An infantile traumatic brain injury with a bright tree appearance detected before the late seizure

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Case Report

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Abstract

Infantile traumatic brain injury (TBI) rarely follows a biphasic clinical course and exhibits a bright tree appearance (BTA) on magnetic resonance imaging (MRI). This is termed infantile traumatic brain injury with a biphasic clinical course and late reduced diffusion (TBIRD). TBIRD has clinical features similar to those of acute encephalopathy with biphasic seizures and late reduced diffusion (AESD). It remains to be clarified which patients with infantile TBI will develop TBIRD and the prevention and treatment of TBIRD. We report a case of TBIRD that exhibited BTA 1 day before the late seizure and review 12 cases of TBIRD. All patients developed a subdural hematoma (SDH), were younger than 2 years, and presented with a biphasic phase within 3-6 days. The median interval between BTA and TBI was 5 days. Of the 5 cases examined with MRI before the biphasic phase, only our case was detected with BTA 4 days after TBI. Therefore, predicting the biphasic clinical course may be possible by examining MR images after TBI in patients under 2 years of age who develop SDH with unconsciousness, seizure, or hemiplegia, and these patients should be strictly followed up for 1 week.

Introduction

Infantile traumatic brain injury (TBI) rarely follows a biphasic clinical course with a bright tree appearance (BTA) on magnetic resonance imaging (MRI), described as infantile traumatic brain injury with a biphasic clinical course and late reduced diffusion (TBIRD) [1]. It remains to be clarified which infantile TBI patients will develop TBIRD. Herein, we report a case of TBIRD that revealed BTA the day before the patient's late seizure and review the literature on TBIRD to clarify its characteristics.

Case Report

A 16-month-old patient was admitted to our emergency department with a few minutes of seizures after falling from a height of 70 cm. The patient's Glasgow Coma Scale (GCS) was E1V1M4, with the presentation of anisocoria (right/left 3.0 / 5.0 mm) and no apparent paralysis. Computed tomography (CT) revealed a left acute subdural hematoma (SDH) with a midline shift (Fig. 1). The patient underwent an immediate craniotomy and decompression. Although MRI did not reveal cerebral edema or diffuse axonal injury on day 2, mild disturbance of consciousness (GCS: E4V3M5) and right-sided paralysis was observed. On day 4, MRI and MR angiography revealed a BTA in the left subcortical white matter and hyperperfusion in the ipsilateral cerebral hemisphere (Fig. 2a, b). The next day, the patient developed status epilepticus, which was treated with anticonvulsants. On day 20, MRI showed residual hyperperfusion on arterial spin labeling, despite the disappearance of the BTA. CT revealed atrophy in the left cerebral hemisphere on day 34. Although the disturbance of consciousness improved, the right-sided paralysis persisted. The patient's Pediatric Cerebral Performance Category (PCPC) scale score was 2 when the patient was transferred to the rehabilitation hospital on day 65.

Discussion

This case followed a biphasic clinical course of status epilepticus 5 days after TBI, revealing BTA and ipsilateral hyperperfusion the day before the late seizure. This case exhibited clinical features similar to those of acute encephalopathy with biphasic seizures and late reduced diffusion (AESD) [1, 2]. AESD is a subtype of acute infectious encephalopathy in infants with a prolonged febrile seizure, following a late seizure after 4–6 days and exhibiting BTA [2]. The pathogenesis of AESD has been considered to involve excitotoxicity with delayed neuronal death [2]. When status epilepticus occurs, excitatory neurons in the cerebral cortex release large amounts of excitatory amino acids, such as glutamate. If the quantity of glutamate released is large, the surrounding astrocytes cannot metabolize it into glutamine. This causes an influx of Ca^{2+} into the postsynaptic neurons, resulting in delayed necrotic cell death or apoptosis [2].

We reviewed 12 TBIRD cases [1, 3–9] (Table 1), excluding cases in which the onset of TBI was unknown or the biphasic clinical course was unclear. The median age was 9 months, and all patients were under 2 years of age. SDH was observed in all cases. The median interval of the biphasic phase was 3 days. Late seizures occurred in 11/12 cases, and 1 case presented only with unconsciousness, although non-convulsive seizures were not ruled out [1]. The median interval between BTA and TBI detection was 5 days. Only our case was detected with BTA 4 days after TBI.

