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Paediatric chronic subdural haematoma: what are the predisposing factors and outcomes in management of these cases?

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Abstract

Introduction Chronic subdural hematoma (cSDH) is a disease of the elderly population. Incidence in paediatric population is relatively uncommon. Child abuse, birth trauma, coagulopathy and shunt surgeries represent major causes. Major impact of the disease on life of patient due to recurrence and repeat surgical procedure is significant, not to mention the burden on health care system.

Material and methods We retrospectively reviewed our institute data for chronic sub-dural cases for the past 10 years (2008–2018) and collected data on the demography, clinical features, metabolic workup, mode of treatment, recurrence rates, predisposing factors, laterality, hematoma characteristics and factors associated with recurrence in all cases with less than or equal to 18 years of age.

Results A total of 30 such cases were found in a period of 10 years (2008–2018). The mean patient age was 7.3 years (range 2 months–17 years), with 20 males (66.67%) and 10 females (33.33%). Raised intracranial pressure (n=9) was the commonest presenting symptom in 30% of cases followed by seizures in 26.67% (n=8). The previous shunt was the commonest predisposing factor seen in 43.33% (n=13). cSDHs were unilateral in 56.67% cases (n=17) and bilateral in 43.33% (n=13). Burr hole craniostomy was done in 27 cases (90%), and conservative management was done in three cases (10%). Follow up was available for 27 cases (90%) with a mean follow up duration of 24 months. Recurrence rate was 30% (n=9). Shunt surgery contributed to 77% of bilateral disease (p=0.009). Child abuse was not reported in our series.

Conclusion Presence of paediatric cSDH is alarming, and the physician should be alerted to look for underlying cause and rule out child abuse. Detailed metabolic, skeletal workup is required. Treatment of primary pathology should be the goal as CSF diversion is not the solution to all problems, but can definitely be the cause of all the problems.

Keywords Chronic subdural hematoma · Shunt · Trauma · Child abuse · Hygroma

Introduction

Chronic subdural hematoma (cSDH) is a common disease of the geriatric population. In the paediatric age group, the incidence of this condition is relatively uncommon, and literature is sparse on this topic. The English literature search

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¹ Neurosurgery, NIMHANS, Hosur Road, Bangalore, Karnataka, India revealed only four major case series in the recent past [1–4]. The common underlying factor of diffuse brain atrophy or anticoagulant, which is predominant in elderly cSDH, is rare in paediatric population. Child abuse, birth trauma, coagulopathy and shunt surgeries are the major predisposing factors in this age group. Long bone and fundus examination along with skin bruises at various stages of healing are invaluable findings pointing to the underlying child abuse.

There are no specific presenting symptoms. The usual presenting symptoms are headache, vomiting, seizure, focal neurological deficit or altered sensorium. In infants, it can present in the form of refusal to feed, increased head circumference and delayed or regression of milestones [4]. Imaging forms the cornerstone for the diagnosis and guides the treatment strategy along with the clinical findings. Various treatment options have been suggested for the management of paediatric cSDH ranging from expectant management, subdural-peritoneal shunt (SPS), subdural-subgaleal shunt, twist drill tapping and burr hole evacuation with or without subdural drain to craniotomy with membrane excision. In neonates/infants, fontanelle tapping remains an important procedure for drainage of sub-dural haematomas. Recently, the role of dexamethasone and atorvastatin has been suggested in the management of recurrent paediatric cSDH [5]. Here, we report our experience of 30 cases of paediatric cSDH managed at our institute over a period of 10 years. The detailed demographic, clinical, imaging, histopathological features and outcome have been studied. We also tried to analyse the factors predicting the recurrence of the disease.

