The common rationale of decompressive craniectomy with or without duroplasty is to let the volume expansion of the swelling brain to extracranial space via removed skull flap and prevent cerebral herniation and secondary damage of brain parenchyme.

In case of traumatic brain injury, decompressive craniotomy for pediatric patient has been reported. However, there has not been a definite evidence or standard guideline for decompressive craniectomy for pediatric patient with non-traumatic acute stroke with uncontrolled refractory high intracranial pressure.

We will introduce our series about the functional and clinical outcome after decompressive craniectomy to control refractory high ICP due to non-traumatic acute stroke in pediatric patient.

INTRODUCTION

Stroke is rare in children, however, a common cause of neurological disease, and it is a major cause of death ranks in the top ten in pediatric period. Documented incidence has been reported as 2.5-8/100000 children/year. Medical treatment includes maintaining cerebral perfusion pressure with hyperventilation and osmotherapy, barbiturate coma and in case of thrombotic ischemia, thrombolytic procedure, and anticoagulant therapy. However, if all medical treatment is fail to drop the intracranial pressure (ICP) and deterioration of patient progressed, alternative treatments, such as surgical decompression could be mandatory.

The common rationale of decompressive craniectomy with or without duroplasty is to let the volume expansion of the swelling brain to extracranial space via removed skull flap and prevent cerebral herniation and secondary damage of brain parenchyme.

In case of traumatic brain injury, decompressive craniotomy for pediatric patient has been reported. However, there has not been a definite evidence or standard guideline for decompressive craniectomy for pediatric patient with non-traumatic acute stroke with uncontrolled refractory high intracranial pressure.

We will introduce our series about the functional and clinical outcome after decompressive craniectomy to control refractory high ICP due to non-traumatic acute stroke in pediatric patient.

Decompressive Hemicraniectomy and Duroplasty in Toddlers and Preschool Children with Refractory Intracranial Hypertension after Unilateral Hemispheric Stroke

Sang-Kook Lee, M.D., Sang-Dae Kim, M.D., Se-Hoon Kim, M.D., Dong-Jun Lim, M.D., Jung-Yul Park, M.D.
Department of Neurosurgery, Korea University Ansan Hospital, Ansan, Korea

Objective : Life-threatening hemispheric stroke is associated with a high mortality and morbidity. Decompressive hemicraniectomy has been regarded as an effective treatment option for refractory intracranial hypertension. Here, we reported the clinical course of 5 children with decompressive craniectomy and duroplasty after non-traumatic refractory intracranial hypertension.

Methods : Four toddlers and one preschool-girl were included in this study; there were 3 boys and 2 girls with a mean age of 34.6 months (range 17-80). Decompressive craniectomy including duroplasty was performed in cases of dilatation of pupil size after intensified standard medical therapy had proven insufficient. All children had a Pediatric Glasgow Coma Scale score <8 at pre-operation state. The mean time-point of craniectomy after stroke attack was 12 hours (range 4-19).

Results : During the long-term follow-up period (mean 47.6 months), no children died. One year later, when we checked their Glasgow Outcome Scale scores, only one toddler received a score of 4 (moderate disability). But the others had good recoveries although they had minor physical or mental deficits. According to the Pediatric Cerebral Performance Category Scale, 4 children received a score of 2 (mild disability).

Conclusion : Despite our small cases, we suggest that decompressive hemicraniectomy and duroplasty is an acceptable and life-saving treatment for refractory intracranial hypertension after unilateral hemispheric stroke in toddlers and preschool children.

Key Words : Decompressive craniectomy · Stroke · Pediatric · Outcome.
MATERIALS AND METHODS

Between March 2004 and December 2006, decompressive-hemicraniectomy and duroplasty was performed in 5 toddlers and preschool children with non-traumatic, malignant refractory high ICP. Patient characteristics and outcome were summarized in Table 1.

