.04$) [4]. Transventricular cystoventriculostomy through a classic precoronal burr hole was used in the treatment of a 10-month-old girl presented by involuntary head movements with postoperative resolution of the cyst and the hydrocephalus [13].

Quadrigeminal arachnoid cysts

Arachnoid cysts occurring inside cisterna quadrigemina are appropriate for endoscopy because they are usually in contact with the ventricular system and/or the subarachnoid cisterns and due to the presence of hydrocephalus. Little information is available in the literature about the role of endoscopy in the treatment of QACs especially in infants. Among 12 infants operated by pure endoscopy, postoperative clinical and radiological improvements were encountered in 91.7% and 83.3%, respectively [8].

Other types of arachnoid cysts

Endoscopy was also used and proved effective in management of other types of arachnoid cysts such as septal, interhemispheric, intraventricular, and infratentorial arachnoid cysts (e.g., cerebellopontine angle, foramen magnum, fourth ventricle). A 3-month-old infant with large interhemispheric arachnoid cyst associated with corpus callosum agenesis underwent endoscopic cystoventriculostomy through a classic precoronal burr hole and showed considerable postoperative clinical and radiological improvements [13]. Another infant (21-month-old girl) with left temporoparietal intraparenchymal arachnoid cyst was operated with endoscopic cystoventriculostomy without any clinical or radiological postoperative improvement [9].

Intraventricular ependymal cysts

Intracranial ependymal cysts are rare neuroepithelial cysts; they are less frequent than arachnoid cysts. They usually occur in periventricular area within the brain parenchyma. It is very rare to be found inside the ventricles, but if it occurs, trigone of the lateral ventricle is the preferred location [29, 30].

The cysts are sometimes discovered incidentally without any symptoms, but if they produce symptoms, surgical treatment will be necessary. These cysts may be communicated endoscopically with subarachnoid space, basal cisterns, or ventricular system, depending on its location. Inserting shunt inside these cysts, which contain proteinaceous fluid, will be usually followed by shunt malfunction and is not recommended.

Two infants (8 and 23 months old) with IVECs were operated by endoscopic cystocisternostomy and cyst-subarachnoid space communication, respectively, with postoperative clinical and radiological improvements [10].

Loculated hydrocephalus

Intraventricular septations result in the formation of isolated cavities or locules, and this condition is called loculated hydrocephalus or complex hydrocephalus. It is classified into two main types; the first type is uniloculated hydrocephalus if there is only one cyst inside the ventricular system. The second type is multiloculated hydrocephalus, if there are multiple cysts inside the ventricular system. Each one of these two types can be either supratentorial or infratentorial [3].

Loculated hydrocephalus is challenging for pediatric neurosurgeons, and there is no well-known treatment that is curative. Classic treatment is placement of multiple shunts which often requires many revisions [31, 32]. However, each consecutive shunt revision stimulates the formation of new membranes and further loculations, which adds to the complexity of the disease and diminishes the incidence of success of any subsequent treatment [33].

Stereotactic aspiration of the cysts was advocated by some authors [31, 34]. It results in cyst decompression and opening of CSF pathways which is beneficial in uniloculated cases. However, devascularization of cyst wall or providing adequate wide fenestrations is not feasible during stereotactic procedures; accordingly, it is associated with high incidence of recurrence and can not be justified in most cases [33].

Open fenestration via microsurgery carries the risk of sacrificing bridging veins which can result in venous infarctions. However, it provides excellent visualization of membranes and compartments, wider and deeper field of view, and better control of bleeding [35]. Microsurgical communication of the cysts can be performed through transcallosal approach [36] or transcortical approach [37].

Endoscopic treatment was found to be a better option than complex shunt systems or microsurgery. Endoscopic cyst fenestration is considered to be the procedure of choice in the treatment of loculated hydrocephalus, especially the multiloculated type which is the most difficult to treat [3]. It combines the ventricular compartments into a minimum number and reduces the number of proximal shunt catheters and shunt revisions [33]. It is a simple minimally invasive procedure with better postoperative recovery than microsurgery; patients recover on the ward and can be discharged next day; however, microsurgery requires a minimum 1 day stay in the intensive care unit [38].

Unliloculated hydrocephalus

It occurs if the lateral ventricle becomes dilated and isolated due to presence of non-colloid neuroepithelial cysts such as arachnoid, ependymal, and choroid plexus cysts obstructing the foramina of Monro [39]. It sometimes occurs in myelodysplastic children treated by low-pressure shunt inserted in the lateral ventricle.

The ipsilateral ventricle continues to over-drain, whereas the contralateral ventricle becomes dilated.

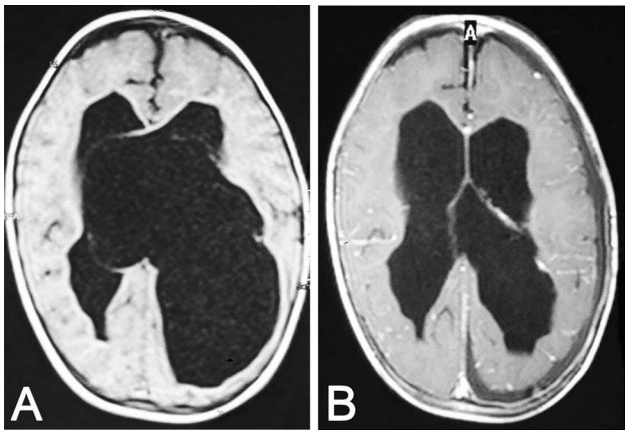

Fig. 1 Axial T1-weighted MR imaging of a case of uniloculated hydrocephalus. Left: preoperative, image showing huge intraventricular neuroepithelial cyst obstructing both foramina of Monro leading to hydrocephalus. Right: postoperative, performed 3 months after endoscopic fenestration, showing significant reduction in both cyst and ventricular size with restoration of ventricular architecture (reprinted from J Neurosurg Pediatr, El-Ghandour 2013) [7]

Implanting another shunt in the contralateral ventricle or upgrading the shunt valve pressure can be used as a treatment option [40]. Endoscopic septostomy can be also performed to treat isolated lateral ventricles, which connects both lateral ventricles in order to avoid inserting double shunts [6].

