

SBU ASSESSMENT • REPORT 255E/2016

Traumatic shaking

The role of the triad in medical investigations of suspected traumatic shaking

A systematic review

Report series This report is an SBU Assessment. It presents the results of a comprehensive systematic review of the available scientific evidence, including economic, social, and ethical impact analyses. The assessment was performed by a team of leading professional practitioners and academics and patient/user representatives supported by SBU staff members. The certainty of the evidence for each finding was systematically reviewed and graded. The report has been reviewed by independent experts and the conclusions have been approved by SBU's Board of Directors.

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Declaration of contents

✓ Assessment of new/ established intervention	✓ Consensus process
✓ Systematic literature search	✓ Produced in collaboration with experts in the field
✓ Relevance assessment (Study selection)	✓ Patient/user involvement
✓ Risk of bias assessment/ quality assessment	✓ Ethical aspects
✓ Synthesis of results	✓ Economic/health economic aspects
✓ Quality of evidence assessed by SBU	✓ Social aspects
✓ Quality of evidence assessed externally	✓ Approved by the SBU quality and priority group
Based on systematic review	✓ Approved by SBU's Board of Directors

Key words Shaken Baby Syndrome, Infant, Child Abuse, Subdural Hematoma, Retinal Hemorrhage, Brain Edema

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Period of validity Results based on strong scientific evidence usually remain valid for a long time. Other results may become outdated over time. This is particularly true for results based on limited or insufficient scientific evidence.

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Summary of the results

The systematic review showed the following graded results:

- There is limited scientific evidence that the triad¹ and therefore its components can be associated with traumatic shaking (low quality evidence).
- There is insufficient scientific evidence on which to assess the diagnostic accuracy of the triad in identifying traumatic shaking (very low quality evidence).

Limited scientific evidence (low quality evidence) represents a combined assessment of studies of high or moderate quality which disclose factors that markedly weaken the evidence. It is important to note that limited scientific evidence for the reliability of a method or an effect does not imply complete lack of scientific support.

Insufficient scientific evidence (very low quality evidence) represents either a lack of studies, or situations when available studies are of low quality or show contradictory results.

Evaluation of the evidence was not based on formal grading of the evidence according to GRADE but on an evaluation of the total scientific basis.

¹ Three components of a whole. The triad associated with SBS usually comprises subdural hematoma, retinal hemorrhages and encephalopathy.

1 Aim

In cases of suspected traumatic shaking, the diagnosis has conventionally been based on three findings, referred to collectively as the triad, namely: subdural hematoma (bleeding between the dura mater and the brain), retinal hemorrhages, and various forms of brain symptoms (encephalopathy). The presenting history is often that of lethargy, seizures and apnea. The purpose of this evaluation was to determine how reliably the triad or its components can be explained by traumatic shaking of children up to one year of age.

2 Background

Child abuse was described in the medical literature as early as in the 1800's [1], but it was only much later that awareness of the practice became more widespread [2,3]. Child abuse can often be concealed within the family and there is a risk of underdiagnosis, in part because the child is unable to speak for itself. At the same time, overdiagnosis can have serious consequences, because families can be split apart on false grounds. Adherence to the healthcare principle that the triad is attributable exclusively to traumatic shaking can lead to overdiagnosis, because of failure to consider other possible causes of the child's condition.

What is traumatic shaking?

Traumatic shaking occurs when a child is shaken in such a way that its head is flung backwards and forwards. In 1971, Guthkelch, a neurosurgeon, hypothesized that such shaking can result in a subdural hematoma, in the absence of any detectable external signs of injury to the skull [4]. The article describes two cases in which the parents admitted that for various reasons they had shaken the child before it became ill. Moreover, one of the babies had retinal hemorrhages. The association between traumatic shaking, subdural hematoma and retinal hemorrhages was described by Caffey in 1972 and referred to as Whiplash Shaken Infant Syndrome [2]. The injuries were believed to occur because shaking the child subjected the head to acceleration-deceleration and rotational forces. In 1987, this theory was queried by Duhaime et al. [5] in a biomechanical study which concluded that isolated shaking, in the absence of direct violence, is probably not of sufficient force to cause the injuries described above.

The name of the condition has since been changed to Shaken Baby Syndrome (SBS). There are a number of studies on the association between various clinical and radiographic findings and injuries caused by violent shaking of a child [6–13].

In recent years the term abusive head trauma (AHT) has been introduced (see section Terminology). The project group decided to apply the term "traumatic shaking" to the trauma mechanism and the "triad" to the actual signs and symptoms [13].

Signs and symptoms

In the scientific literature, various signs and symptoms are described in association with traumatic shaking. The collective name "triad" has been adopted for the most frequently occurring injuries (subdural hematoma, retinal hemorrhage and encephalopathy). The main focus of this report is the triad (see section Terminology). Other signs are occasionally reported in association with traumatic shaking, including bruising to the chest, fractures of, for example, the ribs and shinbone (metaphyseal fractures), but these injuries are not included in the present review.

Presenting medical history

When medical attention is sought for the affected child, the presenting history includes various clinical signs such as seizures, lethargy or other symptoms of encephalopathy. The initial clinical and radiographic examination can disclose the presence of, for example, subdural hematoma, or various symptoms of brain dysfunction. Subdural hematoma, retinal hemorrhages and various forms of encephalopathy can have serious sequelae, with permanent damage to the brain and/or the eyes. Permanent damage can comprise serious impairment of cognitive and/or motor function, with widespread adverse effects on the child's health, development and future quality of life and can ultimately even be fatal.

Healthcare personnel are encouraged to be alert to the findings which comprise the triad and are required by law (Social Services Act: Chapter 14, Section 1) to notify the Board of Social Welfare if they become aware, or suspect, that a child is being abused, or otherwise may need protection. In a frequently quoted article by the American Academy of Pediatrics, 1993 [14] physicians documenting trauma affecting the brain in newborns are encouraged to conduct a thorough examination and to be familiar with the clinical and radiographic findings which can confirm damage caused by traumatic shaking. The regional health care plan published by the Stockholm County Council 2011 stated:

"If there is no history of a traffic accident or a fall from a considerable height, the combination of subdural hematoma and encephalopathy with edema or hemorrhage strongly suggests that the child has been abused. If there are also

retinal hemorrhages then from the medical point of view the diagnosis of abuse is quite clear.” [15].

Other regional care programmes as well as the statement by the Swedish National Board of Health and Welfare on children who are being abused or are at risk of abuse include guidelines on the care of infants in cases of suspected abuse [16,17].

In recent years however, the certainty with which it can be determined that the findings of the triad are in fact attributable to traumatic shaking has been questioned [18–26]. Many articles which have debated the subject of traumatic shaking and the symptoms and signs of the triad have been published in international and national journals and in the media. In this context, it is important to ascertain whether the conclusion that traumatic shaking is the cause of these signs and symptoms is based on evidence of the highest possible scientific quality. However, grading of scientific supporting evidence is based on the assessment of groups – not of individuals. In order for the justice system or social services to make a statement on the association between exposure and disease or injury, assessment of the individual case is required, with other observations and conditions also taken into account.

Terminology

The English term for the triad is Shaken Baby Syndrome (SBS), which refers to the signs and symptoms which allegedly can arise after an episode of isolated traumatic shaking, i.e. shaking without the head impacting on any object.

In 2009 the American Academy of Pediatrics recommended a broader term, Abusive Head Trauma (AHT), which includes also direct trauma to the head [27]. Also to be found in the literature are several other terms, which partly or completely overlap the terms Shaken Baby Syndrome och Abusive Head Trauma (see Chapter 9). The terms are used in a variety of ways in the scientific literature and this contributes to the lack of methodological clarity in studies of the effects of traumatic shaking. The project group has therefore decided to limit the scope of the project to isolated traumatic shaking, thereby including only studies of cases in which there is no evidence of direct trauma (external injury) to the head. Furthermore, the authors have avoided the terms SBS and AHT because they imply both the signs and symptoms and the alleged mechanism behind the findings, even the intent. Instead, the authors chose to make a clear distinction between the injurious mechanism (“traumatic shaking”) and the medical findings (“the triad”).

Investigation of injuries which may be attributable to traumatic shaking

Diagnosis of suspected brain injury is based on computerised tomography (CT) and/or magnetic resonance imaging (MRI). The presence of retinal hemorrhages is determined by examination of the fundus by ophthalmoscopy or fundoscopy.

Other possible causes (differential diagnoses) of the triad and its components

In cases presenting with the triad, it is important to determine whether these can be attributed to causes other than traumatic shaking.

Subdural hematoma, retinal hemorrhages and encephalopathy have been described after delivery and in association with such conditions as various convulsive states, certain hemorrhagic diseases, infectious diseases, metabolic disorders, immunological diseases, skeletal diseases and vascular malformations (see Appendix 1 for details).

The triad – Signs and symptoms

Subdural hematoma

It is well-known that trauma to the head can give rise to subdural hematoma. In an adult, the underlying mechanism is rupture of one or more of the bridging veins, with bleeding into the subdural space. In many such cases there are often also external signs of trauma to the head in the form of soft tissue bleeding, but in other cases a CT scan may disclose internal injury in the absence of any evidence of external trauma.

Shaking an infant causes the movement of the brain to be out of synchrony with the movement of the skull. However, there is lack of consensus about the mechanism underlying the bleeding. It has been proposed that it may result from capillary injury [28,29]. As isolated traumatic shaking does not involve direct trauma to the head, there will be no external signs of head trauma such as swelling of soft tissues, contusions, lacerations or skull fractures. Hence, an incident is not classified as isolated traumatic shaking when soft tissue injuries or skull fractures are detected. Soft tissue injury and skull fracture(s) are therefore findings which exclude isolated traumatic shaking.

Imaging techniques have shown that subdural hematoma can occur in association with vaginal delivery, but is usually resorbed within a few weeks [22]. In the space created by the hematoma, effusion (leakage of fluid) may result in the development of a so-called subdural hygroma, which contains cerebrospinal fluid. It has been proposed that further bleeding in this space could occur spontaneously, or as a result of minor trauma [30–33]. It has also been proposed that an enlarged subarachnoid space could increase the risk of subdural hemorrhage [19,34–36].

Retinal hemorrhages

Retinal hemorrhages associated with traumatic shaking have been attributed to transfer of shearing forces in the vitreous body of the eye to the retina, due to increased pressure in the venous blood vessels in the retina, resulting in rupture of the vessels [37–39]. It has also been proposed that during shaking, repetitive acceleration and deceleration create shearing forces between the vitreous body and the retina, as well as direct injury to the eyeball. However, bleeding in the fundus of the eye has also been demonstrated in association with subdural hematoma considered to be caused by disease and it is therefore possible that retinal hemorrhages can arise as a sequel to subdural hematoma. One possible explanation is that increased intracranial pressure caused by edema of the brain leads to increased pressure in the central optical vein, with congestion in the retina [40,41]. The relationship between subdural hematoma and retinal hemorrhages is supported by studies showing that isolated incidents of retinal hemorrhages are very rare [38,41]. Retinal hemorrhages have also been observed after normal vaginal deliveries [42].

Encephalopathy

Encephalopathy can present with such signs as lethargy, seizures and dyspnea, among others. These signs may be attributable to frictional damage in the brain or the cervical medulla, and/or brain edema. Brain edema and brain hypoxia can cause irreversible brain damage. Increased intracranial pressure, for example due to brain edema or subdural hematoma, can also result in seizures, apnea and lethargy [43,44]. Brain edema can be revealed by both computed tomography (CT) and magnetic resonance imaging (MRI) and appears as effacement of the sulci and compression of the cerebral ventricles. These may be temporary conditions, which resolve without any permanent brain damage. The most serious condition can be revealed by CT and MRI as reduced differentiation between the white and grey matter of the brain, representing a global irreversible ischemic injury.

