Idiopathic bilateral chronic subdural hematoma with left internal carotid artery infarct in a 3 months infant: A rare case report

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ABSTRACT

Spontaneous chronic subdural hematoma (CSH) in infants is extremely rare. A very limited number of cases are known and reported in literature. The clinical presentation can be myriad varying from asymptomatic cases to gross neurological deficits. We report a case of a 3-month-old child who presented to us with repeated episodes of focal seizures of the left upper and lower limb of 1 month duration. Subsequent imaging revealed bilateral CSH (right > left) with left internal carotid artery infarct and midline shift to left by 8 mm. The child improved following burr hole evacuation of the right-sided CSH. The management of such case and a brief review of literature are discussed.

Key words: Burr hole, chronic subdural hematoma, focal seizures

Introduction

Chronic subdural hematoma (CSH) in neonates and infants is very rare. Limited cases have been reported in literature. Commonly associated factors include trauma during pregnancy, maternal coagulopathy, intrauterine infections, use of anticoagulants, metabolic disorders, etc.

However, spontaneous CSH without known etiology is extremely rare. Unrecognized in-utero trauma and a complex interaction of intracranial and intrauterine pressures seem to be a convincing explanation for such cases. Subtle degree of birth asphyxia could also be a possible etiological factor. We present a rare case of CSH in which etiology remained unknown.

Case Report

A 3-month-old female infant firstborn of nonconsanguineous marriage presented with recurrent episodes of focal seizures of the left upper and left lower limb. She was put on tablet phenobarbitone (according to her body weight) by a pediatrician, but the seizures remained uncontrolled.

The antenatal history was insignificant, and there was no history suggestive of birth trauma. However, the child did not cry immediately following birth.
There was no evidence of neonatal sepsis, however social, motor, and language developmental milestones of the child were delayed.

Physical examination revealed head circumference 40 cm and anterior fontanelle open and not tense. There was no apparent cranial nerve paresis. On gross examination, the power and bulk of all limbs were normal.

Preliminary investigations revealed a low hemoglobin concentration (11 g/dl), normal complete blood count picture, normal platelet counts, normal prothrombin time, and a normal partial thromboplastin time.

Magnetic resonance imaging (MRI) brain showed bilateral CSH (right > left) with mass effect to left of 8 mm and left fronto tempo parietal area of encephalomalacia suggestive of the left internal carotid artery (ICA) territory chronic infarct [Figure 1].

Sodium valproate was added to the previous treatment to control the seizures. Bleeding and clotting parameters were repeated.

As mass effect was towards the right with focal seizures of left upper and lower limb, a decision taken to evacuate right-sided subdural haematoma.

The child was taken up for surgery under general anesthesia.

Right sided frontal and parietal burr holes were done and 80cc altered colored blood drained out.

Postoperatively, the child recovered well; subsequently, seizures were controlled on the single antiepileptic drug.

Follow-up MRI after 6 months [Figure 2] showed complete resolution of CSH with encephalomalacia changes in the left ICA territory.

Discussion

Spontaneous CSH without known etiology is an extremely rare presentation in infants.

There are only five case reports in the literature which have described this entity in newborn babies.[1-5]

The diagnosis is established only after excluding known causes of CSH in infants, most common being birth trauma, hemorrhagic diathesis, and metabolic disorders.[6,7]

Various possible etiological factors can be hypothesized for the development of such entity in infants.

Unrecognized in-utero trauma and a complex interaction of intracranial and intrauterine pressures seem to be the most convincing explanation for such cases.[2,4,5,8]

Evidence of these factors coming into play is further stressed by associated cortical infarcts and atrophy in our case.

Although cerebral infarction has been reported in neonates with traumatic acute subdural hematoma, its association with infantile CSH has not been described.

Occlusion of major vessels does occur in head injury leading to cortical infarction.[6]

The occlusion may follow rupture of the vessel, deranged microcirculation, or arterial dissection.

Cerebral atrophy of the ipsilateral hemisphere after acute subdural hematoma (SDH) has been reported in infants.[9]

Other factors associated with the development of idiopathic CSH in infants include sudden resolution of infantile hydrocephalus in cases with intermittent

Figure 1: Preoperative magnetic resonance imaging - chronic subdural hematoma in bilateral frontoparietal region with mass effect toward left and underlying cortical infarct in the left frontotemporoparietal area

Figure 2: Postoperative magnetic resonance imaging after 6 months with complete resolution of chronic subdural hematoma with underlying left cortical infarct
aqueductal stenosis and hydrocephalus, maternal use of anticoagulants, parental history of coagulopathy, cryptic vascular malformations, subtle bleeding diathesis, asymptomatic intrauterine infections, premature closure of fetal foramen ovale causing cardiac and hepatic decompensation with increase in central venous pressure, and secondary coagulopathy and associated arachnoid and neuroepithelial cysts.

Our case is unique in the sense that no exact etiological factors proved, unrecognized in-utero trauma and a complex interaction of intracranial and intrauterine pressures seem to be the most convincing explanation in our case.

The clinical presentation of infantile CSH varies from asymptomatic cases to infants with gross neurological deficits. The presentation could be subtle manifesting as only anemia to the extent of seizures.

Infantile CSHs are often managed successfully with simple burr hole evacuation and drainage. Other surgical options include craniotomy with excision of membrane and subdural peritoneal shunt in recurrent cases.

**Conclusion**

CSH in infants is most commonly due to nonaccidental trauma, while acute SDH in neonates is most commonly due to birth-related trauma.

In contrast, idiopathic CSH in neonates is a rare entity. Unrecognized in-utero trauma seems to be the most common causative factor in such cases.

Review of literature suggests that etiology in these cases is multifactorial, although the final common pathway is tearing of veins and accumulation of blood in subdural space. These infants must be thoroughly investigated to rule out known causative factors.

Prognosis is generally fair in these cases, provided early management is ensured.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

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