Shaken baby syndrome: the quest for evidence

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Shaken baby syndrome (SBS), characterized by the triad of subdural haemorrhage, retinal haemorrhage, and encephalopathy, was initially based on the hypothesis that shaking causes tearing of bridging veins and bilateral subdural bleeding. It remains controversial. New evidence since SBS was first defined three decades ago needs to be reviewed. Neuropathology shows that most cases do not have traumatic axonal injury, but hypoxic–ischaemic injury and brain swelling. This may allow a lucid interval, which traumatic axonal injury will not. Further, the thin subdural haemorrhages in SBS are unlike the thick unilateral space-occupying clots of trauma. They may not originate from traumatic rupture of bridging veins but from vessels injured by hypoxia and haemodynamic disturbances, as originally proposed by Cushing in 1905. Biomechanical studies have repeatedly failed to show that shaking alone can generate the triad in the absence of significant neck injury. Impact is needed and, indeed, seems to be the cause of the majority of cases of so-called SBS. Birth-related subdural bleeds are much more frequent than previously thought and their potential to cause chronic subdural collections and mimic SBS remains to be established.

The diagnosis of shaken baby syndrome (SBS) is characterized by a triad of clinical signs: subdural haemorrhage (SDH), retinal haemorrhage (RH), and encephalopathy. The syndrome remains controversial; this review examines the current evidence for and against the two main hypotheses put forward to explain it. The topic has a bearing on the practice of all paediatricians caring for infants with suspected abuse, as well as those who are asked to give expert opinion.

The ‘accepted’ hypothesis

The term non-accidental head injury (NAHI), which bears no implication for the mechanism of injury, is preferred to the term ‘shaken baby syndrome’. In addition to the triad of signs described above, two further factors of circumstantial evidence are often adduced: (1) that the injuries are invariably inflicted, unwitnessed, by a sole carer; and (2) that the given history is incompatible with the severity of the injuries. The term ‘incompatible’ is highly subjective and open to circular logic. In other words, the history will be regarded as inadequate if it does not correspond to the paediatrician’s notion of the history required to produce the injuries observed. Even the term ‘sole carer’ may be applied when other family members are present in adjacent rooms. Notably, there is no requirement for there to be any objective evidence of trauma.

The association of SDH with child abuse has been recognized since the mid-nineteenth century. In 1962, Kempe et al. described ‘battered child syndrome’, a combination of SDH, multiple skeletal injuries, and bruises. In the 1970s Caffey described the association of long bone fractures and SDH with child abuse. While he admitted that the cerebral lesions of whiplash-shaking had not been studied systematically, and that much of his evidence was circumstantial, he promoted a nationwide education campaign which led to widespread acceptance of SBS as ‘a serious and clearly definable form of child abuse’. It is, however, Guthkelch who is credited with recognizing the importance of whiplash during shaking in causing intracranial injuries. Basing his hypothesis on a study by Ommaya et al., Guthkelch suggested that the rotational forces of shaking would cause tearing of bridging veins and bilateral subdural bleeding. Ommaya subjected Rhesus...
monkeys to whiplash forces that induced concussion, SDH, and white matter shearing injury (diffuse axonal injury) in 18 out of 50 adult animals, 11 of whom also had neck injury. By applying Newton’s second law of motion (Force = mass × acceleration) mass scaling could be applied. In other words, smaller brains required larger rotational accelerations to cause injury than larger brains, and in this way it was thought possible to predict the force required to cause intracerebral damage in infants.

Extrapolation of the findings of these early studies on adult primates to human infants requires careful consideration of the differences in structure and tissue properties of the infant head and neck, including the very specific response of the infant brain and unfused skull to head injury. When considering the implications of his experimental results on the proposed injury mechanism associated with SBS, Ommaya himself noted that: ‘It is improbable that the high speed and severity of the single whiplash produced in our animal model could be achieved by a single manual shake or even a short series of manual shaking of an infant in one episode.’

Nonetheless, the acceptance of shaking as a cause of severe intracranial injury has led to it being considered capable of generating force of enormous magnitude, often equated to a high-speed motor vehicle accident or a fall from a second-storey window.

**Support for the ‘accepted’ hypothesis**

Support for the ‘accepted’ hypothesis is based on a number of sources, particularly the literature, well documented episodes, witnessed shaking events, and confessions and convictions.

‘The literature’ is fraught with problems, including poor case ascertainment and circular logic. These problems were well expressed by Hobbs et al. who said of their study of infant SDH: ‘As the study was retrospective it was inappropriate to apply specific and accepted criteria with which to define non-accidental and accidental injury, the most contentious area of diagnosis. Also there is no absolute or gold standard by which to define NAHI.’

Leestma reviewed the literature from 1969 to 2001 and found 324 cases with detailed individual case information. Of 54 cases in which shaking was admitted, only 11 were without evidence of impact and could, therefore, be considered ‘pure’ shakes. Independently witnessed shakings are even more uncommon; only three cases appear to be recorded in the world literature.

Confessions and convictions are regarded as important in supporting SBS, but confessions are unreliable unless the circumstances in which they are made are known. Confessions may be made by one parent to prevent the remaining children from being taken from the other parent and put into care. Such confessions can result from intense police interrogation of distraught or frightened parents, or they may be part of plea-bargaining. Confessions were demonstrated to be unreliable by the UK Court of Appeal in 2005 where two of the four cases before it had their convictions overturned and one was reduced from murder to manslaughter. One appeal was dismissed.

