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Case Report

Acute subdural hematoma in an infant with a biphasic clinical course and late reduced diffusion

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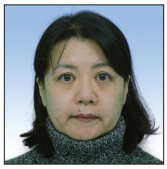

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ABSTRACT

Background: Bright tree appearance (BTA) is a characteristic finding on diffusion-weighted magnetic resonance (MR) imaging with transient high intensity in the white matter. BTA is characteristic of infants with acute encephalopathy with biphasic seizures, but it has also been reported in head trauma cases. In this report, we describe an infant case of traumatic brain injury that demonstrated a biphasic clinical course and late reduced diffusion (TBIRD).

Case Description: A 5-month-old boy suffered from head trauma and developed coma and seizures. Computed tomography scans revealed acute subdural hematoma on the right side. He underwent an emergency operation to remove the hematoma but subsequently had seizure clusters for three days. Diffusion-weighted MR imaging revealed BTA in the right cerebral hemisphere. He was treated with antiepileptic agents and fully recovered to pre-injury condition, and MR imaging no further revealed any BTA 20 days after head trauma. He developed no complications at the 10-month postoperative follow-up.

Conclusion: We reported a case of TBIRD following head trauma in the infant. The pathogenesis remains unclear, but we consider the possibility of biphasic seizures in infant head trauma cases, and we should appropriately administer the anticonvulsants and carefully check for MR imaging.

Keywords: Bright tree appearance, Glutamate, Head trauma, Seizure, TBIRD

INTRODUCTION

Transient high intensity on the white matter is called as a bright tree appearance (BTA). It is known to be a specific finding on diffusion-weighted magnetic resonance (MR) imaging (DWI). This finding was first reported in infants with acute encephalopathy and was named as biphasic seizures and late reduced diffusion (AESD). AESD occurs in infants younger than two years of age whose myelination is incomplete and who follow biphasic clinical courses. Takase *et al.* reported two infantile cases of brain injury that developed a cluster of seizures after calming down an early seizure. These cases demonstrated mimicking AESD on DWI. They reviewed seven previously reported cases that showed very similar findings on DWI and called it as an infantile traumatic brain injury with a biphasic clinical course and late reduced diffusion (TBIRD).[15] Herein, we report an infant case of TBIRD, summarized its characteristics, and discuss its underlying mechanism.

CASE REPORT

A healthy 5-month-old boy collapsed while sitting up and banged the back of his head. He developed repeated seizures and was transferred to our hospital. Neurological examinations on admission revealed consciousness disturbance (Glasgow Coma Scale, E1V1M5), anisocoria (right/left: 5.0/3.0 mm), and left hemiplegia. Computed tomography (CT) revealed an acute subdural hematoma (ASDH) on the right side with the midline shift [Figure 1]. He underwent an emergency operation to remove the hematoma. Intraoperative observations revealed bleeding from the cortical vein and no cerebral contusion.

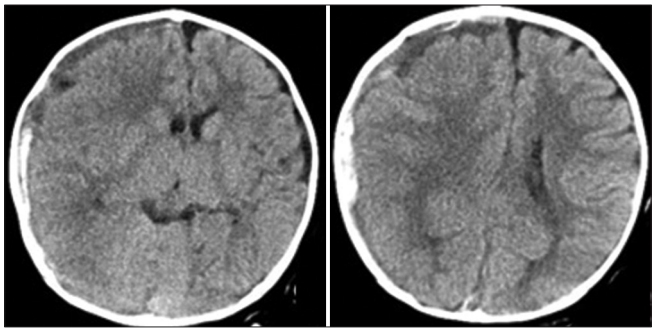

Figure 1: Computed tomography on admission revealed the right subdural hematoma with midline shift.