Materials and methods

This is a retrospective study performed in a single tertiary care referral institute. All the cases of paediatric cSDH with age less than 18 years who were managed surgically or medically at our institute between the years 2008 and 2018 were included. The recurrent cases of cSDH were excluded from the current study. Data was obtained from the hospital's medical record section. The relevant demographic, clinical details, imaging features, surgical details and outcome were studied using chi-square analysis. Computed tomography (CT) or magnetic resonance imaging (MRI) of the brain was done in all the patients. All the attempts were made to find out the underlying predisposing factors especially child abuse. Chest X-ray and spine and extremity X-rays were done along with fundus examination in the suspicious cases of child abuse. Metabolic work up was done when genetic diseases like mucopolysaccharidosis were considered in the differential diagnosis. Routine blood investigations like haemogram, ESR, renal and liver function tests and coagulation profile were done in all the patients.

Decision for surgery was taken based on the clinical presentation and/or radiological progression of the disease. Standard surgical procedure of burr holes with evacuation of chronic SDH was performed under general anaesthesia. The number of burr holes made was decided based on the imaging findings. Wherever membrane was seen during the surgery, biopsy was taken from the membrane and sent for histopathological examination. Decision to keep the subdural drain was left to the surgeon's discretion or lack of brain surfacing at the end of surgery. Drain was removed in 48 h after obtaining a plain CT head. Clinical follow up was obtained at the interval of 3 months to look for recurrence in which re-surgery was performed. Follow up was obtained from the case sheet or, in some cases, telephonic conversation to know their current status.

Results

Clinical presentation

A total of 30 cases of paediatric cSDH were included in the study. The mean patient age was 7.3 years (range 2 months–17 years). The study consisted of 20 males (66.67%) and 10 females (33.33%). Raised intracranial pressure (n=9) was the commonest presenting symptom in 30% of cases followed by seizures in 26.67% (n=8) and headache and altered sensorium in 13.33% each (n=4). Here, raised ICP was defined as any symptom suggestive of increased intracranial pressure such as vomiting, increased head circumference, papilledema or up-gaze restriction. Hemiparesis was seen in two cases: increase in head circumference, regression of milestones and behaviour disturbance in one child each (Tables 1 and 2).

Predisposing factors

Twenty-five cases (83.33%) had underlying predisposing factors. Previous shunt was the commonest predisposing factor seen in 43.33% (n=13). History of trauma was found in 23.33% (n=7) including one birth trauma induced by forceps delivery, followed by one case of coagulopathy, infective (HIV with TB), brain atrophy (mucopolysaccharidosis) (Fig. 1), antiplatelet therapy and post occipital encephalocele surgery each (Table 3).

Our centre usually does not operate on neonates; hence, the usual indications for shunts such as preterm delivery or intraventricular haemorrhage was not contributory. In 11 out of the 13 cases (84.6%), the shunts (Fig. 2) were inserted as a result of obstructive hydrocephalus, i.e. due to tumours either posterior fossa, brain stem or thalamic lesions. All of these shunts were fixed medium pressure ventriculoperitoneal shunts. Interestingly, the period between shunt insertion and presentation as a chronic sub-dural was less than 3 months in all cases and ranged from 1 to 3 months, further lending credence to our hypothesis of shunts leading to hematoma formation. In two cases of congenital hydrocephalus where a medium pressure shunt was inserted, the latency period between the index shunt and presentation of the chronic sub-dural was 28 months and 20 months respectively, with the former having undergone 2 shunt revisions. In both cases, the shunt was ligated and sub-dural evacuated with no symptoms of hydrocephalus at follow-ups.

Imaging features

CT brain was done in 25 cases (83.33%) and MRI in five cases (16.67%). Abnormal skeletal X-rays were seen in three

