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Loculated ventricles and isolated compartments in hydrocephalus: their pathophysiology and the efficacy of neuroendoscopic surgery

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Trapped cerebrospinal fluid (CSF) spaces can, on occasion, complicate the management of hydrocephalus and present the surgeon with a treatment dilemma. This condition can be categorized into one of two types: those arising as a complication of shunting and those that arise as a complication of an inflammatory process within the ventricles. Whatever the cause, the result is a significant escalation in the complexity of the management of the patient. Neuroendoscopy is typically viewed as an attractive treatment alternative in such a setting because of its minimalistic and thus seemingly simplistic nature. We have learned that nothing could be further from the truth. This article reviews the various entities that can arise in the hydrocephalic patient, how they can be managed endoscopically, and what sort of result can be expected.

Shunt-related entrapment

Oi and his colleagues [1–6] have previously reported that excess drainage of CSF via a ventricular shunt system causes morphologic changes in the CSF pathway and possibly leads to the isolation of compartments. Overdrainage, which produces slit-like ventricles, occurs most commonly in young infants and can occasionally lead to the slit ventricle syndrome [6,7]. The presence of slit-like ventricles can, in turn, lead to the development of an isolated ventricle [1]. The mechanism of obstruction at the foramen of Monro in isolated unilateral hydrocephalus and the mechanism of aqueductal obstruction in an isolated fourth ventricle (IFV) occurring after shunt placement are essentially the same [2,5,6]. Both occur in a previously communicating ventricular system, and a reduction in the size of all ventricles is seen initially after shunting in both cases [2,5].

It is well known that although slit ventricles occurring after shunting rarely produce the harmful or life-threatening state known as the slit ventricle syndrome, in some instances, slit ventricles can lead to other complications of shunting, such as isolated unilateral hydrocephalus (IUH) and IFV [1]. Oi et al [2] have reported an extremely high incidence of slit-like ventricles occurring after ventriculoperitoneal (VP) shunt insertion in younger infants and concluded that the main causative factors were high intracranial compliance coupled with overdrainage of CSF by the shunt. They analyzed 87 cases of infantile hydrocephalus, and CT scans done after shunting showed an extremely high incidence of slit ventricles (greater than 90% in premature neonates, 85% in neonates,

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42% in early infants, and 14% in late infants). They also found that there were few occurrences of subdural collection in neonates as compared with infants (10% in premature infants, 14% in neonates, 16% in early infants, and 28% in late infants) [3]. The difference in brain compliance and cranial structure were believed to account for the variance in slit-like ventricles and subdurals in the groups. Furthermore, in a follow-up study of 164 shunted hydrocephalic children of all ages without tumors, 46 (28.0%) developed slit ventricles, 5 (3.0%) developed IFV, and 4 (2.4%) developed IUH. All the patients with IUH and 3 with IFV had associated slit ventricles, 2 of whom had enlarged ventricles as a result of double-compartment hydrocephalus [1]. Fig. 1 shows a proposed classification of entrapments that can be seen in shunted patients [8].

Type 1 or IUH can arise when a shunt overdrains the CSF [1,2]. Functional occlusion of the foramen of Monro may result, which, in turn, leads to a progressive unilateral dilatation of the lateral ventricle [2]. This is seen when bilaterally enlarged ventricles are shunted and one shunted ventricle collapses, leading to an asymmetry in the ventricles. In most situations, the asymmetry equilibrates; however, on occasion, there can be a progressive enlargement of the lateral ventricle opposite the ventricle housing the shunt catheter, leading to IUH. In 1955, Bering [9] clearly showed evidence of pulse pressure as a generator of the force for ventricular enlargement as well as for CSF flow. Kaufman et al [10] found that the pulse pressure on the side of shunt placement was always lower than that of the other lateral ventricle. A diminished pulse wave caused by shunt placement should be considered one of the important causative factors in slit-like ventricle and the related morphologic changes seen after VP shunt placement.

