Decompressive Craniectomy in Children with Severe Traumatic Brain Injury: A Multicenter Retrospective Study and Literature Review

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BACKGROUND: **Severe traumatic brain injury (TBI) is the most common cause of disability in children. Refractory increased intracranial pressure can be a therapeutic challenge. Decompressive craniectomy can be proposed when medical management is insufficient, but its place is not clearly defined in guidelines. The aim of this study was to identify prognostic factors in children with TBI.**

METHODS: **We performed a retrospective, multicenter study to analyze long-term outcomes of 150 children with severe TBI treated by decompressive craniectomy and to identify prognostic factors.**

RESULTS: **A satisfactory neurologic evolution (represented by a King's Outcome Scale for Childhood Head Injury score >3) was observed in 62% of children with a mean follow-up of 3.5 years. Mortality rate was 17%. Prognostic factors associated with outcome were age, initial Glasgow Coma Scale score, presence of mydriasis, neuromonitoring values (maximal intracranial pressure >30 mm Hg), and radiologic findings (Rotterdam score ≥4).**

CONCLUSIONS: **This study in a large population confirms that children with severe TBI treated by decompressive craniectomy can achieve a good neurologic outcome. Further studies are needed to clarify the use of this surgery in the management of children with severe TBI.**

INTRODUCTION

Traumatic brain injury (TBI) is the leading cause of morbidity and mortality in children. In addition, survivors of pediatric TBI may have cognitive and behavioral dysfunction that impairs their ability to reintegrate into social environments. Both the primary brain injury and the management of secondary brain injury are important in determining outcomes of pediatric TBI. Many studies have demonstrated that high intracranial pressure (ICP) is associated with increased mortality. When pharmacologic management of ICP fails, it is unclear whether there is a benefit from decompressive craniectomy (DC).

In the adult population, DC may prove to be beneficial for patients with stroke; however, for adults with TBI, there is no proven benefit compared with maximal medical management. In the pediatric population, Taylor et al.[1] conducted a randomized trial of DC versus maximal medical management for medically refractory intracranial hypertension and reported a potential benefit of early DC. This benefit is supported by 17 retrospective studies (Table 1).[1-17] Guidelines recommend consideration of DC in pediatric patients showing early signs of neurologic deterioration,

Key words
- Brain injuries
- Child
- Decompressive craniectomy
- Traumatic

Abbreviations and Acronyms
DC: Decompressive craniectomy
GCS: Glasgow Coma Scale
GOS: Glasgow Outcome Scale
ICH: Intracranial hypertension
ICP: Intracranial pressure
KOSCHI: King's Outcome Scale for Childhood Head Injury
TBI: Traumatic brain injury

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Table 1. Literature Review

Reference	No DC/ Control	Mean Age (years)	Study Type	Mean Follow-Up (months)*	Evaluation Criteria	Favorable Outcome (%)	Mortality Rate (%)
Dam Hieu et al., 1996[2]	2	8	Case report	8	Recovery of autonomy	100	0
Polin et al., 1997[3]	18	ND	Retrospective controlled study	ND	Good recovery	44	ND
Taylor et al., 2001[1]	14/27	10	Prospective randomized study	7.5	GOS and HUS at 6 months	54	23
Simma et al., 2002[4]	8	10	Case report	ND	GOS score >3	75	25
Figaji et al. 2003[5]	5	8	Case report	28.4 (14–42)	GOS score >3	100	0
Ruf et al., 2003[6]	6	7.8	Case report	6	Normal neurologic status	50	0
Josan and Sgouros, 2006[7]	6	13	Case report	>1 year	GOS score >3	100	0
Kan et al., 2006[8]	6/45	6.6	Retrospective controlled study	18.6	KOSCHI score >3	17	83
Rutigliano et al., 2006[9]	6	14.5	Case report	ND	FIM ambulation and feeding	83	0
Jagannathan et al., 2007[10]	23	11.9	Prospective study	62 (11–126)	GOS score >3	65	30
Adamo et al., 2009[11]	7	1.15	Case report	(1.5–24)	KOSCHI score >3	57	0
Thomale et al., 2010[12]	14/39	8.41	Prospective study	62.4 (12–126)	GOS score >3	86	20
Guresir et al., 2012[13]	34	13	Retrospective study	ND	Rankin Scale score	44	ND
Oluigbo et al., 2012[14]	37	6	Prospective study	ND	KOSCHI score >3	60	16
Khan et al., 2014[15]	25	6	Retrospective study	5	GOS score >3	64	36
Mhanna et al., 2014[16]	7/17	10.2	Retrospective study	48	GOS score >3	42	29
Beuriat et al., 2015[17]	55	7.4	Retrospective study	54	Independent daily life	80	18

