

## Problems of Infant Retino-Subdural Hemorrhage with Minimal External Injury

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This contribution is offered as a reflection - after 40 years' consideration - on a problem of potential child abuse which has caused a great deal of controversy since it was first described (Guthkelch 1971). While controversy is a normal and necessary part of scientific discourse, there has arisen a level of emotion and divisiveness on shaken baby syndrome/abusive head trauma that has interfered with our commitment to pursue the truth.

What follows is a Serious Call – I borrow the phrase from the title of a religious treatise by the 18<sup>th</sup> century Protestant clergyman, William Law - to members of the medical and legal professions *to consider these problems with restraint*. It is, in short, a call for civility in scientific discourse.

### **Problem #1. Is the name 'Shaken Baby Syndrome' justified by the facts?**

There is a serious epistemological difficulty here: one that seems not to have been clearly recognized. Of the several hundred syndromes in the medical literature, almost all are named either after their discoverer (e.g., Adie's Syndrome) or for a prominent clinical feature (e.g., Stiff Man Syndrome). In contrast, *the appellation shaken baby syndrome (SBS) asserts a unique etiology (shaking)*. This term also implies intent since it is difficult to 'accidentally' shake a baby. A newer term, abusive head trauma (AHT), implies both mechanism (trauma) and intent (abusive).

Since subdural and retinal hemorrhages (with or without cerebral edema) may also be observed in accidental or natural settings, *I suggest that the elements of the classic triad of retinal hemorrhage, subdural hemorrhage and cerebral edema would be better defined in terms of their medical features*. Perhaps "retino-subdural hemorrhage of infancy" would be an acceptable name for the primary findings. Other medical findings, e.g., cerebral edema, can be added to the title as appropriate. This would allow us to investigate causation without appearing to assume that we already know the answer.

### **Problem #2. Can shaking cause retino-subdural hemorrhage in infancy with minimal external injury? And, if so, can one reasonably infer shaking (or other forms of abuse) from these findings?**

If shaking is responsible for significant damage to the central nervous system and its coverings, one must ask why the forces generated by humans or laboratory machines shaking a dummy have so often proved insufficient to cause the disruption of these tissues. Uscinski (2004), quoting Ommaya (1968), reasoned that since there is "an injury threshold for neural tissue", and this cannot be attained by shaking alone, there must be an extra feature, such as impact, to explain the findings. This is consistent with the work of Duhaime and others. But this

argument leaves out of account the biophysical differences between the mature and the infant brain, recently stressed by Squier and Mack (2009). These include microscopic or submicroscopic damage, not only directly to the brain itself but also to the control of its blood vessels via the meningeal nerve-tissue. Such effects may be mediated by reflex mechanisms, causing infant susceptibility to hemorrhage from minor trauma or naturally occurring events. Such consequences would be difficult, if not impossible, to replicate in a laboratory model.

Since minor trauma may cause disproportionate harm to infants, it is appropriate to advise parents and caretakers not to shake babies, just as it is wise to advise them not to drop babies or to place them in positions from which they could fall or in which siblings or objects could fall on them. Such precautions recognize that babies are developmentally vulnerable, and that some may be more vulnerable than others.

It does not follow, however, that one can infer shaking (or any other form of abuse) from a finding of retino-subdural hemorrhage in infancy. Tuerkheimer has pointed out the danger of assuming criminal intent simply because the classic triad of retino-subdural hemorrhage and encephalopathy is present and no one can think of any other explanation. While society is rightly shocked by any assault on its weakest members and demands retribution, there seem to have been instances in which both medical science and the law have gone too far in hypothesizing and criminalizing alleged acts of violence in which the only evidence has been the presence of the classic triad or even just one or two of its elements. Often, there seems to have been inadequate inquiry into the possibility that the picture resulted from natural causes. In reviewing cases where the alleged assailant has continued to proclaim his/her innocence, I have been struck by the high proportion of those in which there was a significant history of previous illness or of abnormalities of structure and function of the nervous system, suggesting that the problem was natural or congenital, rather than abusive. Yet these matters were hardly, if at all, considered in the medical reports.

**Problem #3. Can we safely assume that there is a constant relationship between the force of shaking and/or impact and the resulting damage to body tissues, particularly those of the nervous system and its coverings?**

To raise this question does not suggest that *any* violence against a child is acceptable; it is not. But it *is* relevant in considering the extent to which a medical witness can quantify the degree of force required to cause specific medical findings, especially if the question invites a comparison to a high-speed motor vehicle crash or a fall from a high building or asks for an opinion on intent. Any medical expert who answers in the negative questions such as “Given the injuries that you have described in this case, doctor, have you any doubt that they were inflicted with intent to kill, or at least in total disregard of that possibility?” is exceeding his or her authority. New work by Squier and Mack on the neuropathology of the infant brain and its coverings emphasizes the complex relationships between the brain, the dura and the thin-walled bridging veins that lead from the cerebral cortex to the dural venous sinuses. We should not

expect to find an exact or constant relationship between the existence or extent of retino-subdural hemorrhage and the amount of force involved, let alone the state of mind of the perpetrator. Nor should we assume that these findings are caused by trauma, rather than natural causes.

