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The evidence base for shaken baby syndrome

We need to question the diagnostic criteria

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The phrase "shaken baby syndrome" evokes a powerful image of abuse, in which a carer shakes a child sufficiently hard to produce whiplash forces that result in subdural and retinal bleeding. The theory of shaken baby syndrome rests on core assumptions: shaking is always intentional and violent; the injury an infant receives from shaking is invariably severe; and subdural and retinal bleeding is the result of criminal abuse, unless proved otherwise.[1] These beliefs are reinforced by an interpretation of the literature by medical experts, which may on occasion be instrumental in a carer being convicted or children being removed from their parents. But what is the evidence for the theory of shaken baby syndrome?

Retinal haemorrhage is one of the criteria used, and many doctors consider retinal haemorrhage with specific characteristics pathognomonic of shaking. However, in this issue Patrick Lantz et al examine that premise (p 754) and conclude that it "cannot be supported by objective scientific evidence."[2] Their study comes hard on the heels of a recently published review of the literature on shaken baby syndrome from 1966 to 1998, in which Mark Donohoe found the scientific evidence to support a diagnosis of shaken baby syndrome to be much less reliable than generally thought.[3]

Shaken baby syndrome is usually diagnosed on the basis of subdural and retinal haemorrhages in an infant or young child,[1] although the diagnostic criteria are not uniform, and it is not unusual for the diagnosis to be based on subdural or retinal haemorrhages alone.[w1] The website of the American Academy of Ophthalmology states that if the retinal haemorrhages have specific characteristics "shaking injury can be diagnosed with confidence regardless of other circumstances."[4] Having reviewed the evidence base for the belief that perimacular folds with retinal haemorrhages are diagnostic of shaking, Lantz et al were able to find only two flawed case-control studies, much of the published work displaying "an absence of … precise and reproducible case definition, and interpretations or conclusions that overstep the data."[2] Their conclusions are remarkably similar to those of Donohoe, who found that "the evidence for shaken baby syndrome appears analogous to an inverted pyramid, with a very small database (most of it poor quality original research, retrospective in nature, and without appropriate control groups) spreading to a broad body of somewhat divergent opinions."[3] His work entailed searching the literature, using the term "shaken baby syndrome" and then assessing the methods of the articles retrieved, using the tools of evidence based inquiry. Reviewing the studies achieving the highest quality of evidence rating scores, Donohoe found that "there was inadequate scientific evidence to come to a firm conclusion on most aspects of causation, diagnosis, treatment, or any other matters," and identified "serious data gaps, flaws of logic, inconsistency of case definition."[3]

The conclusions of Lantz et al and of Donohoe make disturbing reading, because they reveal major shortcomings in the literature relating to a field in which the opportunity for scientific experimentation and controlled trials does not exist, but in which much may rest on interpretation of the medical evidence.[5]

If the concept of shaken baby syndrome is scientifically uncertain, we have a duty to re-examine the validity of other beliefs in the field of infant injury. The recent literature contains a number of publications that disprove traditional expert opinion in the field. A study of independently witnessed low level falls showed that such falls may prove fatal, causing both subdural and retinal bleeding.[6 w2] A biomechanical analysis validates that serious injury or death from a low level fall is possible and casts doubt on the idea that shaking can directly cause retinal or subdural haemorrhages.[7 w3] An important lucid interval may be present in an ultimately fatal head injury in an infant.[8] Neuropathological studies have shown that abused infants do not generally have severe traumatic brain injury and that the structural damage associated with death may be morphologically mild.[9 10] What is the relevance of the craniocervical injuries to corticospinal tracts, dorsal nerve roots, and so on that have been described?[10 11] We do not know. What is the force necessary to injure an infant's brain? Again, we do not know.