If the aqueduct of Sylvius becomes obstructed in the setting of obstruction of the outlets of the fourth ventricle, isolation and expansion of the fourth ventricle occur (type II or IFV) [1,2]. Although the pathogenesis of IFV, especially after secondary aqueductal obstruction, remains unclear, studies by Oi et al [3–5] strongly suggested that there are two different mechanisms operating in the completion of fourth ventricle isolation. One mechanism is a "functional obstruction," which is created by a pressure differential being established between the supratentorial and infratentorial spaces. This arises when a shunt overdrains the supratentorial space, creating low supratentorial intraventricular pressure, pulse pressure, and brain compliance. Inadequate conveyance of CSF through the aqueduct does not allow for pressure equilibration, and IVF results. Raimondi et al [11] first described this type of isolation of the fourth ventricle caused by functional aqueductal occlusion in 1969. They considered this phenomenon to be the result of such a shunt-induced difference in

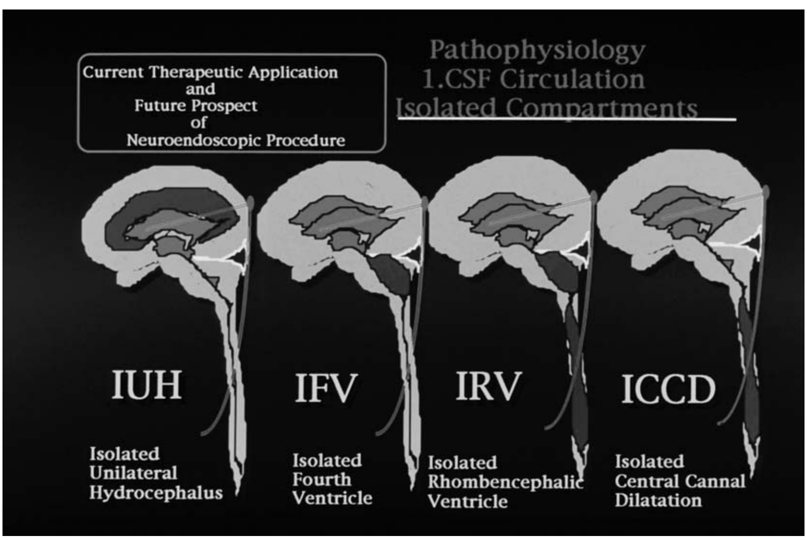

Fig. 1. Diagrams showing the development of postshunting isolated compartments in cases of previously communicating holoneural canal (see text for description). IUH, isolated unilateral hydrocephalus; IFV, isolated fourth ventricle; IRCV, isolated rhombencephalic ventricle; ICCD, isolated central canal dilatation.

pressure between the supra- and infratentorial compartments. It was their opinion that a shunt placed in the lateral cerebral ventricle created an unexpectedly low supratentorial intraventricular pressure by overdraining CSF. Consequently, they emphasized that this condition could be reversed by decreasing the pressure in the infratentorial compartment, which then causes the reopening of the aqueduct. In 1986, Oi and Matsumoto [5] described the phenomenon of reopening the aqueduct in cases of IFV by upgrading the shunt using a higher pressure valve in conjunction with an antisiphon valve to enlarge the slit-like lateral cerebral ventricle. De Feo et al [12] reported a case of "double compartment hydrocephalus" in a patient with a history of cysticercosis meningitis and suggested that the mechanism for the secondary aqueductal obstruction was an upward cerebellar veil displacement caused by increased infratentorial pressure. Oi and Matsumoto [5] reported on a series of patients with IFV and discussed the concepts of secondary obstruction of the aqueduct, analyzing CSF dynamics, pressure measurements, serial CT scan changes, and the outcome after various treatment modalities. Two distinctly different categories could then be identified in these cases: (1) functional obstruction in which the obstructed aqueduct could be reopened either by decreasing the elevated infratentorial pressure or by correcting the overdrainage of the supratentorial system and (2) permanent obstruction with a pathologic occlusion of the aqueduct, necessitating a fourth ventricle VP shunt [3,5].

