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Insufficient evidence for ‘shaken baby syndrome’ - a systematic review.

Short title: The triad in “Shaken baby syndrome”

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ABSTRACT

Shaken baby syndrome has typically been associated with findings of subdural haematoma, retinal haemorrhages and encephalopathy, which are referred to as the triad. However, during the last decade, the certainty with which the triad can indicate that an infant has been violently shaken has been increasingly questioned. The aim of this study was to determine the diagnostic accuracy of the triad in detecting that an infant had been shaken.

The literature search was performed using PubMed, Embase and the Cochrane Library up to October 15, 2015. Relevant publications were assessed for the risk of bias using the QUADAS tool and were classified as having a low, moderate or high risk of bias according to predefined criteria. The reference standards were confessions or witnessed cases of shaking or accidents. The search generated 3,773 abstracts, 1,064 were assessed as possibly relevant and read as full texts and 30 studies were ultimately included. Of these, 28 were assessed as having a high risk of bias, which was associated with methodological shortcomings as well as circular reasoning whenclassifying shaken baby cases and
controls. The two studies with a moderate risk of bias used confessions and convictions when classifying shaken baby cases, but their different designs made a meta-analysis impossible. None of the studies had a low risk of bias. Conclusion. The systematic review indicate that there is insufficient scientific evidence on which to assess the diagnostic accuracy of the triad in identifying traumatic shaking (very low quality evidence). It was also demonstrated that there is limited scientific evidence that the triad and therefore its components can be associated with traumatic shaking (low quality evidence).

Key notes

- Shaken baby syndrome has typically been associated with findings of subdural haematoma, retinal haemorrhages and encephalopathy.
- However, the diagnostic accuracy of this triad in detecting that an infant has been shaken has been questioned.
- This systematic review indicate that there is insufficient scientific evidence on which to assess the diagnostic accuracy of the triad in identifying traumatic shaking (very low quality evidence).

INTRODUCTION

Rationale

The effects of abusively shaking an infant were first suggested by the American neurologist Norman Guthkelch in 1971. Based on a few cases, he introduced the hypothesis that shaking a baby backwards and forwards in a whiplash-like manner might cause certain symptoms and signs, namely subdural haematoma, retinal haemorrhages and encephalopathy, which were eventually referred to as the triad (1). These symptoms and signs could occur without visible signs of impact to the head and were associated to isolated
violent shaking. An inverse version of the hypothesis was also eventually derived: if the triad was identified and no other “acceptable” explanation was provided, the infant had been violently shaken (2).

During the last 40 or so years, a number of studies have been conducted on “shaken baby syndrome”, which is currently a subset of more general labels such as abusive head trauma, non-accidental head injury and similar terms (2, Box 1). It has been maintained by paediatricians and child protection teams (CPTs) that there is a scientifically robust body of knowledge supporting the general assumption that when the triad is observed, the infant has been violently shaken (3, 4). The criteria used to identify shaken baby cases (5) have also been used in criminal trials in order to prosecute and convict suspected perpetrators with the help of expert testimony. If the criteria are not reliable, however, this might result in either under-diagnosis or over-diagnosis, and the classification of shaken baby cases in scientific studies might be mistaken. Under-diagnosis is linked to an increased risk that the infant is not protected sufficiently since he or she is not separated from the perpetrator, while over-diagnosis might carry an increased risk of unjustly separating a family and prosecuting and convicting an innocent parent or guardian. Hence, robust and evidence-based knowledge about the effects of shaking an infant has important medical and societal consequences for the concerned infant, the family, the general public’s trust in the medico-legal system and science in general.

Over the last decade, questions about the validity of the allegedly strong link between the triad and traumatic shaking have successively increased (6-8). Norman Guthkelch, and others, have questioned the manner in which his own original hypothesis, as well as the subsequent inverse version of the hypothesis, became dogma and has claimed that the evidence on which the hypothesis was based is poor (9).
Objectives

The main objective of this systematic review was to determine the diagnostic accuracy of the triad in detecting that an infant had been violently shaken.