**Evidence which undermines the ‘accepted’ hypothesis**

**BIOMECHANICS**

In 1987, Duhaime et al. used dummies (similar to the crash test dummies used in road-safety research) modelled on a typical 6-month-old human infant to test the accepted hypothesis. Using a variety of neck models Duhaime predicted injury thresholds using Ommaya’s data. College students shaking the dummies generated mean acceleration of 9.2G, considered well below the threshold necessary for intracranial damage, while impacting the dummies on to a metal bar generated 482G. Duhaime wrote: ‘It is our conclusion that the shaken baby syndrome, at least in its most severe acute form, is not usually caused by shaking alone. Although shaking may, in fact, be a part of the process, it is more likely that such infants suffer blunt impact. The most common scenario may be a child who is shaken then thrown into or against a crib or other surface, striking the back of the head and thus undergoing a large, brief deceleration. This child then has both types of injury-impact with its resulting focal damage, and severe acceleration/deceleration effects associated with impact causing shearing forces on the vessels and parenchyma.’ In 1979, the term ‘shaken-impact syndrome’ was introduced, despite the finding that impact itself is a sufficient cause of brain damage without any need for prior shaking.

Duhaime’s experiments were reproduced by Cory and Jones. These authors created forces which exceeded the injury threshold for concussion, but noted that there were chin and occipital contacts at the extremes of the shaking motion in their dummies. Their shaker volunteers fatigued after 10 seconds and the authors expressed their concerns regarding the difficulties in extrapolating to human infants the findings in both dummy and animal models. They concluded: ‘It cannot be categorically stated, from a biomechanical perspective, that pure shaking cannot cause fatal head injuries in an infant.’ In the same year (2003), Prange et al. published the results of studies using a dummy modelled on a 6-week-old infant. In these studies, shaking and inflicted impact onto foam were insufficient to cause accelerations considered necessary for SDH or axonal injury, while impact onto a firm surface exceeded the acceleration of a 1.5m fall onto concrete or carpet. Falls onto foam caused much less rotational force to the head as the foam cradled head and neck together. This is an important observation as it is frequently not recognized that falls and impact to the head produce significant rotational forces due to impact forces which are not aligned through the centre of gravity of the head, and the effect of the hinging of the head on the neck. These authors also commented on the problems of modelling the infant skull and neck and extrapolating results from dummy and cadaver studies to human infants.

Neck injuries may be under-reported in infants who die after severe abuse. In Ommaya’s study, 11 of 19 primates had neck injuries; these were adult animals with mature neck structure and musculature. It is likely that the forces required to cause intracranial injury would also damage the weak infant neck. This makes logical sense to all those of us who drive cars fitted with head restraints: they are designed to prevent whiplash, not SDH or RH. In road traffic accidents infants who suffer single severe hyperextension forces have cervical fractures, dislocations, spinal cord injury, and torn nerve roots, not SDH.

**BRAIN PATHOLOGY**

Encephalopathy associated with NAHI was thought to depend on white matter tearing or diffuse axonal injury. However, in 2001 Geddes et al. published a detailed microscopical study of a large series of infants thought to have suffered inflicted brain
The study overturned previous ideas by showing that most of these infants were not suffering from diffuse axonal injury, as seen in adult trauma, instead most had hypoxic–ischaemic injury. Only a minority had axonal injury and this was restricted to specific areas of the brainstem. Her conclusion was supported by subsequent neuroradiological observations.24

This has important clinical implications. Whereas axonal tearing results in immediate loss of function and a lucid interval is unlikely, brain swelling takes place with a speed and severity of huge individual variation, which may allow a lucid interval or a period in which cerebral function is gradually impaired.25,26 This is particularly likely to occur in infants whose skulls still have unfused sutures and are, therefore, distensible.27

The alternative hypothesis
Geddes had observed that infants with NAHI commonly have a thin bilateral film of subdural blood, sometimes so small that it is missed on scans.22,23 Neuroradiological observations have confirmed that these bleeds may be as small as 2 to 3ml and easily missed at autopsy.28 Geddes proposed that these very thin subdural haemorrhages may not be the result of traumatic rupture of bridging veins, which causes thick unilateral space-occupying clots, but may occur when intracranial vessels are damaged by hypoxia in the presence of abnormal haemodynamic forces such as venous hypertension, systemic arterial hypertension, or episodic surges in blood pressure.29

The paper sparked considerable debate,30,31 perhaps somewhat surprisingly as over 100 years ago virtually the same hypothesis was proposed by Cushing: 'The intracranial hemorrhage is usually of venous origin and follows the rupture of some of the delicate and poorly supported venous radicles of the cerebral cortex. Such an injury may be the direct result of undue traumatism during labor or may occur when too great strain has been put upon the vessels by the profound venous stasis of postpartum asphyxia,' just as in later months they may rupture under the passive congestion brought about by a paroxysm of whooping cough or a severe convulsion.32

Support for the Geddes hypothesis
Can infant SDH arise from sources other than traumatic rupture of bridging veins?

Perhaps the best information regarding infant SDH has been derived from the study of neonates. The preceding clinical circumstances are known and the older pathological literature contains detailed descriptions of large numbers of autopsy cases. Bridging vein rupture is uncommon: Craig described 120 neonatal autopsies with 62 showing SDH but only three with torn bridging veins, all with overriding sutures.33 Larroche described 700 autopsies, 18% with SDH.34 She noted a common association with hypoxic–ischaemic injury. She also described venous congestion, particularly with asphyxia and anaemia, and after exchange transfusion causing raised atrial pressure. She did not identify torn veins but referred to the work of Cushing, who both operated on his cases and subsequently did the autopsies. Even he had difficulties: 'In two of the cases I have examined I have satisfied myself that such ruptures were present. A positive statement, however, cannot be given even for these cases, since the dissection and exposure, difficult enough under any circumstances owing to the delicacy of the vessels, is the more so when they are obscured by extravasated blood.'32 The difficulty of autopsy verification of traumatic rupture of bridging veins has been addressed more recently and contrast injection of the veins attempted.35