Diagnostic methods

Intracranial examination

While CT is based on the differences in absorption of x-ray radiation of substances and tissues of varying density, MRI exploits a number of different

properties of substances and tissues and thereby offers a richer and often more specific characterisation of the tissue being examined.

Both techniques allow observation of thin “sections” through the entire brain, with reconstructions in several planes, and also assessment of the intracranial vessels (by using contrast medium in the vessels). Both techniques provide similar information on changes in the brain ventricles and basal cisterns, such as compression of the ventricles in brain edema, widening in hydrocephalus, displacement due to hemorrhage and the risk of brain herniation.

However MRI can provide different information from CT, for example with respect to the presence of fresh blood, deposition of hemosiderin (a decomposition product of hemoglobin) and early ischemic and axonal injuries [45,46].

While an acute subdural hematoma in a small child comprises fresh blood, a subacute subdural hematoma is usually composed of a mixture of an upper layer of fluid and a sediment of coagulated blood [47]. The development of the hemorrhage over time results in different patterns on CT and MRI. The time frames for the development and duration of these patterns can overlap; hence determination of the age of the injury is uncertain [48]. In rare cases, calcifications can be mistaken for fresh blood, particularly in the brain tissue. On a CT scan, hemorrhage has a more robust pattern than that seen on various MRI sequences, which have a varying and partly overlapping appearance, depending on the composition of the bleeding and the time elapsed since the injury. CT assessment of the age of a subdural hemorrhage is therefore considered to be more reliable than assessment by MRI [49,50]. The ability to determine the age of a subdural hematoma can be important for correlation with the alleged time of injury.

Both CT and MRI can be used to determine brain edema, which appears as effacement of the sulci on the surface of the brain and compression of the ventricles and basal cisterns. CT is more reliable than MRI for assessing fractures.

Retinal examination

Two methods can be used for examination of the ocular fundus. The most common is fundoscopy with or without dilatation of the pupil. More recently a photographic method has been developed (Retcam). This method allows subsequent assessment of the findings by other observers who are not aware of the case history or the purpose of the examination [51,52].

At autopsy the entire eye can be examined, and other findings can then be described [53,54].

Hemorrhage in the ocular fundus cannot usually be assessed by CT or MRI. However, in a recently published MRI study, a particular imaging sequence was compared with ophthalmoscopy and it was shown that in 83% of cases retinal hemorrhages could be detected by MRI [55,56].

In this context it is important to be aware that interpretations of CT, MRI and ocular fundoscopy findings are somewhat subjective and the experience of the individual observer can influence the final assessment.

3 Systematic evaluation: method

Question to be addressed

The aim of the present investigation was to address the following question:
With what certainty can it be claimed that the triad, subdural hematoma, retinal hemorrhages and encephalopathy, is attributable to isolated traumatic shaking (i.e. when no external signs of trauma are present)?

PIRO

P (Population): Children ≤ 12 months of age

I (Index test): The triad in cases of suspected traumatic shaking

R (Reference test/gold standard): Admitted or witnessed traumatic shaking or other trauma

O (Outcome measure): Diagnostic accuracy

The Project has been conducted in accordance with the method described in SBU's manual [57].

Criteria for inclusion and exclusion

Inclusion criteria

Study design

Case-control, cohort and registry studies and studies applying qualitative methods of analysis.

Observations

In order to reduce the risk of random errors of selection, only studies comprising 10 or more cases were included. With respect to possible alternative explanations (differential diagnoses), the project group was of the opinion that one published case was sufficient to question the hypothesis that the triad is always caused by traumatic shaking. Articles on differential diagnoses were not quality assessed and are therefore not included in the basis for the results. If a subgroup of children who had been subjected to traumatic shaking and/or a subgroup aged ≤ 12 months (median and/or mean age) was included in AHT studies, then these were included by the project group. The mean age of children subjected to traumatic shaking is stated to be 2–3 months [58] and the project group therefore decided to limit the review to studies of children with a mean or median age of ≤ 12 months.

Language

Articles written in English, German, French, Swedish, Danish and Norwegian were included.

Other criteria

The project group decided to include only cases of traumatic shaking which were witnessed (e.g., video recorded) or in which someone had confessed to shaking the child.

Exclusion criteria

The project group excluded studies of fewer than 10 cases and AHT studies which included external injury to the head and/or fractures and other injuries.

Studies identified in the literature search as biomechanical studies and studies which deal with other possible causes of the triad have been considered separately and are presented in Appendices 1 and 2.

Methodology for selection of studies

Based on the question to be addressed by the project, the literature databases were searched systematically, in close collaboration between the information

specialist and the experts in the project group. The literature search encompassed the databases PubMed, Embase och Cochrane Library through October 15th, 2015. Further studies were searched for manually, through the reference lists of individual studies and systematic reviews. For a detailed description of search terms and limitations (see Appendix 4, www.sbu.se/255e).

Assessment of relevance

The lists of abstracts generated by the literature search were scrutinised independently by two experts. Studies deemed by at least one of the experts to be relevant to the questions to be addressed by the project were retrieved in full text and scrutinised independently by two experts with reference to the project's inclusion criteria. Articles which were scrutinised in full text and did not meet the inclusion criteria were excluded: the main reason for exclusion was recorded (see Appendix 5, www.sbu.se/255e). Disagreements were addressed initially by discussion between the two experts who had read the article. In certain cases the entire project group was involved in the discussion and the decision about inclusion or exclusion was resolved by consensus.

Assessment of the quality of individual studies

Because of the specific field of research, the project group modified SBU's template to assess the quality of the included studies and to determine the risk of bias (circular reasoning: see the section "Circular reasoning in clinical and research settings" in Chapter 5). The template includes i.a. the type of study (prospective, diagnostic, biomechanical etc), the main focus of the study and whether the study addressed subdural hematoma, retinal hemorrhages and/or encephalopathy. In accordance with SBU's guidelines, only studies of moderate or high quality were considered in the results and discussion [11].

Systematic reviews of the field were quality assessed using the AMSTAR instrument [57]. The results in the present report were based on original studies and not on other systematic reviews (see Chapter 5).

Method for synthesis of the results

Meta-analysis is a statistical method for quantitatively appraising the results of several studies in order to obtain data from a larger sample and to achieve a more reliable assessment of the statistical uncertainty. In order to pool the results, the studies must have been conducted using similar methods and it must be possible to adjust the analyses for similar background factors. As only

one of the included studies used a reference group, it was not possible to undertake a meta-analysis.

Assessment of the quality of the evidence

The quality of the evidence indicates the level of reliability of the results and is based on the assessment of study quality (risk of bias), inconsistency, imprecision, risk of publication bias and indirectness.

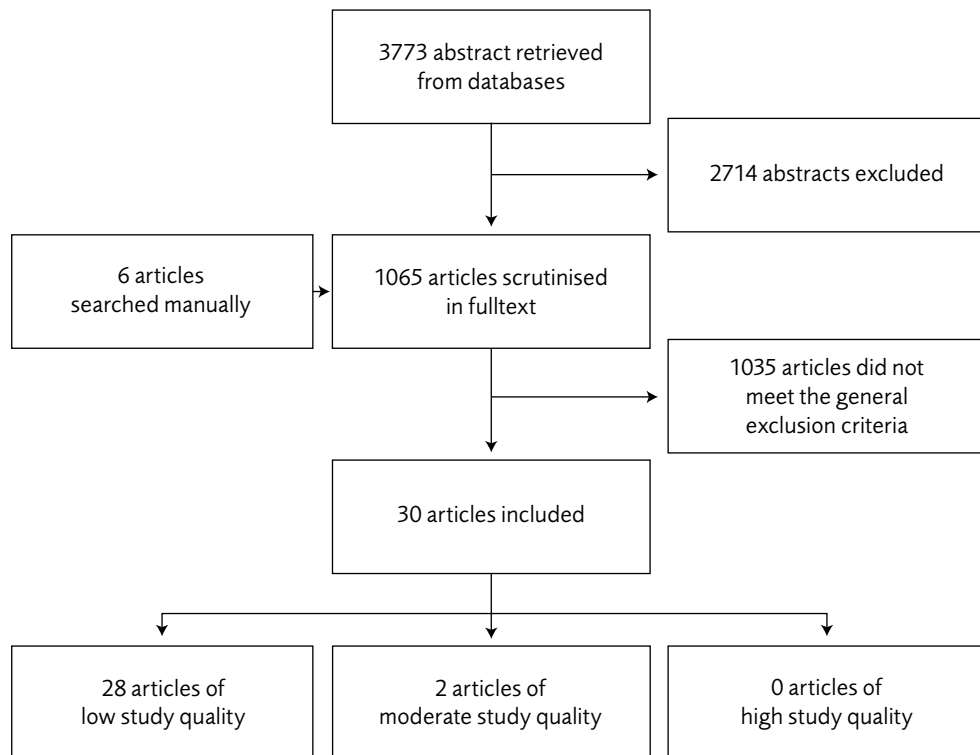
As no meta-analysis was possible, the results were based on a narrative synthesis of the included studies. Evaluation of the evidence was not based on formal grading of the evidence according to GRADE but on an evaluation of the total scientific basis. The quality of the evidence was deemed to be limited (low) when combined assessment of studies of high or moderate quality disclosed factors which markedly weaken the evidence. The quality of the evidence was deemed to be insufficient (very low) when there was a lack of studies, when the available studies were of low quality or when studies of similar quality showed contradictory results. It is important to note that limited evidence for the reliability of a method or an effect does not imply complete lack of scientific support.

4 Results

The literature search yielded 3773 abstracts, of which 1065 were retrieved in full text. Of these 1035 were excluded because they did not meet the inclusion criteria. Of the 30 remaining studies, two were assessed to have moderate quality and none of high quality. The main reason that so few studies met the quality requirements was that the published papers failed to provide information as to whether traumatic shaking was confessed to by the perpetrator or had been witnessed. Thus the results are based on only two studies of confessed traumatic shaking and a meta-analysis was therefore not possible. However, agreement between the results of the included studies was discussed in the project group.

Flow chart of literature search

Figure 4.1
Flow chart of literature search.



Quality of the evidence

The systematic review showed the following graded results:

- There is insufficient scientific evidence on which to assess the diagnostic accuracy of the triad in identifying traumatic shaking (very low quality evidence).
- There is limited scientific evidence that the triad and therefore its components can be associated with traumatic shaking (low quality evidence).

The two included studies of moderate quality, both conducted in France, were based on cases in which the perpetrator confessed to subjecting the child to traumatic shaking. The study by Vinchon et al. was a prospective study. It was based on a register of traumatic head injury in children aged under 2 years, who were admitted to hospital between May 2001 and February 2009, in a catchment area with a population of about 4 million [59]. The material comprised 412 cases, of which 124 were classified as Inflicted Head Injury (IHI) and 288 as Accidental Trauma (AT).

In the group with inflicted injury (IHI group), there were 45 confessed cases: 30 by traumatic shaking and 15 in which the perpetrator admitted to other

external trauma. However, the article does not include detailed descriptions as to how the perpetrator inflicted the injuries, nor the circumstances under which the confession was obtained. This group of children was compared with 39 cases in which accidental trauma was witnessed in a public place (AT group).