METHODS

Protocol and registration

This systematic review was conducted at the Swedish Agency for Health Technology Assessment and Assessment of Social Services and published in Swedish in October 2016 as a report at www.sbu.se/2016. The Agency used a peer-reviewed protocol, including pre-specified objectives in accordance with standards in health and technology assessments. For the used terms traumatic shaking and “shaken baby syndrome”, see Box 1.

Since the present study is based on a literature review no patients or participants were involved.

Eligibility criteria

The eligibility criteria were as follows. The population was infants of 12 month or under 12 months of age and the index test was the presence of the triad in suspected traumatic shaking. The gold standard reference test was either that someone had confessed to shaking a baby, or other documented trauma, and the outcome was diagnostic accuracy.

Case-control and cohort studies with fewer than 10 individuals were excluded to minimize the risk of selection bias. For possible differential diagnoses, also studies of single cases could challenge the hypothesis that the triad always is caused by traumatic shaking. Studies of differential diagnoses were not assessed regarding quality, and were consequently not a
basis for the results. Studies including children older than 12 months of age, or with signs of impact to the head, were included only if a subgroup of 12 month or under 12 months of age, and/or a subset of isolated shaking, was identified.

**Information sources and search terms**

The electronic literature search was performed by an information specialist and included PubMed, Embase and the Cochrane Library up to 15 October 2015. A complementary manual search was conducted among the references in literature reviews and publications not identified in the main search. Studies published in English, German, French, Swedish, Norwegian and Danish were included. Grey literature, such as conference abstracts or dissertations, was not included.

The search terms included, but were not restricted to, infant, subdural haematoma, retinal haemorrhage, cerebral edema, encephalopathy, accidental and non-accidental injury, shaken baby and shaken baby syndrome (2).

**Study selection**

Six reviewers were engaged in the process and were split into three groups of two reviewers. They independently screened the titles and abstracts identified through the search strategy. The full texts of all studies of potential relevance according to the inclusion criteria were obtained and each group of two reviewers assessed one-third of them for inclusion. Any disagreement was resolved by discussion until a consensus was reached.

**Data collection process and data items**

Information concerning the study design, population and results were extracted from the included papers with a low or moderate risk of bias.
Risk of bias in individual studies

Two reviewers independently assessed the risk of bias in individual studies using a modified version of the QUADAS tool (10). Each study was rated as having a low, moderate or high risk of bias. The judgement of the risk of bias focused on the risk of systematic errors due to methodological flaws, including circular reasoning in the classification of shaken baby cases and controls. Systematic literature reviews were assessed using the AMSTAR instrument (11).

Studies were assessed as having a low risk of bias when the study cases, namely shaken babies, were unequivocally confirmed as having been violently shaken and when the shaking preceded the symptoms associated with the triad, for example by a video recording or independent witness information. Furthermore, the control cases needed to have been age-matched and unequivocally subjected to other defined types of trauma.

Studies were assessed as having a moderate risk of bias when the shaken baby study cases were identified as the result of a detailed confession by the suspected perpetrator and/or when there were shortcomings regarding controls, for examples no age match or even a lack of controls. Each individual study underwent an overall assessment with regard to the significance of such shortcomings.

Studies were assessed as having a high risk of bias when additional deficiencies were present and it was judged that the results could not provide reliable information in response to the questions addressed in this systematic review, for example insufficient definition of the study cases and circular reasoning.

Risk of circular reasoning

In many studies, the authors referred to a child protection team (CPT) when classifying shaken baby cases and controls. The CPT and concerned paediatricians often took for granted that if the triad was present, and no other acceptable explanations were provided,
that the infant had been violently shaken (12). The criteria for what was considered an
“acceptable” explanation had also been developed, and if these criteria were not fulfilled, a
case was classified, by default, as a shaken baby case (Table 1). The research question for
the present study concerned the certainty of the conclusion that an infant had been violently
shaken when the triad was observed. But if what was going to be examined had already
been taken for granted by those who were classifying the cases, the result was judged to
have been based on circular reasoning. In order to avoid circular reasoning, only studies in
which someone had confessed to shaking the child were included.