The results of endoscopic treatment of uniloculated hydrocephalus in infants were promising; postoperative clinical and radiological improvements were encountered in 73.7% of infants who had been treated by endoscopic cyst fenestration (Fig. 1) [7].

Isolated fourth ventricle

The fourth ventricle becomes isolated as a result of obstruction of aqueduct of sylvius (AOS) and outlets of the fourth ventricle; this may occur due to previous hemorrhage or meningitis leading to dilatation of the fourth ventricle with cerebellar and brainstem compression. The presence of shunt in the lateral ventricle worsens the condition, because it reduces the pressure required to keep the AOS open, as well as the supratentorial pressure. As a result, the fourth ventricle becomes dilated, whereas both lateral and third ventricles become collapsed [6].

Such condition can be managed by implanting a shunt inside the fourth ventricle, microsurgical canalization of the AOS, or microsurgical fenestration of outlets of the fourth ventricle. Endoscopic procedures such as aqueductoplasty (with or without stenting) have also been used with favorable outcome [41, 42].

Dandy-Walker syndrome

The presence of hydrocephalus associated with posterior fossa cyst and cerebellar dysgenesis is known as Dandy-Walker syndrome. This is a rare developmental anomaly which is usually apparent clinically during early infancy. It is attributed to atresia of the foramina of Luschka and Magendie causing hydrocephalus together with ballooning of the fourth ventricle. Maldevelopment of the cerebellar vermis occurs with posterior migration of the confluence of sinuses and tentorium [43]. The differential diagnosis includes mega cisterna magna and posterior fossa arachnoid cyst. The condition may be sometimes associated with aqueductal stenosis, resulting in lateral ventricular dilatation. Injection of metrizamide into the lateral ventricles at the time of computed tomographic (CT) scanning is important to diagnose if the fourth ventricular cyst is communicating with lateral ventricles or not. If such communication exists, CPS may be used as a treatment option; if no communication exists, a two-compartment shunting will be indicated. Endoscopic third ventriculostomy and choroid plexus cauterization have been also advocated [35].

Multiloculated hydrocephalus

Multiloculated hydrocephalus usually occurs in infants, if the ventricular system becomes encysted following germinal matrix hemorrhage or bacterial meningitis [32, 44]. Predisposing factors include perinatal complications, premature birth, low birth weight, and congenital malformations [45].

The results of endoscopic treatment of multiloculated hydrocephalus were encouraging; postoperative clinical

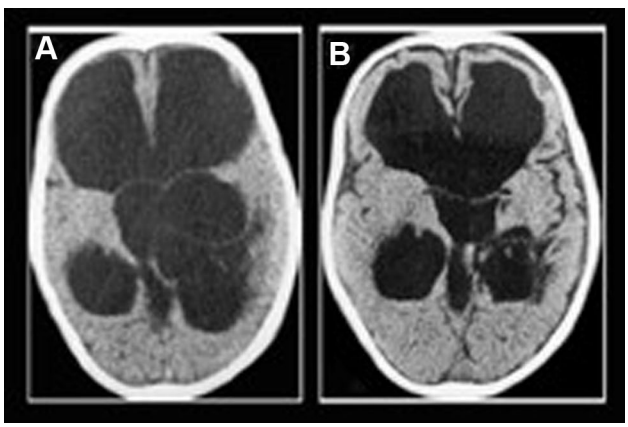

Fig. 2 CT scan axial images of a case of multiloculated hydrocephalus. **A** Preoperative, showing multiple cysts inside the ventricular system. **B** Postoperative, performed 3 months after endoscopic fenestration, showing improvement of hydrocephalus, opening of subarachnoid space, increase in cerebral mantle and restoration of ventricular architecture (reprinted from J Neurosurg Pediatr, El-Ghandour 2008) [3]

and radiological improvements were encountered in 81% of infants who had been treated by endoscopic cyst fenestration (Fig. 2) [3].

Endoscopic cyst fenestration provides wide communication between isolated different locules which avoids the need for additional shunt tubes. Because of the initial inflammatory process and the low-pressure differential existing across the cyst wall, which will consequently increase the incidence of postoperative reclosure, it had been recommended that the fenestration should be as wide as possible, so long that safety is ensured.

Concerning previously existing nonfunctioning shunts, endoscopy plays an indispensable role in dealing with these patients; it is usually very difficult to do shunt retrieval in multiloculated hydrocephalus without using endoscopy. Retained ventricular catheters are usually adherent to surrounding membranes; pulling such catheters without being dissected under endoscopic assistance is dangerous, and it might risk serious intraventricular hemorrhage.

Conclusions

The endoscopic procedure is recommended to be used in the treatment of intracranial cysts in infants; it is associated with favorable outcome and low incidence of morbidity and recurrence among the long-term follow-up. It is considered to be a better treatment option than shunting or microsurgery, because it avoids the invasiveness of open craniotomy and the complications of shunting. However, an important prerequisite is the presence of an area of contiguity with the subarachnoid cisterns and/or the ventricular system.

Declarations

Conflict of interest The author reports no conflict of interest concerning the material or methods used in this study or the findings specified in this paper.

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