Rarely, a trapped fourth ventricle can progress to a situation where there is an associated hydromyelia. Oi et al [8] reported seeing such patients in a series published in 1991. These investigators described patients with an isolated rhombencephalic ventricle (IRV) or type III isolation involving both the metencephalon and myelencephalon. These were patients with functioning ventricular shunts, and the ventriculography confirmed a communication between the fourth ventricle and the hydromyelia. These studies indicated that in some cases, the pathophysiology of hydromyelia was closely related to hydrocephalus. All these patients improved after draining the fourth ventricle with a shunt. Oi et al [8] then proposed a new concept of the development of an isolated compartment after shunting to explain the progression of the hydromyelia seen in these cases. When all the ventricles are in communication and the outlets of the fourth ventricle to the cistern are obliterated, the whole ventricular system expands, including the fourth ventricle, and a disproportionately large fourth ventricle (DLFV) can be seen. If the central canal of the spinal cord is in communication with the fourth ventricle, it too may become involved, and a type IV or holoneural canal dilatation (HNCD) results [8]. In this situation, various forms of ventricular system isolation may develop after a shunt is introduced, and overdrainage of CSF occurs. Most common would be the simpler IUH and IFV, but more in advanced cases, dilatation of both the fourth ventricle and the central canal of the spinal cord could develop, forming an expanded primary IRV. Alternately, only the central canal of the spinal cord could be involved in the dilation with sparing of the fourth ventricle (isolated central canal dilatation: ICCD). This led Oi et al [8] to propose a new clinical category of hydrocephalus, namely, hydromyelic hydrocephalus (type IV in Fig. 1).

Postinflammatory entrapment

Trapped cystic spaces can arise within the CSF spaces as the result of an inflammatory process such as can occur after an infection or hemorrhage. The result can be the entrapment of a ventricle because of occlusion of a tight passage or multiple loculations within the ventricular system caused either by a series of veils developing or, more commonly, by subependymal cyst formation.

The aqueduct of Sylvius and, to a lesser degree, the foramen of Monro are narrow and consequently vulnerable to scarring shut when subjected to an inflammatory process. The aqueduct, because of its greater narrowness and vulnerability to maldevelopment (so-called "forking"), is much more vulnerable to this. As a result, triventricular hydrocephalus is much more common than a unilaterally trapped lateral ventricle. If there is an associated blockage in the outlets of the fourth ventricle, a condition similar to the IFV discussed previously can occur. This "morphologic obstruction" of the aqueduct is the result of a secondary process unrelated to shunt function. The most common cause for this is infection, followed by intraventricular hemorrhage [13–17]. Arachnoidal and ependymal cysts can also occur within the fourth ventricle and lead to an obstruction of outflow of CSF [18–21].

It has also been theorized that a shunt can trigger an inflammatory reaction within the aqueduct of Sylvius, leading to its stenosis with subsequent entrapment of the fourth ventricle in individuals with communicating hydrocephalus [22]. Tumors within the posterior fossa and their treatment can also lead to isolation of the fourth ventricle [17].

A more generalized process can be set in motion after a perinatal intraventricular hemorrhage or a gram-negative ventriculitis occurring in childhood. In the former, parenchymal injury coupled with massive intraventricular blood can lead to scattered cysts, which can exert mass effect on surrounding structures. Gram-negative ventriculitis, conversely, can result in numerous subependymal cysts forming and growing over time, with each potentially acting as a mass lesion.

Treatment

Intracranial endoscopic surgery (neuroendoscopic surgery) has progressed remarkably over the last two decades, and various intracranial pathologic conditions are now considered treatable using this developing "minimally invasive" technology [23–26]. Over a period of more than half a century, the instruments of endoscopic surgery have greatly improved along with the dramatic advances in medical technology, and neuroendoscopic surgery is now a routine technique. Previous reports note that microforceps, lasers, saline torches, microballoons, and fine-rod semirigid endoscopes have been used in neuroendoscopic surgery for hydrocephalus [25–30]. The operative procedures achieved with these instruments included fenestration of septations within hydrocephalic ventricles or the septum pellucidum in isolated ventricles, fenestration of arachnoid cysts and tumor cysts such as that seen with craniopharyngioma, and placement of catheters in ventricles or intratumoral cystic cavities.

Neuroendoscopic surgery has been used to treat patients with various forms of isolated compartments with specific pathophysiology, including IUH (Fig. 2), IFV (Fig. 3), DLFV, IRV (Fig. 4), and loculated ventricles (LVs) [31].

