

Mild-type Infantile Acute Subdural Hematoma Presenting with a Holohemispheric Subdural Hematoma

Nobuhiko Aoki*

Department of Neurosurgery Tokyo Metropolitan Tama Medical Center 2-8-29 Musashidai, Fuchu, Tokyo 183-8524, Japan & Department of Neurosurgery, Bethlehem Garden Hospital 3-14-72 Umesono, Kiyose-shi, Tokyo 204-0024, Japan

*Corresponding author:

Nobuhiko Aoki,
Department of Neurosurgery Tokyo Metropolitan
Tama Medical Center 2-8-29 Musashidai, Fuchu,
Tokyo 183-8524, Japan & Department of
Neurosurgery, Bethlehem Garden Hospital 3-14-72
Umesono, Kiyose-shi, Tokyo 204-0024, Japan

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Keywords:

Infantile acute subdural hematoma; Shaken baby syndrome; Abusive head trauma; Benign enlargement of the subarachnoid space; Large sylvian fissure; Dural border cell layer; Holohemispheric subdural hematoma; Cranio-cerebral disproportion

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1. Abstract

1.1. Background

An acute subdural hematoma (ASDH) in an infant without external signs of head trauma is sometimes thought to be symptomatic of shaken baby syndrome (SBS) / abusive head trauma (AHT) in the United States or of an infantile acute subdural hematoma (IASDH) due to minor head trauma in Japan. The present case report demonstrated that IASDH may occur spontaneously if the dural border cell layer (DBCL) is disrupted in an infant with cranio-cerebral disproportion (CCD), a situation which involves holohemispheric subdural hematoma (HHSDH) development, a neuroimaging finding specific to this condition.

1.2. Case Description

A male infant born after an uncomplicated delivery experienced a febrile convulsion at the age of 1 month. He underwent magnetic resonance imaging (MRI), which revealed an enlarged sylvian fissure (LSF) and benign enlargement of the subarachnoid space (BESS).

At the age of 13 months, he displayed signs of altered consciousness while being driven in a car despite having no history of head trauma. Soon thereafter, he displayed tonic-like movements in both upper extremities and was taken to a nearby hospital, where computed tomography (CT) revealed ASDH. In the emergency room,

symmetrical, bilateral, tonic convulsions without any external sign of trauma were observed. Bilateral, retinal hemorrhages were also noted. He responded well to anticonvulsive medication. Follow-up MRI demonstrated a thin, subdural hematoma surrounding the entire cerebral hemisphere together with BESS and LSF. His later clinical course to his current age of 2 years has been uneventful.

1.3. Conclusion

The present case is invaluable because it demonstrates that mild-type IASDH can occur in the absence of an external impact to the head. The diagnostic criteria for SBS/AHT needs to be reviewed, considering that the presence of CCD, including BESS and LSF, may cause IASDH even without head trauma. Particularly relevant here may be the thin HHSDH on MRI characterizing IASDH localized in the disrupted DBCL.

2. Introduction

Infantile acute subdural hematoma (IASDH) is defined as an acute subdural hematoma (ASDH) in infants caused by minor head trauma without the loss of consciousness or a cerebral contusion [1]. In addition, most patients with IASDH have retinal hemorrhage (RH). In the present study, IASDH was clinically graded as mild (grade I), intermediate (grade II) or fulminant (grade III) in accordance with previous findings (Table 1). Based on the assumption that short falls are unable to cause ASDH, American neurosur-

geons insisted on the automatic diagnosis of subdural hematoma (SDH) + retinal hemorrhage (RH) = shaken baby syndrome (SBS) [2]. Since then, this automatic diagnosis has gained traction in the English-speaking world, where the presence of a subdural fluid collection on CT or MRI leads to the immediate diagnosis of SBS/AHD unless pathological findings demonstrate otherwise [3].

The present case is an invaluable illustration of the fact that IAS-DH has a benign clinical course in the setting of mixture of cerebrospinal fluid (CSF) and holohemispheric subdural hematoma (HHSDH). Furthermore, this report includes previously undocumented neuroimaging findings associated with cranio-cerebral disproportion (CCD), including benign enlargement of the subarachnoid space (BESS) and an enlarged sylvian fissure (LSF) [4-7].

Table 1: Clinical grade on arrival in patients with infantile acute subdural hematoma (Proposed in 1984 [Ref. 2]).