Series	Cases	Presentation	Skeletal findings	Treatment	Shunt	Drain	Recurrence
Parent [3]	28	Macrocephaly Lethargy	NA	Twist drill	NA	None	NA
Ers et al. [6]	33	Convulsions Macrocrania Bulging fontanelle	NA	Continuous external subdural drainage	Recurrences	NA	16
Kyoo et al. [7]	ω	Seizures Motor weakness	NA	Burr holes Subduroperitoneal shunt	3 cases (2 after recurrence)		2 cases in burr hole treatment group 0 in shunt group
Swift and McBride [4]	S	Macrocephaly Trauma	NA	Burr holes	NA	None	NA
Vinchon et al. [8]	244	Intracranial hypertension Epilepsy Macrocephaly Hypotonia Localized deficits Abusive head trauma	Not mentioned	Subduroperitoneal shunt	All cases	NA	15.6% cases had shunt related complication Zero recurrences
Mori et al. [9]	Ś	Headache Vomiting Hemiparesis Ruptured arachnoid cyst (All patients)	NA	Burr holes	NA	None	None
Lin et al. [10]	36	Seizures Bulging fontanelle Consciousness disturbance	Not mentioned	Subdural drainage	Recurrences	NA	2
Zouros et al. [1]	5	Macrocephaly Lethargy	Yes	Burr holes	NA	All cases	1
Kurschel et al. [11]	161	Intracranial hypertension Lethargy Seizures Hemiparesis Abusive head trauma Macrocephaly	Multiple fractures in abusive trauma cases	Unilateral valveless subduroperitoneal shunt	All cases	ΝA	1—symptomatic rebleed; 17—revision of shunt due to shunt-related complications
Blauwblomme et al. [12]	18	Intracranial hypertension Seizures Hemiparesis Abusive head trauma	Not mentioned	Subduro-subgaleal drainage	Recurrences	NA	4
Matsuo et al. [2]	25	Seizure Vomiting	Yes	Burr holes	NA	Recurrences	5
Murat et al. [13, 14]	15	Ruptured arachnoid cyst Focal deficits	NA	Single burr hole	Recurrences	NA	2

Table 1 (continued)							
Series	Cases	Cases Presentation	Skeletal findings	Treatment	Shunt	Drain	Recurrence
Nguyen et al. [14]	50	Seizures Bulging fontanelle Abusive head trauma (All cases)	Not mentioned	Percutaneous trans fontanelle tap Burr holes with or without drains Subduroperitoneal shunt Minicraniotomy	Yes	Yes	45% in trans fontanelle tap required shunt 67% in Burr holes group required minicraniotomy and redo burr holes None in burr holes with subdural drains
Tinois et al. [15]	10	Ruptured arachnoid cyst (all patients) Headache, vomiting Hemiparesis seizures Trauma	NA	Subduroperitoneal shunt	All cases	AN	No recurrence
Wright et al. [16]	×	Abusive head trauma Macrocephaly	Multiple fractures	Subduroperitoneal shunt	All cases	NA	87.5%
Palmer and Albert [17]	11	Abusive trauma Macrocephaly Seizures Lethargy Apnoea/bradycardia	Multiple fractures	Parietal minicraniotomy with subgaleal pocket	None	NA	1 (due to second trauma)
Our study	30	Headache, vomiting, seizures, hemiplegia, behavioural disturbances, regression of milestones, macrocephaly	Linear fracture, thick calvaria, mucopolysaccharidosis	Burr-holes, craniotomy, subduroperitoneal shunt, conservative	2 cases (recurrent)	3 cases	9 (Burr hole in 6 cases; SPS in 2 cases; Craniotomy in 1 case)

Table 2 Demographic and other details with outcome

Mean age	7.3 years (range: 2 months–17 years)
Gender (M/F)	2:1
Presence of predisposing factors	83.33% (<i>n</i> =25)
Use of drain	3
Membrane excision/biopsy	5
Follow up Recurrence	90% (<i>n</i> =27) 9/27 (30%)
Second surgery Re-exploration of burr holes	In all 9 cases 6
SPS Craniotomy	2 1

cases with history of trauma. One case had the acute component on CT brain with history of recent trauma. cSDHs were unilateral in 56.67% cases (n = 17) and 43.33% in bilateral (n = 13) (Table 4).

Treatment

Few patients (n=3) were managed medically, as the symptoms were not severe, and there was no deficit on examination. For those children who underwent surgery, burr hole and evacuation of cSDH were done: 27 cases (90%). Conservative management was done in three cases (10%) with minimal symptoms and no midline shift/mass effect on imaging. Subdural drain was kept in three of the operated cases (Table 5). In five cases, membrane was seen during surgery, and biopsy was performed. All the materials were sent for histopathological examination. None of the biopsy samples revealed evidence of infection or malignancy. All those cases presented with chronic subdural in conjunction with a shunt; the shunt was ligated and primary pathology, i.e., tumour, was dealt with directly.

