KISEP

CLINICAL ARTICLE

J Korean Neurosurg Soc 38 : 273-280, 2005

Surgical Treatment of Subdural Hygromas in Infants and Children

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Objective : There is no acceptable indication and treatment of choice for infantile and child subdural hygroma and there are only a few reports about that in Korea. So the authors studied the clinical findings of infantile and child patients with subdural hygroma to improve the understanding and to suggest a standard treatment method.

Methods : The authors retrospectively evaluated the causes, preoperative symptoms, radiological thicknesses, and postoperative results of 25patients with subdural hygroma who received surgical therapy.

Results : There were 16boys and 9girls whose median age was 6months(range 2~120months). The main clinical manifestations were seizures, increased intracranial pressure, macrocrania and alteration of consciousness. Radiological thicknesses of the subdural hygroma varied from 7mm to 42mm and postoperative changes of thickness(y) could be expressed with the factor of month(x): $y = -1.32 \times +11.8$ in subdural drainage, and $y = -1.52 \times +14.9$ in subduroperitoneal shunts. Of the 25patients, 2 (50%) were successfully treated by aspiration, 13 (59%) by subdural drainage, and 9 (69%) by subduroperitoneal shunt.

Conclusion : It is suggested that the diagnosis and treatment of subdural hygroma in infants and children should be carefully addressed because of its high prevalence in children, and especially in infants. It is also suggested that the subdural drainage could be primary initial treatment method because it is simpler than a shunt, and since our data show that there is no statistical difference in postoperative recovery duration between the two operative methods.

KEY WORDS : Subdural hygroma · Subdural aspiration · Subdural drainage · Subduroperitoneal shunt · Subdural hematoma.

Introduction

Subdural hygroma in infants and children is called by various terms such as subdural hygroma, subdural fluid collection, and subdural effusion according to the pre-existing condition, but it is the opinion of the authors that these are in fact the same disease, as there is no definite diagnostic method to differentiate between them. If the initiation of the treatment for subdural hygroma is not prompt, it may present as hemorrhage and progress into a subdural hematoma in 3~4weeks, commonly resulting in a confusing diagnosis between a subdural hygroma and subdural hematoma. Sohn et al reported in a prospective study that in adult, incidence of traumatic subdural hygroma was 35.6% of patients who admitted for more than 7days in hospital after brain trauma and 45.2% of patients who had traumatic intracranial mass lesion[21]. In adults, reports on the progression rates from traumatic subdural hygroma to chronic subdural hematoma vary from 8~50%[13,18,19,22,27]. In Korea, Lee et al.[12] reported that the progression rate from traumatic subdural hygroma to chronic subdural hematoma in adults and children was 33%, but there are no reports to date regarding the frequency of subdural hygroma or progression rate to subdural hemorrhage only in infants in the worldwide literature.

Since subdural hygromas in infants and children is a condition which compress the developing brain, relief of the pressure allows re-expansion of the brain with subsequent normal development and good outcome. Also, as this condition is markedly different from other disease entities (benign pericerebral fluid collection of infancy, benign subdural collections of infancy, benign extra-axial collections of infancy, benign subarachnoid fluid collection of infancy) which resolve

• Received : December 3, 2004 • Accepted : April 25, 2005
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spontaneously without any therapy, it is essential to arrive at an accurate diagnosis[10,17].

The authors of this study therefore prospectively observed the clinical course and outcome of 25patients who received surgical therapy for subdural hygromas to ascertain the clinical characteristics of this disease.

Materials and Methods

For a period of 4years from January 1st, 1998 to December 31st, 2002, the subjects enrolled in this study were 25patients less than 10years of age who received surgical therapy for subdural hygroma at the neurosurgical department of our

