CLINICAL STUDIES

The spectrum of postmortem ocular findings in victims of shaken baby syndrome

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ABSTRACT • RÉSUMÉ

Background: Ophthalmologists and ocular pathologists are called on to help identify children who have undergone violent shaking. The objective of this study was to describe the spectrum of postmortem ocular findings in victims of shaken baby syndrome and to correlate the ocular findings with the nonocular features found at autopsy.

Methods: The ocular pathology registry at the University of Ottawa Eye Institute was reviewed to identify all victims of fatal shaken baby syndrome whose eyes had been submitted for examination between Apr. 1, 1971, and Dec. 31, 1995. Autopsy reports were accessed from the hospital charts of the identified patients.

Results: Six patients, aged 1 to 34 months, were identified. Intraocular findings ranged from a focal globular hemorrhage at the posterior pole to extensive intraocular hemorrhage involving the entire retina with perimacular folds. All the children had evidence of optic nerve sheath hemorrhage. Nonocular findings included intracranial hemorrhage (in all cases), skull fracture (in two), rib fractures (in three) and high spinal cord hemorrhage (in four). The extent of the intraocular hemorrhage was not consistent with the nonocular findings.

Interpretation: Abused children may display a range of postmortem ocular findings, with intraocular hemorrhage varying from minimal to severe. These findings may not correlate with the severity of the child's other injuries. The presence of any retinal or optic nerve sheath hemorrhage in an infant, in the absence of an appropriate explanation for these findings, should raise suspicion of child abuse.

Contexte : Les ophtalmologistes et les pathologistes oculaires sont appelés à aider à repérer les enfants qui ont été violemment secoués. L'étude a pour objet de décrire la gamme des données oculaires post-mortem chez les victimes du

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syndrome du bébé secoué et d'en établir la corrélation avec les caractéristiques non oculaires trouvées à l'autopsie.

Méthodes : On a examiné le registre de la pathologie oculaire de l'Institut de l'œil de l'Université d'Ottawa pour reconnaître les victimes du syndrome funeste du bébé secoué dont les yeux avaient fait l'objet d'examens entre le 1er avril 1971 et le 31 décembre 1995. Les rapports d'autopsie ont été accessibles à partir des dossiers hospitaliers des patients.

Résultats : Six patients, âgés de 1 à 34 mois, ont été repérés. Les données intraoculaires variaient entre une hémorragie en foyer au pole postérieur et une hémorragie intraoculaire panrétinienne avec replis périmaculaires. Les enfants avaient tous des marques d'hémorragie de la gaine du nerf optique. Les données non oculaires comprenaient : hémorragie intracranienne (tous les cas), fracture du crâne (deux cas), fracture des côtes (trois cas) et hémorragie à la partie supérieure de la moelle épinière (quatre cas). L'étendue des hémorragies intraoculaires ne concordait pas avec les données non oculaires.

Interprétation : Les enfants victimes de violence peuvent présenter une gamme de données oculaires post-mortem, avec des hémorragies oculaires qui varient de minimes à graves. Ces données ne concordent peut-être pas avec la gravité des autres blessures de l'enfant. La présence de toute hémorragie de la rétine ou de la gaine du nerf optique chez un enfant, sans données explicatives pertinentes, devrait éveiller des soupçons de violence.

Ophthalmologists and ocular pathologists are called on to help identify children who have undergone violent shaking as a form of child abuse. These children often experience severe neurologic compromise and sometimes have dramatic intraocular findings, and they frequently die from their injuries. The diagnosis of shaken baby syndrome is commonly based on the clinical picture of subdural hematoma of the brain, occult bone fractures and retinal hemorrhages. It was not until the early 1970s that Caffey[1,2] first defined the clinical symptoms and signs of shaking injuries, although he had observed the association of these findings earlier. The term "shaken–impact syndrome" is preferred by some authors, as often impact is involved in the shaken baby syndrome.[3,4]

Between 1971 and 1995 the ocular pathology laboratory at the University of Ottawa Eye Institute reviewed six pairs of eyes of children whose death was considered to be secondary to shaken baby syndrome. The purpose of this study was to describe the spectrum of post-mortem ocular findings in these children. In addition, we reviewed the nonocular features found at autopsy to determine whether a pattern existed between the severity of the nonocular and the intraocular findings.

