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ORIGINAL PAPER

The efficacy and safety of burr-hole craniotomy without continuous drainage for chronic subdural hematoma and subdural hygroma in children under 2 years of age

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Abstract
Purpose Various treatment modalities have been used in the management of chronic subdural hematoma and subdural hygroma (CSDH/SDHy) in children. However, few studies have examined burr-hole craniotomy without continuous drainage in such cases. Here, we retrospectively evaluated the efficacy and safety of burr-hole craniotomy without continuous drainage for CSDH/SDHy in children under 2 years old. We also aimed to determine the predictors of CSDH/SDHy recurrence.
Methods We conducted a retrospective chart review of 25 children under 2 years old who underwent burr-hole craniotomy without continuous drainage for CSDH/SDHy at a pediatric teaching hospital over a 10-year period. We analyzed the relationship between CSDH/SDHy recurrence and factors such as abusive head trauma, laterality of CSDH/SDHy, and subdural fluid collection type (hematoma or hygroma).
Results CSDH/SDHy recurred in 5 of the 25 patients (20 %), requiring a second operation at an average of 0.92 ± 1.12 months after the initial procedure. The mean follow-up period was 25.1 ± 28.6 months. There were no complications related to either operation. None of the assessed factors were statistically associated with recurrence.
Conclusions Burr-hole craniotomy without continuous drainage for CSDH/SDHy appears safe in children aged under 2 years and results in a relatively low recurrence rate. No predictors of CSDH/SDHy recurrence were identified. Advantages of this method include avoiding external subdural drainage-related complications. However, burr-hole drainage may be more effective for CSDH, which our data suggests is more likely to recur than SDHy, providing the procedure is performed with specific efforts to reduce complications.

Keywords Subdural fluid collection · Abusive head trauma · Recurrence · Irrigation

Introduction

Extra-axial fluid collections, such as chronic subdural hematoma and subdural hygroma (CSDH/SDHy), are pathological conditions that occur more frequently in children, particularly those under 2 years of age, because of physiological craniocerebral disproportion [4, 21]. Various treatment modalities have been used in the management of CSDH/SDHy in children, including percutaneous subdural tapping with or without drain placement, burr-hole craniotomy with or without drain placement, subcutaneous reservoir placement, subdural-subgaleal shunt [3], subdural-peritoneal shunt (SPS), and craniotomy.

SPS is believed to have the lowest recurrence rate of CSDH/SDHy among the various treatments and has been widely used in the treatment of pediatric CSDH/SDHy [13, 15, 21]; however, it frequently requires a second procedure to remove the shunt. Additionally, previous reports have

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indicated complication rates of 8–36 % after SPS and shunt removal surgery [4, 6, 12, 13, 15, 21]. Therefore, there is an apparent need for effective surgical management strategies that can minimize complication rates.

In adults, it has been suggested that burr-hole craniotomy with continuous drainage is superior to that without continuous drainage in adult CSDH patients [1, 14, 16, 20]. By comparison, only one report currently exists of burr-hole craniotomy without continuous drainage in the management of subdural fluid collection in infants [18], and the efficacy of this treatment for CSDH/SDHy in children has not been clarified. Because children are often difficult to keep on bed rest postoperatively, it is expected that the risk of complications, such as accidental removal of the drain and infection, is high. We therefore hypothesized that burr-hole craniotomy without continuous drainage would be more effective and safer for CSDH/SDHy in children than in adults.

Here, we retrospectively evaluated the efficacy and safety of burr-hole craniotomy without continuous drainage for treating CSDH/SDHy in children under 2 years of age. We also aimed to determine the predictors of CSDH/SDHy recurrence following this treatment.

Materials and methods

The records of 25 patients under 2 years of age with CSDH/SDHy treated using burr-hole craniotomy without continuous drainage at Kobe Children's Hospital between November 2004 and May 2015 were reviewed retrospectively. CSDH was defined as a subdural fluid collection surrounded by a typical neomembrane that contained dark red or brown liquid blood. SDHy was defined as a subdural fluid collection that contained xanthochromic or clear cerebrospinal fluid (CSF)-like fluid with or without a neomembrane at the time of the operation. Patients with a subdural abscess or subacute subdural hematoma were excluded. Patients with incomplete postoperative follow-up data were also excluded. All cases of CSDH/SDHy were confirmed by computed tomography (CT) scanning. The location of the CSDH/SDHy was classified as unilateral or bilateral, and the CT appearance of each was classified as hypodense, isodense, hyperdense, or mixed density. Age at the time of surgery, gender, etiology, presenting symptoms and physical findings, CT appearance, treatment, and the patient's clinical course were evaluated. Statistical analysis was performed on the association between CSDH/SDHy recurrence and the following factors: abusive head trauma (AHT), laterality of CSDH/SDHy, use of postoperative hemostatic agents, type of subdural fluid collection (hematoma or hygroma), and presence or absence of an outer membrane surrounding the subdural fluid.