One case which highlights the need for direct management of the tumour was that of a 9-year-old boy who presented to the emergency department with a headache and vomiting for 3 days. On doing a computed tomography (CT) scan, there was a posterior fossa lesion with hydrocephalus. Since the primary was not dealt with directly, the child was shunted. Subsequently, the child returned a month later with similar complaints of drowsiness and poor feeding and the post-ventriculoperitoneal shunt showing collapsed ventricles with chronic subdural haematoma (Fig. 3). The posterior fossa lesion was removed and the shunt ligated in the same procedure. Postoperatively, the plain CT scan showed decompression of the posterior fossa lesion with a ligated shunt in situ; however, the chronic sub-dural remained, and the child did not improve in sensorium. Burr hole and evacuation of the chronic SDH did not result in improvement in sensorium

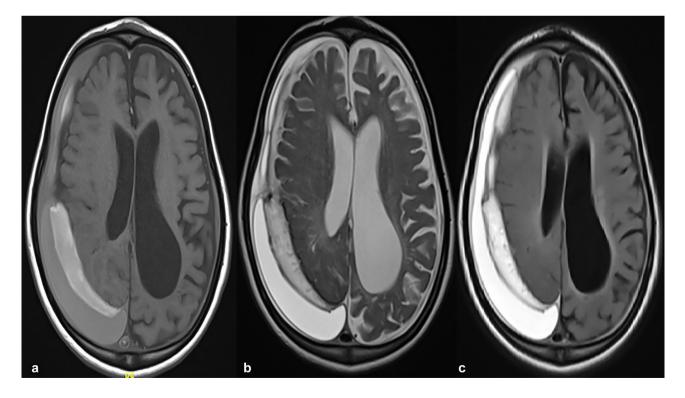


Fig. 1 T1, T2 and Flair axial images of paediatric chronic SDH due to mucopolysaccharidosis with macro-crania

Presentation	Number	In percentage (%)
Raised ICP	9	30
Seizure	8	26.67
Headache	4	13.33
Altered sensorium	4	13.33
Limb weakness	2	6.67
Normal F/U	2	6.67
Milestones regression	1	3.33
Behaviour disturbance	1	3.33
Head circumference	1	3.33
Predisposing factors		
Shunt	13	43.33
Fall	7	23.33
Antiplatelet	1	3.33
Coagulopathy	1	3.33
Atrophic brain (mucopolysaccharidosis)	1	3.33
Infection (HIV, TB)	1	3.33
Encephalocele (post-Surgery)	1	3.33

 Table 3
 Presenting symptoms and predisposing factors of chronic

 SDH in current series of cases

(Fig. 3e, f). A diffusion-weighted imaging and apparent diffusion coefficient MRI images showed multiple venous infarcts secondary to compression by chronic hematoma causing ischemic encephalopathy. The histopathology was that of pilocytic astrocytoma. This case highlights the need for direct management of the lesion or reduction in the time interval between shunting and tumour surgery.

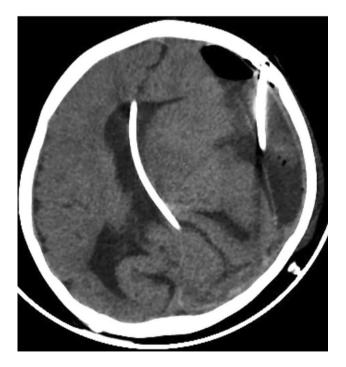


Fig. 2 Axial cuts of plain CT showing evacuated recurrent chronic SDH secondary to a shunt with a sub-dural drain in situ

Table 4 Imaging features used for diagnosis of paediatric chronic SDH

Imaging	Numbers
CT diagnosis	25
MRI diagnosis	5
Laterality	
Unilateral	17
Bilateral	13
Abnormal skeletal X- rays	3
Acute component on SDH	1