Any patient have no trauma history, and refractory high ICP was diagnosed as clinical evidence of acute stroke, massive infarction or hemorrhage with midline shift, compression of basal cisterns in conventional radiologic examination such as computed tomography (CT) and magnetic resonance image (MRI) and, neurological deterioration consisting of decreased level of consciousness or increasing levels of sedation, or somnolence or stupor compared with baseline status at admission. And we regard sustained refractory high ICP as a failure of intensified standard medical therapy.

All operations were performed by one pediatric neurosurgeon and all patients received treatment in pediatric intensive care unit of one medical center.

Medical treatment
In all patients, CT or MRI was obtained immediately, after stabilization of ventilation and hemodynamics at first. Blood sugar, body temperature, ventilation/oxygenation, blood volume and trans-cutaneous oxygen saturation, arterial blood pressure was continuously monitored and controlled in pediatric intensive care unit. Clinical and neurologic status was evaluated with the Glasgow Coma Scale (GCS). To reduce ICP, hyperosmolar therapy is typically begun with use of osmotic diuretics such as mannitol as needed.

Decompressive craniectomy was performed in cases of failure of intensified standard medical therapy.

Surgical protocol
Craniectomy involved the removal of a largebone flap ipsilateral to the involved hemisphere including frontal, parietal, and temporal bone.

A large skin flap was lifted from the skull with meticulous blood control, multiple burr holes were made, large bone flap removed. In the temporal region, the craniectomies were extended toward the floor of the middle fossa to maximize decompression. Multiple openings in the dura were made; a dural patch was placed and sutured. In every case, intra-cerebral pressure monitor probe was positioned epidural space of bone margin.

The bone flaps were stored in a bone bank and then, following improvement of clinical and neurologic status, cranioplasty was performed 3 to 6 months later.

Neurological outcomes were evaluated using neurologicalexaminations. Glasgow Outcome Scale (GOS), and Pediatric Cerebral Performance Category Scale (PCPCS) (Table 2) calculated every 6 months after discharge.

RESULTS

Five pediatric patients were treated with decompressive hemicraniectomy with duroplasty for non-traumatic and refractory intracranial hypertension after unilateral hemispheric stroke. Four patients were toddler and one patient was pre-school girl. Three were boy and two were girl. The mean age was 34.6 months old (range 17-80 months).

Four cases were caused by ischemic stroke, and another one case was hemorrhagic stroke. One case of hemorrhagic stroke, patient had been diagnosed by thrombocytopenia. One case of ischemic stroke, patient had atrial septal defect (ASD). And including this patient three cases of ischemic stroke patients were taken angiography; however there was no abnormal findings in angiography. In all patients, conventional laboratory result and special serum anti-body test, including anti-phospholipid antibody were within normal rage. At the time of operation, all patients had a GCS score <8 (median 7, range 6-8) and all patients had unilateral mydriasis. Surgical decompressive hemicraniec-
abilities (GOS score of 4). And there was no patient shown severe disability or death. The PCPCS scores also had revealed contend results: 4 patients received scores of 2, and only one patient scored of 3.

Patient presentation

A 17-month-old girl was transferred to our emergency department with decreased mentality. Neurological examination revealed a stuporous child without occasional spontaneous eye opening. Her right pupil was 6 mm in size and was not response to light. And her left pupil was 3 mm and was sluggishly reactive to light. Hemiplegia, hyper-reflexia, and Babinski’s sign were observed on the left side. Preoperative GCS score was 7.

In general physical examination, she had an ejection systolic murmur. By pediatric cardiologist consultation, patient was diagnosis with ASD.

Initial brain CT scan was suggestive of acute infarction and hemorrhagic transformation at right temporal area. And hemorrhagic transformation was also noted at same side area. B and C: DWI MRI scan of his brain revealed that lesion was appropriate for acute cerebral infarction with hemorrhagic transformation with significant mid-line shift. CT: computed tomography, DWI: diffusion weighted image, MRI: magnetic resonance imaging.