A comprehensive review of the history and an extensive investigation and interview process was conducted in each case by the hospital's child protection team to determine whether AHT was involved. A fundus examination was also performed in every case as part of the AHT investigation.

Surgery

The decision to operate was made when patients were symptomatic (intracranial hypertension or seizure) or when they were asymptomatic but their CT scans showed the development of subdural fluid collection that presented a midline shift.

All patients underwent surgery under general anesthesia. A small curvilinear incision was made over the subdural hematoma or hygroma, and a single burr hole was created on the lateral aspect of the anterior fontanel along the coronal suture. If the anterior fontanel was closed or small, the burr hole was placed 2.5–5 cm lateral from the midline of where the fluid was especially thick. Irrigation of the subdural fluid was performed with sterile saline solution or artificial CSF (Artcereb; Otsuka Pharmaceutical Factory, Tokushima, Japan), except in cases in which irrigation presented difficulties. To prevent CSF leakage, the periosteum and galea were closed with 4-0 or 5-0 Vicryl (polyglactin 910, Ethicon Inc., Somerville, NJ, USA) when possible, and the skin was closed with 4-0 or 5-0 nylon monofilament. No drainage was used. All patients with bilateral CSDH/SDHy were treated on both sides during the same operation.

Postoperative care

After the surgical procedure, the patient was placed in the supine position, and the head was elevated to an angle of 10–15°. All patients received prophylactic antibiotic therapy before and after surgery. Management of postoperative hemostatic agents for each patient depended on the personal preferences of the surgeon. The follow-up period was calculated from the date of surgery to the last radiographic study, including CT and magnetic resonance imaging. Recurrence rate was defined as the percentage of CSDH/SDHy cases requiring reoperation. Repeated operations were performed when neurological symptoms reappeared due to increasing CSDH/SDHy, or when patients were asymptomatic but increasing CSDH/SDHy was observed on the operated side during CT.

Statistical analysis

All statistical analyses were conducted with EZR version 1.28 software (Saitama Medical Center, Jichi Medical University, Saitama, Japan). Fisher's exact test was used to determine the association between CSDH/SDHy recurrence and the following factors: AHT, laterality of CSDH/SDHy, use of postoperative hemostatic agents, type of subdural fluid collection

(hematoma or hygroma), and presence or absence of outer membrane surrounding the subdural fluid. Statistical significance was assumed at $p < 0.05$.

Results

Patients

During the study period, 25 children under 2 years of age with CSDH/SDHy underwent burr-hole craniotomy without continuous drainage. The mean age was 6.1 ± 4.5 months (range, 1–19 months; Table 1). There were 21 boys and 4 girls. The causes of CSDH/SDHy were as follows: head injury in 10 (drop in 7, fall in 2, traffic accident in 1), post-craniotomy for intracerebral hemorrhage in 1, derived from post-meningitis subdural effusion in 1, following conservative therapy for acute subdural hemorrhage resulting from shock and disseminated intravascular coagulation during abdominal surgery in 1. No specific cause could be identified in 12 patients. AHT was identified in 12 patients, with retinal hemorrhage observed in 10 of these.

Table 1 Characteristics of patients with chronic subdural hematoma and subdural hygroma

Characteristic	Value
Age (month)	
Mean ± SD	6.1 ± 4.5
Range	1–19
Sex	
Male	21
Female	4
Etiology	
Abusive head trauma	12 (10[a])
Head trauma	10
Post-craniotomy	1
Post meningitic subdural effusion	1
After ASDH due to intraoperative DIC	1
Symptoms and signs	
Vomiting	11
Seizure	10
Irritability	4
Lethargy	2
Hemiparesis	1
Laterality of hematoma	
Bilateral	17
Right	4
Left	4
Follow-up period (day)	
Median	613
Range	38–3506

ASDH acute subdural hemorrhage, *DIC* disseminated intravascular coagulation

[a] Retinal hemorrhage was observed in 10 patients with abusive head trauma. Values represent number of children unless otherwise indicated

The main clinical manifestation was vomiting in 11 cases (44 %), and seizure was the second most frequent sign, occurring in 10 cases (40 %). Also noted was irritability in four cases, lethargy in two cases, and hemiparesis in one case. Five patients underwent surgery because the hematoma increased progressively although they showed no signs of increased intracranial pressure.