In the group with inflicted trauma, 37 out of 45 (82%) had a subdural hematoma, compared with 17 out of 39 (44%) in the accidental trauma group; 37 out of 44 (84%) had retinal hemorrhages, compared with 6 out of 35 (17%) in the accidental trauma group and 12 out of 45 (27%) had cerebral ischemia, compared with 1 out of 39 (3 percent) in the group with accidental head trauma.

The study by Adamsbaum et al. was a retrospective observational study, comprising 29 confessed cases of traumatic shaking (in which direct trauma to the head was described in 5 cases) and a comparative group of 83 unconfessed cases [60]. The criteria for inclusion in the study were subdural hematoma disclosed by a CT scan and confession by the suspected perpetrator. As subdural hematoma was one of the criteria for inclusion in the traumatic shaking group, only the results for retinal hemorrhages could be used in this investigation.

In the group in which traumatic shaking was confessed to (Group A) 24 children (83%) had retinal hemorrhages. In all cases where the perpetrator had confessed, the shaking was described as violent (100%) and in some cases (55%) the perpetrator admitted to repeated episodes of shaking. No correlation was established between the density of the subdural hematoma and the number of repeated episodes of shaking. In 14 out of 29 cases in Group A there was a detailed description of how the suspect had committed the act. In the other group (Group B) there were children who had been shaken in an attempt at revival, or had suffered accidental injury and some children for whom no explanation of the condition was presented: thus this group cannot be considered an acceptable reference group.

The studies by Vinchon et al. and Adamsbaum et al. both demonstrate that traumatic shaking can cause subdural hematoma and retinal hemorrhages. In the study by Vinchon et al., the group in which traumatic shaking was confessed to comprised a larger proportion of children with subdural hematoma, retinal hemorrhages and cerebral ischemia than the group of children who had been injured in a witnessed accident. Adamsbaum et al. compared a group of children in which the perpetrators had confessed to traumatic shaking, with a group of children in which the suspects had not confessed: this can result in inclusion bias in one or both groups. As only one of these two studies had a relevant reference group, it has not been possible to conduct a meta-analysis.

There are also other published cases which have been excluded (wrong population, wrong study design), but contain detailed descriptions of confessions which are in accordance with the two studies of moderate quality [61,62]. Because of the low number of studies of moderate or high quality it was not possible to determine the diagnostic accuracy of the triad in identifying traumatic shaking.

Table 4.1 Characteristics of included studies.

Author Year Reference Country	Aims/focus Outcome	Study design Setting Inclusion and exclusion criteria	Number of cases Medical examinations	Results	Methodological considerations/ comments	Study quality
Vinchon 2010 [59] France	<p>Aims/focus To compare the clinical, ophthalmological and radiological features of inflicted head injury (IHI) and accidental trauma (AT) and to test the diagnostic value of these findings</p> <p>Outcome Registered medical findings including SDH (density), RH and brain ischemia</p>	<p>Study design Prospective cohort study collecting medical information on all traumatic head injuries in infants from May 2001 to February 2009</p> <p>Setting Diagnosis in pediatric intensive care unit or neurosurgical unit</p> <p>Inclusion criteria Children under the age of 24 months referred alive to the emergency room due to confessed inflicted head trauma (IHT), or witnessed accidental trauma (AT)</p> <p>Exclusion criteria Obstetric trauma</p>	<p>Number of cases IHI group: 45 cases of confessed inflicted head injury, 29 boys and 16 girls, mean age 3.8 month (range 0.8–18.3). IHI was caused by shaking in 30 cases and beating in 15 cases</p> <p>AT group: 39 cases of witnessed accidental injury, 23 boys and 16 girls, mean age 8.1 months (range 0–23.9)</p> <ul style="list-style-type: none"> • 19 cases where the baby was a car passenger • 3 cases where the baby was in a carriage hit by a vehicle • 5 cases of defenestration • 11 cases injured by a short fall • 1 case without description <p>Medical examinations Clinical examination including neurological symptoms CT scanning Fundoscopy X-ray and/or isotopic bone scanning skeletal survey</p>	<p>IHI group SDH: 37/45 (82%) RH: 37/44 (84%) Brain ischemia: 12/45 (27%)</p> <p>AT group SDH: 17/39 (44%) RH: 6/35 (17%) Brain ischemia: 1/39 (3%)</p> <p>Diagnostic value</p> <p>SDH Sensitivity: 0.822 Specificity: 0.552 PPV: 0.685 NPV: 0.724</p> <p>Severe RH Sensitivity: 0.556 Specificity: 0.974 PPV: 0.961 NPV: 0.655</p> <p>Brain ischemia Sensitivity: 0.267 Specificity: 0.971 PPV: 0.921 NPV: 0.505</p>	<p>The report does not contain a detailed description of the method nor of the IHI cases. It is not possible to extract the results for the shaken baby cases in the IHI group</p> <p>There are limitations in the definitions of the diagnostic criteria</p> <p>The triad used by the authors consists of subdural hematoma, retinal hemorrhage, and absence of sign of impact</p> <p>No details about what the suspect had confessed and under which circumstances the confession was obtained</p>	Moderate

The table continues on the next page

Table 4.1 continued

Author Year Reference Country	Aims/focus Outcome	Study design Setting Inclusion and exclusion criteria	Number of cases Medical examinations	Results	Methodological considerations/ comments	Study quality
Adamsbaum 2010 [60] France	<p>Aims/focus To correlate the history of confessed abusive head trauma (AHT) cases with medical findings and to compare medical findings in confessed cases of AHT with non-confessed cases</p> <p>Outcome Density and location of SDH, RH, fractures and skin ecchymosis</p>	<p>Study design Retrospective observational study with examined forensic evidence, from January 2002 to May 2009</p> <p>Setting Not specified</p> <p>Inclusion criteria Children with AHT (presence of SDH on CT scan with or without traumatic skin lesions) and a perpetrator conviction, with or without confession</p> <p>Exclusion criteria Accidental trauma and metabolic or infectious pathology</p>	<p>Number of cases Group A: 29 cases where the perpetrator confessed violent shaking of the child. In 5 cases a final impact of the infants head on a bed was described by the perpetrator. 22 boys and 7 girls, mean age 4.7 ± 2.9 months. 27 cases were younger than 1 year</p> <p>Group B: 83 cases without confession of a causal relationship between violence and the child's symptoms. 63 boys and 20 girls, mean age 6 ± 5.3 months</p> <ul style="list-style-type: none"> • 19 cases where the children was shaken in order to revive it from a life-threatening event • 28 cases where a minor accident was described • 36 cases where no mechanistic description was given <p>Medical examinations Clinical examination CT scanning Fundoscopy X-ray skeletal survey MRI (only in confessed cases)</p>	<p>Group A RH: 24/29 (83%) SDH hyperdensity: 11/29 (38%) SDH hypodensity: 2/29 (7%) SDH mixed density: 16/29 (55%)</p> <p>9 infants in Group A died. In 4 of the cases the perpetrator admitted head impact</p> <p>Group B RH: 75/83 (90%) SDH hyperdensity: 28/83 (34%) SDH hypodensity: 0/83 (0%) SDH mixed density: 55/83 (66%)</p> <p>16 infants in group B died</p> <p>No statistically significant difference between the two regarding incidence of RH, density of SDH, or location of SDH</p>	<p>A retrospective study design without an appropriate control group</p> <p>The inclusion criterion SDH was part of the triad, why the prevalence of SDH after shaking cannot be evaluated</p> <p>The report contains a detailed description of 14 cases in group A. No details under which circumstances the confession was obtained</p>	Moderate

AHT = Abusive head trauma; AT = Accidental trauma; CT = Computed tomography; IHI = Inflicted head injury; IHT = Inflicted head trauma; MRI = Magnetic resonance imaging; NPV = Negative predictive value; PPV = Positive predictive value; RH = Retinal hemorrhage; SDH = Subdural hematoma

5 Discussion

Although relatively many studies met the criteria for inclusion, the literature search identified only two studies of moderate quality. This is disconcerting, because traumatic shaking is very serious and has dramatic consequences for both the child and its family. The research field is complex, but this does not excuse, for example, circular reasoning and inadequate presentation of data collection. It is important that reviews of the field include consideration of the methodological flaws which characterise this field of research.

The studies by Adamsbaum et al. and Vinchon et al. were deemed to be of moderate quality. Although both studies have methodological limitations, they support the hypothesis that isolated traumatic shaking can give rise to the triad.

The prospective study by Vinchon et al. was based on more than 400 cases: 124 were classified as inflicted and 288 as accidental injuries to the skull. Forty-five were cases of confessed inflicted skull injury, of which 30 were cases of confessed isolated traumatic shaking (IHI group).

Thirty-nine cases were witnessed accidents (AT group). The advantage of this study is that all trauma cases presenting at the hospital were registered prospectively for many years. The study also has a clearly defined reference group, of children who had accidentally sustained injuries in the presence of a witness. However, this group of children is significantly older. One of the limitations of the study is the lack of detailed description of how and when the shaking incident occurred. Vinchon et al. analysed the components of the triad separately, but the authors also introduced a different combination for the triad, namely subdural hematoma, retinal hemorrhages and the "absence of scalp swelling".

However data on effects on the brain are registered in the study, in the form of seizures, lethargy and coma, among others. By definition, the group of children with isolated traumatic shaking will comprise only those cases without signs of external trauma, while the group with accidental injury will include cases with signs of external trauma. At an early stage of the investigation the project group contacted Dr. Vinchon in order to clarify certain ambiguities, but not all queries were answered. The questions included i.a. how retinal hemorrhages were defined, how the authors calculated sensitivity, specificity and the predictive value of the triad, why they chose a different triad component ("absence of scalp swelling" instead of encephalopathy) and under what circumstances the suspected perpetrator had confessed and what had been confessed.

The study by Adamsbaum et al. was a retrospective observational study which included 29 cases in which a suspect confessed to traumatic shaking. While detailed confessions were presented for 14 cases, it cannot be discounted that among the cases for which no detailed confession was forthcoming, there could be some in which shaking occurred after the child exhibited symptoms of brain damage. The study group was compared with a reference group comprising 83 unconfessed cases. However, this is not a "true" reference group, as there may be cases of traumatic shaking among the unconfessed cases. As subdural hematoma is a criterion for inclusion of all cases in the study, only the results for retinal hemorrhages can be considered.

During the literature review the project group identified other conditions or events which can also give rise to the three components of the triad. Some of these conditions or events do not result in permanent disability or are very rare, but it should be noted that the triad or its components can be attributable to causes other than shaking. It is therefore important to consider these possible differential diagnoses in investigations of suspected traumatic shaking. Decisions made by social services or the court system are based not only on medical findings, but also on other evidence.

An analysis of biomechanical studies (Appendix 2) disclosed contradictory results and no conclusions can be drawn as to the minimal forces capable of generating these injuries in children.

Methodological issues

This review of the scientific evidence for diagnosis of traumatic shaking in children under the age of 12 months (mean or median age ≤ 12 months) disclosed a number of methodological issues in the published studies.

Definition of traumatic shaking

The project was limited to studies in which traumatic shaking was considered to be the primary cause of the child's injuries, but several studies have adopted a wider definition, for example inflicted head injury. Thus it has not always

been possible to distinguish between an injury attributable to traumatic shaking and an injury attributable to direct trauma to the head.