While most abused children indisputably show the signs of violence, not all do. No one would be surprised to learn that a fall from a two storey building or involvement in a high speed road traffic crash can cause retinal and subdural bleeding, but what is the minimum force required? "It is one thing clearly to state that a certain quantum of force is necessary to produce a subdural hematoma; it is quite another to use examples of obviously extreme force … and then suggest that they constitute the minimum force necessary."[12]

Research in the area of injury to infants is difficult. Quality evidence may need to be based on finite element

P+ *Additional references w1-w3 are on bmj.com*

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modelling from data on infants' skulls, brains, and neck structures, rather than living animals. Any studies on immature animal models, if performed, will need to be validated against the known mechanical properties of the human infant. Pending completion of such studies, the reviews by Lantz and Donohoe are a valuable contribution and provide a salutary check for anyone wishing to cite the literature in support of an opinion. Their criticisms of lack of case definition or proper controls can be levelled at the whole literature on child abuse. If the issues are much less certain than we have been taught to believe, then to admit uncertainty sometimes would be appropriate for experts. Doing so may make prosecution more difficult, but a natural desire to protect children should not lead anyone to proffer opinions unsupported by good quality science. We need to reconsider the diagnostic criteria, if not the existence, of shaken baby syndrome.

J F Geddes *retired (formerly reader in clinical neuropathology, Queen Mary, University of London)*

London (j.f.geddes@doctors.org.uk)

J Plunkett *forensic pathologist*

Regina Medical Center, 1175 Nininger Road, Hastings, MN 55033, USA

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Shaken baby syndrome

Pathological diagnosis rests on the combined triad, not on individual injuries

Shaken baby syndrome is a form of physical non-accidental injury to infants, characterised by acute encephalopathy with subdural and retinal haemorrhages, occurring in a context of inappropriate or inconsistent history and commonly accompanied by other apparently inflicted injuries.[1 2] Injuries to the neck and spinal cord may also be present. Controversy surrounds the precise causation of the brain injury, the retinal and subdural haemorrhages, as well as the degree of force required and whether impact in addition to whiplash forces is needed.[1 3 4] Although most discussion has concerned fatal injuries of this nature, not all are lethal, but they may be associated with subsequent neurological disability of varying severity.

Expert medical evidence about inflicted injury must have scientific validity, but applying the evidence based criteria appropriate to clinical practice entails some difficulties.[5] In clinical practice medical management of defined clinical problems can be compared and best practice distinguished by clinical outcomes. Conversely, in inflicted paediatric injuries, one is presented with the outcome, investigation follows rather than precedes that outcome, and the history may be incomplete or deliberately misleading. A need exists for an impartial and intelligent assessment, but how may this be achieved in practice?

Because of the serious implications of diagnosing inflicted injury such as shaken baby syndrome, every case must be evaluated in detail, taking account of all the circumstances surrounding the injury and considering the pathological features in full, rather than attempting to evaluate the significance of each component.

In shaken baby syndrome, it is the combined triad of subdural and retinal haemorrhage with brain damage, as well as the characteristics of each of these components that allow a reconstruction of the mechanism of injury, and assessment of the degree of force employed. The application of rotational acceleration and deceleration forces to the infant's head causes the brain to rotate in the skull. Abrupt deceleration allows continuing brain rotation until bridging veins are stretched and ruptured, causing a thin layer of subdural haemorrhage on the surface of the brain. This is not a space occupying lesion; its importance is in indicating the mechanism of injury. The retinal haemorrhages, which are characteristically extensive, occupy much of the circumference of the globe and extend through all the layers of the retina and similarly result from rotational acceleration and deceleration forces.

The mechanism of brain damage is problematic. Traditional wisdom has suggested shearing forces operating within the brain substance with consequent axonal damage.[6] Geddes et al, in a careful neuropathological study of head injuries in children using β amyloid precursor protein immunostaining, observed that the predominant changes in infants with evidence of shaking were hypoxic-ischaemic rather than the diffuse axonal injury seen in older children and adults with fatal head trauma.[7 8] These authors thought that acceleration and deceleration forces might damage the neuraxis to cause apnoea, with consequent ischaemic insult causing diffuse cerebral oedema.

Unfortunately, this logical idea was followed in a second paper by the statement, "Although mechanisms of

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