Are there alternative, non-traumatic, sources of subdural bleeding? The dura, falx, and healing chronic subdural membranes are all potential sources of widespread, thin-film subdural bleeding. Volpe considered that SDH is by no means always traumatic and arises from tributaries of the venous sinuses within the layers of the dura.36 Intradural bleeding is frequently seen in asphyxia,37 and brain scans of infants with severe hypoxic–ischaemic injury frequently show high signal consistent with congestion or haemorrhage in the falx where extensive venous sinuses are to be found.38,39

The natural healing process of SDH is by formation of a membrane containing numerous fragile, thin-walled capillaries which bleed spontaneously.40 Small subdural bleeds are frequent after birth, described in up to 26% of vaginal deliveries,41,42 this may confer vulnerability to rebleeding on these infants which may be exacerbated by hypoxia and hypertension, for example, after coughing and choking.43

Evidence that undermines the Geddes hypothesis
The most commonly cited argument against the Geddes hypothesis is that SDH is not seen in cases of hypoxia, asphyxia, drowning etc., but these cases are rare and very few will be scanned in any one department. Further, as noted above, these bleeds may be small and readily missed on a scan or at autopsy. There has been no detailed prospective study to support this argument. A single retrospective case study gives insufficient clinical detail to assess the cases of the appropriate age group.44

The 2003 Geddes hypothesis paper29 was criticized because only one of the 50 infants who presented with dural bleeding...
had macroscopic SDH. My own experience is that if the dura is adequately sampled, dural and subdural membrane bleeding is frequent in association with macroscopic SDH. Again, careful prospective studies are needed.

Objective assessment of infant head injury

Biomechanics has an increasingly important role in our understanding of the forces involved in infant head injury. It has grown from studies of transport safety and essentially examines the effects of physical forces upon biological systems. With suitable scientific rigour it can determine applied forces in an objective and reproducible way, but the determination of resulting injury is far more complex. Motor vehicle testing laboratories have established injury criteria that are used in the development of car safety devices by detailed reconstruction of accidents.

It has been shown that head impacts onto carpeted floors and steps from heights in the 1 to 3 feet range result in far greater head impact forces and accelerations than shaking and slamming onto either a sofa or a bed (C Van Ee, personal communication 2007; Fig. 1), reproducing the findings from Duhaime and Prange noted above.

Biomechanical studies have shown that both falls and impacts generate rotational forces due to off-axis impact forces and the hinging of the head on the neck. The assumption that rotational forces are indicative only of shaking is clearly unfounded and biomechanically incorrect.

The clinical history

Clinical history is the cornerstone of good diagnostic practice. I am very struck by the numbers of cases of infants with SDH, RH, and encephalopathy with a history of vomiting and choking, given consistently and in detail. Thousands of infants regurgitate and choke every day but they do not develop the triad. Is there any justification for thinking that just sometimes, in a few cases, they might? Inhalation of feed or vomit may play a part in sudden infant death and awake apnoea associated with gastro-oesophageal reflux is well recognized. The physiological response to aspiration may be dramatic; foreign material on the larynx causes laryngospasm which is associated with or without vomiting.

Expert evidence

Giving evidence is in itself daunting and many clinicians are reluctant to become involved. But clear, thoughtful, and balanced input is essential if we are to assist in the promotion of justice. Our responsibility is to examine all the details in every case of unexplained infant collapse and to adopt a rigorous and questioning approach. If we are intellectually honest we will sometimes be forced to admit that we simply do not know the cause. Children must be protected from harm, and parents and families must be protected from hasty and wrongful accusation, which itself can wreak dreadful and lasting damage.

Acknowledgement

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References


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**List of abbreviations**

NAHI Non-accidental head injury

RH Retinal haemorrhage

SBS Shaken baby syndrome

SDH Subdural haemorrhage
Shaken baby syndrome: evidence and experts

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Non-accidental head injury is a topic that attracts controversy in both medical and legal domains and that requires a forensic approach both to the facts and to their interpretation. As this timely article from Dr Squier rightly points out, the evidence that should underpin interpretation of the findings in allegedly 'shaken' babies is often lacking. At the heart of the debate lies the interpretation of the so-called classic triad: subdural haemorrhage (SDH), retinal haemorrhage, and acute encephalopathy. Unfortunately, the review selectively quotes hypothesis supporting hypothesis as though it was evidence.

Dr Squier outlines two hypotheses for the development of SDH in infants: traumatic injury (the 'classic' theory) and raised arterial or venous pressure within the intracranial compartment. She goes on to criticize, on a number of counts, the view that shaking or shaking/impact might be the causative factor. First, it is noted that the literature in this field often makes assumptions that the cause of the triad is non-accidental, because shaking that is actually witnessed is very rare. Second, experiments which seek to model infant brain injury using animals or dummies have themselves generated disagreement among scientists as to the strength and nature of the forces required. Third, it is argued that severe neck injury of the whiplash type should accompany the triad if shaking or impact is the cause. Fourth, the supposition that diffuse traumatic axonal injury is present has been shown to be incorrect by Geddes et al. Finally, the size of the SDH in infants is noted to be small and unlike the space occupying lesion found in adult traumatic head injury, the conclusion being that SDH in the infant does not result from torn bridging veins.

A second scenario is then advanced which is that some natural event, such as choking or coughing, which leads to a rise in intracranial pressure, may be the initiating event for SDH in infancy. This is clearly the favoured view of the author and is presented in order to explain the apparent absence of torn bridging veins in the setting of infant SDH. Parallels are drawn with SDH occurring in the perinatal period, which is often associated with evidence of hypoxic–ischaemic damage in the brain. It is argued, on the basis of the author’s extensive experience, that chronic SDH with membrane formation is the setting for an apparently novel haemorrhage later in infancy.

