REVIEW

Abusive head trauma in Japan

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Abstract

Purpose Abusive head trauma (AHT) is a serious cause of morbidity and mortality in the pediatric population, especially in young infants. This review of the literature aimed to understand the characteristics of AHT in Japan.

Methods PubMed and the Japanese database Ichuushi were searched to understand the differences in AHT between Japan and other countries.

Results Shaking was identified as an uncommon cause of injury, while mothers were the most common perpetrator of pediatric AHT in Japan. Although uncommon in other countries, infantile subdural hematoma caused by an accidental slight head injury was reported in Japan.

Conclusion As in other countries, AHT is a major condition which pediatric neurosurgeons face in Japan. The mechanisms of injury and perpetrators of AHT seemed to differ slightly between western countries and Japan. Additionally, non-accidental infantile acute subdural hematoma was reported mainly in Japan. Therefore, further care and fair judgment is necessary when investigating child abuse in Japan.

Keywords Abusive head trauma · Accidental infantile acute subdural hematoma · Abuse in Japan · Comparison

Introduction

Abusive head trauma (AHT) is one of the main causes of death by child abuse. Even if an AHT victim survives, there is a high possibility that the victim will remain severely disabled. Moreover, as studies have revealed that perpetrators of abuse are more likely to have been abused themselves, child abuse can be interpreted as a "genetic" disease that may affect a family for generations. Child physical abuse (CPA) is considered an international priority by the World Health Organization (WHO), who developed a vast program to prevent this issue [1]. CPA is a malignant problem worldwide, often complicated by larger socioeconomic factors; even though pediatric neurosurgeons are on the frontline to save the victims of CPA, we cannot solve the whole problem. Nevertheless, we are still playing an important role. In this article, we describe the present condition of child abuse

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² Saitama Prefectural Children's Medical Center, Saitama, Japan in Japan and discuss non-abusive infantile acute subdural hematoma (ASDH) reported in our country.

Etiology

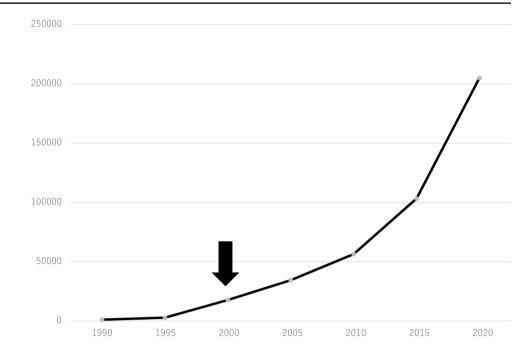
As in other countries, the incidence of child abuse is increasing in Japan. While the government reported around 1000 cases in the early 1990s, this increased to more than 1000 cases each year in the late 1990s. After 17,725 cases were reported, the government finally issued a law after the proclamation of the Child Abuse Prevention Act in 2000. It became a duty for healthcare workers to report cases to childcare centers if there was a suspicion of child abuse. This law may have exposed hidden cases of abuse, leading to an increase in cases over time; in fact, there were more than 200,000 cases reported in 2020 [2] (Fig. 1).

Child abuse is categorized into four types: physical abuse, sexual abuse, neglect, and psychological abuse [3]. When comparing figures from 2010 to 2020 in Japan, the proportion of psychological abuse increased while the proportion of physical abuse decreased (Fig. 2). However, as cases of abuse are increasing overall, it is not possible to conclude that physical abuse is decreasing (Fig. 3). Young mothers,