Disease Grade	Type	Clinical Features
I	Mild	Conscious, no motor disturbance but with vomiting &/or irritability
II	Intermediate	Drowsy, minimal or mild hemiparesis
III	Fulminant	Stuporous to comatose, moderate to severe hemiparesis, with signs of cerebral herniation

3. Case Presentation

The patient, a male infant, was born after an uncomplicated delivery and had an unremarkable history except for the development of febrile convulsions at the age of 1 month. He underwent MRI, which revealed an enlarged sylvian fissure (LSF) on the left side and a benign enlargement of the subarachnoid space (BESS), which was especially conspicuous in the bilateral, parietal area (Figure 1). At the age of 13 months, he displayed signs of altered consciousness while being driven home in a car from kindergarten. The patient had no history of head trauma. Soon thereafter, he presented with tonic-like movement in both, upper extremities and was taken to a nearby hospital, where emergency computed tomography (CT) revealed a mixed-density, subdural hematoma surrounding the inner table of the skull and extending along the interhemispheric fissure on the left side (Figure 2). In the emergency

room, symmetrical, bilateral, tonic convulsions were observed in the absence of any external signs of trauma. Bilateral, retinal hemorrhages were also noted.

The patient was treated conservatively with an anticonvulsant medication and recovered to his premorbid state by the following day. Magnetic resonance imaging (MRI) performed ten days after the onset of the convulsions demonstrated an acute, subdural hematoma in the parasagittal region and subdural fluid collections surrounding the entire left cerebral hemisphere, indicating a holohemispheric subdural hematoma with a tapering configuration. BESS and bilateral LSF were also observed (Figure 3). No parenchymal abnormality, such as a cerebral contusion or diffuse, axonal injury, was observed. Follow-up MRI at 20 days post-onset demonstrated that the HHSDH on the left side had decreased (Figure 4). The patient’s later clinical course to his current age of 2 years has been uneventful.

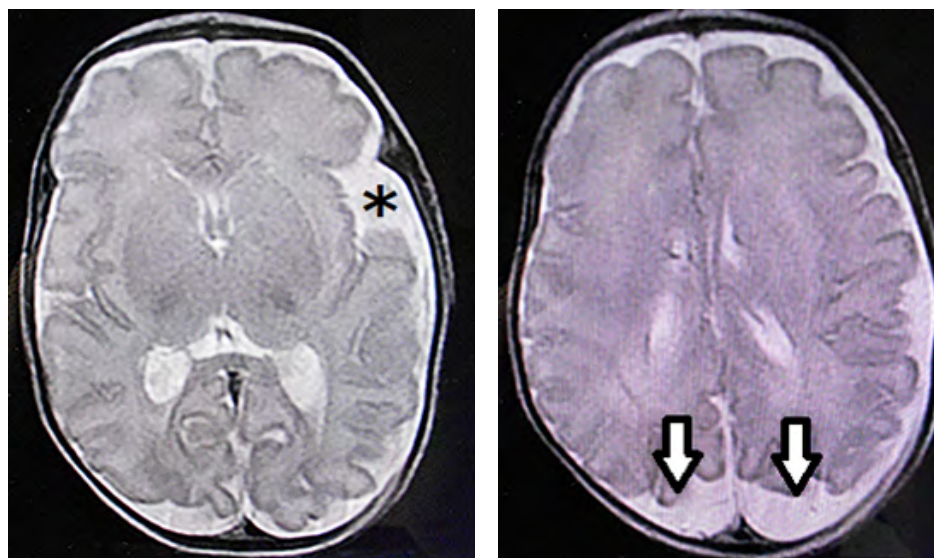


Figure 1: Axial view on magnetic resonance imaging (T2-weighted) at the age of 1 month for febrile convulsion assessment. **Left:** Large Sylvian Fissure on the Left Side (asterisk). **Right:** Benign Enlargement of the Subarachnoid Space, Particularly Conspicuous in the Bilateral Occipital Areas (arrows).