Methods

We reviewed the ocular pathology registry at the University of Ottawa Eye Institute to identify all autopsy eyes from victims of shaken baby syndrome that were received from Apr. 1, 1971, to Dec. 31, 1995. The diagnoses were made by general and ocular pathologists in the involved hospitals on the basis of the findings of subdural hematoma of the brain and retinal hemorrhages, frequently with occult bone fractures. The clinical details of the nonocular injuries of each child were obtained from autopsy reports supplied by the pathology department of the involved hospitals.

The eyes had been removed by deroofing the orbit and cutting the extraocular tissues, including the muscles. The globe and an attached lengthy segment of optic nerve were then placed in 10% formalin. Ocular specimens were studied by gross and microscopic examination.

Results

All the children were boys, with an age range of 1 to 34 months (Table 1). All the cases were considered by the investigating authorities to be secondary to child abuse, and all the alleged perpetrators were subsequently charged by the crown prosecutor; the verdicts are unknown. The alleged perpetrators included a babysitter in two cases, an undetermined parent in two cases, the father in one case and the stepfather in one case.

A history inconsistent with the child's injuries was obtained in three cases: a 34-month-old (patient 3) was said to have fallen from a high chair, a 14-month-old (patient 5) was said to have fallen down stairs and struck a refrigerator, and an 18-month-old (patient 6) was said to have fallen from a bed and struck an ashtray. The history in the remaining three cases was unclear. Often there was uncertainty as to how the injury occurred, as in most cases the alleged perpetrator denied his or her actions.[5]

Pathological findings

The nonocular pathological findings in each case are summarized in Table 1 and the ocular pathological findings in Table 2. Gross and microscopic ocular findings are shown in Fig. 1.

Intraocular findings ranged from a focal globular hemorrhage at the posterior pole to extensive intraocular hemorrhage involving the entire retina with perimacular folds (Fig. 1, A, B, C, D and E). In all cases there was evidence of optic nerve sheath hemorrhage (Fig. 1, F and G).

Nonocular findings included subdural and subarachnoid hemorrhage (in all the children), skull fracture (in two), rib fractures (in three) and high spinal cord hemorrhage (in four).

The concordance between the laterality of the intracranial findings, skull fractures and rib fractures and that of the ocular findings could not be established as no further information was available. However, it is interesting to note that in patient 3, a 34-month-old with known left intracranial hemorrhage, the retinal hemorrhage was greater on the left side than on the right. In general, the extent of the intraocular hemorrhage did not relate to the nonocular findings.

Table 1—Summary of the nonocular pathological findings in six boys who died as a result of shaken baby syndrome

Patient no.	Age, mo	External injuries	Intracranial hemorrhage	Spinal cord hemorrhage	Skull fractures	Rib fractures*	Viral illness
1	95	Multiple bruises over entire body	Small subdural and small subarachnoid hemorrhages (laterality unknown)	Subdural and subarachnoid hemorrhages	Occipital (laterality unknown)	No	Bilateral broncho-pneumonia on autopsy
2	91	Multiple bruises on face, both sides	Diffuse subdural and diffuse subarachnoid hemorrhages bilaterally	Subdural and subarachnoid hemorrhages	No	Yes	No
3	34	Multiple bruises on upper and lower limbs, buttocks, forehead and scalp, both sides	Diffuse left subdural and small left subarachnoid hemorrhages	No	No	No	Recent history of nonspecific viral illness
4	94	No	Diffuse subdural and small subarachnoid hemorrhages (laterality unknown)	Subdural hemorrhage	No	Yes	10-day history of viral gastro-enteritis
5	14	Bruising on chin and face, both sides	Small subdural and small subarachnoid hemorrhages bilaterally	Unknown	Parietal (laterality unknown)	Yes	Broncho-pneumonia on autopsy
6	18	Occipital and abdominal bruises bilaterally	Small subdural and small subarachnoid hemorrhages (laterality unknown)	Subdural and subarachnoid hemorrhages	No	No	No

*Number and location of fractures unknown.

