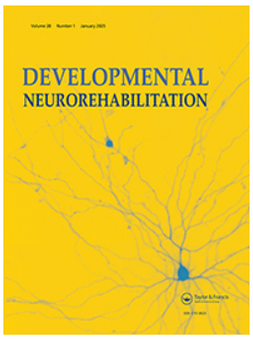

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The 'unified hypothesis' of Geddes *et al.* is not supported by the data*

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Keywords Infant head injury, non-accidental head injury, disability, subdural haemorrhage, retinal haemorrhage, cerebral hypoxia

Summary

Inflicted head injury to the developing brain frequently results in serious disability. The pathogenesis of the neuraxial and ocular findings in infants believed to have suffered inflicted head injury remains the subject of considerable debate. Recent neuropathology studies of fatal cases of inflicted head injury and of a foetal/perinatal non-traumatic model have led to the proposal that there is a 'unified hypothesis', the essential feature of which is hypoxic brain swelling secondary to cervicomedullary injury. It has been suggested that less than violent forces may be involved and even that some cases may not be due to trauma at all. The purpose of this paper is to provide a critical review of the data upon which these suppositions are based on a background of what is already known. It is submitted that there are serious flaws in the methodology; the conclusions reached cannot logically be drawn from the data; and the 'unified hypothesis' is not supported by the evidence. On the basis of the data presented, it is also difficult to sustain the secondary hypothesis purporting to describe a minority cohort with 'infantile encephalopathy with subdural and retinal bleeding' of non-traumatic causation.

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***A response by Geddes *et al.* to this article will be published in the next issue.**

Introduction

Inflicted head injury in infancy is associated with considerable neurodevelopmental disability in later childhood. Small institution-based studies have shown early death rates of 13% [1] and 14% [2]; a larger study drawn from tertiary and quaternary referrals had a death rate of 8% [3], whereas a population-based study of infantile subdural haemorrhage due principally to inflicted injury had a higher death rate of 27% [4]. Of greater concern is the repeated finding that only the minority appear to be left unscathed (table 1) and that disabilities are frequently multiple (table 2). Microcephaly and cerebral atrophy have been found in 94% and 53% of cases, respectively, and underline the need to foresee neuropsychological and language deficits [5]. It is almost certainly the case that the neuropsychological effects are underestimated in the early stages, especially in those who appear to have minimal or no physical sequelae. It is all too easy for this latter group to go undetected, only to surface later with learning difficulties and behavioural problems. Despite natural discomfort regarding the potential for misdiagnosis of child abuse, it is of greater concern that cases of inflicted head injury are very frequently missed. A population-based study [4] of infantile subdural haemorrhage revealed that 18% of infants had been previously admitted to hospital. In an institutional series [3, 6] of 120 children with suspected inflicted head injury, 98% of whom had subdural haemorrhages or haematomas, 52% had been seen previously at a hospital; of those previously seen, 60% had displayed similar symptoms at the first presentation as at the occasion upon which the diagnosis was made and 46% had shown evidence of trauma at the first presentation.

Concurrent or serial abuse in twins has also been encountered in the practice of three of the authors (J. Punt, T. Jaspan and N. McConachie, unpublished data 2004).

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Table 1 Neurodevelopmental outcome in children suffering inflicted head injury

Reference	*Number*	*Age (range)*	*Age (median)*	*Significant morbidity*
[1]	15	1–30 months	3 months	12
[3, 6]	120	3 days–143 weeks	14 weeks	66
[4]	33	3 weeks–17 months	3 months	15
[2]	25	3 weeks–2.8 years	5.8 months	15

Table 2 Pattern of disability following inflicted head injury in childhood

	References		
	[3, 6]	*[4]*	*[2]*
Number	120	33	25
Developmental delay (%)	34	—	—
Speech/language delay (%)	—	3	40
Motor/cerebral palsy (%)	33	21	52
Visual defect (%)	22	27	32
Hearing defect (%)	6	12	—
Seizure disorder (%)	16	24	16

Specialists in paediatric rehabilitation and neurodisability will encounter children who have suffered inflicted head injury in early childhood in a number of clinical and medico-legal settings. As well as playing an indispensable role in long-term management in the community, their expert opinions may be sought in the contexts of causation, prognosis, adoption and claims for relief under the Criminal Injuries Compensation Scheme. Some children may come to light at a later age as 'unexplained' cases of learning difficulties and behavioural disorders disrupting school and family life or even as cases of alleged clinical negligence. Awareness of current debate regarding causation is, therefore, important.