Table 1. Summary of 25patients

Case NO.	Sex/age	Sx & Sign	Cause of SDFC	Tx	Outcome	Cx
1	F/5	vomiting	PostOP state (endodermal sinus tumor)	1st burrhole drainage 2nd S−P shunt 3rd burrhole drainage	Sx improved	
2	M/12m	seizure	Idiopathic	S−P shunt	Sx improved	
3	M/2.5	macrocephaly	Idiopathic	1st S−P shunt 2nd burrhole drainage	death	CNS infection
4	M/6m	seizure (fontanelle bulging)	Idiopathic	1st burrhole drainage 2nd S−P shunt	Sx improved	
5	M/2m	seizure	ICH	burrhole drainage	Sx improved	
6	F/2	seizure	PostOP state (neuroblastoma)	1st aspiration 2nd S−P shunt	Sx improved	
7	M/11m	vomiting	Bacterial meningitis	burrhole drainage	Sx improved	
8	F/2m	vomiting	Head trauma	1st aspiration 2nd burrhole drainage	Sx improved	
9	M/3m	seizure	Idiopathic	burrhole drainage	Sx improved	
10	F/3m	lethalgy	Head trauma	1st burrhole drainage 2nd S−P shunt	Sx improved	pseudocyst formation
11	M/10	stupor mentality	PostOP state (AVM rupture)	1st burrhole drainage 2nd S−P shunt	drowsy mentality	
12	F/4m	macrocephaly	Idiopathic	S−P shunt	fontanelle buging decrease	
13	M/6	headache, vomiting	PostOP state (pilocytic astrocytoma)	burrhole drainage	Sx improved	
14	F/3m	seizure	Idiopathic	burrhole drainage	Sx improved	
15	M/9	headache	PostOP state(T−SAH. SDH)	burrhole drainage	Sx improved	
16	F/5m	seizure	Head trauma	burrhole drainage	Sx improved	
17	M/3m	seizure	Head trauma	burrhole drainage	Sx improved	
18	M/4m	seizure	Head trauma	S−P shunt	Sx improved	
19	M/5m	seizure	Meningitis	1st burrhole drainage 2nd burrhole drainage	Sx improved	
20	F/12m	macrocephaly	Idiopathic	1st burrhole drainage 2nd S−P shunt 3rd burrhole drainage	activity improved	obstruction
21	F/5m	seizure	Idiopathic	1st S−P shunt 2nd S−P shunt	Sx improved	
22	M/4m	seizure	Head trauma	1st burrhole drainage 2nd burrhole drainage	Sx improved	
23	M/6	craniectomy site bulging	PostOP state (T−SAH, EDH)	aspiration	bulging decrease	
24	M/3	seizure	PostOP state (ependymoblastoma)	aspiration	Sx improved	
25	M/7m	vomiting	Idiopathic	1st burrhole drainage 2nd S−P shunt	Sx improved	

m : month, SDFC : subdural fluid collection, Tx : treatment, Sx : symptom, Cx : complication, S−P shunt : subduroperitoneal shunt, CTx : chemotherapy, ICH : intracranial hemorrhage, AVM : arterio−venous malformation, T−SAH : traumatic subarachnoid hemorrhage, SDH : subdural hemorrhage, EDH : epidural hemorrhage

hospital(Table 1).

The diagnosis was determined by either Computed Tomography(CT) or Magnetic Resonance Image(MRI), or both, and the patients were divided into unilateral or bilateral groups. It would have been ideal to determine the volume of the lesion for our study, but since this is very difficult in real practice employing conventional CT or MRI, the authors decided to measure the thickest portion of the lesion as demonstrated by CT or MRI.

Indications for surgical intervention was the presence of symptoms such as increased intracranial pressure, macrocrania, hemiparesis, seizure, mental deterioration, and when the thickest diameter of the lesion was more than 7mm. For simple subdural hygromas, surgery consisted of subdural aspiration or subdural drainage by subdural catheter insertion or subduroperitoneal shunt. However, in those patients with accompanying subdural hematoma, subdural drainage was performed. The authors of this study compared the preoperative and postoperative symptoms, and also the changes in the radiological findings after treatment.

Results

Age and sex distribution

Among the 25patients, the age range was 2~120months (median 6months). Seventeen patients (68%) was less than 1year old, the majority in the 3~5months range. There were more males (16) compared to females (9) (Fig. 1).

Distribution of etiology and symptoms

The most common cause of subdural hygroma was idiopathic, occurring in 9cases (36%). 7cases (28%) occurred following various surgical procedures, 6cases were post-traumatic, 2cases occurred as complications of leptomeningeal infections, and 1case after intracerebral hemorrhage. Among the 9 idiopathic etiology cases, 8patients were under 1year of age (Fig. 2). The main clinical manifestations were seizures in 13cases, increased intracranial pressure (nausea and vomiting) in 6, macrocrania in 4, and alteration of consciousness in 2cases (Fig. 3).