Classification of subjects into groups

Another methodological problem was that traumatic shaking was not always witnessed or confessed to by the suspected perpetrator, hence correct classification of cases into a traumatic shaking group or a reference group was uncertain. Thus there is a risk of incorrect evaluation of the association between the triad and traumatic shaking. Although there is a risk of false confessions, apart from film documentation, this is the only means of gaining an insight into what has actually happened to the infant. Because of the risk of false confessions, all confessions in these studies must be considered with caution.

Thus there are some risks associated with the decision of the project group to include only cases in which someone has confessed. The confession could be false because it was made as part of a plea bargain. It could also be false because the suspect has felt impelled to confess [63–65].

Circular reasoning in clinical and research settings

Under the Social Services Act, the Board of Social Welfare must be notified not only of all cases of (suspected) child abuse, but also of other cases in which a child may be deemed to be vulnerable to harm and in need of protection. Those required to notify suspected child abuse are personnel within the health and medical services, dental, preschool, school, social and criminal services (Chapter 14, Section 1 of the Social Services Act).

In many cases, it is a child protection team which investigates cases of suspected traumatic shaking. Over the years these teams have developed criteria based on certain symptoms and signs which can be associated with isolated traumatic shaking, after exclusion of other possible causes of the child's condition [66,67]. Some of these criteria are associated with the carer's credibility. The carer is not considered trustworthy if he/she cannot provide an "acceptable" explanation for the child's condition, for example that the child had fallen from a low height and had not sustained any external injury. A change of statement – for example, the carer first denies shaking the child and later admits to doing so, but only after the child had stopped breathing or lost consciousness – also reduces the carer's credibility. If the child was shaken because it suddenly showed signs of being unwell (such as dyspnea or apnea), it is however reasonable to assume that the child's condition was already cause for concern before it was shaken and thus the symptoms were not attributable to the shaking. If however, such an explanation of events is not deemed "acceptable", the case is still classified as a case of traumatic shaking.

The child protection team's criteria are based primarily on a clinical approach [66,67]. Problems arise later, when and if these criteria are not tested unconditionally by researchers in systematic studies of the association between the triad and traumatic shaking. This means that the interpretation made by the child protection team characterises the scientific investigation and hypothesis testing

and this, in turn, means that the conventional approach is reinforced instead of being tested. However, if before the study it has already been assumed that the question to be addressed by the study has been answered, i.e. the association between the symptoms and signs of the triad and traumatic shaking has already been described (according to the child protection team's criteria), then circular reasoning occurs. Applied in this context, the reasoning results in a high risk of bias, which in turn results in a situation wherein the researcher does not know what is being compared (the traumatic shaking group may include children who have not been shaken and the reference group may include children who have been shaken). Sensitivity, specificity and predictive values calculated on comparison of such groups will result in incorrect conclusions. It will also result in incorrect calculations of incidence.

In order to avoid such circular reasoning, study cases and control cases must be identified introcontrovertibly. The project group has chosen to accept as study cases only those in which there was a witness to (or video documentation of) an incident of shaking or where someone has made a detailed confession of shaking the child.

Diagnostic methods

There is uncertainty in determining the time at which a subdural hematoma arose. Moreover this uncertainty is greater in children under 12 months of age, because the characteristics of subdural hemorrhage at this age differ somewhat from those in adults. A subdural hematoma in a small child or infant usually consists of an upper layer of fluid and a sediment of coagulated blood: if the subdural hematoma is subacute, this layer can exhibit various degrees of attenuation [47]. The application of CT and MRI scans has recently reduced this uncertainty somewhat [46], but caution must still be exercised in assessing the age of a hematoma because of the existence of different and partly overlapping patterns [48].

In both controlled experimental and observational studies, systematic errors can occur because various observers do not always make the same observations and/or interpret the observations differently. Agreement among different investigators in a study can vary according to how well-trained the observers are. This applies not only in general to observations and assessments, but of course also to examinations and assessments of the symptoms and signs in cases of suspected traumatic shaking.

In one study, for example, there were major variations among the observers' interpretation of retinal hemorrhages, i.e. interobserver agreement was low [51].

Comparison with results of other reviews

The project group identified seven systematic literature reviews addressing the same or partly the same questions as the present report [68–74]. These reviews are not included in the results section of the present paper, but the project group scrutinised and assessed them because they are frequently cited in the scientific literature. All the systematic reviews were assessed by the project group to be of low quality (high risk of bias). Many of them were based on studies in which a team considered that a child had been shaken if it presented with the triad (circular reasoning, see section “Circular reasoning in clinical and research settings”). Another weakness in these reviews was that traumatic shaking was not specified and the more general term AHT was used instead, without a detailed description of what this term included.

6 Issues for future research

It is not possible to conduct randomised experiments in which children of various ages are subjected to various degrees of shaking. Biomechanical studies using dummies or models equipped with various inbuilt measuring instruments have been used to investigate the impact of mechanical forces on a child, but the results are contradictory. Further, for various reasons, it is difficult to extrapolate the results of animal experiments to infants.

The project group was therefore limited to observational studies in which exposure (in this case shaking) was assumed to have occurred. The most reliable are prospective cohort studies and ideally those subjects included in a traumatic shaking cohort should comprise cases in which the perpetrator has confessed in a detailed confession, including documentation of the circumstances under which the confession was obtained.

In many of the scrutinised studies, the children in the reference group were significantly older than those in the traumatic shaking group. The brain, skeleton and neck muscles in a 2-month old baby are different from those of an 8-month old. Hence, at the age of 0–2 months, an infant can be assumed to be more vulnerable to injury from shaking than an older baby. Comparison of two groups of children (traumatic shaking and accidental injury groups) which are not age-matched can lead to selection bias and incorrect conclusions. Studies with matched age groups would allow calculation of sensitivity and specificity and predictive values. In this context, an opinion on the probability that the triad was attributable to traumatic shaking could be expressed with greater certainty.

There is a lack of detailed knowledge about the pathophysiology of the development of subdural and retinal hemorrhages associated with vaginal delivery. Although most bleedings related to delivery are symptomless and disappear (are resorbed) within a few months, occasionally a hemorrhage can degenerate into a hygroma [19,30,36]. This circumscribed collection of fluid is contained by a membrane in which small vessels form and it is considered that this in turn can lead to renewed bleeding (rebleeding) and a chronic subdural pool of fluid. The possibility cannot be discounted that in certain cases, rebleeding can cause symptoms [19,36]. This could be one reason why a child suddenly exhibits signs of encephalopathy (lethargy, apnea and/or seizures), causing the carer to seek medical attention. Hypothetically such rebleeding could occur spontaneously or in response to minor trauma. There is therefore an urgent need for research into the pathophysiology and the natural course of subdural and retinal hemorrhages. Our understanding of the sequelae to traumatic shaking could also be improved by the development of better biomechanical models, for example models which take into account the impact of traumatic shaking on both the brain and the cervical vertebrae.

What measures are required to address the scientific uncertainties?

The reasons for scientific uncertainty in this field vary and should therefore be managed in different ways; from coordination of the entire field of research with respect to the direction future research should take, to conducting studies using correct methodologies and detailed descriptions of how the studies have been conducted.

International coordination

In order to improve diagnosis within the field, broad coordination at international level is required to ensure a study population of adequate size. Researchers in the field should strive to agree on which research questions are most urgent and collaborate to facilitate larger studies and to use similar study designs, allowing the results to be compared more readily. It should also be possible to establish an international register of confessed and well-documented cases.

Priority research topics

Of particular importance are studies intended to improve the diagnostic accuracy of diagnostic imaging of the brain, the cervical spine and the eyes [75]. There is also a need for better methods of studying the natural course of the observed injuries. Differential diagnosis such as bleeding in neonates associated with delivery also needs to be studied in order to identify the natural course of events [22,36,76,77]. Further research is also required in order to improve understanding of the pathophysiology underlying the triad. Refined biomechanical models would also contribute to improved understanding of traumatic shaking.

As far as possible, of course, studies should meet all the predetermined quality criteria. It is also important that the researchers are blinded with respect to the suspected mechanism of origin of the injuries and that the results are presented in such a way as to allow diagnostic accuracy to be calculated. This latter requirement thus means that each individual finding must be assessed in both the study group and the reference group.

One of the reasons that it was difficult to find evidence in this field is that in many studies the method and the results were inadequately described. With respect to future studies, the project group presents the following recommendations of requirements to be met, in order that the quality of the studies can be assessed and that meta-analyses can be conducted:

The studies should:

- Comprise prospective observational studies of confessed and well-documented cases with reliable methodology, in which the risk of false confessions was minimised;
- Be age-matched (study group and reference group);
- Contain detailed presentations of how the study material was collected, including documentation of the examination technique and detailed presentations of any complementary investigations undertaken in order to exclude differential diagnoses;
- Demonstrate that the observers of the symptoms and signs were blinded to (i.e. were unaware of) the suspected or alleged cause of the findings and describe how the blinding was achieved;
- Present raw data, sensitivity/specificity and confidence intervals;
- Be based on a material of adequate size and apply a uniform method of examination throughout;
- Present a detailed account of the confession, what was confessed to and the circumstances under which the confession was made.

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SBU engages external reviewers in its reports. The reviewers contributed valuable comments, which have improved the report. The final version of the report, however, may not include all the alterations or additions recommended by the external reviewers, partly because they were not always in agreement. Therefore the external reviewers do not necessarily support all parts of the report.

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Conflicts of Interest

In accordance with SBU's requirements, the experts and scientific reviewers participating in this project have submitted statements about conflicts of interest. These documents are available at SBU's secretariat. SBU has determined that the conditions described in the submissions are compatible with SBU's requirements for objectivity and impartiality.

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External collaboration

Co-operation with interested parties

The project management held meetings with members of the Swedish National Association for Families' Rights at the beginning of the project in order to inform them of the project and to solicit opinions.

Networking with government authorities

In conjunction with the release of the report, representatives of the following authorities were invited to SBU for information about the results: the Ombudsman for Children in Sweden, the Swedish Health and Social Care Inspectorate, the Swedish Prison and Probation Service, the Swedish National Board of Health and Welfare, the Swedish Association of Local Authorities and Regions, the Swedish Police Authority, the Swedish National Board of Forensic Medicine, and the Swedish Prosecution Authority.

The Swedish National Council on Medical Ethics

The analysis of the ethics was conducted by the Swedish National Council on Medical Ethics.