Follow up and outcome

There was no new post-operative morbidity or mortality in any of the patients. Hemiparesis improved in both the cases, and post-operative period was seizure free for all those presenting with seizures. Follow up was available for 27 cases (90%) with mean follow up duration of 24 months (range: 3 months–10 years) with a median follow up of 15 months. Recurrence was seen in 30% of the available follow up (n=9). All the recurrent cases underwent second surgery. Re-exploration of the burr hole and evacuation were done in six cases, SPS in two cases, and one case required craniotomy and evacuation of SDH.

Recurrence and factors

All the demographic details and other imaging/surgical parameters were analysed to look for the factors predicting the recurrence. Gender, history of trauma, use of subdural drain, laterality and acute component in SDH did not have any significant correlation predicting the recurrence. The trend of recurrence was more in bilateral disease (55.6% vs 44.4%, p=0.075) and those who had undergone shunt in the past (55.6% vs 44.4%, p=0.57); however, the results were not statistically significant, which may be due to the small sample size.

As noted above, a total of 13 patients had a history of undergoing shunt procedure before presentation; the majority (n = 10), approximately 77%, had bilateral disease (p = 0.00943), which is statistically significant, and out of 9 recurrent cases, 5 patients had a history of undergoing shunt, and out of these 5 cases, 4 cases had hematoma as collection, and 1 patient had hygroma which was managed with SPS.

Discussion

Pathophysiology

Among the paediatric age group, cSDH is more prone to occur under the age of 2 years because of physiological craniocerebral disproportion [8, 18] which can cause traversing

Character	<18yrs (current study)	20-40yrs	Older adults [20, 21]
Presentation	Macrocephaly, lethargy, seizures	Raised ICP	Decreasing mental status, severe head- aches, hemiplegia [19]
Occurrence	30 cases/10 yrs	136 cases/10yrs	34,829 cases/43 yrs [20]
Treatment	Burr holes	Burr holes and craniotomy	Twist drill, Burr holes and craniotomy [20]
Recurrence	9/27 (30%)	13/92 (14%)	10.7–11% [20]
Predisposition	Shunts, child abuse	Trauma	Trivial trauma, Anti-coagulants [21]

Table 5 Comparison of the presentation, treatment and outcomes of chronic SDH among all possible age groups reported in literature

veins to tear and form a subdural collection. Arachnoid villi consist of its capsule, fibroblasts and endothelium. These arachnoid villi connect the column of arachnoid cells to venous sinuses. The shear strain at this attachment, which is produced by acceleration/deceleration of the brain, results in disruption of arachnoid column. So, CSF from exposed subarachnoid space communicates freely with the subdural space leading to subdural collection [19].

Predisposing factors

In our study, as noted above, the most common predisposing factor is history of shunt for various pathologies, and it is shown that the presence of shunt predisposes the child to bilateral disease which is responsible for macrocrania, but will cause delay in cerebral development; the presence of shunt has caused maximum number of recurrences in our study, but it was statistically insignificant which may be attributed to the small sample size, which begs the question, should shunt be done when primary pathology can be handled without it?

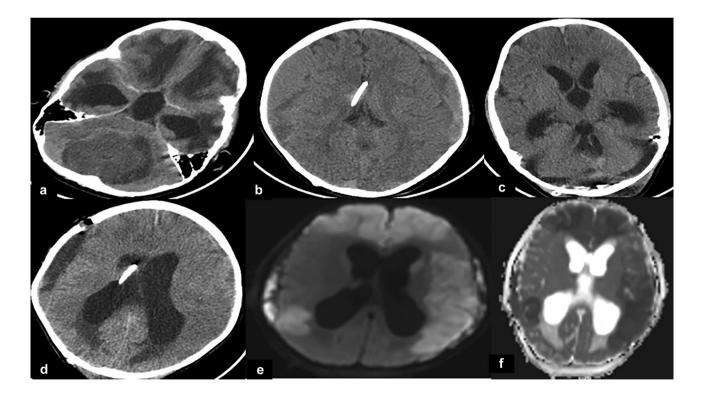


Fig. 3 a Axial cut of a plain CT scan showing a posterior fossa lesion with supratentorial hydrocephalus; **b** post-ventriculoperitoneal shunt showing collapsed ventricles with chronic subdural hematoma; **c** axial cut of a plain CT scan showing decompression of the posterior fossa lesion with a ligated shunt in situ; **d** post-burr hole and evacua-