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

	Value
Age (month), median [IQR]	9.0 [7.25–14.25]
Mechanism of injury, n (%)	8 (67%)
Fall	1 (8%)
Thrown	1 (8%)
Traffic accident	2 (17%)
Unknown	
Initial Symptoms, n (%)	8 (67%)
Convulsive Seizure	3
Status epilepticus	4 (33%)
Unconsciousness	6 (50%)
Hemiparalysis	
Glasgow Coma Scale, n (%)	5 (42%)
≤8	4 (33%)
>8	3 (25%)
Unknown	
Imaging findings, n (%)	12 (100%)
Subdural hemorrhage	4
Midline shift	1 (8%)
Subarachnoid hemorrhage	0
Diffuse axonal injury	
Initial treatment, n (%)	10 (83%)
Conservative therapy	2
Barbiturate coma therapy	2 (17%)
Surgery	
Day of late seizure, median [IQR]	3 [3.0–4.0]

IQR, interquartile range; CT; computed tomography, MRI; magnetic resonance imaging

	Value
Bright tree appearance on MRI	8 (67%)
Bilateral	3 (25%)
Unilateral	1 (8%)
Unknown	
Day of bright tree appearance on MRI, median [IQR]	5 [3.0–5.0]
Second treatment (after the second phase), n (%)	11 (92%)
Conservative therapy	5
Barbiturate coma therapy	1 (8%)
Surgery	
Cerebral atrophy, n (%)	10 (83%)
Yes	1 (17%)
No	1 (17%)
Unknown	
Pediatric Cerebral Performance Category Scale, n (%)	4 (33%)
≤2	7 (58%)
≥3	1 (8%)
Unknown	
IQR, interquartile range; CT; computed tomography, MRI; magnetic resonance imaging	

BTA has been implicated in incomplete myelination until approximately 2 years of age [1]. The developing brain is vulnerable due to immature cerebrovascular autoregulation [10], high concentrations of unsaturated fatty acids, high oxygen consumption, and low concentrations of antioxidants [10, 11]. The high density of N-methyl-D-aspartate receptors [11] may also be associated with the risk of excitotoxicity. MR spectroscopy showed elevated glutamine/glutamate complexes and decreased N-acetyl aspartate levels in AESD [2, 12, 13]. The same findings were observed in 2 cases of TBIRD [1, 7], suggesting that TBIRD is pathophysiologically identical to AESD. Some cases of TBIRD differ from AESD in that status epilepticus is not present in the initial phase. Two cases of TBIRD showed hypoperfusion in the ipsilateral cerebral hemisphere [7, 9], similar to an animal SDH model [14], suggesting a cerebral metabolic blood flow imbalance due to microvessel vasospasm [15, 16]. In contrast, our case showed hyperperfusion, which may suggest ischemia-reperfusion injury associated with impaired cerebrovascular autoregulation.

TBIRD often has a poor neurological prognosis, with PCPC ≥ 3 occurring in 58% (7/12) of cases. Although the prevention and treatment of TBIRD remain to be elucidated, the characteristics of TBIRD were shown to be infantile TBI in patients who developed SDH with unconsciousness, seizure, or hemiplegia in this study. Some patients may exhibit BTA prior to the late seizure, as in our case.

Conclusions

TBIRD rarely develops in infants with TBI. Predicting the biphasic clinical course may be possible by examining the MRIs of patients younger than 2 years who develop SDH with unconsciousness, seizure, or hemiplegia. These patients should be strictly followed up 1 week after TBI.