Table 2—Summary of the ocular pathological findings in the six children*†

Patient no.	Intraocular: Retinal hemorrhages (zone)‡	Symmetry	Macular folds	Posterior vitreous detachment	Retinoschisis	Optic nerve: Length, mm	Intradural hemorrhage§	Subdural hemorrhage§	Subarachnoid hemorrhage§
1	1,2,3	L>R	Yes	Yes	Yes	R 16	2+	2+	2+
						L 12	1+	2+	1+
2	1,2	L=R	No	No	No	R 12	0	2+	1+
						L 17	0	2+	1+
3	1	L>R	No	Unknown	No	R 24	0	2+	1+
						L 12	0	1+	1+
4	1,2,3	R>L	Yes	Yes	No	R 14	0	1+	1+
						L 10	0	1+	1+
5	1,2,3	L=R	No	No	No	R 4	0	2+	1+
						L 4	0	2+	1+
6	1,2,3	R>L	Yes	Yes	No	R 17	3+	3+	2+
						L 19	0	3+	1+

*None of the children had intrascleral hemorrhages.
†L = left, R = right.
‡Grading of intraocular hemorrhages is based on the retinopathy of prematurity classification system:[6] 1 = imaginary circle whose radius is two times the distance from the disc to the macula; 2 = edge of zone 1 to a point tangential to the nasal ora serrata and round to an area near the temporal equator; 3 = residual temporal crescent anterior to zone 2.
§Grading of optic nerve sheath hemorrhages (optic nerve cut 4 mm posterior to sclera–optic nerve junction): 1+ = few erythrocytes; 2+ = erythrocytes present but tissue not saturated; 3+ = tissue saturated with erythrocytes.

Interpretation

When a young child is shaken, the relatively large head oscillates back and forth on the neck, which offers minimal resistance owing to the lack of development of the paraspinal musculature.[7] The acceleration–deceleration forces produced may be responsible for the characteristic findings of shaken baby syndrome: high spinal cord,[8] intracranial and intraocular hemorrhages,[1] and perimacular retinal folds.[9]

High spinal cord hemorrhages, seen in four of our patients, can be explained by compression of the cervical cord caused by rapid flexion and extension of the child's weak neck. Intracranial bleeding, present in all our cases, may be the result of shearing of blood vessels as the brain oscillates in the relatively large subarachnoid space of the young infant.[7,8]

Further systemic autopsy findings that are consistent with shaking include rib fractures, which are often bilateral, as in three of our cases. These are known as grasping fractures and represent the location where the child is held during the shaking episode.[10]

Intraocular hemorrhages were seen in all our patients. Several theories have been advanced to explain the presence of intraocular bleeding in victims of shaken baby syndrome. According to one theory, venous obstruction in the retina is secondary to increased intracranial pressure due to cerebral edema and subdural hemorrhage.[3,11] Terson's syndrome caused by subarachnoid hemorrhage and Purtscher's retinopathy associated with an acute rise in intrathoracic pressure as the child is being grasped about the rib cage may be contributing factors.[12–14]

Another theory postulates that vitreoretinal traction during the acceleration and deceleration of shaking–impact is important not only in the production of retinal hemorrhages but also in the formation of characteristic perimacular retinal folds.[3,15] The vitreous is known to be especially adherent at the ora serrata and optic nerve and in the perimacular area. In the presence of vigorous acceleration–deceleration forces, these firm attachments may explain the globular hemorrhages seen elevating the internal limiting membrane at the ora serrata and at the posterior pole. In addition, the fact that the vitreous can be seen as directly adherent to the crest of retinal folds, as in our case 1, implies that these same forces may be responsible for the folds' formation.