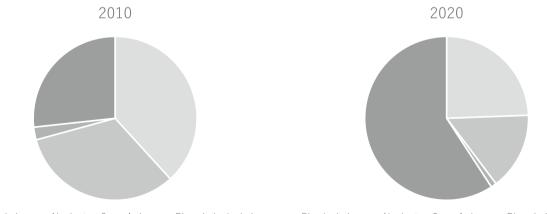
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Fig. 1 The increase of abused cases from 1990 to 2020. There were approximately 1000 cases in the early 1990s. The number of cases reached 17,725 before the government issued a law after the proclamation of the Child Abuse Prevention Act in 2000 (black arrow). This law may have exposed hidden cases of abuse, leading to an increase in cases to more than 200,000 reported in 2020



care by fathers, weak socioeconomic circumstances, drug use, history of psychiatric disorders of the caretaker, unstable marriage, and domestic violence are known social and familial risk factors of abuse. In one study focused on caretakers' confessions, the frequency of abuse varied, and the existence of abuse was difficult to identify [4, 5]. Also, according to a study by Jenny et al., 31.2% of AHT patients under 3 years old were underdiagnosed at their first medical visit and 40.7% worsened after the second visit [6]. Diagnosis of AHT remains difficult. Therefore, it is recommended in Japan to use information from precise history taking, family composition, and contacts in the victims' record to create a flow chart and make a uniform diagnosis of AHT [7].

Most of the clinical characteristics of AHT cases in Japan are similar to those in western counties, though there are some differences. Miki et al. [8] compared 747 cases (Japan, 200 cases; western countries, 547 cases) of abused children. They compared the presence of intracranial lesions, existence of a direct impact, outcome, mechanism of injury, and perpetrator information between countries. There was no difference between Japan and the other countries in terms of intracranial lesions. Subdural hematoma was the most frequent intracranial lesion seen in child abuse cases, making up 75% of the intracranial lesions in both groups. In Japan, direct impact and skull fracture were seen in 64% and 19% of cases, respectively. In western countries, these figures were 58% and 31%, respectively. When investigating the mechanism of injury, in the western countries, there is a tendency for the perpetrator to shake the child [9]. On the other hand, in Japan, the perpetrator tends to batter the

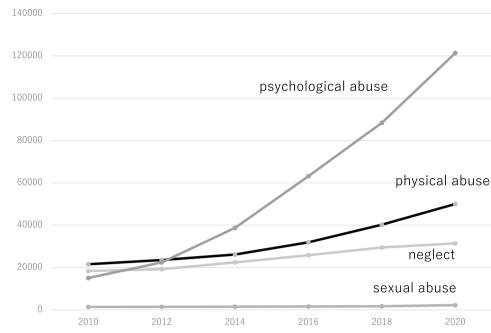


Physical abuse = Neglect = Sexual abuse = Phsychological abuse = Physical abuse = Neglect = Sexual abuse = Phsychological abuse

Fig. 2 The transition of the percentages of psychological, physical, neglect, and sexual abuse in Japan from 2010 to 2020

Fig. 3 Comparison of the distribution of psychological, physical, neglect, and sexual abuse in Japan between 2010

and 2020



child via striking, throwing, and kicking [10–12]. There is limited news coverage regarding a child being "shaken" in Japan. Though it is difficult to identify the mechanism of injury in abuse cases, Fig. 4 shows that only 10% of abused children were "shaken" from Miki's review of the literature [13]. Additionally, there was a clear difference in perpetrator identity between Japan and western countries. In Japan, the

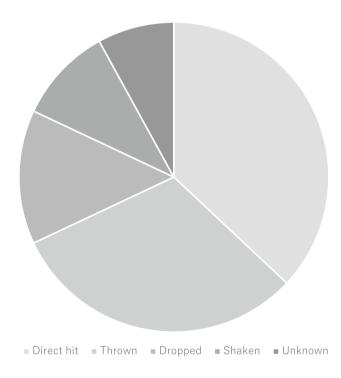


Fig. 4 Mechanism of cases of child abuse based on a review of Japanese literature (n=51), reprinted with permission [13]

most common perpetrator was the victim's mother (44%), followed by their father (30%); their stepmother was the perpetrator in 11% of cases, but this was not seen in western countries. On the contrary, in western countries, almost twice as many fathers (37%) were perpetrators than mothers (13%). Also, the mother's boyfriend (20%) and babysitters (21%) were also identified as perpetrators. Therefore, it can be interpretated that in Japan, 55% of the perpetrators are women, which is higher than in western countries [13]. This tendency has remain unchanged for almost a decade. According to the data published in 2014, the ranking order of the perpetrator frequency was still mothers (52.4%), fathers (34.5%), and stepfathers (6.3%) [2]. To summarize the difference between Japan and western countries in child abuse, children are shaken less frequently, and women are more likely to be the perpetrator of abusive head trauma in Japan.