Method of analysis
Since sensitivity and specificity were not presented, or could not be calculated from the
included studies, it was not possible to conduct a meta-analysis.

RESULTS

Study selection
The literature search generated 3,773 records, of which 1,064 were original papers of
potential relevance and were read as full texts. Of these, 1,034 did not fulfil the inclusion
criteria and were subsequently excluded, resulting in 30 included papers. Of these, 28 were
assessed as having a high risk of bias (13-40), two as having a moderate risk (41, 42) and
none as having a low risk (Fig 1).

The assessed systematic literature reviews were all ranked as being of low quality (43-49).

Study characteristics and risk of bias within studies
The strength of the two included studies with a moderate risk of bias (41, 42) - one
retrospective and one prospective - was the fact that their study groups were based on
confessions. One study provided detailed information about the shaking event in 14 of 29
cases (41), while the control group in the other study entailed witnessed accidents in public

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One methodological weakness of both the included studies was the risk of false confessions, but there were also other methodological flaws.

**Results of individual studies**

In the retrospective case-control study (41), the group of confessed shaking cases was compared to a group containing people who were suspected of, but denied, having shaking the infant. In the confessed shaking group, 13 of the 29 cases were allegedly injured through isolated shaking and detailed information about the shaking event was provided in 14 of the 29 cases. A similar specification was not provided in the denial group, which comprised 82 cases. The authors found no statistically significant differences between the cases in the two groups, with regard to their age, sex, mortality, symptoms, etc.

In the prospective study (42), the authors compared a group of infants in which someone had confessed to and/or, been convicted of having shaken the baby (n=45) with a group in which the infants had been exposed to an accident that was witnessed in a public area (n=39). The authors stated that: “Information on the confessions was obtained by a forensic paediatrician from judicial sources during expertise or after the judicial hearings were made public” (42). No detailed information was provided regarding what had been confessed or under what circumstances the confessions had been obtained. The authors used a triad that comprised subdural haematoma, diffuse retinal haemorrhages, and an absence of scalp swelling. For the applied triad, the authors reported a sensitivity of 0.244, a specificity and positive predictive value of 1.0 and a negative predictive value of 0.534.

Different conditions or events that might have caused the triad or its components included accidental trauma, such as a fall or motor vehicle accident, sequelae of normal delivery, prematurity, macrocephaly and external hydrocephalus, coagulopathies, infections, metabolic diseases, leukaemia, immunological conditions, vascular malformations in the brain, and asphyxia (2).
DISCUSSION

Summary and evidence
The main finding was that 28 of the 30 included studies were assessed as having a high risk of bias, while two had a moderate risk and none had a low risk. There were two main indications of a high risk of bias: methodological flaws and circular reasoning when classifying shaken baby cases and controls. Two conclusions were drawn. The first was that there is insufficient scientific evidence on which to assess the diagnostic accuracy of the triad in identifying traumatic shaking (very low quality evidence according to GRADE measure (50). The second was that there is limited scientific evidence that the triad and therefore its components can be associated with traumatic shaking (low quality evidence according to GRADE (50).

Limitations of the studies identified
The included studies were observational and many of them used comparison groups and were performed as retrospective case-control studies extracted from medical records or registers. Some studies were designed as prospective cohort studies. Apart from the usual methodological bias issues associated with retrospective case-control studies, other issues were also observed. In most studies, the average age of the control group was significantly older than that of the shaken baby group, particularly in accidental falls (51). Furthermore, the radiological and ophthalmological examinations were rarely blinded and, when they were, a poor or moderate inter-rater agreement was reported (52).