No intrascleral hemorrhages were observed in any of our patients. Kivlin and colleagues[3] also found that

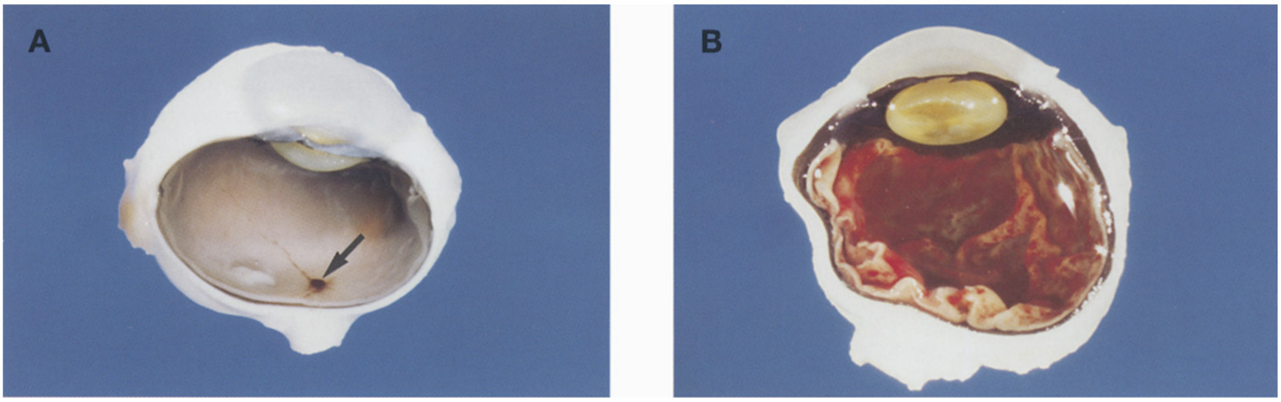
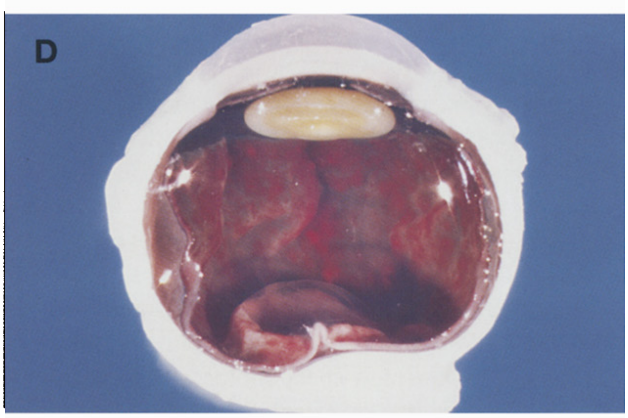
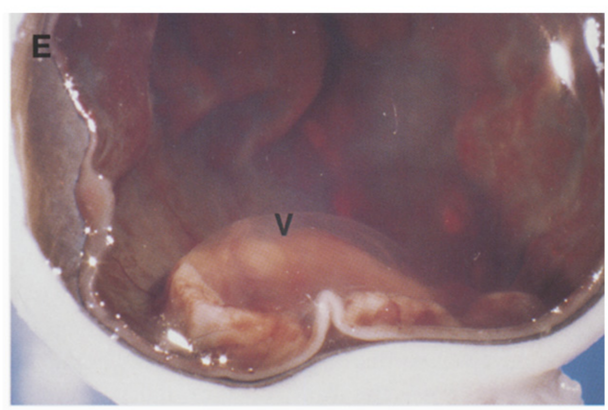
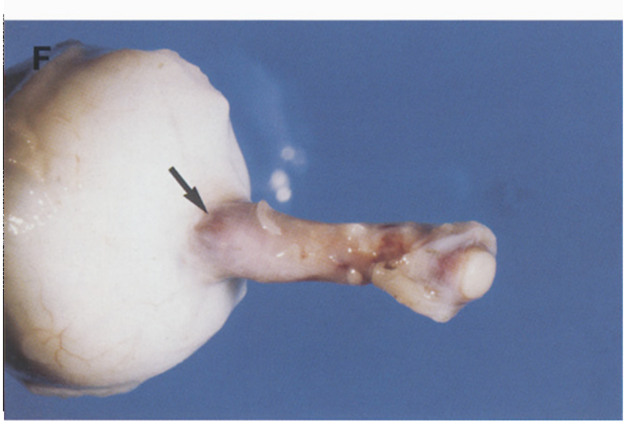
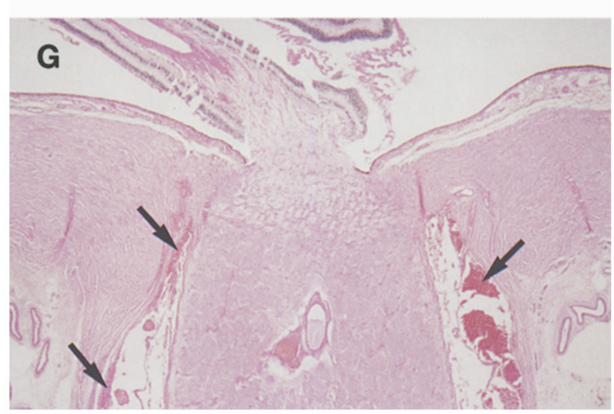