Terminology and mechanism of injury in AHT

Violent shaking is one important mechanism of intracranial hemorrhage in these children. There are many reports in the literature of biomechanical research related to traumatic head injury. Shear force is known to cause tearing of the bridging veins. Namely, the angular acceleration increase by rotation and the sudden stop of the brain caused by violent shaking is known as the classical hypothesis [14]. Duhaime et al. postulated that based on clinical findings, pathological data, and biomechanical models, rotational acceleration/deceleration whiplash injuries do not provide enough force to account for the severe injuries of these children and that in severe cases, blunt trauma must be involved [15]. Despite this, the type and threshold of impact able to cause tearing of the bridging veins remain unknown [16].

In 2009, the American Academy of Pediatrics posted a recommendation to use the terminology AHT. Although violent shaking is the most reported mechanism of injury, blunt impact, or a combination of shaking and blunt impact, causes injury as well. They noted that the use of broad medical terminology inclusive of all mechanisms of injury, including shaking, is required [17].

Costine-Bartell et al. reported that they created an experimental model of AHT using piglets. They succeeded in creating the characteristic findings of AHT by compressing the brain parenchyma caused by ASDH, imposing a blunt impact to the head, adding hypoventilation stress, and inducing seizures. They were also able to create similar pathological findings and found that seizures were found in 70–80% of the subjects. They suggested that seizures may be the driver of AHTs [18].

Most traumatic brain injuries occur due to a combination of the impact to the head and inertial force to the brain. However, it is difficult to ascertain the relevant mechanism due to intensity and vector variation. In the literature, it is known that a child with no other problems will not develop "fetal" brain damage by falling from a height of around 90 cm (3 ft.) [7, 19]. However, in the real world, most cases of head trauma are a result of a mechanism other than a simple fall, such as being pushed from a standing position, shaken off from a carousel, or falling posteriorly with a baby chair. Also, there are reports of ASDH caused by falling from a low position, so it is difficult to judge the mechanism further [20–22]. Moreover, determining if a case is one of AHT or not from the mechanism of injury remains controversial. It is important not to jump to conclusions without prudent discussion with multiple healthcare specialists when diagnosing AHT from the mechanism and severity of the injury [15, 23]. The diagnosis of AHT is difficult and should not be underestimated if there is no firm confession or reliable witness.

Clinically, many AHT patients encounter apnea, hypotension, hypoxia, and seizures, which lead to secondary brain damage. If the patients have additional injuries such as fractures or hemorrhage of the thoracic cavity and/or abdominal space, there will be a risk of anemia, coagulation disorder, systematic inflammation, and shock. These risks are another cause for secondary brain damage, highlighting that the pathophysiology will be more complicated [24–26]. Recently, autoregulation of cerebral blood flow (CBF), dysfunction of the autonomic nerve system, inflammation, and neuronal death have been found in patients with even only slight brain damage [7]. A report from Vavilala et al. found that CBF autoregulation dysfunction was present in all of the AHT cases in their cohort [27].

Hemispheric hypodensity(HH) is also known as big black brain is a well-known findings of CT in AHT. A high signal intensity is revealed in MRI diffusion weighted image, so the mechanism of HH was thought to be a ischemia or hypoxia, but it is still unknown [28, 29]. HH is found in 25-50% in AHT patients, and the mortality rate increases to twice compared to the AHT patients without HH [30]. Costine-Bartell has describe that seizure could be the driver of AHT and HH so that early detection of seizures using continuous EEG monitoring and control of seizures with anti-epileptic drugs are important for the treatment of AHT [28]. The long-term outcome of AHTs is poor. 20-25% of the cases are lost, and other symptoms such as developmental delay and motor dysfunction (15-64%), seizures (11-32%), microcephaly due to brain atrophy (61-100%), visual impairment (18-48%), and language delay (37-64%) and other disabilities remain as a severe disadvantages. Also, there are reports that rehabilitation in both acute phase and long-term phase can improve the status of the less severe cases [28].