ND, not determined; GOS, Glasgow Outcome Scale; HUS, health state utility index; KOSCHI, King's Outcome Scale for Childhood Head Injury; FIM, Functional Independence Measurement.
*Ranges in parentheses.

herniation, or developing intracranial hypertension (ICH) refractory to medical management. However, long-term clinical outcomes after DC have not been widely studied. In this study, we analyzed the long-term neurologic outcomes of children with severe TBI treated by DC in France to identify prognosis factors.

MATERIALS AND METHODS

Our local ethics committee approved this study, and we obtained consent of the families of the patients to be part of the study.We retrospectively analyzed 150 records of children (<18 years old) with severe TBI treated by DC at 10 French neuropediatric centers between January 2006 and December 2016. We excluded non-accidental trauma, rupture of an arteriovenous malformation, and vascular dissection previous to or concomitant with the trauma. The primary endpoint was the neurologic outcome at last follow-up as evaluated by the King's Outcome Scale for Childhood Head Injury (KOSCHI) score (**Figure 1**).[18]

We separated patients into 2 groups according to their long-term neurologic status. Patients with KOSCHI score >3 were considered the good prognosis group (group A), and patients with a score ≤3 were the bad prognosis group (group B). Afterward, we compared initial clinical and radiologic data between both groups to identify prognostic factors. Computed tomography scans were analyzed by a single radiologist to avoid interpretation bias. We used the Rotterdam score[19] to summarize the different lesions for its reproducibility and because we believe that the radiologic score best takes in account cerebral edema, which is a common lesion in children with severe TBI.[20]

The Shapiro-Wilk test was used to analyze data distribution. Normally distributed data were compared using the Student *t* test, and non–normally distributed data were compared using the Mann-Whitney test. Qualitative data were compared using the χ^2 test and Fisher exact test. The significance level was set at 5%. Statistical analyses were performed using XLSTAT software (Addinsoft Inc., Long Island City, New York, USA).

RESULTS

Epidemiologic Data

We included 150 patients from 10 centers. There were 103 boys and 47 girls (male/female ratio: 2.1/1). The mean age was 10.75 years

KOSCHI category	
1. Death	
2. Vegetative	The child is breathing spontaneously and may have sleep/wake cycles. He may have non-purposeful or reflex movements of limbs or eyes. There is no evidence of ability to communicate or respond to commands
3. Severe disability	a) Able to move part of their body/eyes intermittently or make purposeful spontaneous movements; for example, confused child pulling at nasogastric tube; may be fully conscious and able to communicate but not able to carry out any self-care activities b) Implies a continuing high level of dependency, but the child can assist in daily activities; is fully conscious but may have a degree of post-traumatic amnesia
4. Moderate disability	a) The child is mostly independent but needs a degree of supervision/actual help for physical and behavioural problems. Child has overt problems. b) Age-appropriately independent but has residual problems with learning/behaviour or neurological sequelae affecting function. Children with symptoms of PTSD are likely to fall in this category.
5. Good recovery	a) This should only be assigned if the head injury has resulted in a new condition which does not interfere with the child's well being and/or functioning; for example: minor headaches, unsightly scarring, abnormalities on brain scan without any new detectable new problems b) Implies that the information available is that the child has made a complete recovery with no detectable sequelae from the head injury

Figure 1. King's Outcome Scale for Childhood Head Injury score categories. KOSCHI, King's Outcome Scale for Childhood Head Injury; PTSD, posttraumatic stress disorder. (Reprinted from Crouchman M, Rossiter L, Colaco T, Forsyth R. A practical outcome scale for paediatric head injury. *Arch Dis Child.* 2001;84:120-124.[18])

(range, 0.63–17.94 years) with significantly younger patients in group B (9.6 years vs. 11.4 years in group A) (P = 0.0456).

