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Evidence based case report

Perimacular retinal folds from childhood head trauma

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Editorials by Geddes and Plunkett and Harding et al

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A previously healthy 14 month old child was transferred to our medical centre with a severe head injury. The father had collected the boy and his 3 year old brother from their mother at his workplace car park and taken them home while their mother went to work. The children had been watching television while the father prepared dinner. After hearing something fall, the father found the boy on the floor with the television covering the right side of the head and anterior chest. A homemade television stand was partially across the child's lower legs. His older brother stated, "television fell." As soon as the father removed the television, he noticed the child's head beginning to swell. A neighbour drove them to the local hospital. According to the father and the neighbour, the child never stopped breathing and no resuscitative efforts were attempted.

Cranial computed tomography showed extensive head injuries. He had soft tissue swelling of the scalp, diffuse cerebral oedema with a subdural haematoma overlying the frontal convexities and layering along the falx cerebri, a left sided skull fracture adjacent to a widely diastatic coronal suture, cerebral contusions beneath the fracture, and a rightward midline shift measuring 8 mm. The paediatric ophthalmologist described bilateral dot and blot intraretinal haemorrhages, preretinal haemorrhages, and perimacular retinal folds (fig 1).

The child's condition deteriorated, and he died 18 hours after the incident. Child Protective Services removed the 3 year old sibling from the home because the retinal haemorrhages and retinal folds were considered diagnostic of abusive head trauma from shaking. This action was taken despite the father's repeated detailed, consistent account provided to emergency staff, the paediatric child abuse specialist, paediatric intensive care doctors, and law enforcement authorities.

Postmortem evidence

A forensic autopsy showed no direct trauma to the orbits or eyes. There were prominent bilateral scalp contusions with soft tissue and intramuscular haemorrhage, symmetrical parietal skull fractures with coronal sutural diastasis, and a lacerated dura mater with extrusion of brain and blood. In addition to bilateral subdural and subarachnoid haemorrhages, a thin epidural haematoma partially covered the frontoparietal, calvarial lamina interna. The brain showed bilateral cortical contusions, severe cerebral oedema, and diffuse anoxic-ischemic injury. Postmortem ocular examination showed haemorrhages of the optic nerve sheaths with subdural haemorrhage greater than subarachnoid haemorrhage. Both eyes had extensive retinal haemorrhages with perimacular retinal folds (fig 2). Retinoschisis and peripapillary intrascleral haemorrhages were evident, and the retinal haemorrhages extended from the posterior pole to the ora serrata affecting the preretinal, intraretinal, and subretinal layers.

When investigators went to the house to recover the television before the family returned home, it was still on the carpeted floor. The 480 mm screen television with built in videocassette recorder weighed 19.5 kg. The homemade television stand measured 762 mm (height)×635 mm (width)×508 mm (depth) and had a bottom drawer that held videotapes. A greasy smudged area on the glass of the television corresponded with the impact site on the child's head.

A re-enactment in which a 11.4 kg weight (similar to the child's weight at autopsy of 11.8 kg) was placed on the partially opened drawer caused the television and

Details of the included studies are on bmj.com

television stand to readily topple forward. According to investigators, the family home was 7.8 km from the workplace and about 6 km from the local hospital. Based on the distance and estimated driving times plus workplace time clock records, the father was home with the children about 20 minutes when the incident happened. The day after the incident, while in foster care, the 3 year old sibling corroborated the father's account. Despite all this evidence, the paediatric ophthalmologist repeated that perimacular retinal folds coincident with retinal haemorrhages were considered specific for shaken baby syndrome secondary to retinal traction exerted by the oscillating vitreous.

Search for published evidence

We were unable to find a published report of perimacular retinal folds in a childhood non-abusive head injury. We therefore did a systematic review of the medical literature on perimacular retinal folds associated with abusive head trauma in infants and young children. Our background question became: "In infants and young children with an acute intracranial injury, are perimacular retinal folds specific for head injury from vitreoretinal traction occurring during cycles of acceleration and deceleration (shaken baby syndrome)?"

We searched the Medline (1966-2003) database using the terms retinal folds and child abuse and uncovered seven non-comparative case series articles.¹⁻⁷ We also examined references cited in these articles plus review articles and book chapters on ocular findings in child abuse mentioning or discussing perimacular retinal folds relative to non-accidental head injury. Similar searches in the Cochrane Library, ISI Web of Science, and Ovid found no additional articles.

Results

We found 42 articles and book chapters discussing perimacular retinal folds in childhood abusive head trauma. Seventeen mentioned the presence of retinal folds in non-accidental head injury but did not comment on specificity or formative mechanism. A table on bmj.com gives details of the remaining articles. All but two of the articles are non-comparative clinical or autopsy case series, case reports, review articles, or book chapters.