The criteria for classifying study cases and controls varied. Sometimes the composition of the comparison group was explicitly presented, whereas sometimes there was simply a deferral to the judgement of a CPT. Sometimes the criteria for shaken baby cases were
related to controversies concerning the height of a fall. If the fall was below a certain height, for example 1 metre, the case was classified as a shaken baby, but if it was above 1 metre it was classified as a control (see Table 1). Such classifications were applied, despite the fact that several studies have shown that a minor fall could have caused the triad, particularly in cases of increased head circumference due to macrocephaly - benign enlargement of the subarachnoid space in infancy (53-56) - or long-term sequelae of a chronic subdural haematoma after an uncomplicated vaginal delivery (57-60). Such classification criteria resulted in uncertainty as to whether the groups of shaken babies also included accidental injury cases and whether the control groups also contained shaken babies.

The other main reason for the low quality was the issue of circular reasoning linked to the classification criteria. As illustrated in Table 1, in many cases the applied criteria focused more on the suspect’s trustworthiness than on scientifically-based criteria.

The two studies of moderate quality

The two studies of moderate quality included samples in which a person had confessed to and/or been convicted of having shaken an infant (41, 42).

In one study (41), those who had confessed provided detailed information about the shaking event in approximately half of the cases. No significant difference was found between the two groups of those who had confessed and those who had not. Three interpretations seem plausible: either the group who confessed to a shaking event included false confessions, or the group who denied a shaking event actually included shaken babies, or both. The circumstances under which a confession was obtained might have involved false confessions, because of police pressure, or be the result of plea-bargaining procedures,
which also entail an increased risk of false confession (61, 62). It is not known whether police-induced confessions or plea-bargaining procedures were applied in any of the two studies.

In the other study of moderate quality (42), the authors compared a group in which someone had confessed to having shaken an infant to a control group where an accidental trauma had been witnessed in a public area. However, since the authors used a different triad - with encephalopathy replaced by the absence of scalp swelling - it was not possible to calculate specificity and positive predictive value for the traditional triad. The shaking group was compared to a group with accidental injuries, all of which were very likely to have had signs of external impact to the head or skull. Accordingly, it is no surprise that the authors obtained a specificity and predictive value of 100%. Furthermore, since the authors used different ratings of retinal haemorrhages, the modified triad was even more complicated. Moreover, the nature of the confessions was not reported.

Due to the low quality of the reviewed studies, the incidence and prevalence of shaken baby syndrome remains unknown.

**Other conditions and events that could have caused the triad**

The literature search identified a large range of diseases and events that were associated with the triad or its components. The various diagnoses and events were more or less common and the various conditions were more or less controversial, such as re-bleeding after a minor fall in a child with an enlarged head circumference (53-60). Another controversial issue was whether normal vaginal delivery was associated with subdural haematoma and retinal haemorrhages in around 30% of newborn infants (63-66); the
incidence was reported to be higher in assisted deliveries and significantly lower in scheduled Caesarean deliveries. As far as we know, these phenomena were clinically asymptomatic and the haematomas and haemorrhages resorbed within months. In a few cases, however, the subdural haematoma might have developed into a chronic subdural haematoma or hygroma, which might have resulted in symptomatic re-bleeding, either spontaneously or after a minor trauma (57-60). These possibilities complicate the picture when an infant suddenly presents with symptoms such as apnoea and its parent or guardian is unable to provide an “acceptable” explanation for these symptoms.

Ethical considerations

All children must be protected from abuse and it is also important that families are not unnecessarily separated and that innocent parents or guardians are not convicted. From the clinical perspective of a CPT, it might be more important to protect the infant from abuse than to prevent the conviction of an innocent parent or guardian. But it is a problem if scientists base their classifications on the preferences of a CPT. To date, such teams have provided scientists with biased classification criteria, resulting in biased studies that by default support already established but biased evidence. Epidemiologists found that the incidence of homicide among infants from 1980 to 2005 sharply increased from a stable incidence during the period 1940-1979 (67). The authors suggested that the classification of homicides and accidental deaths in recent decades had been influenced by ethical considerations rather than by scientifically-based consideration.