Fig. 1—**A, B:** Eyes removed from two infants, both with nonocular indications of being shaken. **A:** Case 2: Focal globular hemorrhage (arrow) over optic papilla in 1-month-old child. Note macular folding. **B:** Case 4: Diffuse retinal hemorrhages at all layers and macular folds in 4-month-old child. **C:** Case 5: Diffuse retinal hemorrhage (long arrows) to ora serrata with focal globular hemorrhage (short arrow) of nerve fibre layer in 14-month-old child (hematoxylin–eosin; original magnification ×2.5). **D:** Case 1: Eye from 5-month-old child who underwent shaking and direct head impact. Note diffuse retinal hemorrhages and prominent perimacular retinal folds. **E:** Case 1: Higher magnification of perimacular retinal folds shows dome-shaped posterior vitreous detachment (V) with vitreous still adherent to crests of folds. **F:** Case 1: Superior view of optic nerve and globe. Bluish discolouration (arrow), most prominent at sclera–optic nerve junction, represents subdural blood. **G:** Case 1: Subdural hemorrhage (arrows) (hematoxylin–eosin; original magnification ×2.5).









intrascleral hemorrhage was rare compared with other reported series.

Optic nerve sheath hemorrhages were seen to varying degrees in all our cases. Intradural, subdural and subarachnoid hemorrhages may be produced in a similar fashion as intracranial hemorrhages and are especially prominent at the sclera–optic nerve junction, where the nerve is tethered to the globe. Budenz and associates[16] suggested that optic nerve hemorrhages may be helpful in identifying children who have died due to injury. We found that the extent of these hemorrhages does not necessarily display any relation with the severity of the intraocular findings.

We attempted to link the nonocular features found at autopsy to the severity of the ocular findings. Five of our six patients had scalp bruising as evidence of direct head impact; two of the five had skull fractures. Unfortunately, laterality of the skull fractures and external injury could not be established. The remaining child (patient 4) had no external evidence of head injury yet showed dramatic intraocular findings. No history was provided in this case. However, if external injury was indeed absent, this contradicts previous suggestions that direct head impact is necessary to produce characteristic hemorrhages.[11,17]

The presence of retinal hemorrhages in both eyes is associated with a much worse prognosis for survival compared to unilateral retinal hemorrhages.[3] As all our patients had died from their injuries, it is not surprising that all six had bilateral retinal hemorrhages.