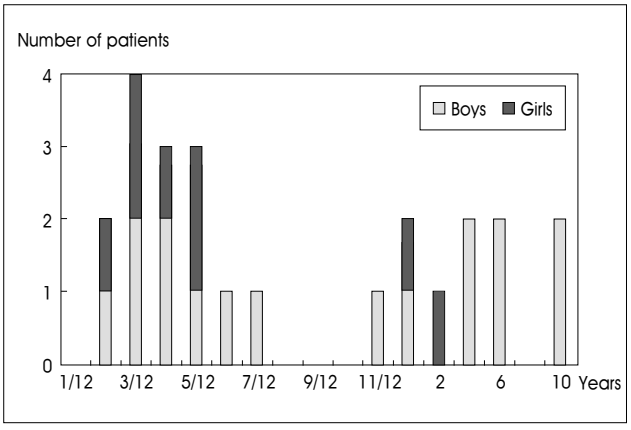

Fig. 1. Distribution of age and sex in 25patients with subdural hygroma.

Radiologic findings and clinical course

Radiological findings showed that subdural hygroma was unilateral in 5cases and bilateral in 20cases. In terms of preceding disease status, it was bilateral in 5cases and unilateral in 2cases after surgery, and was bilateral in 7cases and unilateral in 2cases when the cause are idiopathic (Fig. 2).

The preoperative mean radiological thicknesses of the subdural hygroma was 14.3mm measured by CT or MRI, with a range of 7mm to 42mm. The thickness of the subdural hygroma decreased postoperatively on an average of 1.32mm per month from the mean of 11.8mm after subdural drainage. After subduroperitoneal shunt, the decrease was an average of 1.52mm per month from the mean of 14.9mm. The change of thickness per month could be expressed with the equation of $y = ax+b$ ($a = -1.32 \pm 0.57$, $b = 11.87 \pm 1.27$ in subdural drainage, $a = -1.52 \pm 0.52$, $b = 14.95 \pm 1.51$ in subduroperitoneal shunt) (Fig. 4). However, there was no statistical difference between the two groups (95 % confidence interval, SPSS ver.10)

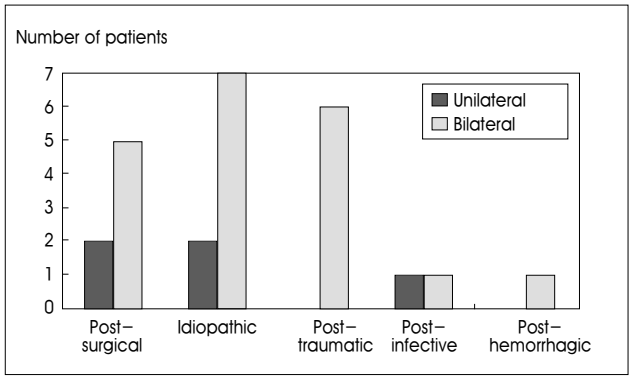

Fig. 2. Predisposing causes of 25patients and laterality of subdural hygroma.

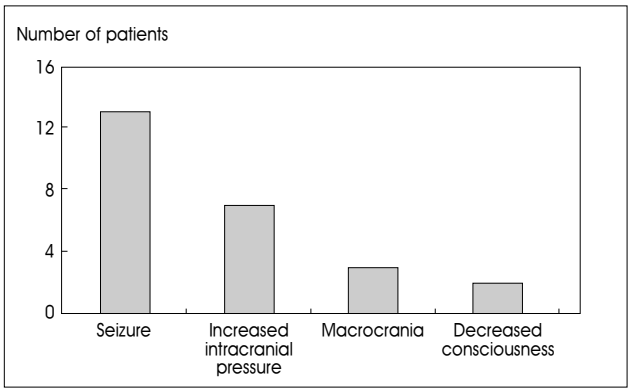

Fig. 3. Clinical presentation of 25patients.