Surgical management

CSDH/SDHy was bilateral in 17 patients and unilateral in 8. Thus, a total of 42 burr-hole craniotomies without continuous drainage were performed. One patient did not receive irrigation because of sufficient brain expansion. Data on irrigation were unavailable in one patient. The operative findings included subdural fluids that were dark red and bloody in 15 patients, xanthochromic in 8, and clear CSF-like in 2. A typical neomembrane was not found in five patients, who all presented with clear CSF-like or xanthochromic fluids. The 15 patients with dark red bloody fluids were diagnosed with CSDH, and the other 10 patients were diagnosed with SDHy. Figure 1 shows the pre- and postoperative CT images from one patient with CSDH and one with SDHy.

Eight patients were treated postoperatively with a hemostatic agent (tranexamic acid or carbazochrome sodium sulfonate hydrate or both). All patients were treated with antibiotics for 1–5 days, mostly with ampicillin or cefazolin. Bloody fluid from CSDH was found in 3 of 3 cases that appeared hyperdense on CT, 1 of 1 case that was isodense, 8 of 17 cases that were hypodense, and 3 of 4 cases with mixed density.

Recurrence and complication

The mean follow-up period was 25.1 ± 28.6 months (range, 1.25–115 months). There were no complications related to the surgical procedure. Five of the 25 patients (20 %) had recurrence of CSDH/SDHy that required a second operation at 0.92 ± 1.1 months (range, 0.20–2.9 months) after the initial burr-hole craniotomy (Table 2). Among the 42 hematomas and hygromas, 5 (11.9 %) recurred. Of these patients, case 1 was a 5-month-old boy with bilateral CSDH with mixed density on CT, who presented with convulsions. The recurrence occurred 16 days after initial surgery and he underwent SPS for right CSDH. It recurred again 97 days after the second surgery, and the patient underwent SPS revision. Case 2 was a 2-month-old boy with bilateral CSDH with mixed density on CT, who presented with lethargy and vomiting, and was subjected to abuse. The recurrence occurred 6 days after initial

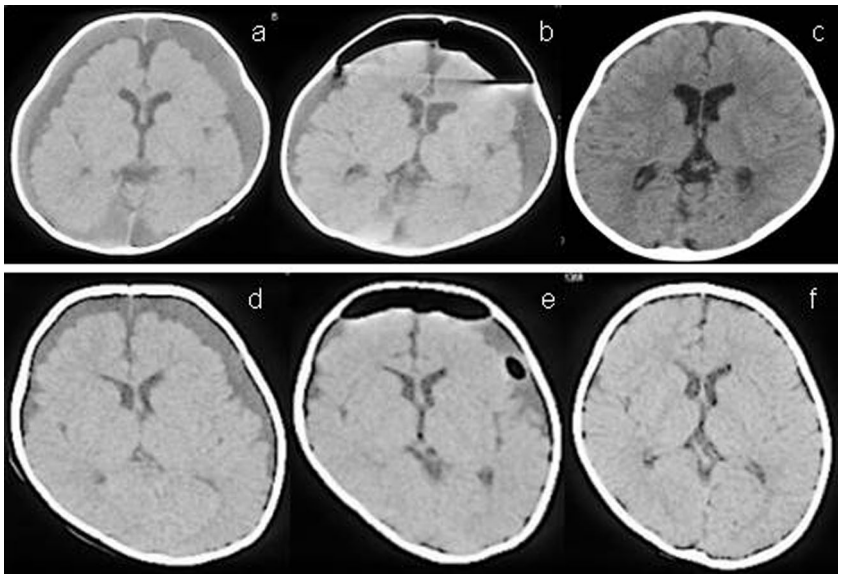

Fig. 1 Computed tomography images of a 3-month-old boy with bilateral chronic subdural hematoma (**a** preoperative; **b** postoperative; **c** last follow-up image 33.2 months after the operation) and a 2-month-old girl with bilateral subdural hygroma (**d** preoperative; **e** postoperative; **f** last follow-up image 10.3 months after the operation)

surgery and he underwent SPS for left CSDH. Case 3 was an asymptomatic 19-month-old boy with right CSDH with hypodensity on CT. The recurrence occurred 20 days after initial surgery and he underwent SPS. Case 4 was a 4-month-old girl with bilateral CSDH with hypodensity on CT, who presented with convulsions and was subjected to abuse. The recurrence occurred 88 days after initial surgery and she underwent burr-hole surgery for left CSDH using the same burr hole used in the initial surgery. Case 5 was a 17-month-old boy with bilateral SDHy with hypodensity on CT, who presented with vomiting. The recurrence occurred 10 days after initial surgery and he underwent SPS for left SDHy. All of these patients were treated successfully.

