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External Hydrocephalus: A Probable Cause for Subdural Hematoma in Infancy

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Subdural hemorrhage is common in infancy, particularly in the first year of life. The most common cause is nonaccidental (child abuse), with accidental in second place.

We present three healthy infants, ages 4, 5, and 7 months that, during an evaluation for macrocephaly, were found to have frontal subdural hematoma in association with prominent extracerebral cerebrospinal fluid spaces (external hydrocephalus). There was no history of trauma or risk factors for child abuse. Skull surveys and ophthalmologic examinations were normal. All infants were neurologically intact and achieved normal developmental milestones in one-year follow-up.

We suggest that some infants with external hydrocephalus may be at risk for development of subdural hematoma with minimal or no trauma, most likely secondary to stretching of the bridging veins in the unusually widened subarachnoid spaces. Child abuse, although it should always be kept in mind and should be excluded, may not be the most common cause in this specific context.

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Introduction

Subdural hemorrhage is common in infancy, particularly in the first year of life, and presents a major diagnostic challenge. Most cases are due to child abuse, and only in a few cases is the cause unknown [1,2].

We present three healthy infants ages 4, 5, and 7 months old that, during an evaluation for increased head circumference, were found to have frontal subdural hematoma in association with prominent extracerebral cerebrospinal fluid (CSF) spaces (external hydrocephalus). In the absence of any evidence of previous trauma, coagulopathy, or child abuse, we suggest that the enlarged subarachnoid spaces in those infants may have been a predisposing factor for the developing of subdural hematoma.

Case Reports

Case 1

A 4-month-old infant was referred to our clinic for evaluation of increased head circumference. Pregnancy, labor, and delivery were normal. Head circumference at birth was 35.5 centimeters, which is at the 70th percentile. There was no history of postnatal infection or trauma, and no factor that suggested abuse. The physical examination demonstrated weight and height at the 50th percentile. The head circumference was 45 centimeters, which is at the 95th percentile. The anterior fontanel was soft and flat. The infant was alert and was responsive appropriately to visual and auditory stimuli. Eye examination including dilated fundus examination was normal with no evidence of papilledema or retinal hemorrhage. Neurologic examination was normal, and developmental milestones were appropriate for age. Brain magnetic resonance imaging (MRI) demonstrated bilateral prominent extracerebral CSF spaces with moderate ventriculomegaly, and small right frontal subdural hematoma (Fig 1). Laboratory studies including hemogram, serum electrolyte, liver function tests, and coagulation studies were all normal. Skeletal surveys were normal. A follow-up MRI at 1 year of age showed disappearance of the hematoma. The infant remained neurologically intact and achieved normal developmental milestones in one-year follow-up.

Case 2

A 5-month-old healthy boy was evaluated for increased head circumference, which was first noticed at age 3 months. He was born after an uncomplicated pregnancy and had been developing appropriately. There was no history of any recent trauma, vomiting, change in behavior, or lethargy. Physical examination revealed weight and height at the 75th percentile, with head circumference of 46 cm, which is at the 95th percentile. The infant was otherwise alert and playful. Complete eye examination was normal. He had a normal tone with no asymmetry, and his development was intact. Brain MRI revealed prominent subarachnoid

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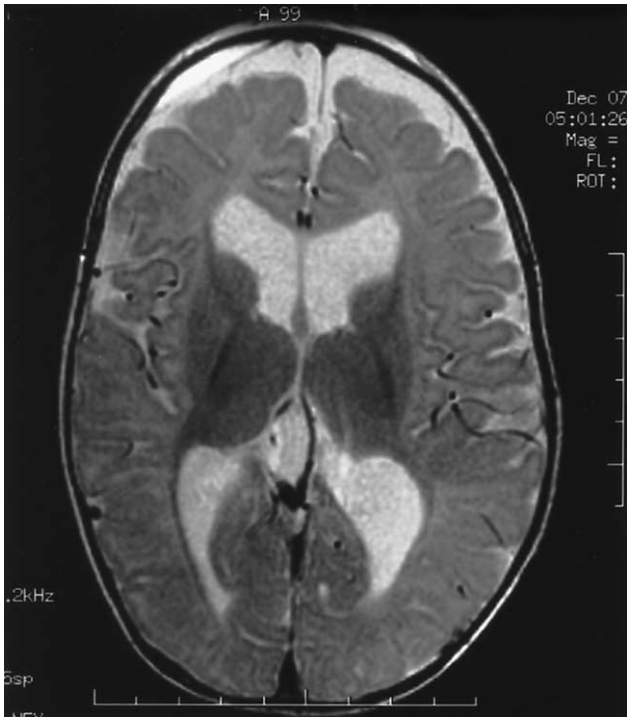