In order to obtain valid knowledge, future research must avoid circular reasoning when classifying shaken baby cases and controls.
CONCLUSION

This review showed there is insufficient scientific evidence on which to assess the diagnostic accuracy of the triad in identifying traumatic shaking (very low quality evidence).

Furthermore, there is limited scientific evidence that the triad and therefore its components can be associated with traumatic shaking (low quality evidence).

Since valid knowledge is necessary in order to determine whether or not an infant has been violently shaken, future research requires that circular reasoning be avoided when classifying shaken baby cases and controls.

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FINANCE

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CONFLICTS OF INTEREST

None of the authors have any conflicts of interest to declare.


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**Box 1**

The term “shaken baby syndrome” (SBS) signifies a constellation of symptoms and signs, viz. subdural hematoma, retinal hemorrhages and encephalopathy, often referred to as “the triad” as caused by violent shaking.

The present review demonstrates that there is *insufficient* scientific evidence to support claims that the triad indicate that an infant has been violently shaken (*very low quality* evidence), and that there is *limited* scientific evidence to support the assumption that shaking an infant can cause the triad (*low quality* evidence). The term “shaken baby syndrome” is thus not justified, since it includes both the medical findings and the alleged, but scientifically unproven, injurious mechanism - and even the intent behind this mechanism. The same applies to a number of other ill-defined terms used in the literature, for example “Abusive head trauma” (AHT), “Non-accidental head injury” (NAHI), “Inflicted head injury” (IHI), or “NAHT”, which can symbolize two completely opposite meanings, viz. Non-Abusive Head Trauma and Non-Accidental Head Trauma.

Hence, the authors have in this paper avoided the acronyms above, and chosen to differ distinctly between *the injurious mechanism* (“traumatic shaking”) and *the medical findings* (the symptoms and signs, “the triad”). Intent is not, for obvious reasons, for the medical community to decide.
Table 1. The Child Protection Teams’ and the scientists’ criteria for cases classified as shaken babies and controls.

<table>
<thead>
<tr>
<th></th>
<th>Shaken babies</th>
<th>Controls</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lack of explanation</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Accidental fall &lt; 1 meter</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Accidental fall &gt; 1 meter</td>
<td></td>
<td>Yes</td>
</tr>
<tr>
<td>Not witnessed accidents</td>
<td>Yes</td>
<td>Yes</td>
</tr>
<tr>
<td>Witnessed accidental fall</td>
<td></td>
<td>Yes</td>
</tr>
<tr>
<td>Witnessed shaking</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Confessed shaking + details</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>False confession + details</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Confessed milder resuscitation shaking</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Cases in which someone is convicted</td>
<td>Yes</td>
<td></td>
</tr>
<tr>
<td>Cases in which caretakers change story</td>
<td>Yes</td>
<td></td>
</tr>
</tbody>
</table>
Table 2. Recommendations and cautions when conducting future research within the field of shaken baby.

**Prospective cohort and case-control studies**

When classifying shaken baby cases, demand information about:
- police interrogation methods (risk of false confession)
- presence of plea bargain (risk of false confession)
- role of Child Protection Team
- what the suspect has actually confessed
- whether and how differential diagnoses were excluded

When classifying controls, demand information about:
- witnessed events in a public area
- age matching
- role of Child Protection Team

Avoid circular reasoning when classifying cases and controls!

**Other requested studies**

- Screening of newborns for subdural hematoma and retinal hemorrhage
- Natural course of subdural hematoma and retinal hemorrhage among newborns
- Vulnerability of infants with macrocephaly
- Blinding of observations of subdural hematoma and retinal hemorrhages
- Physiological mechanisms of shaking
Figure 1. Flow chart illustrating the literature search.