Following surgery, he was neurologically normal and was free from seizures under phenobarbital administration. However, on day 3, he presented with a cluster of seizures in his left upper extremity with consciousness disturbance. MR images revealed a high signal intensity in the white matter of the right frontal and parieto-occipital lobes on DWI, which was consistent with the so-called BTA [Figure 2a]. The apparent diffusion coefficient (ADC) value in the white matter was reduced [Figure 2b]. However, the intensity in the white matter was normal on fluid-attenuated inversion recovery

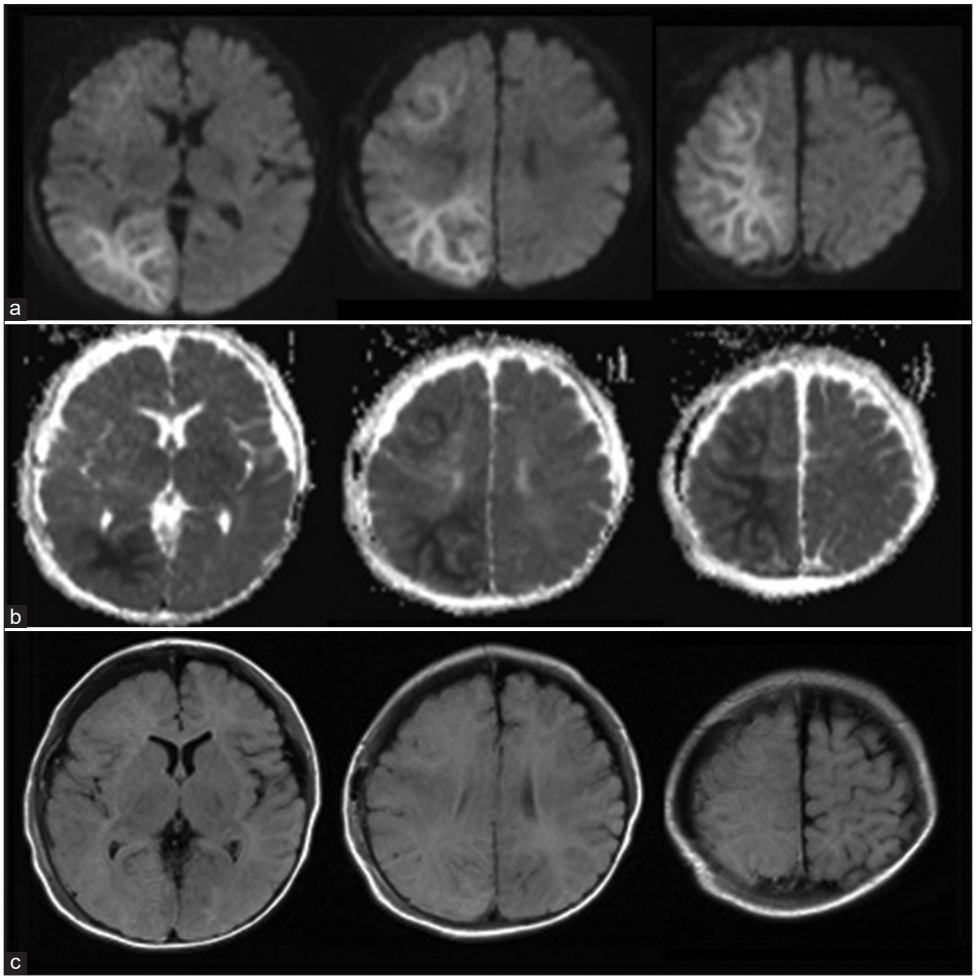

Figure 2: Magnetic resonance images on day 3 revealed the bright tree appearance on the right frontal and parietal lobes at diffusion-weighted images (DWI). That lesion demonstrated low intensity at the apparent diffusion coefficient map image and iso-intensity at the fluid-attenuated inversion recovery (FLAIR) image. (a) DWI. (b) Apparent diffusion coefficient map image. (c) FLAIR image.

(FLAIR) images [Figure 2c]. Electroencephalogram (EEG) demonstrated the spike and wave in the right frontal lobe even during the nonconvulsive state. He was diagnosed with status epilepticus and treated with continuous infusion of barbiturates under EEG monitoring. Follow-up MR images on day 20 showed that the abnormal signal intensity in the white matter of the right cerebral hemisphere completely disappeared on DWI [Figure 3a], although the high-intensity area in the right parietal lobe with atrophy was observed on FLAIR images [Figure 3b]. He was discharged from the hospital on day 26 without any neurological deficits. His development was normal ten months after the injury.