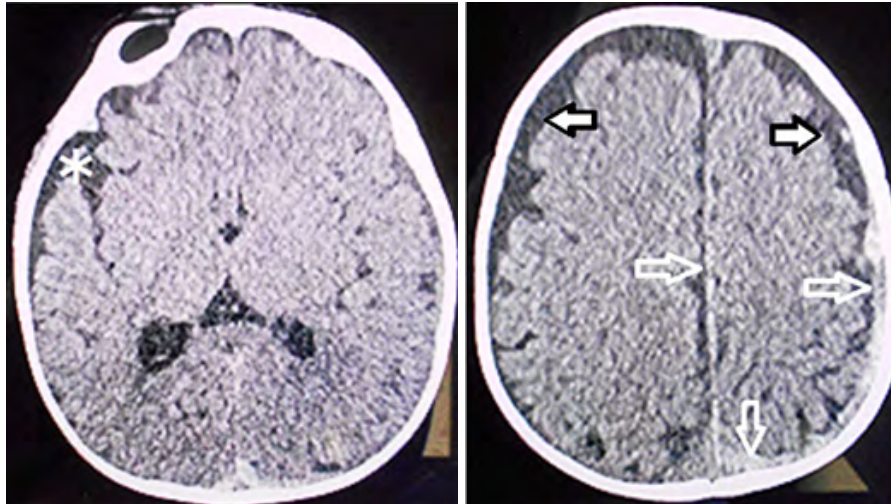


Figure 2: Axial View on Computed Tomography at Presentation.

Left: Large Sylvian Fissure on the Right Side (Asterisk).

Right: Mixed Density Subdural Hematoma Surrounding the Inner Table of the Skull and Extending Along the Interhemispheric Fissure on the Left Side (White Arrows).

Benign Enlargement of the Subarachnoid Space (Black Arrows).

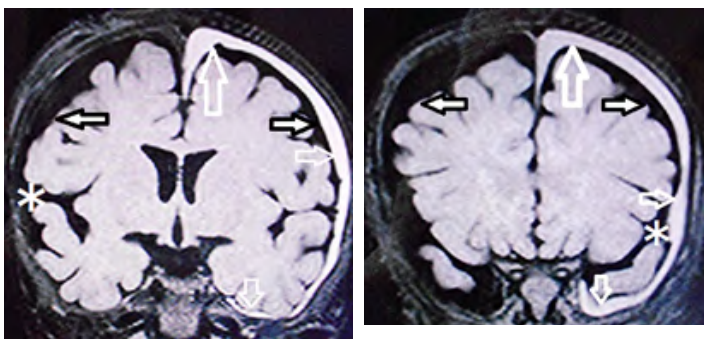


Figure 3: Coronal View on Magnetic Resonance Imaging (Fluid-Attenuated Inversion Recovery) 10 Days After Presentation.

Left & Right:

Prominent acute subdural hematoma in the parasagittal region on the left side (large white arrow).

Homogeneous subdural hematoma surrounding the entire cerebral hemisphere (i.e., a holohemispheric subdural hematoma) with a tapering configuration on the left side (small white arrows).

Benign enlargement of subarachnoid space (black arrows).

Enlarged, bilateral sylvian fissures (asterisk).

No parenchymal abnormality, such as a cerebral contusion or diffuse axonal injury, was observed (not shown in these images).

4. Discussion

The present patient experienced rapid, neurological recovery following episodes of altered consciousness and seizure. His favorable clinical course indicated mild-type IASDH [4,5,7]. Furthermore, the BESS and LSF seen on CT and MRI indicated intracranial, structural vulnerabilities predisposing the patient to IASDH development [4-7]. Based on the anatomical characteristics of infants, IASDH was originally defined in 1984 as acute, infantile subdural hematoma apparently caused by minor head trauma

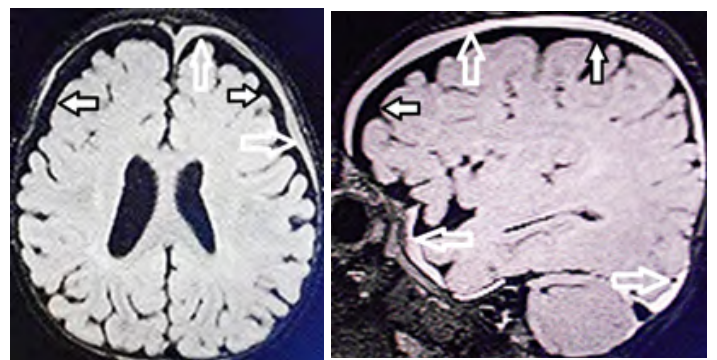


Figure 4: Magnetic Resonance Imaging (Fluid-Attenuated Inversion Recovery) 20 Days After Presentation.

Left (Axial View), Right (Sagittal View)

The Subdural Hematoma Decreased in Volume (White Arrows).