Table 2. Pediatric Cerebral Performance Category Scale (PCPCS)

<table>
<thead>
<tr>
<th>Score</th>
<th>Category</th>
<th>Clinical feature</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Normal</td>
<td>• Normal at age appropriate level</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• School age child attends regular school classroom</td>
</tr>
<tr>
<td>2</td>
<td>Mild disability</td>
<td>• Conscious alert and able to interact at an age appropriate level</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• School age child attending regular school classroom but grade perhaps not appropriate for age</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• May have a mild neurologic deficit</td>
</tr>
<tr>
<td>3</td>
<td>Moderate disability</td>
<td>• Conscious</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Sufficient cerebral function for age-appropriate independent activities of daily life</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• School age child attending special education classroom</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• May have learning deficit</td>
</tr>
<tr>
<td>4</td>
<td>Severe disability</td>
<td>• Conscious</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Dependent on others for daily support because of impaired brain function</td>
</tr>
<tr>
<td>5</td>
<td>Coma or vegetative state</td>
<td>• Any degree of coma without any of the criteria for brain death</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Unawareness even if awake in appearance without interaction with the environment</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Cerebral unresponsiveness</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• No evidence of cortical function and not aroused by verbal stimuli</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Possibly some reflexive responses spontaneous eye opening and/or sleep-wake cycles</td>
</tr>
<tr>
<td>6</td>
<td>Brain death</td>
<td>• Apnea OR</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Areflexia OR</td>
</tr>
<tr>
<td></td>
<td></td>
<td>• Electroencephalographic (EEG) silence</td>
</tr>
</tbody>
</table>

Fig. 1. A: Non enhanced brain CT revealed an acute right-sided middle cerebral artery territory infarct with mass effect manifested as right lateral ventricular compression, attenuation of the basal cisterns and right convexity sulci effacement. Because of midline shift due to severe brain parenchymal swelling, posterior horn of lateral ventricle was not seen. And hemorrhagic transformation was also noted at same side area. B and C: DWI MRI scan of his brain revealed that lesion was appropriate for acute cerebral infarction with hemorrhagic transformation with significant mid-line shift. CT: computed tomography, DWI: diffusion weighted image, MRI: magnetic resonance imaging.

The mean follow-up period was 47.6 months (36-68 months). GOS and PCPCS for 5 patients had been measured to evaluate postoperative neurological outcome. Based on the regular follow up GOS scores, 5 patients had shown satisfied recoveries: 4 had good recoveries (GOS score of 5), and 1 had moderate disability (GOS score of 4). And there was no patient shown severe disability or death. The PCPCS scores also had revealed contend results: 4 patients received scores of 2, and only one patient scored of 3.
loss of consciousness. After surgery, she was treated in pediatric intensive care unit with be kept intubated for 7 days.

A brain CT at the time of post-operative 7 days revealed sustained brain swelling, but midline shift did not noted anymore. The immediate post-operative ICP was 28 mm Hg. The ICP dropped to normal gradually within one day after surgery. At the time of postoperative 3-month, she returned for an elective cranioplasty. The bone flap was secured without any complications.

At the end of follow-up, 41 months after surgery, this patient maintained a PCPCS of 2, and GOS of 5, and she had showed mild left side hemiparesis, mild behavioral disorder and strabismus.

**DISCUSSION**

In stroke patient, most common cause of death is due to uncontrolled ICP associated with large hemispheric infarctions\(^{10,12}\). When it is present, it is associated with a dramatic increase in mortality. In addition, early ICP elevation in patients with large hemispheric infarctions is highly concluded with high mortality\(^{40}\). It means that the one of most important goal of acute phase stroke treatment is control the ICP and prevent a secondary damage due to brain swelling.

In spite of many clinical and experimental trials on medical therapeutic method, most of them have not been shown to benefit pediatric stroke patient. There has been a widely accepted definite guideline for oxygen saturation, temperature, serum glucose level, blood pressure for adult stroke patient\(^{40}\). However, for pediatric stroke patient, even in many of mostly basic and elementary medical guideline has not been established yet\(^{14}\).