The two studies that included controls both showed bias in selection of controls and contained no cases with perimacular retinal folds but discussed the postulated causal mechanism.^{8,9} In the prospective controlled study, the authors reported on 79 children younger than 3 years who had sustained head injuries.⁸ The manner of injury in one case was indeterminate. Three children, including one who died, had non-accidental head injury diagnosed, all of whom had retinal haemorrhages; 72 of the 75 children with non-abusive injuries were managed by observation alone. No perimacular retinal folds were observed; however, the presumed causative mechanism of traumatic retinoschisis and retinal folds was discussed.

The second controlled study was a prospective autopsy study that examined the presence and location of ocular findings in 169 childhood deaths.⁹ Ocular haemorrhages (retinal, peripheral retinal, optic nerve sheath and intrascleral) were more likely in craniocer-

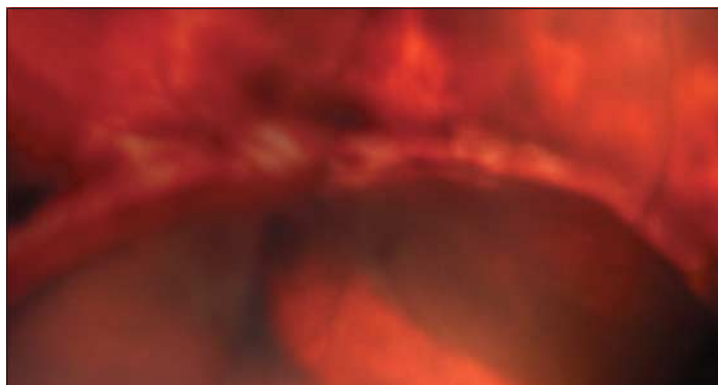


Fig 1 Clinical image highlighting temporal portion of perimacular retinal fold at 2-3 o'clock area in left eye with a blood vessel bending over the fold (magnification $\times 6$)

bral trauma than in non-head injuries and natural diseases. Although case selection was purportedly random, the study contained a disproportionately high number of deaths from child abuse compared with natural and non-abusive causes. Case selection depended on the pathologist's willingness to participate in the study, and we were told by one of the authors that pathologists were more willing to participate when they believed that the deaths were abusive or suspicious (M Gilliland, personal communication, 2002). Perimacular retinal folds were not noted, but the authors concluded that acceleration-deceleration injury to the retina accounts for peripheral retinal haemorrhages and retinal folds.

Supporting evidence

The references cited to support statements about the specificity or causal mechanism of perimacular retinal folds and abusive head injury in the articles we found are all non-comparative observational reports, unsystematic review articles, and book chapters. Seventy per cent of the articles cited four non-comparative case series.^{1,2,3,10} We assessed the quality of this evidence.

Gaynon et al reported on two infants with presumed shaken baby syndrome who had retinal folds and concluded that these folds may be a hallmark

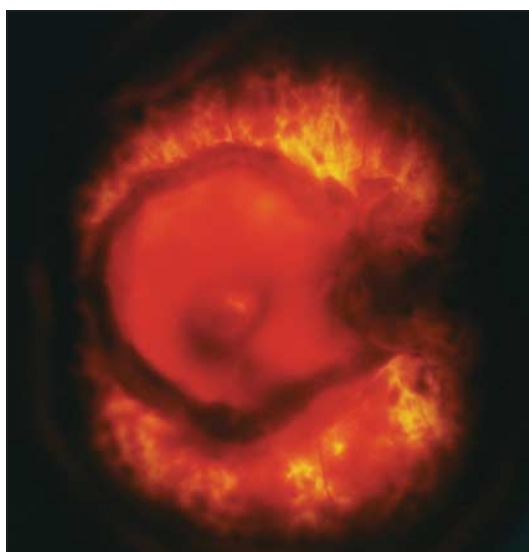


Fig 2 Transilluminated retinal image of right eye at autopsy showing circinate, elevated, perimacular retinal fold and extensive retinal haemorrhages

of shaking injuries in child abuse victims.¹ One infant reportedly fell 1.5 m to the floor while being carried down a stairway.

Massicotte et al reported the ocular findings at autopsy of three children with perimacular retinal folds.² Two infants had sustained direct head trauma, but in the other there was no physical or forensic evidence of direct head trauma. They observed that the vitreous had partially separated from the retina but remained attached to the internal limiting membrane at the apices of the folds and the vitreous base. They concluded that their study confirmed the role of vitreous traction in formation of perimacular folds and proved that shaking alone caused these folds and shaking was never an accidental phenomenon.

Elner et al reviewed the ocular and autopsy findings in 10 consecutive children who died of suspected child abuse.³ Perimacular retinal folds were observed in three children, all of whom had evidence of blunt head injuries.