Neurologic Outcome

During the initial hospitalization (0–49 days), 23 patients died, mostly during the first week after trauma. One more patient died 5 years after trauma during surgery for splenorenal anastomosis. Eleven patients were lost to follow-up. The total mortality rate was 17.2%.

Among the 115 survivors available for follow-up (mean 3.55 years [range, 0.09–12.96 years]), 87 patients (75.6%) had a KOSCHI score >3 and were included in group A, and 28 patients (24.4%) had a KOSCHI score between 2 and 3 and were included in group B. Deceased patients were also included in group B (Figure 2).

Initial Clinical Data

The median initial Glasgow Coma Scale (GCS) score was 6 (range, 3–15) and was significantly lower in group B (median score = 4 [range, 3–15]) compared with group A (median score = 7 [range, 3–15]) (P = 0.003). As expected, the initial GCS score strongly impacted the prognosis. Initial GCS score ≤6 was significantly associated with a poor neurologic outcome (P = 0.009). Prehospital pupillary abnormalities were found in 47 patients (31.4%): 26 with anisocoria and 21 with bilateral mydriasis. This finding was significantly associated with bad prognosis (P = 0.027). On admission to the intensive care unit, 36 patients had a pupillary abnormality (24%): 24 with anisocoria and 12 with bilateral mydriasis. Pupillary abnormality at admission was also associated with bad prognosis (P = 0.002).

ICP Monitoring

ICP was monitored in 70 patients. The mean initial ICP value (i.e., the first value recorded after placement of the monitor) was 31 mm Hg (range, 2–90 mm). The mean maximal ICP value (i.e., the highest value recorded before surgery) was 41 mm Hg (range, 5–100 mm Hg). Maximal ICP >30 mm Hg was associated with a bad prognosis (P = 0.0280), especially if the threshold was 40 mm Hg (P = 0.006). Finally, the best ICP value related to the long-term outcome was postoperative ICP. The mean postoperative ICP decreased to 10 mm Hg in group A but remained high at 30 mm Hg in group B (P = 0.0000012). We considered the threshold of postoperative ICP >15 mm Hg as a bad prognostic factor (P = 0.0003).

Radiologic Data

There were 81 preoperative computed tomography scans available for analysis. The radiologic parameters that statistically correlated with bad prognosis were Rotterdam score (P = 0.001), absence of cisterns (P = 0.0002), and median shift >5 mm (P = 0.019).

Surgical Data

The mean elapsed time between trauma and surgery was 1.45 days (range, 0–9 days). There was no evident correlation between the delay of surgery and prognosis (P = 0.2857). Hemicraniectomy was performed in 124 patients (83%), mostly in cases of unilateral lesions with median shift, and bifrontal craniectomy was performed in the 26 (17%) remaining patients who presented with diffuse edema without median shift. The technique did not impact the prognosis.

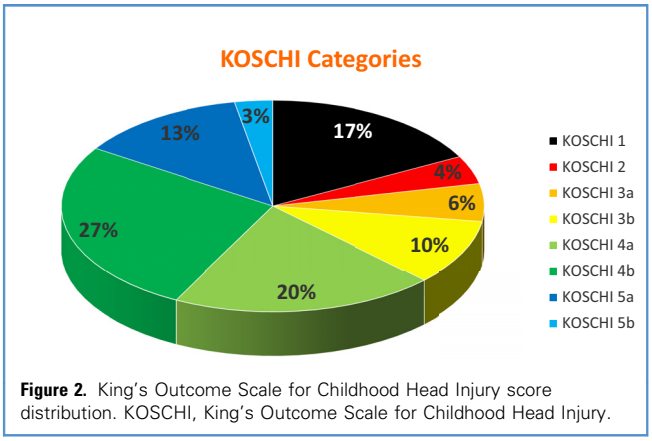

Figure 2. King's Outcome Scale for Childhood Head Injury score distribution. KOSCHI, King's Outcome Scale for Childhood Head Injury.