DISCUSSION

Post-traumatic seizures are more common in children than in adults. Early seizure in traumatic brain injury is reported in 0.4% of patients over 18 years of age,[9] while the frequency is much higher, at 0.83–12% in children.[2,8] The threshold for seizure is speculated to be lower in infants than in adults. Among children, the incidence of early seizure is more common in those aged 0–5 years, and ASDH is known to be an important risk factor for post-traumatic seizures.[2] A cluster of seizures is unlikely to occur within a week of achieving early seizure control. However, recent reports have shown that the seizures may recur once again despite the appropriate control with anticonvulsants and become severe within a week with BTA[1,3-5,11-13, 15,16,18] [Table 1]. Cases of TBIRD have some common characteristics: Age <2 years, initial CT revealing ASDH in all but one case, no cerebral contusion, and BTA presentation on DWI. The BTA was bilateral or unilateral, but it almost always coincided with the side of the hematoma. Most TBRID cases have poor prognoses.

Glutamate metabolism may be a factor for TBIRD pathogenesis. In AESD, that clinical course is similar to TBIRD; glutamate increases on days 1–4, and glutamine increases on days 4–12 by MR spectroscopy in the presenting BTA,[13] indicating that glutamate metabolism is one BTA pathogeneses. Increasing glutamate and glutamine levels have also been reported in the case of TBIRD.[15,17] Increasing glutamine concentration in the extracellular fluid or parenchyma has been reported in adult head trauma cases,[6,7,10,19] including in an ASDH case.[7] This increase may also occur in infants with ASDH and may be highly relevant to TBIRD pathogenesis. However, it is unclear whether the same pathophysiology occurs in AESD and TBIRD because the causes of elevated glutamate levels are different. Excessive glutamate is thought to injure the white matter as well as the gray matter. A histological study by Takanashi, *et al*. reported a marked decrease in myelinated axons in the BTA lesion