tion of chronic SDH with a shunt in situ; \mathbf{e}, \mathbf{f} axial cuts of diffusionweighted imaging and apparent diffusion coefficient MRI images showing multiple venous infarcts secondary to compression by chronic hematoma causing ischemic encephalopathy It is pertinent to mention the 5 cases which had no predisposing factors. One possible explanation could be an unnoticed trauma which was not reported. Many infants with TBI have no clinical symptoms of brain injury [20–29], and even if these are present, the child may essentially be asymptomatic. It is only after weeks have passed and the hematoma has converted into a chronic sub-dural do these children present with headache, seizures or altered sensorium. Another explanation can be rupture of an unnoticed arachnoid cyst which has presented directly as a chronic sub-dural hematoma [30]. Minor head trauma often precedes complications causing rupture of an arachnoid cyst. Burr hole and evacuation done initially may relieve the child of the symptoms while the arachnoid cyst may go unnoticed [30].

Clinical feature

Wang et al. have reported a case of cSDH in a 9-year-old child with history of dodge ball injury [20]. The above study has suggested that diagnosis of cSDH should be suspected for any patient with or without a history of trauma presenting with the following: (1) any change in the neurological state, (2) focal neurological deficit and (3) progressive severe headache. In the series from the most recent decade [14, 17, 25–27], some cases had minimal headaches or were asymptomatic shunted hydrocephalic patients scanned during routine follow-up. Our series has reconfirmed the male preponderance similar to other study [3]. Most cases presented with symptoms of seizures, bulging fontanelles, anorexia, and lethargy. At least 40% of cases from the study by Parent [3] were related to child abuse, which is in contrast to many authors [21, 22]. Birth trauma, on the other hand, accounted for only a few cases in other series. We had one such case of birth trauma related to forceps delivery.

Imaging

MRI is superior to CT scanning for differentiating subdural from subarachnoid collections. Zourous et al. have explained the theory of dynamic CSF hemato-hygroma of traumatic origin based on the MRI findings [1]. Suh et al. demonstrated that diffusion-weighted MRI was more sensitive in detecting white matter injury as compared to standard MRI and also for predicting the outcome [23]. In our series, MRI was done in only 5 cases, mainly in those with suspicion of metabolic cause or non-traumatic origin. CT was predominant modality of imaging done in 25 cases. CT provides useful information for calvarial fracture in case of traumatic brain injury especially in cases of child abuse. Ultrasound through the anterior fontanelle has important role in infant when undergoing evaluation of macrocephaly. It can reveal the amount of fluid in the subdural fluid and also the septations within it. Its utility might increase in the future because of ease of availability, can be done bedside, better logistic aspects and also lack of sedation as compared to MRI [4]. The role of ultrasound in normal-sized infants for the detection of subdural hematomas is limited by its lateral location which hampers its correct visualization. Indeed, ultrasound has a limited value for both initial diagnosis and postoperative assessments.

Treatment

The various treatment options described in the literature are percutaneous tapping, burr hole drainage with or without subdural drain, craniotomy and evacuation, SPS or subduralsubgaleal shunt. SPS has been supposed by many to have the least recurrence for the paediatric cSDH; however, it often requires second look surgery to remove the shunt at a later date [8, 24, 25]. The use of modern high-viscosity programmable valves offers the possibility to avoid the second surgical procedure, which is not risk-free. In a systematic review by Xu et al. [31], programmable shunts had significantly lower rate of revision ((RR = 0.56, 95%CI (0.45,0.69), p < 0.01) in favour of programmable valves) in hydrocephalus as compared to fixed pressure shunts due to the lower incidence of blockage in presence of high-viscosity fluids. Hence, the same may also be applied in drainage of chronic sub-durals. There are few studies which mention that burr hole with subdural drain placement is a better option for management of cSDH in adults [26-29]. The implementation of the same procedure is not feasible in the paediatric age group especially infants because of difficulty in keeping the patients restricted to bed. This leads to accidental removal of the drain and also increasing the risk of infection [2].