Surgical Complications

Apart from conditions commonly observed in patients admitted to the intensive care unit (e.g., urinary tract infection, pneumonia), 21% of patients had a surgical complication. Six (4%) patients had meningitis, and 14 (9.3%) patients developed a wound infection. Empyema was found in 3 (2%) patients. One patient with an external ventricular drain developed a cerebral abscess. Meningitis was the only complication significantly related to bad prognosis (P = 0.03). Ten patients presented with hemorrhagic complications (4 epidural hematomas, 4 subdural hematomas, 1 intraparenchymal hematoma, and 1 intraventricular hemorrhage after ventriculoperitoneal shunt placement).

DISCUSSION

In this series, we demonstrate good long-term outcome (mean follow-up 3.5 years) in 62% of children with severe TBI treated with DC and a mortality rate of 17%. These results are similar to results observed in previous series. Mortality rates varied between 0% and 36% in different publications.[5,7,8,11] Most of these studies describe good neurologic outcomes in >50% of children with severe TBI who underwent DC.[1-17,21-25] However, the low numbers of subjects (range, 2–55) as well as the single-center and retrospective nature of most of these studies do not allow statistically significant results. They are mostly case studies or series. Beuriat et al.[17] reported good neurologic outcomes for 65% of patients. The main assessment criterion also varies among studies. It is often subjective (i.e., not based on a formal evaluation or using a reproducible and validated tool). In the study by Polin et al.,[3] the evaluation criterion was a favorable neurologic progression, which was reported to be the case for 47% of children. According to Ruf et al.,[6] the neurologic condition is considered normal for 50% of children who sustained a severe TBI that required a DC.

More recent series are based on the Glasgow Outcome Scale (GOS)[1,4,5,7,10,12,15,16] or KOSCHI score.[8,11,14] We decided to use the KOSCHI score to evaluate long-term functional neurologic outcomes, as we consider it more suitable for the pediatric population and more discriminatory.[26] Five studies compared outcomes with a control group.[3,8,11,12,15] Josan and Sgouros[7] reported satisfactory neurologic outcomes (1-year GOS score ≥4) in 100% of children

with craniectomy compared with 50% in the control group; the mortality rates were 0% and 33%, respectively. Thomale et al.[12] used the same assessment criteria and noted that only 50% of the patients had a 1-year GOS score ≥4, with a mortality rate of 21% in the craniectomy group. In a retrospective case-controlled study of 34 patients, Mhanna et al.[16] concluded that although DC does not impact the survival of patients with TBI, it does considerably change the neurologic progression, with 42% of patients achieving a GOS score of 5 in the surgical group compared with 0% in the control group. Only one study was carried out in a prospective and randomized manner.[1] The authors showed that patients who underwent a DC had a favorable neurologic progression in 54% of cases compared with 14% in the control group ($P = 0.046$). However, the statistical significance of this study can be questioned because the threshold was lowered to 0.0022 to account for multiple analyses. Moreover, although the study was prospective and randomized, it was only a pilot study with no calculation of the required number of participants carried out a priori. Although it yielded encouraging results, it does not provide sufficient evidence to make formal conclusions about the indications for DC.

Studies have suggested that children with TBI treated by DC have better outcomes than adults owing to greater cerebral plasticity and a lack of systemic comorbidities.[3] Controversy remains, however, and results within the pediatric population are contradictory. Some authors report worse results, especially among older children, whereas others claim that the high vulnerability of the developing brain leads to irreversible damage in infants.[27] Güresir et al.[13] found no obvious difference in terms of long-term outcomes in children according to their age group (≤2 years, 2–6 years, 6–12 years, and 12–18 years). In our series, patients with bad prognosis (group B) were significantly younger ($P = 0.0456$), but we failed to define an age that would be a suitable threshold. We had only a few patients <2 years ($n = 12$) because we excluded nonaccidental trauma, which accounts for most of the etiologies in this age group.

A low initial GCS score in TBI is known to be a poor prognostic factor. In our series, the GCS score was statistically correlated with neurologic outcomes ($P = 0.0003$). Other pediatric series on DCs concluded that low GCS score was significantly associated with poor outcomes.[15] In other series, GCS score was not correlated with outcome.[12,13] The GCS score is difficult to assess in pediatric patients, especially if the GCS is not administered by a specialized pediatric team.[28-30] This is why our series did not include numerical GCS scores for 8 patients; rather, the state of consciousness of the patient was described as coma or agitated coma.