In approaching these lesions, the first consideration is what type of scope to use. The advantage of a lens scope is the superior visualization of structures within its field of view, but its disadvantage is that because it is a rod, the area available for viewing is defined by the angulation of the lens at its tip and the limited amount of movement of the shaft that the surrounding brain parenchyma will tolerate. The flexible scopes with their deflectable tip allow for larger fields of view, but the conduction of the image from the tip to the camera by fiberoptics degrades image quality, resulting in a poorer image as compared with a lens scope. One option is to use both as advocated by Oi, who prefers to use the rigid-rod endoscope (5.6 mm in diameter) first to observe the intraventricular morphology and then the flexible steerable-rod endoscope for further therapeutic maneuvers.

Potentially, all the surgical instruments, including the laser, microforceps, microballoon, and monopolar coagulator, may be needed during the

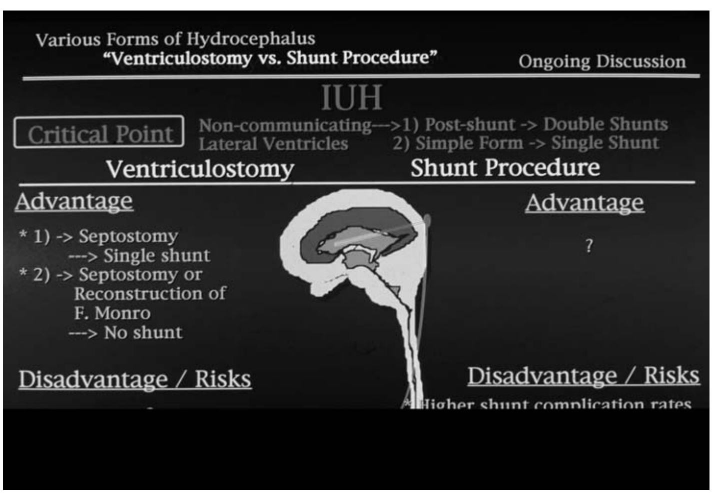

Fig. 2. Surgical procedures in the treatment of isolated unilateral hydrocephalus: neuroendoscopic ventriculostomy versus shunt procedure (advantages and disadvantages).

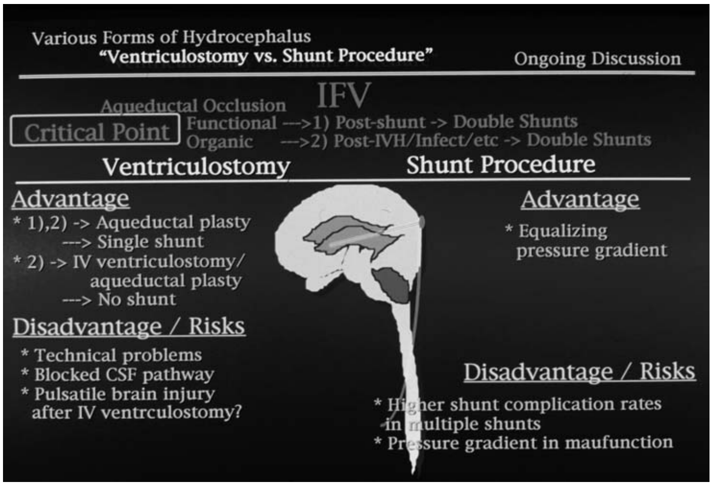

Fig. 3. Surgical procedures in the treatment of isolated fourth ventricle: neuroendoscopic ventriculostomy versus shunt procedure (advantages and disadvantages).

procedure; thus, steps should be taken to ensure that they are compatible with the scopes planned for use [23,24]. The presently available neuroendoscopic instruments for hydrocephalus include KTP/YAG lasers, monopolar and bipolar coagulators, microforceps (grasping and biopsy), scissors, knives, and microballoons. There are also assisting or supporting systems, including self-retaining retractors, endoscopic ultrasonography, and stereotactic systems [23,24]. The selection of instruments should be based on the surgeon's familiarity with the instrument and the goal of the procedure and should consider problems and needs that could occur during the procedure, such as the need to cut or coagulate tissue or to biopsy the tissue [23,24]. Supporting personnel should be familiar with the equipment to be used, including the camera and illumination system, and they should be competent in troubleshooting problems in the equipment, such as, for example, during procedures when malfunctions and equipment failure occur. Thought should have been given beforehand to location of back-up equipment, and contingency plans should have been made for irretrievable failure of equipment.