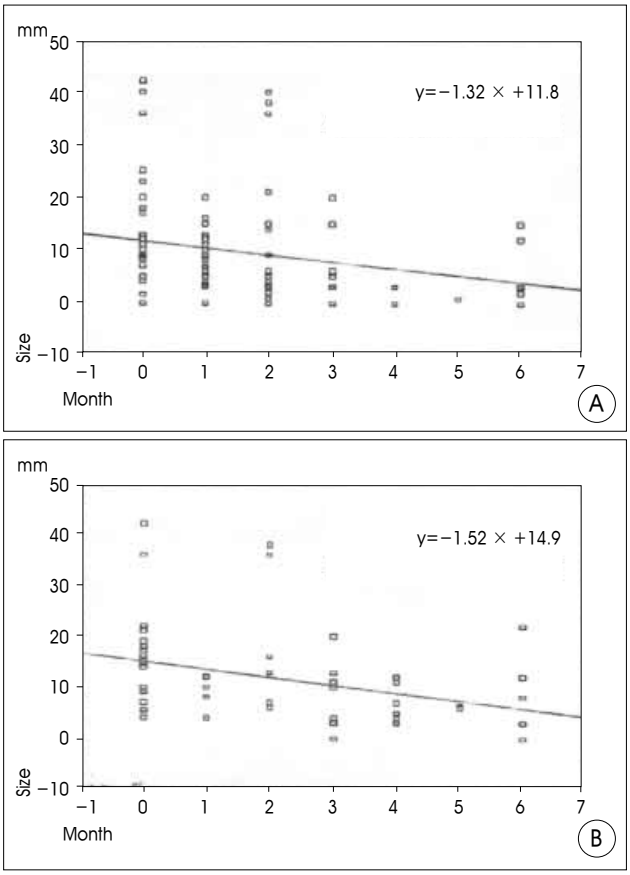

Fig. 4. Change of maximum thickness of subdural hygroma after subdural drainage(A), and after subduroperitoneal shunt(B). A line means interpolated average thickness of subdural hygroma after operation that is expressed as an equation. There is no statistical difference of the maximum thickness between both methods.

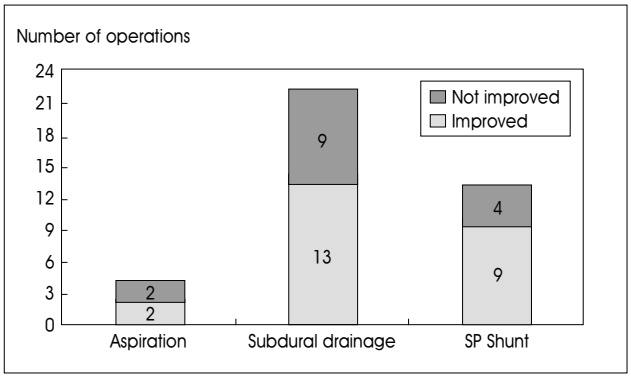

Fig. 5. Result of a total of 39 operations in 25patients. There is no statistical difference of the success rate between subdural drainage and subduroperitoneal shunt.

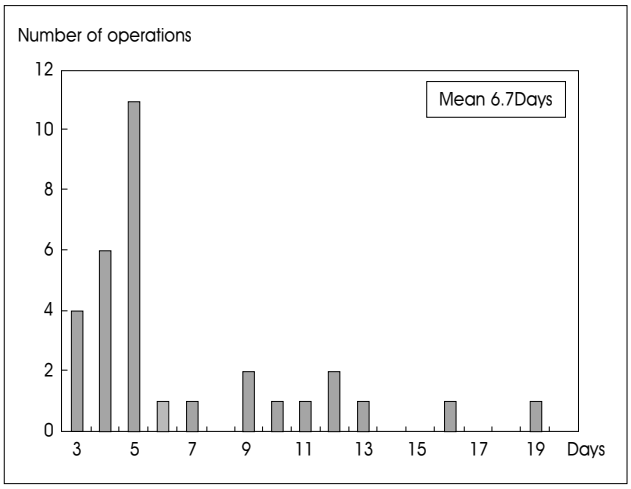

Fig. 6. Durations of 22 subdural drainages.