9 Glossary

AHT	Abbreviation for Abusive Head Trauma: damage to the skull caused by maltreatment of the child
AT	Abbreviation for Accidental Trauma
Attenuation	Attenuation: absorption of radiation in the body, which varies in accordance with the density of the tissues.
BE	Abbreviation for Brain Edema/Edema
Child protection team	Interdisciplinary team which investigates cases of suspected child abuse
CT	Abbreviation for Computed Tomography
Hydrocephalus	Increased volume of cerebrospinal fluid in the cavities of the brain
Hygroma	Accumulation of fluid, possibly arising after an earlier episode of bleeding
IHI	Abbreviation for Inflicted Head Injury or Intentional Head Injury: head injury caused by abusive maltreatment of the child
IHT	Abbreviation for Inflicted Head Trauma: injury to the head resulting from abusive maltreatment of the child
MF	Abbreviation for Metaphyseal Fracture: a fracture in the growth zone of a long bone, e.g in the shinbone just below the knee
MRI	Abbreviation for Magnetic Resonance Imaging
MRT	Abbreviation for Magnetic Resonance Tomography (see MRI)
RB/RH	Abbreviation for Retinal Bleeding/hemorrhage, intraocular bleeding
SAH	Abbreviation for Subarachnoidal Hemorrhage: bleeding in the subarachnoid space, i.e. between the soft meninges of the brain)
SBS	Abbreviation for Shaken Baby Syndrome; a syndrome comprising three components, the triad

SDH	Abbreviation for Subdural Hemorrhage, Subdural Hematoma: bleeding under the dura
Subarachnoid space	The space between the soft meninges
Traumatic shaking	The injurious mechanism when a child is shaken violently (not to be confused with the medical findings, "the triad")
Triad	Three components of a whole. The triad associated with SBS usually comprises subdural hematoma, retinal hemorrhages and encephalopathy

10 References

1. Tardieu A. Etude médico-légale sur les sévices et mauvais traitements exercés sur des enfants. *Annales d'hygiène publique et de médecine légale*; 1860.
2. Caffey J. On the theory and practice of shaking infants. Its potential residual effects of permanent brain damage and mental retardation. *Am J Dis Child* 1972;124:161-9.
3. Kempe CH, Silverman FN, Steele BF, Droegemueller W, Silver HK. The battered-child syndrome. *Jama* 1962;181:17-24.
4. Guthkelch AN. Infantile subdural haematoma and its relationship to whiplash injuries. *Br Med J* 1971;2:430-1.
5. Duhaime AC, Gennarelli TA, Thibault LE, Bruce DA, Margulies SS, Wisner R. The shaken baby syndrome. A clinical, pathological, and biomechanical study. *J Neurosurg* 1987;66:409-15.
6. Minns RA, Jones PA, Tandon A, Fleck BW, Mulvihill AO, Elton RA. Prediction of inflicted brain injury in infants and children using retinal imaging. *Pediatrics* 2012;130:e1227-34.
7. Fujiwara T, Okuyama M, Miyasaka M. Characteristics that distinguish abusive from nonabusive head trauma among young children who underwent head computed tomography in Japan. *Pediatrics* 2008;122:e841-7.
8. Myhre MC, Groggaard JB, Dyb GA, Sandvik L, Nordhov M. Traumatic head injury in infants and toddlers. *Acta Paediatr* 2007;96:1159-63.
9. Tung GA, Kumar M, Richardson RC, Jenny C, Brown WD. Comparison of accidental and nonaccidental traumatic head injury in children on noncontrast computed tomography. *Pediatrics* 2006;118:626-33.
10. Vinchon M, Defoort-Dhellemmes S, Desurmont M, Dhellemmes P. Accidental and nonaccidental head injuries in infants: a prospective study. *J Neurosurg* 2005;102:380-4.
11. Keenan HT, Runyan DK, Marshall SW, Nocera MA, Merten DF. A population-based comparison of clinical and outcome characteristics of young children with serious inflicted and noninflicted traumatic brain injury. *Pediatrics* 2004; 114:633-9.

12. Bechtel K, Stoessel K, Leventhal JM, Ogle E, Teague B, Lavietes S, et al. Characteristics that distinguish accidental from abusive injury in hospitalized young children with head trauma. *Pediatrics* 2004;114:165-8.
13. Harding B, Risdon RA, Krous HF. Shaken baby syndrome. *BMJ* 2004; 328:720-1.
14. American Academy of Pediatrics Committee on Child Abuse and Neglect: Shaken baby syndrome: inflicted cerebral trauma. *Pediatrics* 1993;92:872-5.
15. Flodmark O. Stockholms läns landsting regionalt vårdprogram: Vid misstanke om fysisk misshandel av späda barn. Hämtad från: <http://bit.ly/294D4O4>, 16-06-28. 2011.
16. Socialstyrelsen. Barn som far illa eller riskerar att fara illa. ISBN: 978-91-7555-209-5. Hämtad från: http://www.socialstyrelsen.se/publikationer2014/2014-10-4_160817. 2014.
17. Västra Götalandsregionen. Regional riktlinje: Spädbarnsmisshandel. Hämtad från: <http://bit.ly/2bxdajq> 160817. 2014.
18. Bode-Janisch S, Bultmann E, Hartmann H, Schroeder G, Zajaczek JE, Debertin AS. Serious head injury in young children: birth trauma versus non-accidental head injury. *Forensic Sci Int* 2012;214:e34-8.
19. Gabaeff SC. Investigating the possibility and probability of perinatal subdural hematoma progressing to chronic subdural hematoma, with and without complications, in neonates, and its potential relationship to the misdiagnosis of abusive head trauma. *Leg Med (Tokyo)* 2013;15:177-92.
20. Geddes JF, Tasker RC, Adams GG, Whitwell HL. Violence is not necessary to produce subdural and retinal haemorrhage: a reply to Punt et al. *Pediatr Rehabil* 2004;7:261-5.
21. Geddes JF, Tasker RC, Hackshaw AK, Nickols CD, Adams GG, Whitwell HL, et al. Dural haemorrhage in non-traumatic infant deaths: does it explain the bleeding in 'shaken baby syndrome'? *Neuropathol Appl Neurobiol* 2003;29:14-22.
22. Kelly P, Hayman R, Shekerdemian LS, Reed P, Hope A, Gunn J, et al. Subdural hemorrhage and hypoxia in infants with congenital heart disease. *Pediatrics* 2014;134:e773-81.
23. Lantz PE, Couture DE. Fatal acute intracranial injury, subdural hematoma, and retinal hemorrhages caused by stairway fall. *J Forensic Sci* 2011;56:1648-53.
24. Miller R, Miller M. Overrepresentation of males in traumatic brain injury of infancy and in infants with macrocephaly: further evidence that questions the existence of shaken baby syndrome. *Am J Forensic Med Pathol* 2010;31: 165-73.
25. Guthkelch AN. Problems of infant retino-dural hemorrhage with minimal external injury. Hämtad från: <http://bit.ly/29b5qqn> 160629. *Houst J Health Law Policy* 2012;12.
26. Högsta domstolen. Dom. Mål nummer B 3438-12. 2014.
27. Christian CW, Block R; Committee on Child Abuse and Neglect; American Academy of Pediatrics. Abusive head trauma in infants and children. *Pediatrics* 2009;123:1409-11.
28. Squier W. The "Shaken Baby" syndrome: pathology and mechanisms. *Acta Neuropathol* 2011;122:519-42.
29. Geddes JF, Hackshaw AK, Vowles GH, Nickols CD, Whitwell HL. Neuropathology of inflicted head injury in children. I. Patterns of brain damage. *Brain* 2001;124:1290-8.
30. Feldman KW, Bethel R, Shugerman RP, Grossman DC, Grady MS, Ellenbogen RG. The cause of infant and toddler subdural hemorrhage: a prospective study. *Pediatrics* 2001;108:636-46.
31. Greiner MV, Richards TJ, Care MM, Leach JL. Prevalence of subdural collections in children with macrocrania. *AJNR Am J Neuroradiol* 2013;34:2373-8.
32. McKeag H, Christian CW, Rubin D, Daymont C, Pollock AN, Wood J. Subdural hemorrhage in pediatric patients with enlargement of the subarachnoid spaces. *J Neurosurg Pediatr* 2013;11:438-44.

33. Ravid S, Maytal J. External hydrocephalus: a probable cause for subdural hematoma in infancy. *Pediatr Neurol* 2003;28:139-41.
34. Drubach LA, Johnston PR, Newton AW, Perez-Rossello JM, Grant FD, Kleinman PK. Skeletal trauma in child abuse: detection with ¹⁸F-NaF PET. *Radiology* 2010;255:173-81.
35. Ghosh PS, Ghosh D. Subdural hematoma in infants without accidental or nonaccidental injury: benign external hydrocephalus, a risk factor. *Clin Pediatr (Phila)* 2011;50:897-903.
36. Hymel KP, Jenny C, Block RW. Intracranial hemorrhage and rebleeding in suspected victims of abusive head trauma: addressing the forensic controversies. *Child Maltreat* 2002;7:329-48.
37. Howard MA, Bell BA, Uttley D. The pathophysiology of infant subdural haematomas. *Br J Neurosurg* 1993;7:355-65.
38. Kivlin JD, Currie ML, Greenbaum VJ, Simons KB, Jentzen J. Retinal hemorrhages in children following fatal motor vehicle crashes: a case series. *Arch Ophthalmol* 2008;126:800-4.
39. Levin AV, Christian CW, Committee on Child A, Neglect SoO. The eye examination in the evaluation of child abuse. *Pediatrics* 2010;126:376-80.
40. Firsching R, Muller C, Pauli SU, Voellger B, Rohl FW, Behrens-Baumann W. Noninvasive assessment of intracranial pressure with venous ophthalmodynamometry. Clinical article. *J Neurosurg* 2011;115:371-4.
41. Lashutka MK, Chandra A, Murray HN, Phillips GS, Hiestand BC. The relationship of intraocular pressure to intracranial pressure. *Ann Emerg Med* 2004;43:585-91.
42. Laghmari M, Skiker H, Handor H, Mansouri B, Ouazzani Chahdi K, Lachkar R, et al. [Birth-related retinal hemorrhages in the newborn: incidence and relationship with maternal, obstetric and neonatal factors. Prospective study of 2,031 cases]. *J Fr Ophtalmol* 2014;37:313-9.
43. Friden HG, Ekstedt J. Volume/pressure relationship of the cerebrospinal space in humans. *Neurosurgery* 1983;13:351-66.
44. Shapiro K, Fried A. Pressure-volume relationships in shunt-dependent childhood hydrocephalus. The zone of pressure instability in children with acute deterioration. *J Neurosurg* 1986;64:390-6.
45. Robinson RJ, Bhuta S. Susceptibility-weighted imaging of the brain: current utility and potential applications. *J Neuroimaging* 2011;21:e189-204.
46. Schelhorn J, Gramsch C, Deuschl C, Quick HH, Nensa F, Moeninghoff C, et al. Intracranial hemorrhage detection over time using susceptibility-weighted magnetic resonance imaging. *Acta Radiol* 2015;56:1501-7.
47. Vinchon M, Noule N, Tchofo PJ, Soto-Ares G, Fourier C, Dhellemmes P. Imaging of head injuries in infants: temporal correlates and forensic implications for the diagnosis of child abuse. *J Neurosurg* 2004;101:44-52.
48. Bradford R, Choudhary AK, Dias MS. Serial neuroimaging in infants with abusive head trauma: timing abusive injuries. *J Neurosurg Pediatr* 2013;12:110-9.
49. Adamsbaum C, Morel B, Ducot B, Antoni G, Rey-Salmon C. Dating the abusive head trauma episode and perpetrator statements: key points for imaging. *Pediatr Radiol* 2014;44 Suppl 4:S578-88.
50. Vezina G. Assessment of the nature and age of subdural collections in nonaccidental head injury with CT and MRI. *Pediatr Radiol* 2009;39:586-90.
51. Mulvihill AO, Jones P, Tandon A, Fleck BW, Minns RA. An inter-observer and intra-observer study of a classification of RetCam images of retinal haemorrhages in children. *Br J Ophthalmol* 2011;95:99-104.
52. Tandon A, McIntyre S, Yu A, Stephens D, Leiby B, Croker S, et al. Retinal haemorrhage description tool. *Br J Ophthalmol* 2011;95:1719-22.