Greenwald et al reported five cases of children in whom definite or probable physical abuse during infancy was associated with traumatic retinoschisis.¹⁰ They hypothesised that when an infant is shaken, the head is subjected to repetitive accelerations and decelerations causing the relatively dense lens to move forward and back within the ocular fluids. Transmission of force through firm attachments between the lens, vitreous gel, and particularly the macular retina presumably would result in appreciable traction on the retina causing it to split and creating the surrounding folds.

Discussion

Statements in the medical literature that perimacular retinal folds are diagnostic of shaken baby syndrome are not supported by objective scientific evidence. Non-comparative observational reports and unsystematic narrative review articles contain insufficient evidence to provide unbiased support for or against diagnostic specificity, and inferences about associations, causal or otherwise, cannot be determined. Clinical and autopsy evidence of ocular lesions must therefore be considered alongside other physical findings and a thorough investigation before concluding whether a head injury is caused by abuse. The child in our case had ocular haemorrhages (peripheral retinal, optic nerve sheath and intrascleral) and retinoschisis, which again some people consider specific for child abuse. Unfortunately, the evidence for these assumptions has similar problems to

that for perimacular retinal folds. An evidence based analysis of indexed medical publications on shaken baby syndrome from 1966-1998 uncovered a weak scientific evidence base.¹¹ Selection bias, inappropriate controls, and the lack of precise criteria for case definition were identified as important problems with the data. Many studies committed a fallacy of assumption, selecting cases by the presence of the clinical findings that were sought as diagnostically valid. Unsystematic reviews and consensus statements often mingled opinion with facts and added no original supporting evidence.

Perimacular retinal folds are associated with increased neurological morbidity and mortality in infants and children with abusive head injuries.⁶ The reported incidence of perimacular retinal folds in shaken baby syndrome varies from 6% in a consecutive clinical case series to 50% in a sequential autopsy case series.^{5,12} Clinical and autopsy studies with appropriately matched controls are needed to determine the causal mechanism of perimacular retinal folds and their specificity for abusive head injury. Until good evidence is available, we urge caution in interpreting eye findings out of context.

Contributors: PEL conceived the idea, collected the articles, and wrote the initial draft. All authors contributed to the review process, writing, and final editing of the paper. PEL is the guarantor. Competing interests: None declared.

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Table

Articles referring to diagnostic specificity or causal mechanism of perimacular retinal folds (PRF) in childhood non-accidental head injury

Reference	Publication date	Study type (No of cases)	Reference(s) cited re PRF
Gaynon MW et al ¹	1988	Clinical case series (2; 2 with PRF)	Case series ¹⁰
Han DP et al. ⁴	1990	Clinical case series (6; 2 with PRF)	Case series, ¹ Abstract ^{w18*}
Greenwald MJ ^{w1}	1990	Review article	Case series ^{1;3;10} Case report ^{w19} Abstract ^{w18}
Massicotte SJ et al ²	1991	Autopsy case series (3; 3 with PRF)	Case series ^{1;3}
Kaur B et al ^{w2}	1992	Review article	Case series ^{1;2}

Buy's YM et al ⁸	1992	Prospective clinical study; 0 with PRF	Case series ^{1;2}
Munger CE et al ⁷	1993	Autopsy case series (12; 5 with PRF)	Case series ^{1;10}
Keithahn MAZ et al. ^{w3}	1993	Clinical adult case series (2; 2 with PRF)	Case series ^{1;2;3}
AAP Committee on Child Abuse & Neglect ^{w4}	1993	Position paper – Review article	Case series ¹⁰ Review article ^{w12}
Gilliland MGF et al ⁹	1994	Prospective autopsy study; 0 with PRF	Case series ^{1;2;7}
Meier P et al ^{w5}	1996	Clinical case series (2; 2 with PRF)	Case series ^{1;2;w3}
Andrews AP et al ^{w6}	1996	Review article	Case series ^{1;2}
Rohrbach JM et al ^{w7}	1997	Autopsy case report	Case series ^{1;2;3;10}
Ellis PS ^{w8}	1997	Review article	Case series ^{1;2}
Mills M ⁶	1998	Clinical case series (10; 4 with PRF)	Case series ^{1;2}
Drack AV et al ^{w20}	1999	Clinical case series (4; 1 with PRF)	Case series ^{1;2}
Ophthalmology Child Abuse Working Group ^{w9}	1999	Consensus review article	Case series ^{1;2;3;7}
Levin AV ^{w10}	2000	Book chapter	Case series ^{1;2;6}
Taylor D ^{w11}	2000	Review article	Case report ^{w7} Case series ^{1;2;3;7}
Levin AV ^{w22}	2001	Book chapter	Case series ¹⁰ Book chapter ^{w10}
Kivlin JD ^{w21}	2001	Review article	Case series ^{1;2;6} Review article ^{w1;11}
AAP Committee on Child Abuse & Neglect ^{w13}	2001	Technical report – review article	Case series ¹⁰ Review article ^{w12}