Although a general paediatrician with special responsibility takes the lead role in any child protection procedure, the complexities of the findings are such that it is only correct that they are addressed by specialists. The principal issues are those of timing, mechanism, force and the likelihood or otherwise of there being an innocent explanation for the medical findings. The articles of Geddes *et al*. [7–9] are of relevance to the last three matters. In view of the importance of the issues to the child and to the family and the contested nature of the proceedings, it is unsurprising and entirely appropriate that a debate has arisen. Publication of the scientific articles in 2001 and 2003 was associated with considerable media coverage that predicted major consequences in the field of child-care and medical jurisprudence [10–13].

The matter has reached further pre-eminence following the quashing on appeal of the convictions of Sally Clarke and Angela Cannings and the decision of the Attorney-General to review all convictions for fatal and non-fatal offences against children arising from alleged inflicted head injury [14].

This article addresses the basis of the suppositions advanced.

Discussion

The 'unified hypothesis' is explicitly declaimed in the abstract of the most recent publication [9]: an infant may suffer a craniocervical injury that causes respiratory disturbance leading to global cerebral hypoxia. The resultant hypoxia, brain swelling and raised central venous pressure causes subdural leakage of blood from intracranial veins and retinal haemorrhages. No impact or other 'considerable force' is required. It is suggested that, in some cases, there may not be any trauma at all.

This hypothesis challenges the most generally accepted position regarding the degree of force required to produce subdural bleeding and brain injury as expressed in a current international textbook of paediatric neurosurgery: namely, that 'while controversy still exists as to the exact mechanism, most authors now agree that the forces necessary to cause this type of injury are far from trivial and in fact are considerable', and 'that this sort of injury is unlikely to be inflicted "accidentally" by well-meaning caretakers who do not know that their behavior can be injurious' ([15], p. 375).

The supposition regarding the forces involved is central to the 'unified hypothesis'. It is, therefore, appropriate to visit the earlier study by Geddes *et al*. in depth. Unless otherwise stated, the concepts and commentaries discussed in this review refer to infants in the first 2 years of life, although some aspects may also apply to older children.

GEDDES *ET AL*. [7, 8]

The second of these two papers [8] is particularly relevant as it concerns babies and infants aged less than 9 months, the age group that is most frequently the subject of clinical, forensic and social investigation.

With regard to the degree of force required from shaking, it is the following contention that has given rise to most debate: 'nobody really knows how babies are injured, it may not be necessary to shake an infant very violently to produce stretch injury to its neuraxis'. The first statement is erroneous; the second is supposi-

tion that is not justified from the data. Both propositions require consideration.

'. . . NOBODY REALLY KNOWS HOW BABIES ARE INJURED. . .'

Those clinicians who have treated the whole range of accidental and inflicted head injuries in babies can draw upon considerable experience of both surviving and fatal cases in reaching an opinion regarding the forces required to produce harm. Evidence comes from three sources:

First, there is the negative evidence of what does not cause harm. Trivial head injuries are commonplace in the first 2 years of life, even in the best-regulated households. The outcomes from such trivial head injuries are almost always benign. This is the common experience not only of doctors, but also of parents. It is supported by a large population-based study [16] and by observation of children falling from heights of 3–4 feet whilst in hospital [17, 18].

Secondly, there is a smaller body of experience regarding the consequences of major accidental trauma in the first 2 years of life. Experience is much smaller for the simple reason that in the first 2 years of life major accidental head injuries are rare, because babies are not engaging in the type of behaviour that predisposes to major accidental head injury. In those cases that are encountered, it is quite clear from the histories and mechanisms of the injuries that major forces must have appertained and that the cause and nature of the accident are readily apparent [19, 20]. One much quoted study did not demonstrate abnormalities of the kind found in inflicted head injury in five children aged less than 2 years who died following falls from playground equipment [21].

Finally, in the course of clinical and forensic experience, cases are encountered in which, in non-adversarial settings, perpetrators have acknowledged how they have handled babies and infants who have suffered non-accidental injury. The methods of handling have always been described in ways that make it quite clear that the forces involved were considerable and represented self-evidently inappropriate and potentially damaging handling. It is noted that, in a statement to the lay press that appeared before publication of the papers in the scientific press, Geddes provided a significantly different version of the assertion, namely '. . . no one *except the perpetrator* [our emphasis] really knows how children are injured . . .' [10: p. 4, 11: p. 6].