Surgical technique

Among the total of 25patients, 4cases underwent subdural aspiration, 22 underwent subdural drainage, and 13patients underwent subduroperitoneal shunt. The clinical outcome according to each type of surgical procedure demonstrated that subdural aspiration succeeded in 2cases, while the other 2 did not clinically improve, resulting in a success rate of 50%. Subdural drainage succeeded in 13cases, while it failed in 9cases showing a 59% success rate. Subduroperitoneal shunt succeeded in 9cases and failed in 4, marking down the success rate to 69 % (Fig. 5).

Primary surgery was successful in 13 of 25cases (success rate 52%), while 9cases received further secondary surgery which succeeded among 12patients (success rate 75%). Two patient received a third operation which turned out to be successful. As for the side effects of each type of surgical method, reoperation was necessary after aspiration in 2cases due to no improvement, and an infection that occurred in 1case after subdural drainage. In those patients who received subduroperitoneal shunt, there were 2 complication that one was pseudocyst formation in the peritoneal cavity and the other was obstruction of the catheter.

Among the 16cases of subdural hygroma that were bilateral, unilateral subdural drainage was conducted in 6cases, and bilateral subdural drainage was conducted in 10cases. The duration of subdural drainage raged from 3 to 19days, with a mean of 6.7days (Fig. 6). Among the 1cases of bilateral subdural hygroma, unilateral subduroperitoneal shunt was performed in 7cases, and bilateral subduroperitoneal shunt was performed in 4cases. For the 2case of unilateral subdural hygroma, unilateral subduroperitoneal shunt was performed. The shunt valves utilized were regular low pressure valves in 2cases, small low pressure valves in 7cases, and small low-low pressure valves in 4cases.

Discussion

The progression of subdural hygroma in infants and children leads to hemorrhage and eventually to subdural hematoma formation. Consequently, previous literature pertaining

to the treatment of subdural hygroma has been addressed in the same concept as that of subdural hematoma, and which therefore makes the standardization of the diagnosis and treatment for the subdural hygroma difficult. There is as yet no accurate report to date on the incidence of subdural hygroma in infants, but it has been suggested that subdural hematomas occur every 21 per 100,000 infant population[9].

The causative mechanisms involved in the formation of subdural hygromas is known to be idiopathic, and as a secondary phenomenon after brain damage. In idiopathic cases cells in the granulation tissue of the arachnoid membrane does not develop adequately, which then interferes the normal cerebrospinal fluid absorption process. Ultimately, subdural hygroma ensues when the outer arachnoid membrane breaks up or when the cap cells of arachnoid membrane proliferates. Nevertheless it is known that subdural hygroma improves when the cerebrospinal fluid is absorbed normally as time goes on[7,14]. On the other hand, if the subdural hygroma grows too large while waiting for spontaneous resolution, it has been reported that it may influence brain development and may also further develop in to a subdural hematoma, which needs surgical treatment[25].

When the etiology is secondary, it has been demonstrated that when treatment consisted of subdural drainage or subduroperitoneal shunt, the success rate is high due to the brain expansion in the developmental age. Therefore, the authors of this study considers it acceptable that all subdural hygromas in infants and children should be treated taking into consideration of size of the lesion and the presence or absence of any symptoms.

It has been reported that idiopathic cases are the most frequent in 3~5months old infants[1,25], and the results of this study are in agreement which also showed that the idiopathic cause is most common in patients under the age 1, and especially at 3~5months. When and how often infants or children develop subdural hygromas is not yet accurately known, but it has been shown that subdural hematoma usually develops 3~4weeks after the appearance of subdural hygroma[15]. This study also showed 11cases which were initially subdural hygroma in the early stage which developed into a subdural hematoma as time progressed, and who received surgical treatment, accounting for 44% of the 25cases.

According to a report by Caldarelli et al.[2], which studied 85cases of infantile subdural hygroma, the main symptoms were increased intracranial pressure, macrocrania, mental retardation, hemiparesis, seizure, and decreased consciousness, in decreasing order of frequency. However, our study showed that seizure was the most common symptom, while the frequency of increased intracranial pressure, macrocrania, and decreased consciousness was similar to the Caldarelli's study. In aspect that seizure usually begins from a chronic cause, we presume that this may be the result of less attention to diagnosis and treatment of subdural hygroma within the our country.