In my 1953 article, I reviewed 24 cases of infantile subdural effusions, primarily subdural hematoma. In the article, I emphasized that infantile subdural effusions are not rare and that “the frequency with which [they] are found is proportional to the intensity with which they are sought.” Most of these cases occurred in the first few months of life (11 before age 3 months, 5 between 3 and 6 months, and 2 thereafter). Seventy-five percent were associated with abnormal or difficult labor, and two cases were associated with a head injury two weeks prior. Subdural effusions were also found in association with meningitis (5 cases) and venous sinus thrombosis, which may complicate any infective focus (1 case). Of those with known histories, there were two sets of twins and two premature babies. In 75% of the cases, the hematoma was surrounded by a membrane that was adherent to the dura, and in almost all instances the fluid obtained from subdural tap was xanthochromic, with a variable quantity of fresh blood, confirming that this process had been ongoing for some time, in many cases possibly from birth.

In my 1971 article, I described two of my own cases involving potential shaking – one in which the mother had shaken the child to clear his throat during a fit of coughing since she feared he might choke, and another in which the mother admitted she and her husband “might have” shaken the baby when he cried at night. I also mentioned an earlier case in which Caffey described a mother who grabbed the arm of a baby who was rolling off a table to prevent his fall, jerking him in the process. In none of these cases was there any apparent malicious intent or evidence that the shaking was severe. While these events may have triggered a subdural hemorrhage or rehemorrhage, it is unwarranted to go from this possibility to the assumption that unexplained subdural hemorrhages, with or without retinal hemorrhage or encephalopathy, are caused by violent shaking or other forms of abuse.

Today, advanced radiological imaging is finding more and more subdural hemorrhages, with one study indicating that 46% of asymptomatic infants have subdural hemorrhages following normal births. This percentage is likely higher in symptomatic infants, following difficult births, or on pathology. While most birth-related subdural hemorrhages appear to resolve without symptoms, the babies who later become symptomatic may be the ones in which the birth hemorrhages reflected more serious underlying damage or became chronic, developing membranes that were subject to rebleeding. In such cases, the focus should not be on finding a “culprit” – or blaming the last person with the baby – but rather on the early identification of babies with pre-existing conditions and the development of treatment options.

**Problem 4. Why is it important to get it right, and how should we proceed?**

When we make incorrect medical diagnoses, our advice and treatment is likely to be suboptimal or even harmful. This is particularly true in cases involving retino-subdural hemorrhage of infancy, where a wrong diagnosis may send innocent parents and caretakers to prison. Since the reported conviction rate in fatal child abuse cases is 88%, this is not a trivial concern. A recent study further notes that five separate reports have found rates of AHT in the U.S. that are approximately double those in Canada, the U.K. and New Zealand, a striking difference that is not obviously attributable to differences in the actual incidence of abuse and may reflect a lack of objectivity in diagnosis. My own experience in reviewing a dozen cases in which the alleged perpetrator continued to assert his/her innocence “through thick and thin” suggests that a less critical assessment of the data in difficult – and often imperfectly documented – cases may explain some of this disparity.

As medicine and science evolve, “getting it right” requires that we distinguish between hypotheses and knowledge. SBS and AHT are hypotheses that have been advanced to explain findings that are not yet fully understood. There is nothing wrong in advancing such hypotheses; this is how medicine and science progress. It *is* wrong, however, to fail to advise parents and courts that these are simply hypotheses, not proven medical or scientific facts, or to attack those who point out problems with these hypotheses or who advance alternative hypotheses. Often, “getting it right” simply means saying, clearly and unequivocally, “*we don’t know.*”

In evaluating individual cases, it is also important not to rely on statistics, alluring as that might seem. While statistics may be helpful when we are dealing with relationships between well-defined populations, this stage has not been achieved in the study of SBS/AHT. Instead, cases involving retino-subdural hemorrhage of infancy encompass varying age groups, genetic characteristics, underlying conditions and potential causes, including birth injuries, dehydration, metabolic disorders, illness and seizure disorders.

We must also consider the impact of even a relatively small percentage of misdiagnoses. Even if we knew that 90% of infant retino-subdural hemorrhages were caused by abuse, the assumption of abuse could result in false accusations or convictions involving a large number of innocent parents or caretakers (up to 1 in 10 of the accused). This figure will increase if many or most retino-subdural hemorrhages of infancy are attributable to accidents or natural causes, including birth injuries.

Given the importance of “getting it right,” the evidence base for SBS/AHT should be carefully and independently evaluated by scientists who are not involved in this controversy. Since the issue is not what the majority of doctors (or lawyers) think but rather what is supported by reliable scientific evidence, the evidence should be reviewed by individuals who have no personal stake in the matter, and who have a firm grounding in basic scientific principles, including the difference between hypotheses and evidence. It will not be easy to select such a group, but the effort will be justified in terms of justice and happiness.

In closing, may I be so bold as to suggest that as members of learned professions, we should never forget two dicta attributed to the great Canadian physician, William Osler, and his colleague Thomas McCrae? The first is this: "As is our pathology, so is our practice." The second is equally succinct: "More is missed by not looking than not knowing." Today, we need to develop a better understanding of the pathology of the infant brain and its coverings, and we need to look much more rigorously at the evidence. Only then will we be able to confidently correlate the medical findings with clinical symptoms and causes.

As CP Scott, the revered editor of the Manchester Guardian newspaper for most of the first half of the 20<sup>th</sup> century, used to say: "comment is free, facts are sacred." Over the past forty years, there has been much comment on retino-subdural hemorrhage in infancy, but we have not yet determined the facts. For that, we need new research on the pathology rather than a vain repetition of hypotheses. In obtaining a better understanding of the pathology, such research will assist in the earlier diagnosis and treatment of children; it will also serve the cause of justice.

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