The youngest child in our series (patient 2, aged 1 month) had several classic findings of shaking, including grasping rib fractures and high spinal cord hemorrhage, yet showed minimal intraocular findings. This observation is consistent with other studies in which some children had minimal or no hemorrhages.[3,5] Thus, an absence of retinal hemorrhages should not rule out shaking as the mechanism of injury, even in a very young child.

The oldest child in our series (patient 3, aged 34 months) had numerous scalp bruises along with bruises on his upper and lower limbs. Globular intraocular hemorrhages were confined to the posterior pole. In this case, perhaps because of more developed paraspinal musculature, the child was better able to support his head against shaking. As a result, the shaking mechanism was decreased, which may account for the reduced intraocular findings.

Four of the six children had evidence of recent viral infections before the shaking episode, based on the clinical history and autopsy reports. A history of continuous crying before the shaking episode was obtained in several of our patients. Thus, the child may have been shaken out of frustration by a caregiver faced with an inconsolable infant. It is important to differentiate whether the signs and symptoms of the viral disease appeared before or after the shaking episode: in many cases the shaken baby presents with irritability, lethargy and vomiting and is commonly believed to have gastroenteritis or other infection because the history of injury is withheld.[3,7]

There are many causes of fundus hemorrhages in infants, including coagulopathies, other blood dyscrasias, severe hypertension, sepsis, endocarditis, meningitis and vasculitis.[18] Such conditions are much less likely to be confused with child abuse because of the systemic status of the child and lack of other injuries suggesting abuse.

Retinal hemorrhages occur after normal vaginal delivery in up to 40% of infants.[19] They typically appear at the posterior pole and resolve within a few days.

Purtscher's retinopathy can mimic the appearance of child abuse but would presumably be accompanied by a history of severe chest injury.[19]

A child with Terson's syndrome caused by subarachnoid hemorrhage may present findings similar to those in a deliberately injured infant.[12,19] Spontaneous subarachnoid hemorrhages are rare in infants but have been confused with child abuse.[20] The presence of vascular anomalies that could spontaneously rupture must be carefully excluded at autopsy.

Buys and coworkers[21] found that retinal hemorrhages were not present in children with accidental head injury who presented to the emergency department for assessment. Most of the patients, however, were not severely injured. An autopsy study showed that head injury associated with rapid deceleration, such as occurs in a high-speed motor vehicle accident, can cause retinal hemorrhages that include the retinal periphery.[22] In these cases the mechanism of injury was obviously not child abuse.

In conclusion, any retinal or optic nerve sheath hemorrhages in an infant without a systemic condition or history appropriate to explain them should raise a high index of suspicion for child abuse. The severity of the hemorrhage may be quite minimal and may not correlate with the severity of the child's other injuries.

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Key words: child abuse, head injury, infants, intraocular hemorrhage, optic nerve, perimacular folds, retinal hemorrhage, shaken baby, subdural hemorrhage, subarachnoid hemorrhage

Discussion

Dr. Marshall and colleagues bring to the medical literature six additional cases of shaken baby syndrome. The autopsy findings are consistent with much of what has previously been described in the now-extensive literature on the ocular manifestations of this disorder:[1] 1) the prevalence of retinal hemorrhages in victims who die approaches 100%; 2) although the severity of retinal hemorrhage may be wide-ranging, it is more common to see extensive retinal involvement out to the ora; 3) retinal hemorrhages may be asymmetric (and in other studies even unilateral); 4) macular folds and retinoschisis occur secondary to traction exerted by the shaking vitreous; and 5) optic nerve sheath hemorrhage is often found. There is new information as well. The authors note a remarkably high prevalence of signs or symptoms of viral illness that may have left children irritable and crying, factors that are well recognized to be an impetus for parental loss of control and violent outburst. The authors use what I believe to be a superior method for removal of the globe using an intracranial approach with unroofing of the orbit. Although I would prefer that the orbital tissues be removed en bloc to yield a better view of orbital tissue injury at histologic section, a posterior approach is necessary to identify the full extent of optic nerve sheath injury.[1] In particular, the presence of injury due to vitreous traction and optic nerve sheath pathology, especially the intradural hemorrhage, support the importance of the shaking acceleration–deceleration forces on the globe and orbit in the generation of hemorrhage. This has forensic significance. As reflected accurately in the article, we are moving away from theories that hypothesized that retinal hemorrhage is an indirect effect of systemic and intracranial events, and are moving increasingly toward the realization that these findings are indicative of the fact that shaking forces must have been applied. The results of studies conducted at our centre continue to reinforce this line of reasoning.[1]