Radiological characteristics demonstrated by CT and MRI showed that subdural hygroma can be distinguished from external hydrocephalus. Subdural hygromas shows expansion of the subdural space and not the subarachnoid space due to accumulation of CSF in the subdural space. Therefore, the enhanced vessels by contrast media are compressed to adhere to the brain, and the hygroma is placed outwards[10,16,26]. When a subdural hygroma appears on the inside of a cerebral cortical vessel, it should be differentiated from external hydrocephalus. If the patient is under age 1, it should also be differentiated from transitory diseases, such as benign perisylvian fluid collection and benign familial subdural fluid collection. Of course the subarachnoid membrane can be seen without enhancement on the MRI, and therefore it can be certain that the location of the hygroma is not in the subarachnoid space, but in the subdural space[3,7]. The development of a chronic subdural hematoma after a traumatic injury is shown as a high density in the entire subdural space, but the subdural hematoma that originates from a subdural hygroma is shown as a partial density in a part of the subdural space.

There has been a previous reports that suggests that the operative indication for subdural hygromas is a lesion larger than 7mm with accompanying symptoms[1], or larger than 10mm without symptoms[23]. But since exact measuring methods are still unavailable, further study involving larger number of cases is required to develop the precise measurement of the lesion size. It is expected that surgical indication can be easily made with the symptoms rather than size estimation because the three-dimensional size can not be measured accurately on CT or MRI. In the past, many medical institutions attempted subdural aspiration first in infantile patients with subdural hygroma, and then subdural drainage or subduroperitoneal shunt in those patients where the previous treatment was ineffective. This is still not an uncommon surgical procedure, but subdural aspiration is only a temporary measure, and is rarely effective. Also, because of the serious brain hemorrhage by subdural aspiration, it has been largely discontinued as a mode of treatment nowadays. In the authors' study, subdural aspiration were performed in 4cases in which one was an infant with a large anterior fontanelle. The other 3cases were postoperative craniectomy state. Among the 4cases, 2cases improved and there was no incidence of the most serious complication, bleeding.

Subdural drainage through subdural catheter insertion may be tried for subdural hygromas, but it is most effective when a subdural hematoma develops, and the success rate has been variably reported as 43~100%, although a recent report sug-

gested that it would be more appropriate to consider the success rate as being 60~70%[2,4,6,8,11,23,24].

In this study, the success rate of subdural drainage was 59%, and this is similar to that of recent reports (Fig. 7). With regard to the choice of bilateral or unilateral drainage in bilateral subdural hygroma, it has been reported that subdural drainage on the contralateral side was performed because the contralateral subdural hygroma did not improve after unilateral subdural drainage. Similarly in this study, among the 4cases that underwent unilateral subdural drainage at first, there were 3cases in which the contralateral side did not improve, which supports the principle of bilateral drainage when a subdural hygroma is large. The drainage period is usually accepted as 3~8days, but Van Calenbergh et al reported that they drained for as long as 29days[2,4,6,24].

According to a previous report, it is more effective when the catheter is removed slowly, controlling the amount of subdural fluid by adjusting the height of the subdural catheter to increase the success rate instead of removing the subdural catheter immediately after drainage[24]. In terms of complications of subdural drainage, there was 1case of infection (4.5%) in this study, which is similar to a previous report of 0~9%. As to the relationship between infection and the period of drainage, Ersahin et al.[4] reported 0% infection rate after drainage for a mean of 7days, while Van Calenbergh et al.[24] reported it as 3% after drainage for a mean of 12days. Caldarelli et al.[2] reported it as 9% after drainage for a mean of 5days. The above results do not suggest that there is a relationship between the period of drainage and infection rate.

In spite of the fact that the success rate is higher after subduroperitoneal shunt compared to subdural drainage, it is not the more preferred mode of treatment for subdural hygroma. This is because of the complications of the abdomen due to the abdominal operation, though infection rate is similar to that of subdural drainage while obstruction is frequent, which does not occur with subdural drainage, and finally the shunt catheter needs to be removed after recovery[24]. In addition, subduroperitoneal shunt cannot be performed in the presence of subdural hemorrhage because of the high risk of obstruction. As a last resort, it has been suggested that if the subdural drainage is not effective, the patient should be observed until the appearance of the subdural hemorrhage, and then perform subdural drainage one more time[15].