We take issue with most of the tenets of the review and would argue that trauma remains the most likely cause of SDH in infancy, while accepting that other causes, such as bleeding disorders, do exist. The argument in relation to shaking is much like that applied to short falls. Peer-reviewed literature exists in support of both shaking and short falls as a cause of infant death, and Dr Squier does quote literature in which shaking was observed to have been associated with infant death. Presumably, this literature negates the discussions within the biomechanical literature; regardless of the data produced by animal models and dolls, real life has demonstrated that shaking can produce a sufficient force to kill an infant. However, what is clear from the literature quoted is that observed fatal shaking of an infant is a rare occurrence. In this review, much of the discussion of shaking focuses around biomechanical analysis of animals and models. When discussing the data generated by Ommaya et al., Dr Squier rightly reminds the reader of the differences between a rhesus monkey and a human infant. Similar statements should be applied to the dolls used in the studies cited in the later discussions. Many biomechanical studies are still using diffuse traumatic axonal injury as a benchmark for analysis of force generation. As far as we are aware, we still have no indication of the forces required to produce focal cervicomedullary axonal injury, which may underpin the observed hypoxic–ischaemic brain injury and, as such, cannot provide definitive statements.

Injuries to the soft tissues of the neck, as well as to the cervico-medullary junction and cervical spinal roots, are undoubtedly apparent in some infants who present with the triad and these injuries have been documented. They may be missed if they are not looked for in each case. It is clear from the work of Geddes et al. and others that knowledge regarding brain injury in the infant is far from complete and it cannot be assumed that an infant brain responds to injury in the same way as that of an adult.

That trauma can cause torn bridging veins and, therefore, SDH is beyond doubt and has been demonstrated at autopsy in cases of alleged non-accidental injury. In neonatal SDH, torn bridging veins are rare but trauma is the cause of the majority, through dural tears involving sinuses. Vitamin K administration is a recognized non-traumatic cause of intracranial haemorrhage in neonates but we are unaware of any other non-traumatic causes seen in clinical practice in this group. Therefore, it is difficult to argue against trauma as a mechanism for SDH in infants. Is hypoxia another likely cause of macroscopically visible SDH? There are papers based on observation at autopsy which refute that hypothesis but none supporting. Dr Squier dismisses this paper although we are uncertain which aspects of the clinical details would make the observations either more robust or unfounded. Therefore, until there is a definite body of evidence in support, this must remain a hypothesis only. Indeed this hypothesis was considered by the UK Court of Appeal and found to be wanting. The papers presented in the review actually refer to microscopic intradural bleeding (a common occurrence) or to a mathematical-computer model.
The former cannot be assumed to be relevant for macroscopic subdural haemorrhage. The studies papers are of considerable interest and require investigation in human tissues. If they can be confirmed by observation in human infants they would greatly change our view of this field.

We do have to take issue with the statement that ‘bleeds may be small and readily missed on scan and at autopsy’. If this is indeed the case, at what point are these bleeds picked up and at what point does non-accidental injury enter the differential diagnosis?

A third possibility, which is not discussed in this review, is that infant SDH may differ from those seen in adults due to acute brain swelling. Both trauma and hypoxia may be involved in the pathogenesis of the triad. The sequence of events may be as follows; initial trauma causes both SDH and stretch injury in the brainstem, followed by hypoxic–ischaemic brain swelling. If brain swelling develops rapidly, the pressure may be sufficient to limit bleeding from torn veins and blood may be compressed within the subdural space in the typical thin film. It is important to emphasize that the final distribution of subdural blood tells us little about how the brain was injured.

Dr Squier makes interesting comments in relation to choking. Paediatric pathologists and neuropathologists sometimes have the opportunity to review tragic cases in which choking is the clear mechanism of death. Macroscopic SDH is not a feature of such cases. We, therefore, have to consider whether there is a subgroup of infants who choke and aspirate leading to hypoxic–ischaemic encephalopathy (but display no pulmonary pathology). Dr Squier has drawn attention to the lack of firm evidence which currently exists for the triad as being pathognomonic of deliberate and inflicted injury. However, the evidence for her alternative hypothesis, that a significant number of cases might be attributable to a completely accidental cause such as choking, is also lacking.

It remains our view that the main differential diagnosis for causation of infant SDH lies between accidental and inflicted injury. It is important to stress that the triad is not pathognomonic of inflicted injury. Accidental trauma can produce the same spectrum of injuries and it is then for the professional, be it medical or legal, to assess the carer’s story of how the injury developed. As Dr Squier suggests, it would be unfortunate for professionals to immediately adopt the starting point that all carers are guilty and, indeed, that would be rather at odds with the legal system in the UK. When applying a scientific basis to the assessment of the triad it is important to accept that we do not know what the lowest thresholds are to produce a potentially fatal injury. Until inflicted injury can be differentiated with certainty from accidental, both on clinical or pathological grounds, the courts will be required to base their judgements both on the carer’s version and on presentations put forward by expert witnesses. In this fraught area it is important to keep an open mind about the history of events as provided by the carer. The reasons advanced by Dr Squier for dismissing confessions of abuse seem to us wholly unconvincing.

Finally, we believe that there is an urgent need for research to be undertaken in these cases. It is somewhat ironic that the legal profession expects a level of certainty from expert witnesses in court which may not be possible based on current pathology evidence. At the same time, legislation has been introduced which precludes the possibility of future pathology-led research aimed at achieving that evidence, since consent for research is required from parents who may themselves be accused of causing the infant’s death.

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Shaken baby syndrome: the quest for evidence

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There have been two recent excellent reviews of non-accidental head injury which focus on the controversial issues that are also highlighted in the Squier review in this issue. These include an incomplete understanding of the biomechanics of the disorder, the legal implications of interpretation of the clinical trial and neuropathological findings, and the accuracy or appropriateness of the terminology applied - shaken baby syndrome versus the shaken-impact syndrome, non-accidental head injury, or inflicted traumatic brain injury.