A differential diagnosis: infantile ASDH

In 1965, Dr. Nakamura and colleagues classified children's intracranial hemorrhage accompanied with head injury into 3 types according to the severity of the impact and coloration of the intracranial lesions. This was during the era before CT was introduced in Japan. Type I was defined as a slight injury from normal everyday events such as falling at home or falling down less than 90 cm from the balcony of the first floor. Type III was defined as the most severe injury, such as traffic accidents or falling from a high altitude. Type II was defined as injuries between type I and III, such as falling from a jungle gym. The intracranial lesions in type I were ASDH without contusions. In the 18 cases included in their report, the mean age was around 1 year old, which was when the patients just started to walk. Most cases received an impact at the posterior area of the head. The children hit their heads on tatami (a type of mat used as a flooring material in traditional Japanese-style rooms and dojos. It is covered with a soft rush weave and has a core with sewn-together rice straw or extruded polystyrene foam, with a characteristic firmness.) and carpets, and none of them hit their heads on paved roads. Thirteen cases had seizures after the impact followed by a deterioration of consciousness. Ten cases were associated with retinal bleeding. It was explained that the impact to the occipital area induced the laceration of the bridging veins, causing the subdural hematoma. They concluded that if an infant around 1 year old hit their head, ASDH should be suspected if four of the following criteria were met: (1)

impact to the occipital area, (2) repeating vomiting, (3) seizures, (4) hemiparesis, (5) disturbance of consciousness, (6) cyanosis, (7) a tight fontanelle, and (8) anisocoria and associated retinal hemorrhage [31]. Though the original article did not mention abuse, this report published in the 1960s led to recognition by Japanese neurosurgeons that there are cases of ASDH associated with retinal hemorrhage caused by minor head injury that closely mimic the pathophysiology of AHT.

Aoki and Masuzawa analyzed 26 cases of infants with ASDH apparently due to minor head trauma without loss of consciousness or cerebral contusion; the most common age was 7–10 months old, most of their cases had generalized tonic seizures, and surprisingly all of their cases were associated with retinal or preretinal hemorrhage. It was interesting that 16 cases (62%) involved a fall onto a tatami [32].

In 2006, Nishimoto and Kurihara published their data. They analyzed children who met the following inclusion criteria: (1) ASDH caused by trivial household trauma and a history of trauma corroborated by a caretaker, (2) absence of physical injuries consistent with child abuse, (3) fundoscopic examinations performed by a pediatric ophthalmologist, (4) absence of fractures on general bone survey, and (5) child abuse ruled out by long-term follow-up (more than 5 years). Twenty-five patients met these criteria. Regarding the episode of injury, 17 cases fell on the floor while sitting or standing, and the other eight cases fell from a short distance (50-90 cm). Most of the cases (68%) involved impact to the occipital area. The main symptoms were convulsion (56%) and disturbance of consciousness (24%). Retinal hemorrhages were seen in 60% of cases. Three cases underwent surgery to remove an ADSH, while burr hole drainage surgery was necessary in four cases. Since most of the cases were treated conservatively, none of the cases were lost and most developed normally, except for three cases with epilepsy [33].

Akutsu et al. conducted a multicenter retrospective analysis of infantile subdural hematoma. Two university hospitals, two children's hospitals, and two general hospitals participated in this study. A total of 452 patients were included, consisting of 58 cases in the non-accidental group and 394 cases in the accidental group. The mean age of the nonaccidental group was 5.8 months, and the mean age of the accidental group was 12.1 months. Subdural hematoma was the second most common radiological finding in the whole cohort, with 51 non-accidental cases and 107 accidental cases. The mean age of patients with ASDH was 5.9 months and 10.2 months in the non-accidental and accidental groups, respectively. Retinal hemorrhage was present in 45 of the 58 patients in the non-accidental group and 40 out of 394 patients in the accidental group. Unlike in other countries, subdural hematoma was found to be frequently associated with, but not characteristic of, AHT in Japan [34].