We have used burr hole evacuation method for 27 patients, and three patients were managed medically. Subdural drain was used only in three of our patients, and all three had recurrence. The decision to put drain during surgery was decided intra-operatively by the treating physician depending on whether or not the brain was surfacing. Interestingly, recurrence with the use of drain was higher in our series (100% vs 30%). This may be due to the selective use of the same in cases where the brain was atrophic and was not surfacing after hematoma evacuation. Matsuo et al. have reported that burr-hole craniotomy without subdural drain has a higher recurrence rate but lower complication as compared to SPS [2].

Huang et al. have used atorvastatin (daily 2.5–5 mg) and dexamethasone (tapering dose for 4 weeks) to treat recurrent paediatric cSDH with good results [5]. There was no further recurrence at 4 years of follow up. No adverse events were noted during the medical management in the above recurrent cases. According to the above authors, atorvastatin can promote

cSDH absorption by reducing local inflammation and promoting angiogenesis. In a recent randomised controlled trial [32] in adults with chronic sub-durals which included 748 patients — 375 were assigned to the dexamethasone group and 373 to the placebo group — a favourable outcome was reported in 286 of 341 patients (83.9%) in the dexamethasone group and in 306 of 339 patients (90.3%) in the placebo group. It was thus concluded that treatment with dexamethasone resulted in fewer favourable outcomes and more adverse events than placebo at 6 months, but fewer repeat operations were performed in the dexamethasone group. The same idea may be worth exploring in future cases of paediatric sub-durals.

Recurrence and outcome

The recurrence rate in our study was 30% irrespective of the use of subdural drain in three cases. Matsuo et al. have reported recurrence rate of 20% in their study of children under age of 2 years [2]. The above authors also could not find any predictive factors of recurrence in their study. Zouros et al. have studied 5 infants of cSDH, who were treated with burr hole and subdural drain with good outcome [1]. So, there is no large study in paediatric cSDH to guide the use of subdural drain after burr hole drainage. Clearly, these patients need to be followed for a longer period to determine socio-economic and academic outcome; not only the surgical and radiological outcome, prospective longitudinal study is the need of these patients.

Conclusion

cSDH in the paediatric population is rare and should always alert the physicians to look for the underlying cause especially child abuse. Detailed haematological, coagulation profile, skeletal X-rays and fundus examination should be performed whenever child abuse is suspected. Surgery forms the main stay of intervention. However, recent study has shown the promising result by use of dexamethasone and atorvastatin for selected group of recurrent cSDH. Recurrence rate is quite high irrespective of use of subdural drain. Gender, laterality, coagulopathy, previous shunts or none of the other predisposing factors were predictive for the recurrence in the current study, but larger sample size is required to be certain. Shunt patients had significant number of bilateral cases; hence, primary tumour excision with avoidance of shunt surgery should be the goal, and more enquiry should be done specifically on shunt-induced cSDH as these are essentially iatrogenic; timely intervention in this age group is as vital as their adult counterpart or may be more in case of infants where the brain is still developing to achieve the best outcome and to prevent any permanent long-term sequelae.

Author contribution HD, AJ: collection of data and writing of manuscript; HD, RG: concept and idea and critical revisions; DS: study supervision and critical revisions; VV: supervision and critical revisions; SK: supervision and critical revisions; ARP: supervision and critical revision.

Declarations

Ethics approval Study design is retrospective; hence, approval of ethical committee was not sought.

Conflict of interest The authors have no conflicts of interest to declare.

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