Our group at the Hospital For Sick Children in Toronto recently published a text on head injury in children and adolescents with a section devoted to a discussion on the current state of knowledge about non-accidental traumatic brain injury. We summarized the issues and accepted factual information as follows:

1) Shearing rotational forces are necessary to produce the array of significant intracranial, brainstem/cervical, and ocular pathology that create this syndrome – translational or linear forces (those produced by simple falls) do not result in catastrophic injury.

2) Shaking alone is thought to be insufficient to result in severe intracranial injuries based on biomechanical studies – there is good evidence for a requirement of associated angular deceleration produced by head impact on a firm or solid surface.

3) Post-mortem neuropathological studies show macroscopic swelling with microscopic neuronal hypoxic-ischaemic injury, and seldom is there evidence of diffuse axonal injury.

4) A unifying hypothesis is that the critical factor in producing injury is prolonged hypoxemia – a result of brainstem and upper cervical torsion injury with resultant apnea of variable duration.

The debate on nomenclature deserves further comment. Whether or not the term ‘shaken baby syndrome’ is accurate in the scientific or evidence-based sense, there has been an opportunity to research societal and parenting risk factors with construction of educational programs on the theme of opportunity to research societal and parenting risk factors in the scientific or evidence-based sense, there has been an opportunity to research societal and parenting risk factors.

The Squier article notes the need for continuing study. Research can be focused in a number of areas which include:

1) Evaluation of the individual and unique biochemical properties and characteristics of the injured infant brain which can include serum and cerebrospinal fluid protein and enzyme markers (S100B, neuron-specific enolase), and the relationship of outcome to the apolipoprotein E genotype. Further study of individual differences in neuroimmunological repair mechanisms and apoptotic gene upregulation in response to injury will add to the provision of neuroprotective strategies.

2) Recognizing that, at present, there is no satisfactory biomechanical model for this syndrome complex and no accurate knowledge of the mechanical forces involved in creating this type of brain injury, it is necessary to construct a research model that will address these issues.

3) There needs to be continued neuroradiological, clinical, and research efforts which will result in better temporal and mechanistic delineation of the injury patterns.

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References
Letters to the Editor

‘Shaken baby syndrome’

SIR – I am grateful to Drs Smith and Bell, and the editors of the Journal for the opportunity to continue this debate. One thing that is certain is that conflicting and difficult medical hypotheses cannot be properly examined in the stultifying adversarial atmosphere of the Court of Law, where worn out arguments are bandied back and forth by a very small band of ‘experts’. The only way that these arguments can be rigorously evaluated, and either supported or rebutted, is by open debate in the wider clinical and scientific community.

Several points in Drs Smith and Bell’s response invite further discussion. They say, without citing evidence, that ‘real life has demonstrated that shaking can produce a sufficient force to kill an infant’ but that ‘observed fatal shaking of an infant is a rare occurrence’. If shaking has not been witnessed, how can any one be sure that it happened? The standard for diagnosis of accidental injury is that it is independently witnessed; the standard for diagnosis of shaking appears to be lower and based on surmise.

Smith and Bell have a dismissive attitude to biomechanical studies indicating that most use traumatic axonal injury as the benchmark for force analysis and have not generated figures for the force required to cause cervicomедullary axonal injury. Most studies, in fact, use concussion, subdural haemorrhage (SDH), and traumatic axonal injury as injury criteria.3,4 As SDH is perhaps the most critical of the diagnostic features of the triad this, if any, is the relevant benchmark; indeed focal cervicomедullary injury was not seen in two-thirds of infants thought to have non-accidental head injury.5 Their word ‘dolly’ does not do justice to the very sophisticated anthropomorphic dummies used (the same models used in laboratory tests which allow us all to drive our cars in relative safety).

On the issue of SDH, Drs Smith and Bell baldly state, without evidence, that the majority of neonatal SDH is due to traumatic dural tearing involving the sinuses. This is not the experience of Larroche,6 based on over 700 autopsies (‘dural tears are exceptional’, ‘tentorial tears are rare’), nor of Volpe,7 the most erudite of paediatricians, whose textbook notes that this kind of bleeding is often not traumatic and arises from the tributaries of the dural sinuses.8 This makes perfect sense as infant dural folds contain extensive venous sinuses which gradually recede in the first year of life.4 During childbirth, compressive forces on the head and the chest will obstruct cerebral venous drainage and raise pressure in these sinuses, so it is likely that some may rupture, as Cushing suggested in 1905.9 Smith and Bell do not recognize an association of hypoxia with SDH, an association noted four decades ago by Larroche6 and currently supported by post-mortem magnetic resonance imaging and autopsy correlation studies.10 The study they cite to refute this association11 failed to use stringent histological criteria for hypoxic brain injury, or to note whether infants had been subject to cardiopulmonary resuscitation or prolonged ventilation, as most ‘shaken baby syndrome’ (SBS) infants are. Most significantly, infants up to 3 years of age were grouped with no specific data relating to infants under 6 months, the most common age for a diagnosis of SBS. The anatomy and pathophysiology of the brain, dura, skull, and their blood supply, as well as cerebrospinal fluid dynamics, are quite different at this age from the older infant or child. This, in itself, is surely reason to pause for thought. It is clear from the response to the paper by Byard et al.12 and from ongoing studies that paediatric pathologists in day-to-day practice do see SDH in infants with hypoxia.12

‘That trauma can cause torn bridging veins and therefore SDH is beyond doubt’ cites Maxeiner,13 who in fact came to the paradoxical conclusion that he could demonstrate torn bridging veins in infants who had no SDH. If Smith and Bell had read Maxeiner’s conclusions, on which they depend, then why should they ‘have to take issue with the statement that bleeds may be small and readily missed on scan and at autopsy’? Their question ‘at what point are these bleeds picked up and at what point does non-accidental injury enter the differential diagnosis?’ is chilling indeed if one of the most significant aspects of the triad can be discarded yet the diagnosis of shaking remain viable.