53. Levin AV. Retinal hemorrhage in abusive head trauma. *Pediatrics* 2010;126:961-70.
54. Wygnanski-Jaffe T, Levin AV, Shafiq A, Smith C, Enzenauer RW, Elder JE, et al. Postmortem orbital findings in shaken baby syndrome. *Am J Ophthalmol* 2006;142:233-40.
55. Zuccoli G, Panigrahy A, Haldipur A, Willaman D, Squires J, Wolford J, et al. Susceptibility weighted imaging depicts retinal hemorrhages in abusive head trauma. *Neuroradiology* 2013;55:889-93.
56. Hughes LA, May K, Talbot JF, Parsons MA. Incidence, distribution, and duration of birth-related retinal hemorrhages: a prospective study. *J AAPOS* 2006;10:102-6.
57. SBU. Assessment of methods in health care. Stockholm: Swedish Agency for Health Technology Assessment and Assessment of Social Services (SBU). http://www.sbu.se/globalassets/eng_metodboken_160808.pdf. 2016.
58. Parks S, Sugerma D, Xu L, Coronado V. Characteristics of non-fatal abusive head trauma among children in the USA, 2003–2008: application of the CDC operational case definition to national hospital inpatient data. *Inj Prev* 2012;18:392-8.
59. Vinchon M, de Foort-Dhellemmes S, Desurmont M, Delestret I. Confessed abuse versus witnessed accidents in infants: comparison of clinical, radiological, and ophthalmological data in corroborated cases. *Childs Nerv Syst* 2010;26:637-45.
60. Adamsbaum C, Grabar S, Mejean N, Rey-Salmon C. Abusive head trauma: judicial admissions highlight violent and repetitive shaking. *Pediatrics* 2010;126:546-55.
61. Bartschat S, Richter C, Stiller D, Banschak S. Long-term outcome in a case of shaken baby syndrome. *Med Sci Law* 2016;56:147-9. Epub 2015 Jun 8.
62. Bell E, Levin A, Shouldice M. A perpetrator confesses. *Child Abuse Negl* 2011;35:74-7.
63. Esernio-Jenssen D, Tai J, Kodsi S. Abusive head trauma in children: a comparison of male and female perpetrators. *Pediatrics* 2011;127:649-57.
64. Gertner N, Brower B, Shechtman P. Why the Innocent Plead Guilty: An Exchange. *New York Review of Books* 2015;January 8:23.
65. Kassin S, Drizin S, Grisso T, Gudjonsson G, Lee R, Redlich A. Police-induced confessions: Risk factors and recommendations. *Law Hum Behav* 2010;34:38.
66. Reece RM. What are we trying to measure? The problems of case ascertainment. *Am J Prev Med* 2008;34:S116-9.
67. Duhaime AC, Alario AJ, Lewander WJ, Schut L, Sutton LN, Seidl TS, et al. Head injury in very young children: mechanisms, injury types, and ophthalmologic findings in 100 hospitalized patients younger than 2 years of age. *Pediatrics* 1992;90:179-85.
68. Bhardwaj G, Chowdhury V, Jacobs MB, Moran KT, Martin FJ, Coroneo MT. A systematic review of the diagnostic accuracy of ocular signs in pediatric abusive head trauma. *Ophthalmology* 2010;117:983-992.e17.
69. Kemp AM, Jaspan T, Griffiths J, Stoodley N, Mann MK, Tempest V, et al. Neuroimaging: what neuroradiological features distinguish abusive from non-abusive head trauma? A systematic review. *Arch Dis Child* 2011;96:1103-12.
70. Maguire S, Pickerd N, Farewell D, Mann M, Tempest V, Kemp AM. Which clinical features distinguish inflicted from non-inflicted brain injury? A systematic review. *Arch Dis Child* 2009;94:860-7.
71. Maguire SA, Kemp AM, Lumb RC, Farewell DM. Estimating the probability of abusive head trauma: a pooled analysis. *Pediatrics* 2011;128:e550-64.
72. Maguire SA, Watts PO, Shaw AD, Holden S, Taylor RH, Watkins WJ, et al. Retinal haemorrhages and related findings in abusive and non-abusive head trauma: a systematic review. *Eye (Lond)* 2013;27:28-36.

73. Piteau SJ, Ward MG, Barrowman NJ, Plint AC. Clinical and radiographic characteristics associated with abusive and nonabusive head trauma: a systematic review. *Pediatrics* 2012;130: 315-23.
74. Togioka BM, Arnold MA, Bathurst MA, Ziegfeld SM, Nabaweesi R, Colombani PM, et al. Retinal hemorrhages and shaken baby syndrome: an evidence-based review. *J Emerg Med* 2009;37:98-106.
75. Hadley MN, Sonntag VK, ReKate HL, Murphy A. The infant whiplash-shake injury syndrome: a clinical and pathological study. *Neurosurgery* 1989;24: 536-40.
76. Rooks VJ, Eaton JP, Ruess L, Petermann GW, Keck-Wherley J, Pedersen RC. Prevalence and evolution of intracranial hemorrhage in asymptomatic term infants. *AJNR Am J Neuroradiol* 2008;29:1082-9.
77. Looney CB, Smith JK, Merck LH, Wolfe HM, Chescheir NC, Hamer RM, et al. Intracranial hemorrhage in asymptomatic neonates: prevalence on MR images and relationship to obstetric and neonatal risk factors. *Radiology* 2007;242:535-41.
78. Stray-Pedersen A, Omrand S, Nedregaard B, Klevberg S, Rognum TO. An infant with subdural hematoma and retinal hemorrhages: does von Willebrand disease explain the findings? *Forensic Sci Med Pathol* 2011;7:37-41.
79. De Leeuw M, Beuls E, Jorens P, Parizel P, Jacobs W. Delta-storage pool disease as a mimic of abusive head trauma in a 7-month-old baby: a case report. *J Forensic Leg Med* 2013;20:520-1.
80. Mansour AM, Jaroudi MO. Recurrent vitreous haemorrhage and epidural haematoma in a child with hypofibrinogenaemia. *BMJ Case Rep* 2012 Jul 9;2012.
81. Botte A, Mars A, Wibaut B, De Foort-Dhellemmes S, Vinchon M, Leclerc F. Two children with cerebral and retinal hemorrhages: do not diagnose shaken baby syndrome too rapidly. *Arch Pediatr* 2012;19:42-6.
82. Pinto FC, Porro FF, Sukanuma L, Fontes RB, de Andrade AF, Marino Jr R. Hemophilia and child abuse as possible causes of epidural hematoma: case report. *Arq Neuropsiquiatr* 2003;61:1023-5.
83. Ermis B, Ors R, Tastekin A, Orhan F. Severe congenital factor X deficiency with intracranial bleeding in two siblings. *Brain Dev* 2004;26:137-8.
84. Kato K, Kobayashi C, Katayama Y, Moriyama N, Shiono J, Kudo K, et al. Forty-two-day-old boy with acute idiopathic thrombocytopenic purpura. *Pediatr Int* 2010;52:485-7.
85. Carrim ZI, Arbabi EM, Long VW. Presumed non-accidental injury with retinal haemorrhages--findings from a tertiary referral centre in the United Kingdom. *Brain Inj* 2012;26:1716-22.
86. Guddat SS, Ehrlich E, Martin H, Tsokos M. Fatal spontaneous subdural bleeding due to neonatal giant cell hepatitis: a rare differential diagnosis of shaken baby syndrome. *Forensic Sci Med Pathol* 2011;7: 294-7.
87. Ambade VN, Malani AP, Kukde HG, Meshram RN. A rare case of head injury associated with Albers Schonberg disease. *J Forensic Leg Med* 2007;14:92-5.
88. Innis MD. Vitamin K deficiency disease. *J Orthomol Med* 2008;23:15-20.
89. Demiroren K, Yavuz H, Cam L. Intracranial hemorrhage due to vitamin K deficiency after the newborn period. *Pediatr Hematol Oncol* 2004;21: 585-92.
90. Brousseau TJ, Kissoon N, McIntosh B. Vitamin K deficiency mimicking child abuse. *J Emerg Med* 2005;29:283-8.
91. Visser DY, Jansen NJ, Ijland MM, De Koning TJ, Van Hasselt PM. Intracranial bleeding due to vitamin K deficiency: Advantages of using a pediatric intensive care registry. *J Intensive Care Med* 2011;37:1014-20.
92. Shemie S, Cutz E. Late hemorrhagic disease of the newborn: A fatal presentation of hepatobiliary disease masquerading as shaken baby syndrome. *J Intensive Care* 1995;10:315-8.

93. Wetzel RC, Slater AJ, Dover GJ. Fatal intramuscular bleeding misdiagnosed as suspected nonaccidental injury. *Pediatrics* 1995;95:771-773.
94. Nassogne MC, Sharrard M, Hertz-Pannier L, Armengaud D, Touati G, Delonlay-Debeney P, et al. Massive subdural haematomas in Menkes disease mimicking shaken baby syndrome. *Childs Nerv Syst* 2002;18:729-31.
95. Agrawal S, Peters MJ, Adams GG, Pierce CM. Prevalence of retinal hemorrhages in critically ill children. *Pediatrics* 2012;129:e1388-96.
96. Speidel B, Allbery S, Love T. MR of infant subdural hemorrhage: Incidence, etiology, and imaging differentiation. *Pediatr Radiol* 2015;45:137-8.
97. Salvatori MC, Lantz PE. Retinal haemorrhages associated with fatal paediatric infections. *Med Sci Law* 2015;55:121-8.
98. Adeleye AO, Shoshan Y, Cohen JE, Spektor S. Ruptured middle cerebral artery aneurysm in an infant presenting as acute subdural hematoma: a case report. *Pediatr Neurosurg* 2008;44:397-401.
99. Kemp AM, Joshi AH, Mann M, Tempest V, Liu A, Holden S, et al. What are the clinical and radiological characteristics of spinal injuries from physical abuse: a systematic review. *Arch Dis Child* 2010;95:355-60.
100. Wirtschafter J, Weissgold DJ, Budenz DL, Hood I, Rorke LB. Ruptured vascular malformation masquerading as battered/shaken baby syndrome: A nearly tragic mistake. *Surv Ophthalmol* 1995;39:509-12.
101. Reddy AR, Clarke M, Long VW. Unilateral retinal hemorrhages with subarachnoid hemorrhage in a 5-week-old infant: is this nonaccidental injury? *Eur J Ophthalmol* 2010;20:799-801.
102. Fledelius HC. Retinal haemorrhages in premature infants: a pathogenetic alternative diagnosis to child abuse. *Acta Ophthalmol Scand* 2005;83:424-7.
103. Abedzadeh-Kalahroudi M, Talebian A, Jahangiri M, Mesdaghinia E, Mohammadzadeh M. Incidence of neonatal Birth injuries and related factors in Kashan, Iran. *Arch Trauma Res* 2015;4:e22831.
104. Smith WL, Alexander RC, Judisch GF, Sato Y, Kao SC. Magnetic resonance imaging evaluation of neonates with retinal hemorrhages. *Pediatrics* 1992; 89:332-3.
105. Schoppe CH, Lantz PE. Are peripapillary intrascleral hemorrhages pathognomonic for abusive head trauma? *J Forensic Sci* 2013;58:228-31.
106. Galaznik JG. A case for an in utero etiology of chronic SDH/effusions of infancy. *J Perinatol* 2011;31:220-22.
107. McNeely PD, Atkinson JD, Saigal G, O'Gorman AM, Farmer JP. Subdural hematomas in infants with benign enlargement of the subarachnoid spaces are not pathognomonic for child abuse. *AJNR Am J Neuroradiol* 2006;27:1725-8.
108. Piatt JH, Jr. A pitfall in the diagnosis of child abuse: external hydrocephalus, subdural hematoma, and retinal hemorrhages. *Neurosurg Focus* 1999;7:e4.
109. Miller D, Barnes P, Miller M. The significance of macrocephaly or enlarging head circumference in infants with the triad: further evidence of mimics of shaken baby syndrome. *Am J Forensic Med Pathol* 2015;36:111-20.
110. Bishop FS, Liu JK, McCall TD, Brockmeyer DL. Glutaric aciduria type 1 presenting as bilateral subdural hematomas mimicking nonaccidental trauma. Case report and review of the literature. *J Neurosurg* 2007;106:222-6.
111. Hartley LM, Khwaja OS, Verity CM. Glutaric aciduria type 1 and nonaccidental head injury. *Pediatrics* 2001;107:174-5.
112. Juul K, Andersen J, Cvitanich VB, Lund AM. Case 2: Suspected nonaccidental injury – Quest for the diagnosis. *Acta Paediatrica, Int J Pediatr* 2006;95:1323-5.
113. Fitzgerald NE, MacClain KL. Imaging characteristics of hemophagocytic lymphohistiocytosis. *Pediatr Radiol* 2003;33:392-401.