Figure 1. Axial T_2-weighted image, from brain MRI (TR/TE –4000/80) revealing bilateral prominent extracerebral CSF spaces with moderate ventriculomegaly, and small right frontal subdural hematoma.

spaces over both frontal lobes with 1 cm subdural hematoma over the left frontal convexity (Figure 2). Laboratory studies including hemogram, serum electrolyte, liver function tests, and coagulation studies were all normal. Skull surveys were normal. The infant continued to develop appropriately. A follow-up MRI done 6 months later was normal.

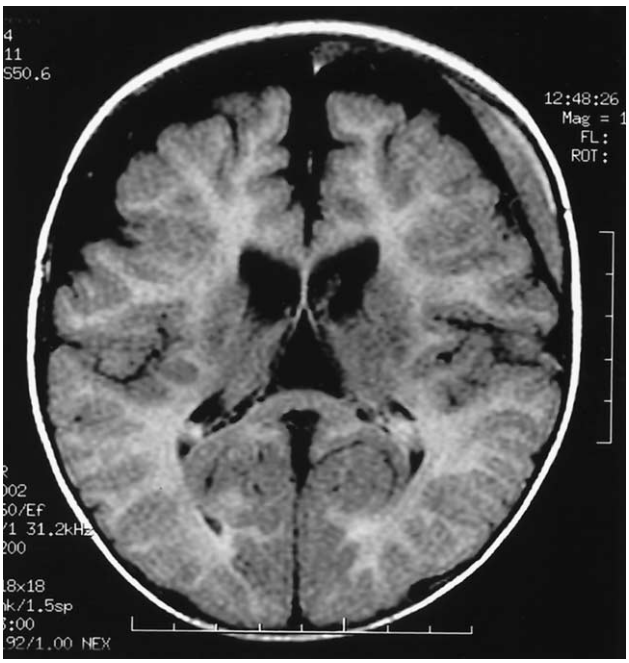

Figure 2. Axial T_1 weighted image from brain MRI (TR/TE - 9002/160) demonstrating prominent subarachnoid spaces over both frontal lobes with 1 cm subdural hematoma over the left frontal convexity.

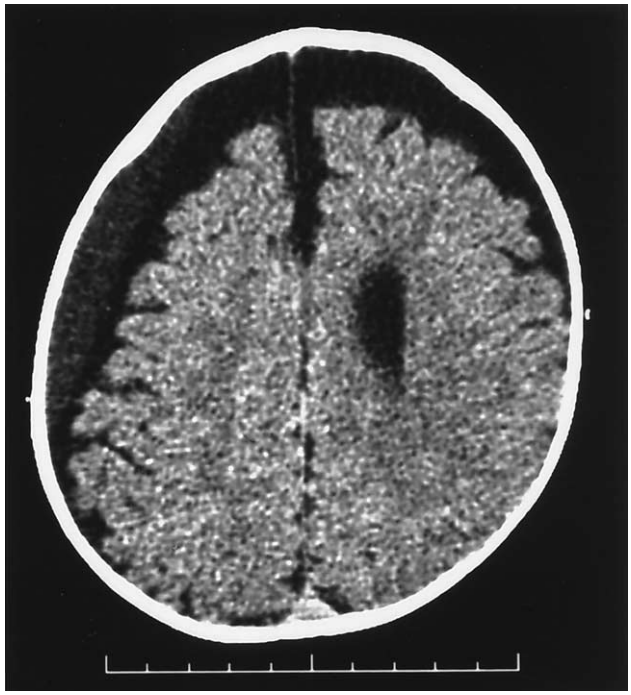

Figure 3. Unenhanced head CT shows benign enlargement of the subarachnoid spaces, with right frontoparietal subdural hematoma. There is flattening of the left parieto-occipital region.