Having regretted that my article ‘selectively quotes hypothesis supporting hypothesis as though it was evidence’, it is perhaps surprising that Smith and Bell should add another of their own: ‘Initial trauma causes both SDH and stretch injury in the brainstem, followed by hypoxic-ischaemic brain swelling. If brain swelling develops rapidly, the pressure may be sufficient to limit bleeding from torn veins and blood may be compressed within the subdural space in the typical thin film’.

This hypothesis defies logical analysis. The blood supply to the infant brain is some 55ml per 100g of brain per minute; in a 6-month infant about 330ml per minute. If only one-third of this blood flow drains via the bridging veins, this represents 110ml of blood in 1 minute. Brain swelling would have to be virtually instantaneous to reduce leakage from these vessels to a thin film. This kind of bleeding is more likely to produce the thick space-occupying SDH well recognized after impact to the head. Any trauma is itself a potent cause of brain swelling and hypoxia.14 Are we seriously to consider that the brain will respond by swelling at a different rate and degree following accidental trauma than following inflicted trauma?

As I proposed in my review, this is a difficult and controversial area. Uncritical acceptance of the literature is fraught with danger. Careless opinions have devastating effects on the lives of infants and their families.

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References


‘Shaken baby syndrome’

SIR — I read with interest the article ‘Shaken baby syndrome: the quest for evidence’ by Dr Squier1 and the two attached commentaries by Dr MacGregor2 and Drs Smith and Bell.3

In this review article, Dr Squier is reviewing the ‘accepted’ shaken baby hypothesis by covering the challenges to the traditional hypothesis and presenting evidence for alternative explanation. Her analysis stands on its own. However, I feel the commentaries by Dr MacGregor and Drs Smith and Bell raise specific points for comment.

First, the title of the article clearly indicates that the subject is ‘Shaken baby syndrome: the quest for evidence’, not ‘Shaken impact’ or vague, all-inclusive terms of ‘non-accidental head injury’, or ‘inflicted traumatic brain injury’, or ‘inflicted blunt force head injury’, which are non-specific as to mechanism of injury. Since the publication by Goldsmith and plankett4 of a video taped fall that resulted in the trial of retinal hemorrhage (RH), subdural hemorrhage (SDH), and brain encephalopathy at autopsy, there can be no question that impact from a short distance fall can be fatal and such a history allows for a lucid interval and the trial. This irreducible evidence is consistent with and confirms the predictions of the biomechanical research. However, the commentary by Drs Smith and Bell appears to muddy the water by shifting the subject from pure shaking, the mechanism of controversy, to include impact, a mechanism of known validity. I hope this was not an intentional tactic on their part to avoid dealing with the real issues at hand. If so, this attempt to shore up the shaking mechanism and shield it from challenge, should be recognized as an acknowledgment by Drs Smith and Bell that the pure shaking mechanism cannot be trusted to stand on its own. Second, I would point out that Drs Smith and Bell and Dr MacGregor failed to address Dr Squier’s reference to the old pertussis literature that documented intracranial bleeding and ocular bleeding in infants dying of whooping cough. This literature was more fully cited in the Geddesand Talbert 2006 article5 and documents the three findings of the trial without impact or impulse-loaded forces. Any response challenging the tenets of Dr Squier’s article must account for this pertussis literature, if it is to be credible.

The validity of the traditionally accepted hypothesis that abusive shaking can cause a unique and recognizable pattern of physical injury has been seriously challenged. To appropriately explore this controversy, one must focus on the shaking mechanism of injury. To maintain this focus, I will use the terminology of inflicted impulse loaded rotational acceleration/deceleration without impact (ILRADWI), rather than allow distracting vagary to creep into this discussion. The fundamental issues at the heart of this controversy are:

1. Is inflicted ILRADWI (pure shaking) a valid injury mechanism capable of causing the injury pattern of primary RH, SDH, and diffuse axonal injury (DAI), the triad, without structural neck failure?

2. Is inflicted ILRADWI a unique primary cause of the entire triad?

3. If inflicted ILRADWI were not a valid primary cause of the entire triad, then is it a unique primary cause of any single feature of the triad and, therefore, a necessary component of an alleged abusive injury?

4. If inflicted ILRADWI is not a valid primary cause of all three elements of the trial, or if it is not a unique cause of at least one of the three elements of the trial, then the trial as a whole and each of its elements must have alternate mechanisms of causation. Presently, proposed alternatives center on some combination of hypoxic injury (hypoxemic or ischemic), increased intravascular pressure from any cause, and/or coagulopathy, either as primary events or as secondary injuries that come into play in the time between the acute life-threatening event (ALTE) and physical death autopsy, which mayfollow the initial ALTE by hours to days. Primary blunt force impact can result in the triad but when history and physical evidence of impact is lacking, one cannot just assume that injuring impact on a soft, non-bruising surface occurred with any degree of forensic medicai certainty.

If the answer to the first three questions above is ‘no’, then inflicted ILRADWI should be dropped from consideration, and all previous convictions where inflicted ILRADWI was presented as valid or as a necessary component of the allegations should be reviewed as potentially unsafe.