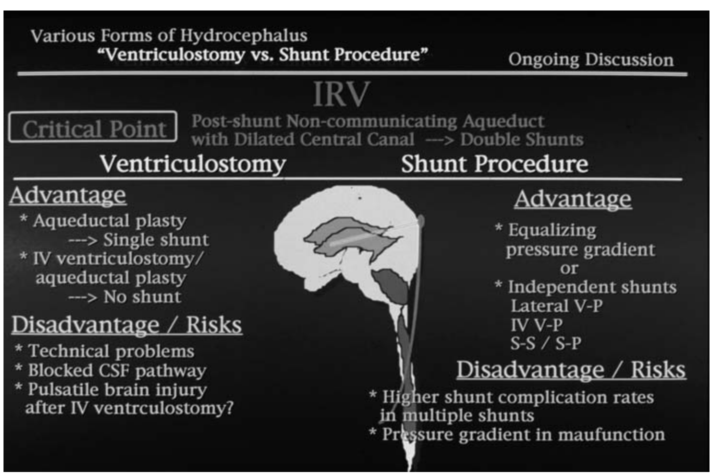

Fig. 4. Surgical procedures in the treatment of isolated rhombencephalic ventricle: neuroendoscopic ventriculostomy versus shunt procedure (advantage and disadvantages).

Time should be spent studying radiologic images of the patient's intraventricular anatomy. First, goals for the surgery are set. It may be too much to expect to fenestrate every subependymal cyst in a child with postinfectious multiloculated ventricles through a single burr hole or to safely fenestrate an enlarged lateral ventricle into an adjacent collapsed lateral ventricle. Location of the burr hole(s) is then determined with the surgical goals being kept in mind. Important here is to consider one's ability to remain oriented during the surgery. Moving in a straight line is far easier than in an intended path with many turns. Use of computer-assisted navigational equipment can be helpful in surgical planning, because one can establish a series of targets and then manipulate the three-dimensional image to establish the optimal location of the burr hole(s) and trajectory to accomplish the goals of the surgery (Fig. 5). It is of primary importance that the surgeon identify anatomic landmarks that are available to him/her during the surgery, which can be used for orientation.

Solitary cysts or entrapped ventricles

The goal in treating these solitary lesions is to drain them effectively so that they no longer exert a mass effect. This can be accomplished either by fenestrating them into an adjacent CSF space that is being drained effectively or by placing a catheter into them and shunting them to another space within the body.