Case 3

A 5-month-old healthy boy was evaluated for increase in his head circumference. Pregnancy and delivery were normal without complications. Head circumference at the age of 2 months was at the 95th percentile, and a head computed tomography scan (CT) done at the same age indicated only prominent extra-axial fluid spaces. In the 2 months before his visit, there was a further increase in his head circumference. There was no history of head trauma, and he was otherwise asymptomatic. Physical examination revealed weight and height at the 25th percentile, with head circumference of 46.5 cm, which is just above the 95th percentile. The head was asymmetric with flattening of the left parietal occipital region. The infant was alert and maintained good eye contact. Funduscopic examination was normal with no evidence of papilledema or retinal hemorrhage. Neurologic examination was normal and his development was appropriate. A second head CT, including three-dimensional imaging, followed by a brain MRI revealed benign enlargement of the subarachnoid spaces, with right frontoparietal subdural hematoma. There was flattening of the left parieto-occipital region but with no evidence of craniosynostosis (Fig 3). Hemogram, serum electrolyte, liver function tests, and coagulation studies were all normal. Skeletal surveys were normal. The subdural collection was surgically drained. The infant remained intact neurologically and continues to achieve normal developmental milestones.

Discussion

Many studies in the past illustrate that the majority of cases with subdural hemorrhages in children under 2 years of age are due to child abuse [1,2].

The clinical presentation in those infants is nonspecific [3]; the prognosis is poor with high percentage of deaths and long-term neurologic sequel [2,4,5].

In a study of 84 infants under 1 year of age with head injury, Billmire found child abuse as a cause in 18 (95%)

out of 19 children with intracranial hemorrhage [6]. Duhaime, in a study of 100 children under 2 years of age with head injury, found 16 infants with subdural hematoma, 13 (81%) of which were due to inflicted injuries [7]. Jayawant et al. performed a population-based study of children under the age of 2 years who presented with subdural hemorrhage. Twenty-seven (82%) out of 33 cases were highly suggestive of abuse [2].

The finding of subdural hematoma resulting from a nontraumatic cause is a rare event, especially in infants with no underlying coagulopathy. In our patients the subdural hematoma was found incidentally, during an evaluation for increased head circumference. All infants were healthy and completely asymptomatic at presentation with no history of head trauma. There was no history of an underlying coagulopathy and no findings by history, physical examination, or x-rays to suggest child abuse. The MRI findings of bilateral prominent extracerebral CSF spaces suggested the diagnosis of external hydrocephalus in our patients.

External hydrocephalus is regarded as a benign condition, consisting of bilateral decreased densities over the frontal convexities, prominent cerebral sulci, slightly enlarged ventricles, and normal brain size [8].

The infants are usually asymptomatic and are being studied with a CT or a MRI as part of an evaluation of the large head. The syndrome is benign and the radiologic findings usually resolve spontaneously by the age of 2 years [9].

The combination of external hydrocephalus and frontal subdural hematoma in an otherwise asymptomatic infant is rare and only few cases were reported [10,11]. Azais and Echenne, in a series of 41 infants with external hydrocephalus, describe three in which subdural hematoma was found as an incidental finding without any symptoms of high intracranial pressure. Two of those infants had mild axial hypotonia at the time of presentation, but all of them had a normal neurologic examination and normal development in further follow-up [12]. The question of whether the external hydrocephalus is responsible for the subdural hematoma or is secondary to it is often raised. In a series of patients with enlarged extracerebral spaces and associated subdural hematomas by Kapila et al., radionuclide cisternography results were not indicative of communicating hydrocephalus therefore suggesting that the enlarged subarachnoid spaces were the predisposing factor [13]. At least one of our patients (Case 3) had a previous head CT revealing external hydrocephalus only without the subdural hematoma. These findings may suggest that in this group of infants stretching of the bridging veins probably occur inside the enlarged subarachnoid spaces, and the absence of an appropriate support makes them vulnerable to bleed either spontaneously or with only a minimal trauma. Recently a theoretical mathematical model of the cranial vault by Papasian and Frim, produced a relationship between venous stretch and the width of the extra-axial spaces, supporting the predisposition towards extra-axial bleeding with only minimal trauma in infants with benign external hydrocephalus [14].

The prognosis of this group of infants that are otherwise healthy is usually better compared with other cases with subdural hemorrhage [13]. In most infants the hematoma will resolve spontaneously. In two of our patients the hematoma resolved spontaneously, and the infants remain neurologically intact and achieved normal developmental milestones in 1-year follow-up. One infant (Case 3), although completely asymptomatic, was sent for neurosurgical consultation and had a surgical drainage of the hematoma. He also had no neurologic sequel and continued to achieve normal developmental milestones.

We suggest that in infants with external hydrocephalus, the enlarged subarachnoid spaces and the secondary stretching of the bridging veins may be predisposed to the development of subdural hematoma with minimal or no trauma. Child abuse should always be kept in mind, and should be ruled out in infants with subdural hematoma, but in the specific context of young infants with external hydrocephalus, subdural hematoma may be spontaneous in nature with probably good prognosis.

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