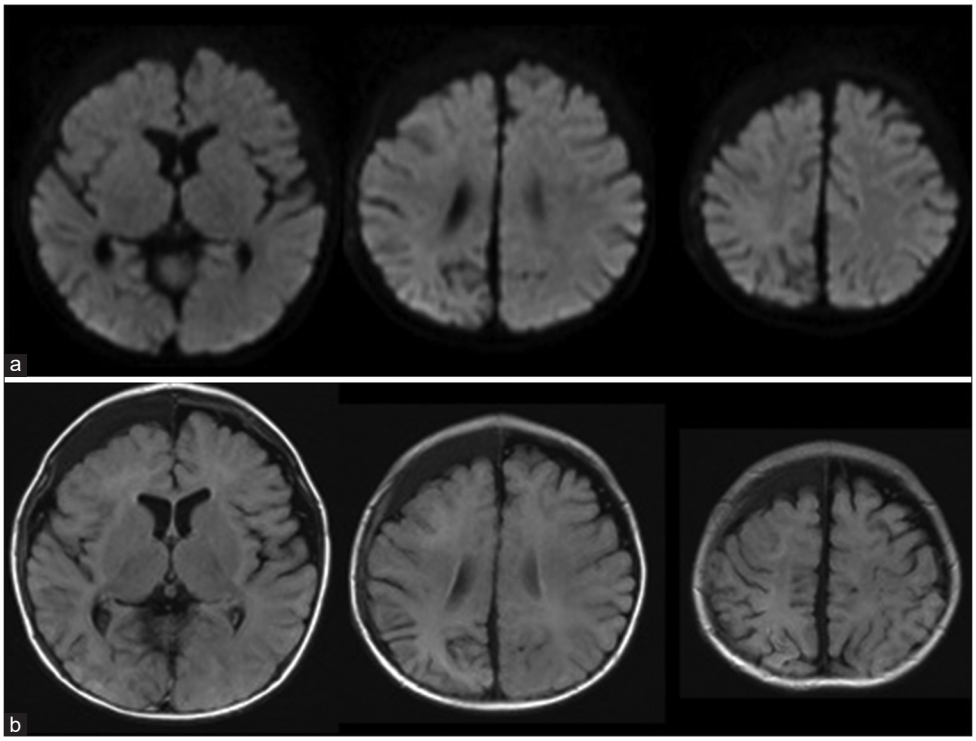

Figure 3: Magnetic resonance images on day 20 showed the abnormal signal intensity in the white matter of the right cerebral hemisphere completely disappeared on diffusion-weighted imaging (DWI), although the high-intensity area in the right parietal lobe with atrophy was observed on fluid-attenuated inversion recovery (FLAIR) images. (a) DWI. (b) FLAIR image.

Table 1: Summary of infantile traumatic brain injury with a biphasic clinical course and late reduced diffusion

	BTA	Outcome	No.	Author	Year	Age	Sex	Duration until second seizure (day) side	CT on admission day	Location of hematoma
1	Kanno[5]	2012	8m	M	3	ASDH	lt	lt	3	MD
2	Nishiyama[11]	2014	9m	M	4	ASDH	falx	bil	5	SD
3	Nishiyama[11]	2014	9m	F	4	ASDH	falx	bil	5	SD
4	Tasaki[16]	2014	8m	M	3	ASDH	lt	bil	6	SD
5	Okizuka[12]	2014	4m	F	3	ASDH	falx	bil	3	MD
6	Inoue[3]	2014	9m	M	not applicable(intubation with muscle relaxant)	ASDH	lt	lt	3	SD
7	Yokochi[13]	2016	12m	M	6	ASDH	rt	rt	6	SD
8	Takase[15]	2018	15m	F	4	ASDH	lt	rt	5	GR
9	Takase[15]	2018	6m	M	3	normal	-	-	5	GR
10	Arai[1]	2018	11m	M	6	ASDH	lt	lt	6	GR
11	Yoshino[18]	2021	7m	M	2	ASDH	rt	rt	2	SD
12	Yoshino[18]	2021	8m	M	3	ASDH	rt	rt	4	SD
13	Kaneko[4]	2022	16m	M	5	ASDH	lt	lt	4	SD
14	Present case	2022	5m	M	3	ASDH	lt	lt	3	GR

m: Months, M: Male, F: Female, CT: Computed tomography, ASDH: Acute subdural hematoma, lt: Left, rt: Right, BTA: Bright tree appearance, bil: Bilateral, MD: Moderate disability, SD: Severe disability, GR: Good recovery.

in an AESD case,[14] indicating a strong effect of glutamate excitotoxicity on astrocytes and myelin in infants younger than two years of age with incomplete myelination, causing cytotoxic edema, indicating BTA.

Unfortunately, this case had no glutamate and glutamine measurement by MR spectroscopy. Therefore, although the possibility that damage to the cortical vein affected the seizures might be considered since the MR imaging showed BTA in areas other than the perfusion area of the cortical vein that was the source of the hemorrhage, we considered it likely that elevation of glutamate was affecting the seizures. In this case, even during the nonconvulsive state, EEG revealed spikes and waves. Nonconvulsive seizure due to elevation of glutamate is likely to have continued after the control of the first seizure, and appropriate anticonvulsant administration might prevent a second seizure and white matter damage.

Here, we reported a case of TBIRD in an infant. Infants with ASDH under two years may have a biphasic clinical course, even without brain contusion. We speculated that increased glutamate levels in ASDH may play an important role in this condition. Head trauma cases with early seizures in infants under two years of age should be treated with appropriate anticonvulsants and follow-up MR imaging, including DWI and EEG.

CONCLUSION

Herein, we reported a case of TBIRD with a BTA. The TBIRD is a relatively new concept. The details of its pathogenesis and clinical features are unclear. The excitotoxicity of glutamate, which is increased in subdural hematoma, has been suggested as one of the factors of TBIRD, such as in patients with AESD. Careful attention should be paid to late seizures in cases of head, especially in infants under two years of age with ASDH.

Ethical approval

Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript, and no images were manipulated using AI.

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