Declarations

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Statements and Declarations:

All authors contributed to the study's conception and design. Material preparation, data collection, and analysis were performed by Naoki Kaneko. The first draft of the manuscript was written by Naoki Kaneko, and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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Figures

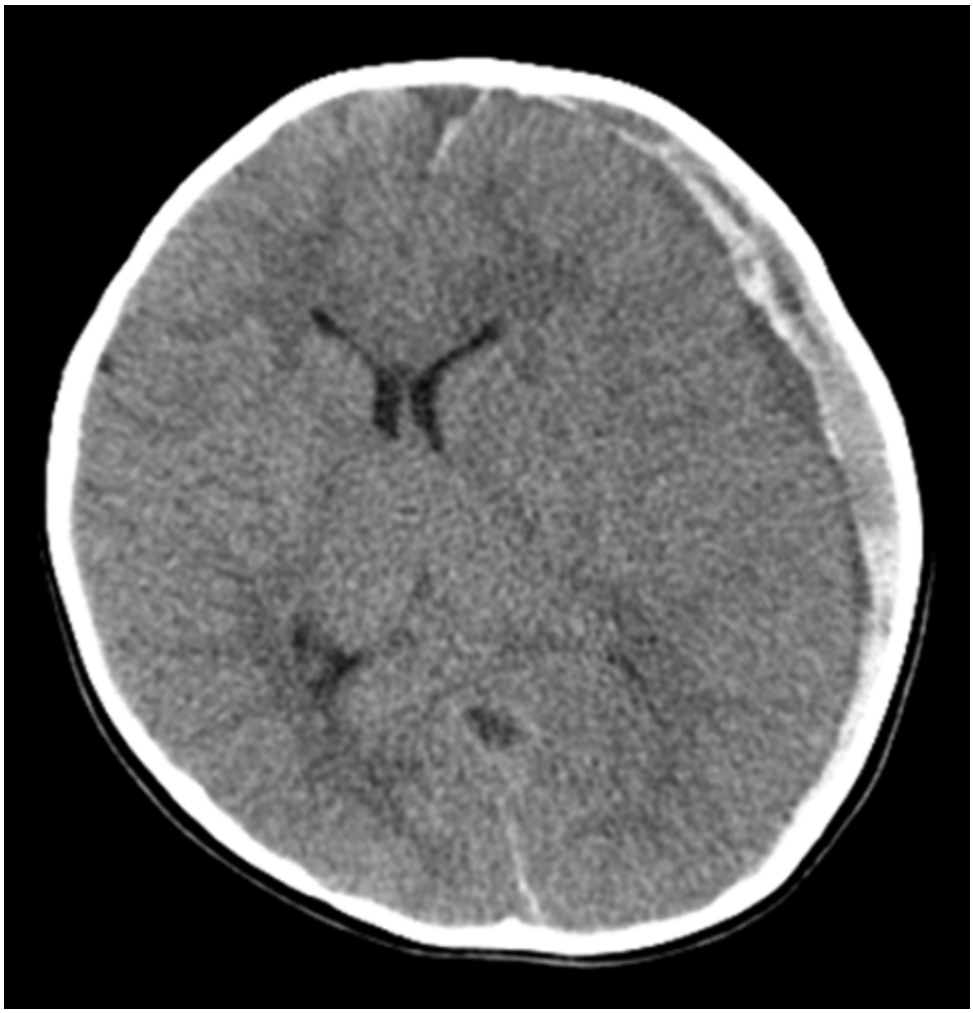

Figure 1

Computed tomography at arrival reveals a left subdural hematoma with a midline shift.

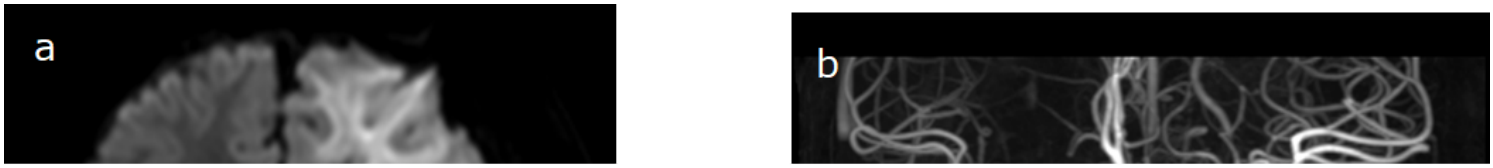

Figure 2

Magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) on day 4. (a) MRI shows left bright tree appearance in the left subcortical white matter. (b) MRA shows hyperperfusion in the ipsilateral cerebral hemisphere.