In Japan, it has been reported that some cases of subdural hematoma have a severe outcome despite occurring due to a slight head injury. Yoshimura et al. reported a case of an accidental ASDH where the patient underwent removal of the hematoma and developed hemispheric hypodensity, known as a "big black brain" [35]. They found five cases of hemispheric hypodensity cases upon a review of 67 cases of ASDH in the Japanese literature [10, 33–37].

Although reports from Japan have accumulated data on ASDH caused by minor head injury, they have been criticized for not being fully investigated for child abuse [38]. Conversely, similar reports have been published from other countries [39–43]. In a study by Akutsu et al., there was no difference in the age range of non-accidental cases compared to other reports; however, the peak age of accidental cases in their cohort was 8–10 months, which is when infants begin to sit or pull to stand and ambulate independently [34]. This age range is not highlighted in other reports.

As most of the reports mention, it is true that accidental cases cannot be clearly distinguished from AHT cases and that AHT should not be underestimated. However, it seems that there is an entity of subdural hematoma due to mild head injury in infants that shows similar findings to AHT. The reason why these cases are reported mostly in Japan is not clear. Potential explanations include the presence of a specific impact surface, such as tatamis and futons, and ethnicity. It could be an underestimation in the detection of abuse; however, recent reports from Japan has been carefully determined because of the awareness of this discrepancy.

The role of pediatric neurosurgeons in Japan

In Japan, there are 6000 certified neurosurgeons. Among these neurosurgeons, there are 240 certified pediatric neurosurgeons after the certification system started in 2016. The role of neurosurgeons in Japan is diverse. They start treating patients from the emergency room as an emergency doctor, perform surgery, take care of patients postoperatively, and follow the patients for a long time as a family doctor. Pediatric neurosurgeons undertake similar roles, so they have a high probability of encountering AHT cases from their initial presentation. They also act as a leader in the multidisciplinary child protection team.

Regarding AHT in Japan, the Ministry of Health, Labor and Welfare published the "Guide to Responding to Child Abuse" as a standard for child guidance centers to take temporary custody; the 2014 revision states that an infant with subdural hematoma is highly likely to have been abused and should be treated with suspicion. Because of this guideline, if a physician (mainly a pediatrician or neurosurgeon who does not specialize in pediatrics) treats a child who has the "triad" of subdural hematoma, retinal hemorrhage, and encephalopathy, they may jump to a conclusion that there is abuse.

According to a systematic review from Sweden, there is insufficient scientific evidence on which to assess the diagnostic accuracy of the triad in identifying traumatic shaking (very low-quality evidence) and limited scientific evidence that the triad and its components are associated with traumatic shaking (low-quality evidence) [44]. After this review, 15 societies including the Japanese Pediatric Society posted a counterargument [45]. This review does not deny the relationship between the triad and AHT, as it is obvious that the triad is seen in AHT; the triad is an important finding to diagnose AHT. However, these reports suggest that careful identification is necessary to diagnose AHT.

When compared to western countries, there are fewer head traumas sustained by shaking and a higher proportion of female perpetrators of abuse in Japan. Also, it seems that there are cases of infantile subdural hematomas caused by a minor head injury, at least in the Japanese population. Therefore, it is necessary, especially for Japanese pediatric neurosurgeons, to carry out a fair and careful investigation when they encounter suspected AHT cases.

Author contribution Kazuaki Shimoji conceived the idea of the study, conducted literature search, and drafted the original manuscript. Eiichi Suehiro conceived the idea of the study and conducted literature search. Akira Matsuno supervised the conduction of the study. Takashi Araki supervised the conduction of the study. All authors reviewed the manuscript draft and revised it critically on intellectual content and approved the final version of the manuscript to be published.

Availability of data and material This article is a review paper so that there are no data and material to provide.

Declarations

Ethics approval and consent to participate This article was approved by the International University of Health and Welfare IRB committee.

Consent of publication We give our consent for the publication of identifiable details, which can include photograph(s) and/or videos and/or case history and/or details within the text ("Material") to be published in this journal.

Conflict of interest None.

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