Note the Subdural Hematoma Surrounding the Entire, Cerebral Hemisphere (i.e., The Holohemispheric Subdural Hematoma) on The Sagittal View.

Benign Enlargement of The Subarachnoid Space (Black Arrows)

No Parenchymal Abnormality, such as a Cerebral Contusion or Diffuse Axonal Injury (not Shown in These images), was Noted.

without the loss of consciousness or any association with a primary brain injury [1]. Cases of IASDH have been reported in Japan since the 1960's [8]. However, because most of these cases were published in Japanese-language journals and accusations that the diagnosis was an attempt to conceal child abuse were frequently raised, the concept of IASDH has not been widely accepted in the English-speaking world. Nonetheless, recent Japanese reports have demonstrated that patients with IASDH can be distinguished from those with SBS/AHT through multidisciplinary assessment,

including an evaluation by child abuse pediatricians and pediatric neurosurgeons [4, 5, 8, 9] (Table 2). Furthermore, the absence of a primary brain injury in IASDH indicates a fundamental difference from SBS/AHT. The findings of nation-wide, multicentric, retrospective studies in Japan (2022) are particularly valuable in that they provide reliable corroboration of this finding [10-12]. The dural border cell layer (DBCL), a distinct, soft-tissue layer at the dura-arachnoid interface [13], is composed of a loose conglomeration of cells having enlarged, extracellular spaces and no extracellular collagen. A SDH occurring in this environment results from the disruption of the DBCL by pooled blood. Furthermore, in the superficial compartment of the DBCL, there is a well-developed dural venous plexus which fills the venous sinuses. Because

the DBCL is easily disrupted, a hemorrhage originating in the inner dural plexus may be chiefly responsible for the non-traumatic symptoms of this condition [14, 15]. In the present patient, the parasagittal venous plexus, which communicates with the superior sagittal sinus, was the likely source of the bleeding, as suggested by the conspicuous hemorrhages seen on CT. The parasagittal hemorrhage continued peripherally in a tapering fashion, forming the characteristic appearance surrounding the entire hemisphere (i.e, the holohemispheric subdural hematoma) (Figure 2). This hemorrhagic lesion may directly affect the fragile DBDL. Thus, the age-specific vascularity of the dura together with intracranial, structural vulnerabilities secondary to CCD, including the BESS and LSF, are considered to be the etiological factors of IASDH occurring spontaneously or after a minor impact to the head.

Table 2: Comparison of infantile acute subdural hematoma (IASDH) and shaken baby syndrome (SBS) \ abusive head trauma (AHT).

	IASDH	SBS/AHT
Applied force	Minor head trauma	Abuse (high energy impact)
Main etiology	Disruption of bridging vein	Cerebral contusional tears
Primary brain injury	None	Common
Age distribution	Peak at 6 ~ 10 Months	Widely distributed (including less than 3 months)
Sex	Marked male preponderance	None
Recurrence	Rare	Not rate
	Depends on hematoma size	
Prognosis		Proof
	(Mostly benign clinical course)	
Retinal hemorrhage	Frequent	Occasional

(Cited from ref. 8 with the permission of the Society of Japanese Neurosurgery).

5. Conclusion

The present case is invaluable because it demonstrates that IASDH can occur in the absence of an external impact to the head. The diagnostic criteria for SBS/AHT, particularly in terms of their application to very young infants, need to be reviewed, considering that the presence of CCD, including BESS and LSF, may cause IASDH even without head trauma. Particularly relevant here is the holohemispheric appearance on neuroimaging studies characterizing IASDH localized in the disrupted DBCL. The present study proposes the term, holohemispheric subdual hematoma, to describe this phenomenon.

6. Author contribution:

Nobuhiko Aoki (the corresponding author with no coauthors) conceived the idea of the study, conducted a search of the literature, and drafted the original manuscript.

The author has reviewed the manuscript draft, revised it critically for intellectual content, and approved the final version for submission.

7. Availability of data and materials:

This article does not include any data or material that can be provided.

Declarations, ethics approval, and the consent to participate:

This article was approved by the ethics committee of Bethlehem Garden Hospital and Tokyo Metropolitan Tama Medical Center. No funding was obtained for this study. The parents/legal guardians provided consent to publish the details of this case.

8. Consent to publish:

The author consents to the publication of all identifiable details, including photographs, case history, and other details.

9. Conflicts of interest: None

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