Predictors of recurrence

Recurrence occurred in 2 of 12 patients with AHT, compared with 3 of 13 without AHT (16.7 vs. 23.1 %, respectively). The relationship between AHT and recurrence was not statistically significant (Table 3). Recurrence occurred in 4 of 17 patients with bilateral CSDH/SDHy, compared with 1 of 8 patients with unilateral CSDH/SDHy (23.5 vs. 12.5 %, respectively). The relationship between bilateral hematoma and recurrence was not statistically significant. Recurrence occurred in 4 of 15 patients with CSDH, compared with 1 of 10 patients with SDHy (26.7 vs. 10.0 %, respectively). There was no significant difference between CSDH and SDHy in terms of recurrence. Use of postoperative hemostatic agents and presence of outer membrane surrounding the subdural fluid were not related to recurrence.

Discussion

We found that burr-hole craniotomy without continuous drainage for CSDH/SDHy appeared safe in children under 2 years old and was associated with a relatively low recurrence rate. We also found that AHT, bilateral CSDH/SDHy, use of postoperative hemostatic agents, subdural fluid collection types (hematoma or hygroma), and presence or absence of an outer membrane surrounding the subdural fluid were not significantly associated with recurrence.

Tolias et al. reported on 18 patients under 2 years old with subdural fluid collection who had been followed for a median of 12 months [18]. Recurrence of subdural fluid collection was observed in 22 % of these patients. However, the investigators excluded all patients with a history of AHT, and some patients required subdural drainage but the rate was not

Table 2 Surgical treatment of chronic subdural hematoma and subdural hygroma

Initial operation	Reoperation	Third operation	No.
Burr-hole craniotomy without drain			20
Burr-hole craniotomy without drain	Burr-hole craniotomy without drain		1
Burr-hole craniotomy without drain	Subdural-peritoneal shunt		3
Burr-hole craniotomy without drain	Subdural-peritoneal shunt	Subdural-peritoneal shunt	1
Total			25

Table 3 Factors potentially related to recurrence of chronic subdural hematoma and hygroma

Factor	No. of patients (%)		*p* value 95 % CI
	Recurrence	No recurrence	
Abusive head trauma ($n = 12$)	2 (17 %)	10 (83 %)	$p = 1$ 95 % CI 0.05–7.33
Bilateral hematoma ($n = 17$)	4 (24 %)	13 (76 %)	$p = 1$ 95 % CI 0.01–6.21
Subdural fluid is hematoma ($n = 15$)	4 (27 %)	11 (73 %)	$p = 0.62$ 95 % CI 0.25–179
Postoperative hemostatic agents ($n = 8$)	1 (12.5 %)	7 (87.5 %)	$p = 1$ 95 % CI 0.01–6.21
Existing outer membrane ($n = 20$)	4 (20 %)	16 (80 %)	$p = 1$ 95 % CI 0.07–61.4

recorded. To our knowledge, the present study is the largest and most comprehensive assessment of the efficacy and safety of burr-hole craniotomy without continuous drainage in the treatment of children under 2 years of age with CSDH/SDHy.

We observed a recurrence rate of 20 %, which is unsatisfactorily high. However, it is comparable to previously published data for burr-hole craniotomy without drainage. In adults, the recurrence rate of CSDH after burr-hole craniotomy without drainage is 17–33 % [5, 14, 16, 20, 22]. Together, these results indicate that the recurrence rate is similar in pediatric and adult patients.

In previous reports, recurrence occurred after SPS in 0–12 % of patients, and postoperative complications were observed in 8–36 % of cases [4, 6, 12, 13, 15, 21]. Compared with these results, our findings suggested that burr-hole craniotomy without continuous drainage had a slightly higher rate of recurrence but lower rate of complications than SPS. However, these previous reports examining SPS may have underestimated the rate of recurrence because most of them included shunt obstruction as a complication rather than a recurrence. Because previous reports indicated that 38–93 % of cases required removal of the shunt after SPS [4, 12, 13, 15, 21], the burr-hole craniotomy performed in the present study had a higher rate for completion of treatment in a single surgery than SPS. Additionally, complications related to removal of the SPS, such as subdural hemorrhage due to adherence of the proximal catheter, may occur in up to 18.6 % of cases [12].