To prevent and control ICP, thrombolytic therapy such as clot lysis has an accepted role in acute stroke treatment. In adult stroke patient group, there are antithrombotic management of adult ischemic stroke protocol and guideline published by the American Heart Association\(^{1}\), such as intravenous tissue plasminogen activator, intra-arterial thrombolysis and oral administration of aspirin. But, for pediatric stroke patient, there is no guideline of this useful treatment method either\(^{40}\).

To the best of our knowledge, the first report about the benefits of decompressive hemicraniectomy in pediatric patient was described by Carter et al.\(^{3}\) at 1997. However, from this time, only a few report deals with decompressive craniectomy for pediatric patient were published.

In adult acute stroke patient group, the effect of decompressive craniectomy also has been established. Vahedi et al.\(^{21}\) reported that in a meta-analysis combining the data of 93 subjects from three small, randomized, controlled trials, mortality for conservative management in adult ischemic stroke patients with early brain edema was 71% as compared with 22% for decompressive surgical intervention.

Nowadays, some case reports and review has been published favorable outcome after decompressive surgery in childhood acute stroke\(^{1,19}\). It is not often however in some article, decompressive craniotomy have been regarded as emerging procedure can save life, for pediatric stroke patient\(^{6,11}\), even guideline for craniectomy was introduced\(^{41}\). In point of fact, there is some debate indications and optimal timing of decompressive craniectomy\(^{12,19}\). Widely accepted proper operation timing is immediatly after signs of herniation and a midline shift which was defined as a more than equals 5-mm contralateral shift of the midline structures on the preoperative cranial CT scan on septum pellucidum level, compressed basal cisterns, or uncal herniation--had to be present on cranial CT had been noted. Recent studies in adult stroke group have shown that this procedure not only reduces mortality but also improves neurological functional outcome. Especially when operation is performed in the early hours\(^{12,18}\). This procedure may be life-saving if done early in cases of impending herniation, is associated with good outcomes in pediatric stroke patient\(^{6,13}\). Maybe if operation was performed earlier, it may minimize the vicious circle of brain swelling, increased ICP, ischemia, and infarction\(^{22}\).

In our series, four toddlers and one pre-school girl with refractory high ICP due to non-traumatic, ischemic and hemorrhagic stroke were treated with decompressive craniectomy. All
had GCS scores <8 at operation. All of them were survived and have a relatively satisfied recovery. Four patient received PCPCS score of 2 (mild disability) and only one received that of 3 (moderate disability) after 3 years later post operation. These results showed that decompressive craniectomy cannot only play a rescuer therapeutic role but also guaranty acceptable clinical outcome in the treatment regimen in non-traumatic, refractory high ICP in pediatric field.

In some articles, decompressive craniotomy has been tried to apply more alternative pediatric field. Meningitis andencephalitis could be not very rare diseases of pediatric period. And these diseases could make a malignant intracranial hypertension. Even in adult, effectiveness of decompressive craniectomy has not been established firmly. Aghakhani et al.\textsuperscript{2} report that decompressive craniotomy was performed in pediatric patient with malignant intracranial hypertension due to infectiousencephalitis. In adult patient group, there had been not infrequently reported that malignant intracranial hypertension due toencephalitis treated with decompressive craniectomy. Now, even in case of pediatric patient with infectious disease in central nerve system, decompressive craniectomy could be considered as life-saving-treatment options.

Gordon et al.\textsuperscript{3} report that pediatric stroke can affect not only patient itself, but also his or her whole family. Over half of stroke survivors have suffer from decrease in quality of life and affects the entire family, and relates to both neurological deficits and psychosocial factors\textsuperscript{4,5}. Hence, if all medical treatment is fail to drop the ICP in non-traumatic pediatric acute stroke patient, without hesitation, a decompressive craniectomy could be regarded as next step treatment option.

CONCLUSION

In this study, we describes the young pediatric patients have undergone decompressive hemicraniectomy for a non-traumatic acute hemispheric stroke. It also shows that decompressive hemicraniectomy can be lifesaving and can besafely performed in toddler and pre-school children. And young children can get a survival and relatively independence and family satisfaction. Decompressive hemicraniectomy should be considered as an alternative therapy for patients with life-threatening brains swelling refractory to medical management.

References