			Book chapter ^{w10}
Nadel FM et al ^{w14}	2001	Case report; 0 with PRF	Case series ^{w15} Review article ^{w12;w16}
Marshall DH et al ⁵	2001	Autopsy case series (6; 3 with PRF)	Case series ^{1;2} Review article ¹¹
Levin AV ^{w17}	2002	Review article	Case series ²

PRF = perimacular retinal fold.

*Reference w18 could not be verified as cited.

Interpreting shaken baby evidence difficult

Report casts doubt on reliability of markers used to diagnose syndrome

By Susannah Benady

MONTREAL – Were the authors "misdirected" in their interpretation of the evidence when they reported the puzzling case of a 14-month-old baby in North Carolina who had suffered massive retinal hemorrhage after a television allegedly fell on his head at home?

Researchers from the Hospital for Sick Children in Toronto think so, after studying similar cases of 17 television tips-over. The child's injuries first appeared to be consistent with shaken baby syndrome, but after extensive investigation of the circumstances, the medical team decided the injuries were the result of an accident.

In reporting the case in the British Medical Journal earlier this year, researchers from Wake Forest University in Winston-Salem, N.C., suggested severe retinal hemorrhage and retinal folds could be seen in accidental crush injury. They then concluded that if investigators relied on the existence of retinal folds to diagnose shaken baby syndrome, it could result in caregivers being accused falsely.

"Statements in the medical literature that perimacular retinal folds are diagnostic of shaken baby syndrome are not supported by objective scientific evidence," wrote Dr. Patrick Lantz and co-investigators. In an editorial accompanying the article, forensic pathologist Dr. John Plunkett and brain damage expert Dr. Jennian Geddes argued that the finding suggested the criteria used to diagnose shaken baby syndrome ought to be reassessed.

The articles prompted a flurry of correspondence casting doubt on the reliability of the ophthalmological evidence used to diagnose the shaken syndrome. The result has been, particularly in the United Kingdom, that confidence in the diagnostic criteria is in danger of eroding.

But now, specialist Dr. Alex Levin, who also examined the histological evidence in this case, said the authors were "misdirected" in their interpretation of the clinical evidence and ended up drawing mistaken conclusions. Dr. Levin was a presenter here at the first North American Conference on Shaken Baby Syndrome.

This was creating an unwarranted backlash against solid evidence that has been building up over the past three decades, he warned the conference.

Dr. Levin, chairman of the International Advisory Board of the National Centre on Shaken Baby Syndrome, said while the case is puzzling, it is probably an outlier case and that general

extrapolations about shaken baby syndrome should not be drawn from it." After examining the histology on this case, I can tell you that while this child had a fold in the retina, it was not the typical kind you see in shaken baby syndrome," said Dr. Levin, a staff ophthalmologist at the Hospital for Sick Children in Toronto.

"The fold in this child's eye was very pointed. I've never seen anything like it so you cannot draw conclusions about shaken baby syndrome from it.

"Another puzzling aspect of the case is that the injuries the child suffered have never been reported in crush accidents, said Dr. Levin." I am puzzled by the evidence of severe retinal hemorrhage and perimacular folds in this child.

"These have never been reported in crush injury." The case prompted Dr. Levin and his team to set up a study to look at the type of retinal injuries children suffer in similar accidents. "We looked at the cases of 17 children at the Toronto Sick Kids' Hospital who had been injured by TV tips-over and none of them had retinal hemorrhage or folds," he said in an interview at the conference here.

Another set of crush cases from a centre in North Carolina headed by Dr. Mary Gilliland also failed to show findings similar to those reported by Dr. Lantz's group.

Dr. Levin said he believes the authors of the BMJ article asked the wrong questions. Their questions should have focused on the case itself, rather than using this, an idiosyncratic case, to cast doubt on decades of peer-reviewed evidence, said Dr. Levin." There are more than 900 papers and chapters in the medical literature that provide evidence relevant to an understanding of retinal hemorrhage. "On top of that, there is the radiology literature and outlier general literature on shaken baby syndrome.

"The authors should have asked the kind of questions researchers put forward in outlier cases, such as: Why is this child different from everything ever reported in the literature?" Either the authors have the wrong diagnosis or there is something special about this situation that sets this child apart," he said. "Either this child was not the victim of the alleged TV crush or there is something unique about this child to allow him to exhibit this unusual eye finding."