114. Edwards RJ, Allport TD, Stoodley NG, O'Callaghan F, Lock RJ, Carter MR, et al. External hydrocephalus and subdural bleeding in infancy associated with transplacental anti-Ro antibodies. *Arch Dis Child* 2012;97:316-9.
115. Ganesh A, Jenny C, Geyer J, Shouldice M, Levin AV. Retinal hemorrhages in type I osteogenesis imperfecta after minor trauma. *Ophthalmology* 2004;111:1428-31.
116. Paterson CR, Monk EA. Temporary brittle bone disease: association with intracranial bleeding. *J Pediatr Endocrinol Metab* 2013;26:417-26.
117. Odom A, Christ E, Kerr N, Byrd K, Cochran J, Barr F, et al. Prevalence of retinal hemorrhages in pediatric patients after in-hospital cardiopulmonary resuscitation: a prospective study. *Pediatrics* 1997;99:E3.
118. Pham H, Enzenauer RW, Elder JE, Levin AV. Retinal hemorrhage after cardiopulmonary resuscitation with chest compressions. *Am J Forensic Med Pathol* 2013;34:122-4.
119. Barnes PD, Galaznik J, Gardner H, Shuman M. Infant acute life-threatening event – dysphagic choking versus non-accidental injury. *Semin Pediatr Neurol* 2010;17:7-11.
120. Korkmaz A, Yigit S, Firat M, Oran O. Cranial MRI in neonatal hypernatraemic dehydration. *Pediatr Radiol* 2000;30:323-5.
121. Innis MD. Vaccines, apparent life-threatening events, Barlow's disease, and questions about "shaken baby syndrome". *Journal of American Physicians & Surgeons* 2006;11:17-9.
122. Margulies S, Prange M, Myers BS, Maltese MR, Ji S, Ning X, et al. Shaken baby syndrome: a flawed biomechanical analysis. *Forensic Sci Int* 2006;164:278-9; author reply 282-3.
123. Cory CZ, Jones BM. Can shaking alone cause fatal brain injury? A biomechanical assessment of the Duhaime shaken baby syndrome model. *Med Sci Law* 2003;43:317-33.

Appendix 1

Other possible causes of the components of the triad

In determining the clinical diagnosis, differential diagnoses are considered. In cases where a child presents with symptoms and signs suggesting brain damage, further investigation is required. This report has therefore taken note of differential diagnoses, disclosed in the database searches, which offer alternative explanations for the various symptoms and signs of the triad, either separately or as the complete triad. These articles are usually in the form of case reports of isolated patients without a reference group and have therefore not been included in the quality assessment. However, the project group considered that it would still be of interest to present these potential alternative explanations for the triad.

Table 1
Other possible causes
(differential diagnosis)
of the triad and its
components.

	Disease/condition	Reported findings from the triad	Reference number (number of cases, or cases/study population size) Reported finding from the triad
Diseases or conditions causing hemorrhagic symptoms	von Willebrand's disease	SDH, RH	[78] (1)
	Delta storage pool disease	SDH, BE, RH	[79] (1)
	Hyperfibrinogenemia	RH (including vitreous hemorrhage)	[80] (1)
	Hemophilia A	SDH/RH	[81] (2) ^{RH} [82] (1) ^{SDH}
	Factor X deficiency	SDH	[83] (2)
	Idiopathic thrombocytopenic purpura	ICH	[84] (1)
	Kasabach-Merrit syndrome thrombocytopenia	RH	[85] (1)
	Hepatitis	RH, BE, SDH	[86] (1)
	Albers-Schönberg disease	SDH	[87] (1)
	Vitamin K deficiency	SDH (ICH)/BE/RH	[88] (3) ^{SDH} [89] (17) ^{SDH} [90] (1) ^{SDH} [91] (16) ^{SDH} [92] (1) ^{SDH, BE, RH} [93] (1) ^{SDH, BE}
	Menkes disease (Copper deficiency)	SDH	[94] (1)
	Unspecified	RH, SDH	[95] (1) [96] (3)
	Infections	Infection	RH
Infection with or without hypoxia		SDH (intradural bleeding)	[21] (10/30)
Vascular malformations	Aneurysm, Arterio-venous malformation	SDH (SAH)/BE/RH	[98] (1) ^{SDH} [99] (1) ^{SDH, BE} [100] (1) ^{SDH, BE} [101] (1) ^{SDH, RH}
Prenatal and birth-related injuries	Prematurity	RH	[102] (11)
	Delivery injury	SDH (ICH)/RH	[18] (2) ^{SDH} [103] (3) ^{ICH} [96] (3) ^{SDH} [56] (53) ^{RH} [104] (10) ^{RH}
	Normal delivery (or prenatal)	SDH/RH	[77] (17/97) ^{SDH} [39] (94/252) ^{RH} [76] (32/63) ^{SDH}
	Prenatal trauma	RH	[105] (2)
	Congenital SDH	SDH	[106] (1)
	Congenital heart disease	SDH	[22] (66/152)

The table continues on the next page

Table 1
continued

	Disease/condition	Reported findings from the triad	Reference number (number of cases, or cases/study population size) Reported finding from the triad
Large head size	Enlarged SA space/ External hydrocephalus/ Benign enlargement of the subarachnoid spaces	SDH	[31] (6/108) [32] (4/177) [107] (7) [33] (3) [96] (6)
	External hydrocephalus	RH, SDH	[108] (1) [109] (6)
Metabolic diseases	Glutamic aciduria	SDH/RH	[110] (1) ^{SDH} [111] (1) ^{SDH} [112] (1) ^{SDH, RH}
Immunological diseases	Hemo-phagocytic lymphohistiocytosis	SDH, SAH (ICH), BE, RH	[113] (1)
	Transplacental acquisition of anti-Ro antibodies	SDH	[114] (2)
Skeletal diseases	Osteogenesis imperfecta	SDH, RH	[115] (3)
	Brittle bone disease	SDH/RH	[116] (20/20) ^{SDH} (11/20) ^{RH}
Other	Hypoxia and resuscitation	RH	[117] (1/33) [118] (1)
	Hypoxia	SDH/intradural bleeding	[21] (20/30)
	Choking and resuscitation	SDH (SAH), RH	[119] (1)
	Resuscitation in patients with retinopathy of prematurity	RH	[118] (2) [117] (1/33)
	Hypnatremia and dehydration	ICH, BE	[120] (1)
	Leukemia	RH	[95] (3)
	Vaccine-induced vitamin C deficiency	SDH, BE	[121] (2)

BE = Brain edema; **ICH** = Intracranial hemorrhage; **RH** = Retinal hemorrhage;
SAH = Subarachnoid hemorrhage; **SDH** = Subdural hematoma

Appendix 2

Biomechanical studies

In order to study the biomechanisms underlying injuries due to traumatic shaking and related questions, various physical and virtual models have been created to simulate shaking of an infant, with the aim of analysing certain effects of shaking. Thirty scientific articles on biomechanics which were identified in the literature search were studied more closely. Several review articles present a good understanding of the field and also understanding of general traumatic brain injuries independent of age. Most of the articles present experiments using models and simulations of biomechanical forces and many also present preliminary data, but few of the experiments have been repeated. The results presented in the articles are very diverse, from case reports to construction of various models intended to explain the mechanisms involved in traumatic shaking. A few articles comprise comments about another article. One such example pointed out that the authors of a previous article had made a 10-fold error in calculations as to whether or not an injury could occur [122]. Some studies present clearly contradictory results. One example of this is presented below.

Duhaime et al. present a biomechanical model for traumatic shaking [5]. The work is regarded as a reference article and has served as the basis of many other experiments and the method has been further developed. The article concludes that it is not possible to achieve damaging effects by shaking.

Cory och Jones' article [123] is based on a biomechanical model modified from Duhaime's [5]. This article shows that Duhaime's model is flawed and the results show that the forces generated by shaking of a child can in many cases exceed the minimum forces needed to cause injury.

Thus the scientific basis of these studies is divergent and no definite conclusions can be drawn with respect to the minimum forces required to result in injury.

Appendix 3

Ethical analysis of traumatic shaking¹

The Swedish National Council on Medical Ethics has conducted an ethical analysis of ‘traumatic shaking’ in connection with the Swedish Agency for Health Technology Assessment and Assessment of Social Services’ report on the subject.

The Council has not undertaken any scientific assessment of the basic material. As such, the analysis is based on the Agency’s results as presented in the report, which can be summarised as follows:

- “There is insufficient scientific evidence on which to assess the diagnostic accuracy of the triad in identifying traumatic shaking (very low quality evidence).”
- “There is limited scientific evidence that the triad and therefore its components can be associated with traumatic shaking (low quality evidence).”
- “The triad or its components can be attributable to causes other than shaking.”

¹ This is an unofficial translation of an ethical analysis performed by The Swedish National Council on Medical Ethics (Smer). In the translation process, some linguistic nuances may have been lost. To comply with the SBU terminology, Smer is using the term ‘traumatic shaking’ in this translation.

Outline

The analysis begins with an analysis of the term traumatic shaking. The rest of the ethical analysis is structured according to two dimensions. The first identifies the parties with an interest in the issue, and the second identifies the ethical values that come into play in connection with a possible shaken baby situation. Finally, the various crucial values are weighed up and the value conflicts that can arise in this process are considered.

Conceptual problems

The term ‘traumatic shaking’ has been used in cases when the triad of subdural bleeding, retinal bleeding and various forms of brain injury are found in an infant. The Swedish Agency for Health Technology Assessment and Assessment of Social Services’ review of the scientific literature found limited scientific evidence that the triad and therefore its components may occur due to traumatic shaking, but it was also found that the triad or its constituent parts may also be due to causes other than shaking.

According to the Swedish Agency for Health Technology Assessment and Assessment of Social Services’ report, there is insufficient scientific evidence “to assess the diagnostic accuracy of the triad in identifying traumatic shaking”.

The doctor dealing with the family of the child with the triad may also have sources of information available other than those offered by medical imaging, neurological examinations and retinal examinations. There may be other injuries to the body that support the suspicion of abuse, or observations made in discussions with the custodial parents. It is an ethical requirement that all of this is considered in the doctor’s assessment before any concerns are reported to the social welfare committee.

The doctor has a duty to precisely describe everything that has emerged in the examination, both injuries that have emerged and the information that the custodial parents provide concerning the course of events and any other circumstances. It is also essential that all injuries are documented meticulously, both for medical professionals’ use and in case of future legal proceedings.