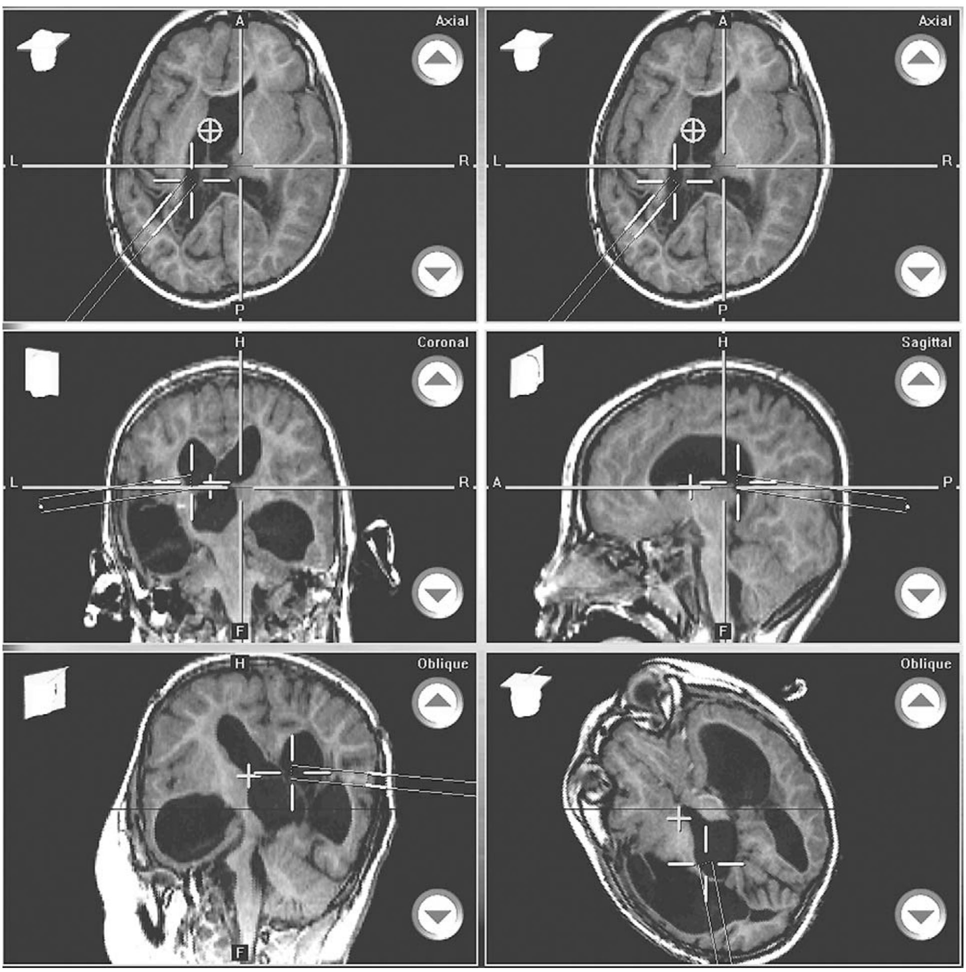

Fig. 5. Computer-assisted navigation of the working sleeve of the endoscope showing the sleeve just after it has been advanced through the cyst wall into body of the lateral ventricle (best seen on bottom two images).

Fenestration of a solitary cyst or entrapped ventricle requires that there be an accessible CSF space immediately adjacent to the space and that the endoscope can be safely maneuvered to a location where a fenestration can be done. These needs mandate that the surgeon carefully study the situation and plan out a safe approach that will accomplish the goals of the surgery. With regard to IUH, the safest approach is to fenestrate into the trapped ventricle and not out from it. It is vital to appreciate what is in the lateral wall of the lateral ventricle, namely, the internal capsule. When a ventricle is being compressed by a trapped contralateral ventricle, its width can narrow. The septum can deviate many millimeters before giving way to an instrument trying to fenestrate it. McLone [32] has reported on injury to the internal capsule on the contralateral hemisphere while attempting to fenestrate the septum pellucidum. The odds of such a mishap happening are dramatically lessened when coming from the normal ventricle into the enlarged trapped ventricle. Thought should also be given to structures and pathways when planning where to do the fenestration. With regard to the septum pellucidum, 1 cm above and anterior to the foramen of Monro should keep instrumentation safely away from the fornices.

Recently, there have been several reports of using aqueductoplasty to communicate a trapped fourth ventricle (either IVF or IRV) with the third ventricle [31,33–41]. An extremely ballooned fourth ventricle can sometimes be safely dealt with by fenestrating it into the cerebellar-pontine cisterns (Fig. 6). Key here is there being a wide enough point of abutment of the ventricle onto the cistern with no intervening brain parenchyma. Also key to success are anatomic landmarks that can guide the surgeon. Once again, it is easier to work from the normal to the abnormal, from the cistern into the ventricle in this case, because there are many more landmarks available for guidance in the cistern. Thought must be given to how these landmarks will be oriented and appear to the endoscope so as to avoid confusion. Care must be taken when doing the fenestration, with the surgeon bearing in mind the trajectory the instrument is taking and what is downstream. In many cases, study of the scans may lead the surgeon to determine that surgical fenestration is not the wisest approach. A shunting of the space may be preferred. In 1995, Lee et al [42] reported on the difficulty in maintaining a posterior fossa shunt, and in 1997, Arginteanu et al [43] reported on endoscopically placing catheters to drain trapped fourth ventricles using computer-assisted navigation from coronal burr holes. The stated advantage of this approach was better orientation of the catheter and confirmation of entry into the space.