Dr MacGregor’s commentary is straightforward and clearly presents the dilemma for proponents of the classic shaken baby hypothesis. She starts by laying out three seemingly contradictory statements ‘accepted factual information’ and then tries to offer a ‘new unifying hypothesis’ to somehow preserve the validity of the inflicted ILRADWI mechanism of injury. She states, ‘We summarized the issues and accepted factual information as follows:

1. Shearing rotational forces are necessary to produce the array of significant intracranial, brainstem/cervical, and ocular pathology that create this syndrome – translational or linear forces (those produced by simple falls) do not result in a catastrophic injury.’ [This statement appears to assert that shearing rotational forces are necessary for the generation of the trial and to deny the possibility that short falls and pertussis can in fact also produce the trial. Goldsmith and Plumkett4 and the old pertussis literature clearly contradict MacGregor’s first statement of accepted factual information. This first statement also contradicts point 2 and offers no explanation for point 3.]

2. Shaking alone is thought to be insufficient to result in severe intracranial injuries based on biomechanical studies – there is good evidence for a requirement of associated angular deceleration produced by head impact on a firm or solid surface. [Note this statement avoids mention of the eyes. It contradicts point 1 by asserting a rotational component to impact. It offers no explanation for point 3 below. When taken together with
Goldsmith and Plunkett, it invalidates the claim that inflicted ILRADWI is a unique cause of the triad or that inflicted ILRADWI is a valid cause of the triad.

3. Post-mortem neuropathological studies show macroscopic swelling with microscopic neuronal hypoxic-ischemic injury, and seldom is there evidence of diffuse axonal injury. [Note this statement of uncontested pathological findings, is not explained by the inflicted ILRADWI in point 1 or by point 2.]

In this maze of contradiction, Dr MacGregor, the only potential resolution is a new hypothesis, that ‘the critical factor in producing injury [the triad] is prolonged hypoxemia – a result of brainstem and upper cervical torsion injury with resultant apnea of variable duration.’ By proposing a new hypothesis, MacGregor is acknowledging that all presentations of the shaking mechanism prior to 2001 were factually in error. It should also be noted that if this new unifying hypothesis were not to be supported by clinical observations in general or in specific cases of alleged inflicted ILRADWI, then presently there is no other purposed hypothesis to explain how inflicted ILRADWI would be a valid primary cause of the triad. In that event, alternate injury mechanisms, such as the ones purposed by Dr Squier, are what remain and must be taken as the best that are presently available.

Des Smith and Bell’s commentary is not straightforward and is open to several specific challenges. They start by stating, ‘We take issue with most of the tenets of [Dr Squier’s] review and would argue that trauma remains the most likely cause of SDH in infancy, while accepting that other causes, such as bleeding disorders, do exist.’ The subject of the article is shaken baby syndrome, not the vague general term ‘trauma’. Inflicted ILRADWI is the classic/traditional hypothesis – the hypothesis that has come under challenge. Goldsmith and Plunkett and the biomechanical literature validate blunt force impact as a cause of SDH and the full triad. With regard to impact, it is no longer a ‘quest for evidence’ – we have that evidence. Des Smith and Bell then state, ‘Peer-reviewed literature exists in support of both shaking and short falls as a cause of infant death, and Dr Squier does quote literature in which shaking was observed to have been associated with infant death.’ Dr Squier did cite Leestma’s background, but Dr Squier did so outlining the challenges to this literature, not affirming its validity. I would point out that ‘shaking... associated with infant death’ is not the same as establishing that shaking, ILRADWI, was the primary mechanism producing a unique pattern of injury. Then relying entirely on this challenged literature, Drs Smith and Bell state, ‘Presumably, this literature negates the discussions within the biomechanical literature; regardless of the data produced by animal models and dolls, real life has demonstrated that shaking can produce a sufficient force to kill an infant.’ Remember, the forensic issue is not whether violent shaking can kill but rather whether inflicted ILRADWI is a unique and valid cause of the triad. Des Smith and Bell’s out-of-hand dismissal of all experimental biomechanical and animal research, justified by reliance on challenged literature is quite remarkable. They then undercut the foundation of his unshakable convictions by adding, ‘However, what is clear from the literature quoted is that observed fatal shaking of an infant is a rare occurrence.’ My question is: has there been any independently witnessed or videotaped evidence of an abusive shaking without impact or pre-existing issues that has produced the triad? Remember, the issue is not whether shaking can kill but whether it can result is some unique pattern of findings that is sufficient to validate a diagnosis of inflicted ILRADWI in a criminal case. Smith and Bell, like MacGregor, then attempt to shore up inflicted ILRADWI as a valid mechanism of injury by calling forth the hypothesis of shaking-induced cervico-medullary junction and cervical spinal root injury sufficient to produce brain lethal hypoxic injury from a primary central apexa. MacGregor and Smith and Bell are not the only ones retreating to this ‘new hypothesis’. In the winter 2007 issue of the Official Newsletter of the American Society of Pediatric Neuroradiology, Dr Boos stated, ‘There is a growing consensus that distortion of the brainstem and upper spinal cord occurs during shaking, and that resulting dysfunction produces apexa, with secondary hypoxic–ischemic brain injury. Within such a model, rotational thresholds for coma and diffuse axonal injury need not be exceeded to explain the findings of the SBS.’

With brain injury now being attributed to hypoxia from central apexa, rather than the primary shearing from acceleration/deceleration, the issue becomes whether inflicted ILRADWI is a unique primary cause of either RH or SDH.

Recent literature has has found that ILRADWI is not only not a unique primary cause of eye findings, but that it is not even a valid primary cause of eye findings. Forbes et al. reported RH in infants with epidural hemorrhage from accidental injury concluding a role for increased intracranial pressure in the generation of RH. Obi et al. documented similar RH, schisis, and folds in accidental injury as those alleged to occur in inflicted injury. Brown et al. failed to find any eye pathology in two kittens and a rabbit killed by witnessed shakings by a large dog. Binenbaum et al. failed to produce any eye findings in piglets subjected to rapid acceleration followed by immediate deceleration to levels 40 times what Prange et al. predicted could even be achieved in inflicted ILRADWI. On analysis of human specimens, Emerson et al. could find no support for the hypothesized vitreous traction mechanism of injury that would be unique to inflicted ILRADWI. This new evidence leaves one looking to alternate mechanisms of hypoxia, increased intravascular pressure, and coagulopathy to account for ocular pathology in cases of alleged abuse.