Every decision made by medical professionals, whether diagnostic or therapeutic in nature, is based on both fact and values. In this context, ‘fact’ refers to a description of all relevant findings made through physical, radiological, laboratory-based and other medical examinations of the child. However, it should be borne in mind that ‘fact’ may also include judgments, e.g. assessments of medical imaging findings. The next stage in the doctor’s work is to evaluate the medical findings and the substance of the custodial parents’ account of events. This is a different kind of task to the factual description. Here, the doctor has an important ethical responsibility to ensure that assessments are based only on science and tried and tested experience.

Parties

The starting point for the analysis is a scenario in which an infant, accompanied by one or two custodial parents, arrives at a health care facility with injuries that give rise to a clinical suspicion that abuse may be a cause of the child's injuries. If the child's injuries include the triad of symptoms and findings, the question of an eventual traumatic shaking arises. Already at this stage, there are several parties with a legitimate interest in how the situation is handled. These are the child, its custodial parents and various health care professionals. Where applicable, the child's siblings may also be affected by the process. At a later stage, the situation may also involve social services staff and political officials (e.g. in the social welfare committee), as well as police, prosecutors and the judicial authorities at various levels.

Values

The child has a unique status in this situation due to various considerations based on ethical values. In this context we are talking about very young children. This means that the child itself is entirely incapable of explaining what has happened, and therefore cannot, for obvious reasons, safeguard its own interests. The injuries in question in situations in which traumatic shaking is suspected may be serious in nature, both in acute terms and also in the longer term. The injuries may be life-threatening, or entail a risk of permanent consequences in terms of the child's development, health and future quality of life.

For these reasons, an ethical analysis of traumatic shaking should be primarily based on a child perspective. The key ethical question is how the child's interests can best be safeguarded, as it can never be acceptable that a young child is subjected to abuse.

It is an ethical duty that the young, unprotected child's interests are safeguarded by somebody else. It would normally be the duty of the child's custodial parents to safeguard its interests. In a situation in which traumatic shaking is suspected, however, it is often one (or both) of the custodial parents who may have caused the injuries. This means that they may not have discharged their parental responsibilities.

In the scenario outlined here, the immediate responsibility for safeguarding the child's crucial values falls to the medical professionals dealing with the family at the hospital. In such situations, staff must act based on their professional ethics and applicable legislation.

The first step may be taking vital, acute medical measures required by the child's state of health. All necessary medical measures must be taken to remedy and alleviate the child's acute injuries and prevent future after-effects. Naturally, this is the top priority in handling the case.

If the suspicion arises that the injuries may have occurred due to violence, it is the doctor's duty to investigate this suspicion on the basis of science and tried and tested experience. It is also the doctor's duty under Chapter 14, Section 1 of the Social Services Act (2001:453) to report to the social welfare committee any suspicions of risk of harm to the child.

Society has an explicit responsibility to protect children in a number of respects. This is clear from various laws, including the Social Services Act and the Care of Young Persons Act (1990:52). The former provides opportunities for society to intervene in consultation with the custodial parents, while the latter provides opportunities for society to take measures to protect the child without the custodial parents' consent. As a last resort, the social welfare committee can take the child into care outside the home.

The Convention on the Rights of the Child, which is currently being incorporated into Swedish law, outlines a number of fundamental rights enjoyed by all children, including the right to protection of their life and health, the right to grow up in good conditions and the right to good care. The Convention was drawn up based on a rights perspective, but it also rests on central ethical principles of adult society's responsibility for children's life situation, to protect what are crucial values for all children.

When a doctor asks a child's custodial parents whether the child's injuries may have been caused by some external event of which they are aware, it is uncommon for them to admit it immediately (Lowenstein 2004). In this situation, it is important that the doctor does not take on the judicial system's role of determining whether an offence has taken place or accusing a particular individual. The custodial parent(s) has/have a legitimate interest in ensuring that certain values that are crucial to them are considered in the situation. These include the right to good care, which the custodial parents are generally anxious to ensure regardless of the cause or any intent (Leuthner 2001). Moreover, it is an important value for them that they are listened to adequately and that the hospital's handling of the situation has an impartial and unbiased starting point with respect to all conceivable causes of the injuries observed.

For medical staff, it is a crucial value to be met with respect for their professional duties from both a medical and an ethical perspective. It is usually the doctor who is responsible for assessing the likelihood that the injuries observed in the child may have been caused by an adult, usually one of the custodial parents, and thus could be a sign of traumatic shaking. For the doctor, it is of considerable value to be allowed space to consider the decision of whether or not to report any concerns. A decision to report is associated with considerable consequences for both the child and the custodial parents, and must therefore be well-founded and well-considered. Such a decision should always be taken in consultation with at least one other doctor.

The doctor also has an interest in having sufficient training and expertise in the area of child abuse to be able to handle these ethically and psychologically very difficult situations in a professional manner.

Social services have a radically different division of responsibility compared to medical professionals. The decision-making mandate for measures without custodial parents' consent rests with the political officials in the social welfare committee, represented in urgent situations by their delegated chair. The basis for the decision is, however, produced by social services staff. They have professional ethical rules for their work that must be taken into account in situations of this nature. For social services staff it is a crucial value to safeguard the child's interests and protect the child from threats to its life, health and development. It is a crucial value for social services that the information that they receive from medical professionals is medically correct, well-founded and formulated in such a way that conclusions about the cause of injuries observed are not reported without a solid basis.

If the case – immediately or at a later stage – is subsequently transferred to police, prosecutors and courts, those authorities will have a similar interest with respect to information from medical professionals. If and when a case comes to court, it is important for the court to have access to scientific expertise to express an opinion in accordance with the professional ethical principles and applicable legal rules concerning certificates and opinions.

Value conflicts

There are several significant value conflicts with respect to traumatic shaking. One of the most important concerns whose interests should take precedence – the child's or the custodial parents'. From a child perspective there cannot be any doubt that the child's interests have the highest priority in several respects. Firstly, the child needs to have its injuries examined and treated professionally and competently in a medical setting. If it is suspected that the injuries may have been caused by abuse, there is an additional obvious need for protection of the child's life and health.

On the other hand, the custodial parent(s) suspected of shaking a child has/have a legitimate interest in not being condemned when innocent. Here we see a potential value conflict that can be described as an ethical dilemma in the sense that there is no entirely problem-free solution.

This dilemma can also be expressed in terms of the risks of underdiagnosis and overdiagnosis. Underdiagnosis refers to children who really have been subjected to shaking not being identified and thus not receiving society's protection against further abuse or growing up in conditions that are otherwise inadequate. Such underdiagnosis may occur due to a lack of competence or vigilance among medical professionals, or a lack of willingness or ability to investigate suspicions of traumatic shaking in a professional manner.

Overdiagnosis may occur if doctors who encounter children presenting the diagnostic triad immediately assess this as evidence that shaking and shaking alone is the cause of the injuries observed. This is thus a matter of confusing a hypothesis of a possible cause for a child's injuries with a claim of certain

knowledge that there is such an unambiguous and certain link between cause and effect.

This process thus creates a risk that the continued treatment in such a case will mainly be characterised by a 'validation strategy' (Melzer et al, 2013). This means that further measures are taken purely to confirm the hypothesis, and that insufficient account is taken of information that could disprove the hypothesis.

Both under- and overdiagnosis are extremely problematic from an ethical point of view. Overdiagnosis protects many children, both those in whom traumatic shaking is established as cause and a number of others. Nonetheless, it leads to families being split up, some of them on false premises. Separating children from their custodial parents is a serious intervention that should only be implemented when a child runs a clear risk of abuse at home. The fact that other children in the family may be taken into care may further exacerbates the situation.

The value conflict outlined above between the interests of the child and the custodial parent(s) needs to be related to the legal principle that no innocent person should be convicted of a crime. Overdiagnosis of traumatic shaking results in a number of children being protected, some of whom really are victims of such shaking, but this is at the expense of a number of custodial parents being deprived of their liberty without having committed an offence. However, underdiagnosis of traumatic shaking leads to children who are being mistreated remaining in a harmful home environment, at risk of future acts of violence.

The medical controversy that has surrounded traumatic shaking in Sweden and around the world is largely about whether there is established scientific support for the claim that the symptomatic triad of subdural bleeding, retinal bleeding and brain injury is caused by shaking and shaking alone. The Swedish Agency for Health Technology Assessment and Assessment of Social Services' report shows that there is scientific evidence – albeit limited – for the idea that the triad may be caused by shaking, but that there are other illnesses and events that can cause the triad or its constituent parts.

This raises the question of when doctors can and should express an opinion when it comes to traumatic shaking. Ethically, it is particularly important that doctors and other medical professionals are observant with respect to injuries in young children that could conceivably have been inflicted by human hands, even if the custodial parents deny anything of the sort. The clinical examination and treatment of injuries must be entirely robust. The question is whether a doctor can express an opinion about the cause of the observed injuries with scientific certainty at a later stage. The doctor has, as previously outlined, a range of different information to take into account when assessing the possible causes of the injuries. To state on the basis of the mere existence of the triad that it was definitely caused by shaking must, however, be considered incompatible with both doctors' professional ethics and the regulations concerning legal certificates (Albert et al, 2012).

This observation does not mean that there cannot be grounds to report concerns in spite of this uncertainty, as the child's need for protection is a broader issue than the question of the cause of the injuries.

Conclusions

The Swedish National Council on Medical Ethics has based this ethical analysis on the observation in the Swedish Agency for Health Technology Assessment and Assessment of Social Services' report that scientific evidence concerning traumatic shaking is limited. There is limited scientific evidence that the 'triad' of symptoms or its constituent parts may occur due to shaking, but the report states that there are differential diagnoses that can also cause the three symptoms/findings in the triad.

Given this observation, the term 'shaken baby syndrome' itself is ethically problematic, as it encompasses an aetiological observation. The Council considers that it is ethically problematic for medical professionals to establish with certainty that certain specific injuries in infants are automatically evidence that they were caused by shaking. Such overdiagnosis of traumatic shaking should not occur when the state of scientific knowledge is so limited (Riggs & Hobbs, 2011).

The Council also considers that underdiagnosis is ethically problematic, in the sense that it means that children who really have been subjected to shaking are not identified and examined by medical professionals. This risk can, however, be limited through improved professional training on child abuse in general and traumatic shaking in particular, within both health care services and social services.

The Council would like to emphasise the importance of medical professionals observing their duty to report to the social welfare committee cases in which it is suspected that children have been mistreated in any way. This is particularly applicable in cases where any kind of child abuse is suspected. Medical professionals must be able to combine high vigilance of suspected traumatic shaking with caution with respect to expressing an opinion on the cause of the injuries observed, since the state of scientific knowledge does not permit any clear conclusions in this area.

References

Albert DM, Weisberger Blanchard J & Knox BL. Ensuring appropriate expert testimony for cases involving the 'shaken baby'. *JAMA* 2012; 308: 39-40.

Leuthner SR. Ethical challenges in the care of the shaken baby. *Journal of Aggression, Maltreatment & Trauma* 2001; 5: 341-347.

Lowenstein LF. Recent research and views on shaking baby syndrome. *International Journal of Psychiatry in Medicine* 2004; 34: 131-141.

Meltzer CC, Sze G, Rommelfanger KS, Kinlaw K, Banja JD & Wolpe PR. Guidelines for the ethical use of neuroimages in medical testimony: report of a multidisciplinary conference. *American Journal of Neuroradiology* 2014; 35: 632-637.

Riggs JE & Hobbs GR. Infant homicide and accidental death in the United States, 1940–2005: ethics and epidemiological classification. *Journal of Medical Ethics* 2011; 37: 445-448.

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On behalf of the Council,

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