Multiloculated ventricles

Ventricles containing numerous growing cysts is an extremely difficult situation to manage effectively. Simple drainage of the loculations using catheters is frustrating because of the multiplicity

Fig. 6. Entrapped ballooned fourth ventricle with adjacent giant cisterna magna.

and noncommunication of the cysts, their tendency to collapse down around the catheters obstructing them, and the tendency for new cysts to arise over time. One is guaranteed many trips to the operating room if it is elected to manage the condition with shunting alone. In the past, we were ultimately forced to consider open craniotomy for fenestration after playing a holding game waiting for the process of new cyst formation to mature and stabilize. This seemingly radical approach led many of us to welcome endoscopy as the ultimate answer for treating the condition, giving rise to an inappropriate hastiness in applying the technology to the condition. Surgeons entered the operating room with great expectations and a firm surgical plan based on naiveté only to find reality once the scope had been introduced into the cyst—there were no landmarks. Based on this early experience, most endoscopists will admit that this is one of the hardest conditions we are called on to deal with and that special strategies are needed to be effective.

Of primary importance is a strong sense of humility when setting out to deal with this condition. There is nothing wrong in telling a family that the procedure may need to be repeated several times to stabilize the condition. This is a result of the reality that this is an evolutionary condition with new cysts developing over time and that it is sometimes difficult to develop a plan where all cysts will be fenestrated along a single trajectory. Also, it can be difficult to appreciate what has been accomplished with the surgery. In this case, postoperative metrizamide CT can be illuminating (Fig. 7).

Good preoperative imaging is essential. MRI is superior, given the ability to reformat the orientation to appreciate the three-dimensional nature of the problem. Use of computer-assisted planning stations such as are available with the navigational systems can prove to be extremely helpful in visualizing the problem. Once a plan has been formulated, the patient is taken to the operating room. Positioning should ideally place the burr hole at the apex of the surgical field to prevent CSF egress from the head with a resultant distortion of the brain anatomy. Consideration should be given as to how computer-assisted navigation can be employed to accomplish the surgical goals. This can provide confirmatory information to lend credence to the impression of location formed by the appearance of the surgical field as viewed by the endoscope. Tracking systems can be used to either place a guidance tube for the scope on target or to actually track movements of the scope itself. Other tools that can be used to assist in orientation during the procedure include ultrasonography, whose use via a separate burr hole has been described and, more recently, intraoperative CT and MRI scanning. In the not too distant future, we will hear of image guidance coupled with such equipment in the operating room to improve the integrity of the guidance image set.

There are several methods for accomplishing the fenestration. A monopolar cutting current can be used either to make a series of punctuate cuts through the wall or to scroll-cut the surface. The laser can also be used, as can such specialty instruments as the saline torch [29]. Important to consider when using all these tools is what is on the other side of the wall and whether the energy of the device is confined to the surface being cut. Once a large enough opening has been made so that one can see through to the other side, and thus can navigate the scope through the opening, the scope is advanced through to the other side of the cyst wall and then moved side to side to further enlarge the opening. Work then proceeds as indicated.

Families should be warned when treating this condition that there can be a major pressure decompression with an attendant sense of disequilibrium and accompanying nausea for several days. Intravenous fluid might be required as well as bed rest until such symptoms resolve. Finally, they should be reminded that multiple surgeries may be required, this being the price of avoiding the major open craniotomy.

Results

In our early experience with these forms of hydrocephalus treated neuroendoscopically, reconstruction of the CSF pathway was attempted in patients with various forms of noncommunicating hydrocephalus. With regard to isolated unilateral hydrocephalus and loculated lateral ventricles, all patients experienced either stabilization or improvement after their endoscopic procedure, with most (9/13) showing a decrease in ventricle size. All were improved therapeutically. Trapped fourth ventricles proved more problematic. Only 1 of 4 patients was successfully managed with endoscopic fenestration. Shunting seemed to be the preferred technique for most of these cases as a result of the difficulty of the anatomy. Placement of the ventricular catheter from a coronal burrhole seems to improve the survival of the system, and there may be some advantage to doing this with endoscopic assistance. Multiloculated ventricles