Percutaneous subdural tapping for subdural fluid collection in children has a higher recurrence rate (42–100 %) than that found using the present treatment strategy [2, 12, 13, 15, 18]. In addition, percutaneous subdural tapping has been associated with a high infection rate (25 %) [18]. The major differences between percutaneous subdural tapping and burr-hole craniotomy are in the amount of hematoma that can be evacuated and in the ability of craniotomy to be combined with irrigation. Some reports have shown that significantly greater levels of tissue plasminogen activator and interleukin-6 are detected in recurrent hematomas than in non-recurrent cases; a similar tendency has been noted for the α2-plasmin inhibitor–plasmin complex in hematomas [10, 11]. Therefore, we speculate that the reduction of these factors by irrigation may effectively prevent recurrence. Indeed, irrigation of the hematoma cavity is accepted as an effective treatment for CSDH in adults [5, 7, 14]. However, some authors have stated that the rapid decrease in intracranial pressure obtained by irrigation can lead to adverse effects [7], although none of these adverse effects were observed in any of the patients in the present study.

In our study, no relevant complications were observed after burr-hole craniotomy without continuous drainage. According to a previous meta-analysis of adult CSDH, the complication rate is 15.6 % in cases with drainage and 10.6 % in cases without drainage [14]. Complications following drain placement in children may be higher than those in adults owing to more delicate skin and difficulty in maintaining postsurgical bed rest in children. Tolias et al. previously reported that complications occurred in 17 % of patients after burr-hole craniotomy without drainage for infant subdural fluid collection [18]. However, the results of our study indicated that a lower complication rate may result when care is taken to prevent CSF leakage, with adequate closure of the periosteum and galea, and sufficient infection control is provided.

Because there is a higher incidence of recurrence in CSDH compared with SDHy as shown in this study, different treatment strategies could be considered for each condition. Burr-hole drainage might be more effective for CSDH, providing efforts are taken during the procedure to reduce complications.

AHT, bilateral CSDH/SDHy, use of postoperative hemostatic agents, subdural fluid collection type (hematoma or hygroma), and presence of an outer membrane surrounding the subdural fluid were not significantly associated with recurrence in this study. Children with AHT have a high risk of poor outcome [17]. Cerebral atrophy following abuse may increase the recurrence rate of CSDH/SDHy [8, 24]; however,

AHT was not related to recurrence in the present study. Although bilateral CSDH has been considered a risk factor for recurrence of CSDH in adults [19], no significant correlation was found for children under 2 years in the present study.

It has been reported that the recurrence rate is higher in patients with lower protein content in the subdural fluid [24], so recurrence could occur more frequently in SDHy than CSDH. Our study showed no significant difference in recurrence rates between CSDH and SDHy, but we found that cases of CSDH showed a higher tendency for recurrence than did cases of SDHy. The exact pathogenesis and diagnosis of SDHy are still a matter of debate and uncertainty, particularly in infants. However, CSDHs are commonly shown to be derived from SDHys [23]. SDHys can develop neomembranes from the proliferating dural border cells, with neovascularization. It has been suggested that repeated microhemorrhages from these fragile new vessels convert an SDHy into an expanding CSDH [23]. If SDHys develop into CSDHs, it may be difficult to clearly distinguish between these two pathophysiological states, especially in cases with conditions that border CSDH and SDHy.

The limitations of this study are that a small number of patient records were examined and that this is a retrospective study. However, the number of cases is not necessarily considered small for our local population because subdural hematoma and effusion have been shown to occur with an annual incidence of 12.54 per 100,000 infants under 2 years [9], and the population under 2 years of age in Kobe City in 2011 was only 2465.

Conclusions

Burr-hole craniotomy without continuous drainage for CSDH/SDHy in children under 2 years of age appeared safe, with a relatively low recurrence rate. Additionally, AHT, bilateral CSDH/SDHy, use of postoperative hemostatic agents, type of subdural fluid collection (hematoma or hygroma), and presence or absence of an outer membrane for the subdural fluid were not significantly associated with recurrence.

The advantages of burr-hole craniotomy without continuous drainage include avoiding external subdural drainage-related complications, such as infection and accidental removal of the drain, and preventing SPS-related complications. However, if efforts are taken during the procedure to reduce complications, external drainage could be a more effective treatment for CSDH, which our data indicate is more likely to recur than SDHy. A larger prospective and randomized control study of burr-hole craniotomy without continuous drainage and SPS or external subdural drainage will be needed to confirm our findings and to address remaining issues, including differences in treatment strategies for CSDH and SDHy.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflicts of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent For this type of study, formal consent is not required.

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