Several studies reported that pupillary anomaly in TBI is associated with poor prognosis.[31,32] We confirmed that the presence of a unilateral or bilateral mydriasis was strongly associated with poor prognosis, especially if it persisted after resuscitation maneuvers. However, in 16 patients with initial mydriasis, 6 (37.5%) had a favorable neurologic outcome with a KOSCHI score of 4a or 4b. In the series by Güresir et al.,[13] 60% of patients with unilateral mydriasis and 45% of patients with bilateral mydriasis had a favorable neurologic outcome.

There were 80 computed tomography scans available for the analysis. Only a few pediatric series have studied the impact of DC in children after assessing radiologic lesions with a validated score. Cerebral edema, which is the most frequent lesion in children with TBI (2–5 times more common in children compared with adults), used to be associated with bad prognosis.[19,33] However, in patients treated with DC, children seem to have a more favorable outcome than adults, even with diffuse post-traumatic brain edema. Therefore, we think that DC is particularly effective in this situation, and indications for this surgery must be considered after analyzing the degree of brain edema.

Not all studies described DC surgical protocols, and none of the studies compared different techniques. In the prospective randomized study of Taylor et al.,[1] a temporal craniectomy was performed without opening the dura mater. In other pediatric series, the technique and size of the craniectomy depended on the lesion side and presence of cerebral edema.

Our emphasis was on identifying the optimal timing for decompression. According to the different studies, the time frame varies from a few hours to several days. Nevertheless, all pediatric series evaluating the benefit of DC in children with cranial trauma agree on early surgery (within 48 hours after the trauma) to avoid the adverse effects of ICH.[1,12,22] Few groups have been able to demonstrate a significant effect of surgical delay on neurologic outcome. Josan and Sgouros[7] showed that DC surgery within 24 hours of trauma was associated with better outcomes compared with the control group. Figaji et al.[5] also reported better results in children with early decompression. For Polin et al.,[3] DC performed after the second day after trauma was a poor prognostic factor.

Our surgical complication rate was 21%. In previous series, the complication rate ranged from 0% to 47%. The difference in complication rates could be due to definition differences. We analyzed the specific complications associated with surgery rather than the overall complications. We observed hemorrhagic complications in 10 patients. Whether extradural or subdural, approximately 75% were contralateral to the craniectomy. Based on this, we propose that DC can decompensate contralateral lesions. Other complications were predominantly infectious and were reported in 25 (17%) patients. Meningitis occurred in 6 (4%) patients. Cerebrospinal fluid leakage from the scar was observed in 4 patients, but none of them had meningitis. In contrast, 14 (9.3%) patients had scar infections, 3 of whom developed meningitis. Three patients developed empyema. A patient with an external ventricular drain developed a cerebral abscess. Only the occurrence of meningitis was associated with neurologic outcome ($P = 0.03$). Few series describe infectious complications. Kan et al.[8] reported an 8.6% rate of infectious complications. This was similar to the 9% rate described by Beuriat et al.[17] Jagannathan et al.[10] described only 1 case of meningitis in their series of 23 patients.

Among frequently described complications, the persistence of a cerebrospinal fluid resorption disorder (hydrocephalus or hygroma) was reported in 40% of cases in the series by Khan et al.[15] Beuriat et al.[17] reported a 16% rate of subdural collections requiring subduroperitoneal shunting, and 10% of patients required ventriculoperitoneal derivation. The presence of an

external ventricular drain increases the risk of developing this complication, especially in patients with TBI treated with DC. It was discussed that DC, all the more so when associated with a duraplasty, could alter cerebrospinal fluid hydrodynamics.

CONCLUSIONS

No study has been able to adequately demonstrate the benefit of DC in children with severe TBI. Our study did not include a comparison group, which precludes us from reaching a conclusion about the effectiveness of this intervention. Several authors have reported good outcomes with children (50%–60% having good neurologic progression) and low rates of complications in the medium term, despite serious pathology. The overall risk/benefit ratio indicates that this surgical intervention is often warranted. Managing refractory ICH remains controversial and varies by center and surgeon training and experience. Children have a long life expectancy, making it important to provide them with suitable and intensive treatment. The benefit of including DC in the therapeutic arsenal for refractory ICH warrants a thorough investigation. A prospective randomized study with univariate and multivariate analyses may allow identification of better candidates for DC.

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