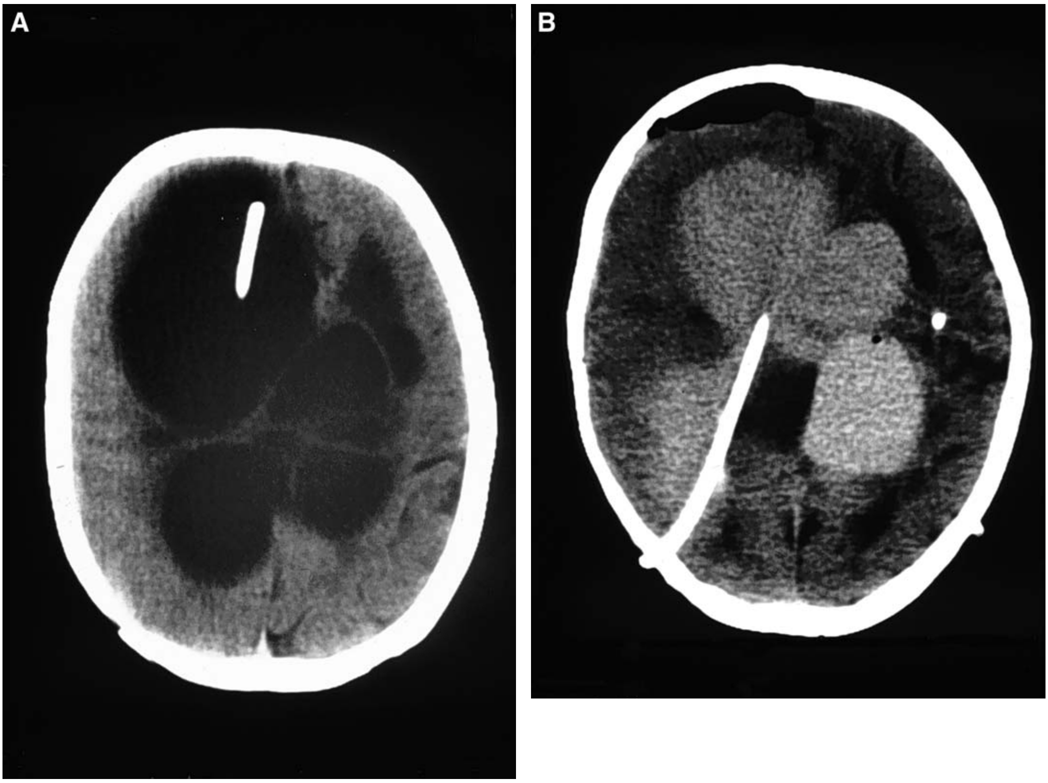

Fig. 7. Multiloculated ventricles. (*A*) Multiloculated ventricle in a patient experiencing symptoms caused by raised intracranial pressure. (*B*) Same patient after endoscopic fenestration. Metrizamide injection into the shunt shows that all but one of the cysts have been communicated and that the cerebrospinal fluid spaces are decompressed as indicated by air over the convexity.

continue to be troublesome. There does not seem to be a preferred technique with regard to therapeutic outcome. Endoscopic management is advocated not for its superior success rate but rather for its lower morbidity rate. For the 12 children managed in this manner, three or more procedures were needed to stabilize their clinical condition.

Further deliberation is necessary before we can decide what radiographically describes a clinical success (ie, what radiographic changes are predicative of resolution of symptomatic complaints). It remains confusing as to why some children's symptoms improve, although their ventricles remained unchanged on imaging studies.

Summary

The neuroendoscopic procedures applied in patients with isolated compartments, including IUH, LV, IFV, IRF, and multiloculated ventricles were foramen of Monro reconstruction, septostomy, septal wall removal, cyst wall fenestration, fourth ventriculostomy, and endoscopic shunt placement. It was found that the operative goal, creating a state of arrested hydrocephalus, could be achieved by communicating the trapped space to the rest of the ventricular system, opening the ventricular isolation. The associated hydrocephalus could not always be managed endoscopically, however, and shunting of the ventricular system was frequently required, especially in infants. This may be because of the immaturity of the subarachnoid CSF dynamics in infants. What does seem to be logical is to continue to consider managing these conditions with the assistance of the endoscope.

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