Owing to the small sample, and in some instances the absence of data, the authors are unable to use statistical analysis to draw conclusions about possible positive or negative correlations between variables such as severity of ocular injury, severity of systemic injury, evidence of blunt head injury, optic nerve sheath hemorrhage, viral illness or rib fractures. They cannot tell us whether the laterality of the ocular injuries and that of the intracranial injuries are related. Yet they specifically note that in one case the asymmetry was ipsilateral, a statement made presumably to suggest the inference of a conclusion. They write, "In general, the extent of the intraocular hemorrhage did not relate to the nonocular findings." This assertion runs contrary to the literature and may mislead a casual reader to incorrect conclusions.[2,3] They show us elevation of the internal limiting membrane in Fig. 1, C, but in Table 2 say that patient 5 did not have retinoschisis. Elevation of the internal limiting membrane is indeed a form of retinoschisis,[4] an entity that has been reported only due to shaking in children in this age range. The importance of this semantic difference has enormous forensic significance. Likewise, there were children with no physical evidence of direct head impact. This does not prove a "shaking only" mechanism of injury. The impact may have been against a soft surface, such as a pillow, leaving no physical signs of impact but creating an acute elevation of the G-forces induced in acceleration–deceleration.[5] Shaking alone can injure and kill children with or without retinal hemorrhage.[1] But one might incorrectly ascribe a physical finding to shaking alone when indeed it was caused by impact. Such conclusions have been used in many courtrooms around the world to try to support or refute allegations of abuse. Five of the children had marked evidence of battering to their body, a finding that is uncharacteristic for shaken baby syndrome, in which external signs are usually absent. Perhaps this is a different subset of shaken children about whom forensic and scientific interpretation of the medical data should be considered not applicable to children who are shaken without being physically beaten.

I note these discrepancies and nuances not just to educate the reader about the ocular manifestations of shaken baby syndrome. There is another lesson here. Information is powerful. This power increases dramatically in arenas where there are higher-outcome stakes. Child abuse in one of those arenas. Information, in the form of scientific investigation and report, becomes a critical player in the adjudication of cases: whether an alleged perpetrator goes to jail or is set free, a family is reunited or torn apart, a child is protected or reintroduced into a potentially lethal environment. Although great strides are being made in understanding the ophthalmic manifestations of shaken baby syndrome, the lack of information in the past, prior reliance on theories that are now being turned aside and high stakes make it increasingly important that we use the scientific method of inquiry and strict attention to detail to ensure valid interpretations of data and correct application.

In the end, the authors' concluding paragraph is not supported by their data *even though it is true*. When reporting small case series we must guard against extrapolations without statistical underpinnings. Every case added to the literature, like these, has value to increase our pool of collective knowledge and raise new questions. But we must also work together as a medical community to take a purposeful and accurate application of the scientific method of inquiry to allow us to develop a meaningful understanding of the ocular findings of this disorder. This is especially true in the area of child abuse. Through multicentre collaborations, animal models, pathophysiologic experimentation, prospective randomized masked trials and observation of large numbers of cases we can ensure that the power of our information is best applied in the advocacy of children who may have been victimized.

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