With regard to SDH, the issue is not whether SDH ever results from a torn bridging vein, but rather whether all SDHs are the result of bleeding from torn bridging veins. The evidence that impact is a valid primary cause of large volume SDH in infants, is irrebuttable given Goldsmith and Plunkett. However, the pertussis literature and Des Smith and Bell’s own citing of coagulopathy as a cause for intracranial bleeding clearly establish a multiplicity of causes and mechanisms for extra-axial intracranial bleeding, since neither pertussis nor coagulopathy involve impact or ILRADWI or external trauma, either accidental or inflicted. Dr Squier further amplified this list by citing documentation of birth-related SDH and raising the issues of benign external hydrocephalus, chronic SDH, and neomembranes as predisposing to both new and/or recurrent extra-axial bleeding. This is compelling evidence that inflicted ILRADWI is certainly not a unique cause of SDH. The issue is whether inflicted ILRADWI is a valid primary cause of SDH at the levels of rotational acceleration that can realistically be achieved in inflicted ILRADWI in normal human infants without preexisting extra-axial pathology. If not, then allegations of inflicted ILRADWI must resort to mechanisms other then tensile stress failure of bridging veins to account for thin films of SDH at autopsy. The Klinich et al. article cited by Dr Squier would indicate that inflicted ILRADWI is not a valid primary mechanism for subdural bleeding. In this study, Klinich et al. reconstructed six auto accidents where human infants were in rear facing infant seats (neck protected) installed in the front passenger seat. The three frontal collisions
with airbag deployment were fatal with findings of SDH; in the three collisions without airbag deployment, infants survived without injury. On reconstruction, SDH and death resulted from accelerations that exceeded 100 resultant G of acceleration while reconstructions of accidents where infants survived indicated that 25 to 45 resultant G of acceleration produced no injury. This human infant data correlates nicely with the available human adult data gathered by Funk and Duma14 on human adult American football players, who analyzed 27,319 impacts that resulted in only four mild traumatic brain injuries (MTBI).

None of these four players lost consciousness and none were reported to have subural bleeding. They found nominal values of 85G and 6000 rad/sec2 were associated with approximately a 1% or lower risk of mild traumatic brain injury based on the risk curves developed in their study. When shaking models of 1-month-old infants, Duhaime et al.16 achieved maximal accelerations of only 13.85G with a single-hinged neck and impact of the head on the chest and back. Prange et al. predicted that the maximal rotational acceleration achievable in an abusive shaking would not significantly exceed 2600 rad/sec2.12 Wolfson et al.17 using computer modeling indicated that any neck modifications toward creating a more human-like infant model would produce progressively lower levels of damaging acceleration with vigorous shaking. Using a more human-like realistic model with two hinges and allowing for some lateral motion (Aprica 3.4 ATD), Carole Jenny18 could produce maximal accelerations of only 1456.5 rad/sec2 equating to only 9.9 resultant G of acceleration, confirming Prange et al. and Wolfson et al.’s predictions. When one compares this biomechanical experimental data and human adult data with the human infant data from Klinich, it would now appear that any claim that inflicted ILRADWI is a cause of primary brain injury or a cause of primary subdural bleeding from a torn bridging vein in an infant without pre-existing extra-axial abnormalities, that would predispose to bleeding from minimal trauma, should now be required to provide solid experimental, objective, and reproducible evidence to establish its validity. To my knowledge such evidence is lacking.

The title of the review article by Dr Squier is ‘Shaken baby syndrome: the quest for evidence’. In my opinion, after review of the current evidence, there is no evidence that:

1. Inflicted ILRADWI can cause primary eye findings by a unique vitreous traction hypothesis.

2. Inflicted ILRADWI can cause primary brain injury by the shearing hypothesis at the levels of acceleration achievable in an abusive shaking.

3. Inflicted ILRADWI can cause SDH by the primary tearing bridging veins at the levels of acceleration achievable in an abusive shaking of a previously normal infant.

In recognition of this present state of medical science, there is a growing consensus acknowledged by Dr Boos, Dr MacGregor, and Dr Smith that, if inflicted ILRADWI is to remain a valid mechanism of injury, it must resort to mechanical cervico-medullary disruption and immediate profound central apnea for its mechanism of injury. This new hypothesis rests on the fundamental assertion that a brain-lateral hypoxia resulting from a central apnea is responsible for the findings of the trial. Ironically, this is the same argument being presented by Dr Squier. A brain-lateral hypoxia is not unique to this new hypothesis for inflicted ILRADWI. It is part of the Geddes et al. hypothesis19 and it is embodied in the Geddes and Talbert20 article cited by Dr Squier. It would also be the fundamental injury in a number of other causes of potentially fatal AITE. Obstructive airway compromise from allergic reaction, aspiration, GERD, choking/aryngospasm /bronchospasm, infection, and congenital issues and central apnea from impact trauma, infection, and metabolic and developmental neurological issues could also cause a brain-lateral hypoxia, and all set off the same subsequent chain of events resulting in the trial at autopsy. While inflicted ILRADWI remains in this differential for central apnea, it is not alone. If the clinical data in any given case supports an alternate cause of brain-lateral hypoxia, or if the clinical data in any given case does not support cervico-medullary trauma as the cause of a brain lateral central apnea, then inflicted ILRA DWI should be removed from the differential, and alternate mechanisms of injury, such as those outlined by Dr Squier, should be considered before any legal action is initiated, either criminal or civil.

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References