Nevertheless, the reason why the use subduroperitoneal shunt is being widely reported is that it may be performed when subdural drainage is ineffective, and its success rate is as high as 70~90% (Fig. 8). With regard to the choice of unilateral or bilateral subduroperitoneal shunt in bilateral subdural hygroma, Aoki[1] reported that the course of subdural hygromas was imporved after unilateral subduroperitoneal shunt only, but Ersahin et al.[5] reported that the contralateral side shunt was necessary because there was no improvement of the unilateral shunt in 5 of 31cases. But Vinchon et al.[25] reported that obstruction occurs more frequent than infection, so bilateral shunt would be more favorable because of a lower possibility of obstruction. Overall, the choice of unilateral or bilateral shunt is as yet controversial. But data

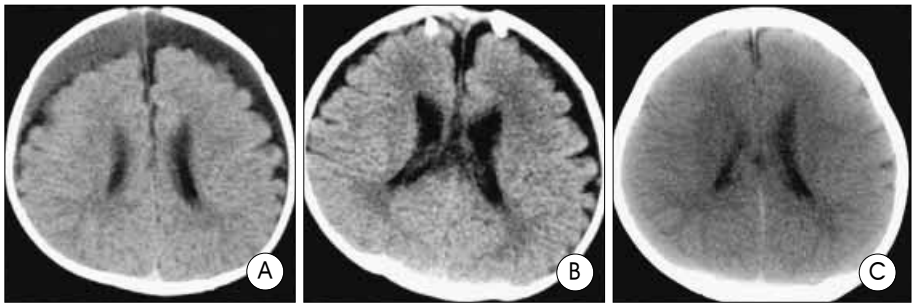

Fig. 7. Computed tomography(CT) image of 4month-old boy with subdural hygroma who received subdural drainage operation. A : Preoperative CT image. B : Postoperative image of bilateral subdural drainage reveal decreased subdural hygroma. C : Follow up CT image shows that the subdural hygroma has completely improved.

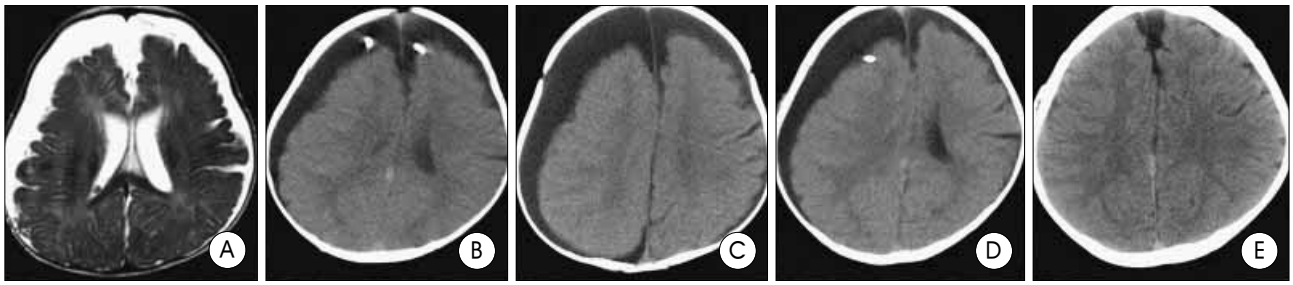

Fig. 8. A 10-month-old boy with subdural hygroma improved by subdural drainage and subsequent subduroperitoneal shunt. A : Bilateral subdural hygroma is noticed on magnetic resonance image. B : Two tubes are noticed in the frontal subdural space for bilateral subdural drainage on computed tomography(CT) image. C : Subdural hygroma has not improved on CT image after bilateral subdural drainage. D : Follow-up CT image immediately after subduroperitoneal shunt shows unilateral subdural catheter in the right frontal area. E : Follow-up CT image 6 months after subduroperitoneal shunt shows that the subdural hygroma has improved completely.

from another series have indicated that obstruction occurs more frequently in unilateral shunts, as shown by Ersahin et al[5] and Sakka et al[20]. These reports further proposed that if the cell and protein components of the CSF are high risk factors of obstruction, it seems that the choice of a bilateral shunt would be preferable. Following this concept, the authors performed bilateral shunt in 4cases and unilateral shunt in 7cases among the 11cases of bilateral subdural hygroma. Although there were more cases in which unilateral shunt was conducted, there was only 1case of obstruction, which is a lower rate compared to other reports.

The valves used in shunts were mostly small sized and low pressure valves of 3~5cm, or low-low pressure valves under 3cm, as the patients were infants and children. There were 2 infantile patients in whom small size low pressure type valves was employed but they did not improve. So the valve was exchanged for a regular size low pressure valve, and the reservoir was regularly made pump to force the fluid out, and this was successful.

The most common reported complications of subduroperitoneal shunt are infection and obstruction, and the incidence has been cited as 2.7~9%[2,11,20,25] and 9~23%[5,11,20,25], respectively. The reason why shunt for subdural hygromas show less complications compared to hydrocephalus is that subdural hygroma generally improves after 6months, and needs no further treatment. In this study there were 2 complication cases among 13cases, which comprised of an obstruction case and an abdominal pseudocyst formation case. Thus, our complication rate was similar to previous reports.

It is well known that subduroperitoneal shunt is the most effective treatment modality for subdural hygromas, but because in many patients subdural hematomas occur concomitantly, the shunt alone cannot be applied uniformly to all patients in the early stage of the disease. Therefore, it would be advisable to determine the choice of treatment according to the degree of hematoma formation accompanying the subdural hygroma. To further establish the optimal treatment modality for the various degrees of the disease, wide spread studies pertaining to the prognosis for each different result is warranted.

Conclusion

Children, especially infants under the age of one, acquire subdural hygromas of idiopathic etiology in many cases. It is therefore important to prevent damage of the brain in the developmental period through accurate diagnosis and appropriate treatment. In this study, there was no statistical difference in terms of the recovery period between subdural drainage and subduroperitoneal shunt for treatment. Thus, in early stage subdural hygroma patients, it is suggested that subdural drainage is preferred due to the relative simplicity of the procedure rather than shunt. Subduroperitoneal shunt should be considered when the subdural hygroma does not improve with subdural drainage.

• Acknowledgement
This article was reported at 2003 Annual Autumn Meeting of the Korean Neurosurgical Society.

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Commentary

Since the advent of computed tomography and magnetic resonance image into neurosurgical practice the diagnosis of subdural fluid collection(SDFC) or subdural hygroma (SDHG) became easy and accurate in diferentiation with so called external hydrocephalus (subarachnoid fluid collection). The latter entity, which is common in developing infantile brain, is asymptomatic self-limiting benign lesion and usually resolve spontaneously until age of 2years.

Sympomatic SDFC has been managed with ① repeated subdural tap and aspiration, ② external subdural drainage (ESD), and ③ subduroperitoneal shunt(SPS). But selection of the treatment modalities is still controversial because of their differences in effectiveness(success rate), risk of complication(infection rate, obstruction rate, bleeding incidence etc), and cost of the treatment(duration of hospital stay, imaging studies etc). In case of failure with repeated subdural taps and/or ESD to resolve the SDFC, the last resort is to do SPS.

The authors studied 25cases of infantile and childhood SDFC, and concluded that ESD is preferable procedure in early stage of SDHG just because of simpler procedure than SPS. But recent trend of the management of infantile SDFC is in favor of the primary SPS instead of ESD. The reasons are definite treatment modality with far superior success rate, less infection rate and shorter hospial stay of the SPS than those of ESD.

To achieve a good result with continuous ESD for SDFC, 51.5~87.1% success rates, the drain should be in place for 7~20days with infection rates of 3~17% on the literatures. It is known that unilateral SPS for the bilateral SDFC is usually successful ; the valve of SPS should be low pressure or unvalved and distal open-ended one may be used to accelerate the drainage of SDFC and to prevent shunt obstruction; shunt removal is not necessarily needed.

Some authors insist that bilateral SPS is betther than unilateral SPS because of decreased chances of shunt obstruction. The authors pointed out this and favored the bilateral shunting for bilateral SDFC. The authors reviewed the literature appropriately and the lengthy discussion give detailed informations about SDFC.

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