Furthermore, the clinical presentations, radiological features and pathological findings are indistinguishable from those cases in which no acknowledgement is made [22, 23].

The contention by Geddes *et al*., as it appeared in the medical press [8], that 'nobody really knows how babies are injured' does not, therefore, correspond to the experience of those who care for babies in life, either as doctors or as parents.

'. . . IT MAY NOT BE NECESSARY TO SHAKE AN INFANT VERY VIOLENTLY TO PRODUCE STRETCH INJURY TO ITS NEURAXIS'

Geddes *et al*. [7, 8] performed the largest published study of the histopathological features of changes in the brain and spinal cord in 53 cases of fatal inflicted head injury in children. The essential finding was of evidence, in 13 out of the 37 (35%) cases, of microscopic damage to the lower brainstem and upper cervical spinal cord in babies aged less than 1 year who had died as a consequence of inflicted head injury. A second finding was the relative absence of microscopic changes in the brain of a type usually associated with diffuse traumatic brain injury.

This observation is central to the supposition that followed. The entire basis of the authors' hypothesis relies upon the detection of a particular pattern of immunohistochemical changes that the authors themselves acknowledge 'was not always easy'. The design of the study did not employ either any review of the material by an observer blinded to the clinical details or a system of double-reporting of slides or any similar consensus panel review. Concerns regarding this aspect of the study are not assuaged by sight of a number of pathology reports submitted in contested cases, subsequent to the publication of the learned articles, in which very experienced pathologists, including one of the authors of the learned articles in question, have been unable to distinguish between hypoxic and traumatic axonal injury with any degree of certainty. These difficulties have been encountered despite the very considerable experience gained whilst conducting the study of the 53 cases that comprise the core data of the hypothesis. In view of these persisting difficulties with interpretation, the apparent absence of any blinded review undermines the methodology of the work, the scientific validity of the fundamental observation and the credibility of the conclusions.

On the basis of their microscopic findings, Geddes *et al*. [8] hypothesized that babies aged less than 1 year who died as a result of inflicted head injury sustained fatal brain damage not as a direct consequence of trauma to the brain tissue, but as an indirect conse-

quence of failure of breathing secondary to damage to the lower brainstem and upper cervical spinal cord.

It should be noted, however, that of the 13 infants in their series with evidence of a neck injury, four were dead at presentation. Seven of the remaining nine (78%) presented with apnoea. Even if the four who died at presentation were assumed to have been apnoeic, the proportion only rises to 85%. Of the remaining 24 who did not suffer a neck injury, 20 (83%) had apnoea at presentation. Therefore, the likelihood of apnoea occurring in infants sustaining a putative stretch injury to the cervicomedullary axis is at least no greater than in those who did not sustain such an injury.

Geddes *et al.* ([8], p. 1299) cited the reporting of 'an episode of significant apnoea at presentation ... in 75% of cases' as corroborative evidence for their hypothesis. It is, however, also the case, all too familiar to clinicians, that many perpetrators allege that they just happened to find the baby in an apnoeic state or observed a previously healthy baby to suddenly become floppy with poor respiration. Frequently the baby has been crying or feeding normally, activities that are indicative of excellent respiratory and brain stem function, shortly before the change in health status. In those infants who rally to an extent, it is not unknown for crying to be described by a carer or observer after a putative causative event such as a low level fall. This is behaviour that is hardly consistent with lethal neurogenic apnoea. Geddes *et al.* do not suggest in their published articles that there may be a latent interval between the causative injury and the apnoea during which the child may be remotely normal. Both perpetrators and non-perpetrating carers may regard coma as sleep [22].

It is noteworthy that Geddes *et al.* ([8], p. 1300) were at pains to have available 'full documentation, including clinical histories and witness statements, ... for all cases'. This being the case, it is highly significant that there is no single case in which Geddes *et al.* [8] can relate a history of less than violent trauma (the cause) with fatal outcome (the effect). This is all the more remarkable given the forensic nature of the investigations, a setting in which trivial, everyday, minor injuries and events are very regularly proposed as the cause for severe or fatal outcomes.

Even in the three cases in which an infant was allegedly dropped and in the one case in which an infant allegedly fell, Geddes *et al.* [8] cannot show a connection between less than violent trauma and death. Both the senior author and the lead author have, individually and independently, acknowledged this understanding to be correct in response to open questions at national or international meetings held, respectively, in Nottingham on 1 March 2002 and in London on 29 November 2002. The injuries sustained by the three infants who were allegedly dropped included bilateral skull fractures (case 4); cerebral contusional tears, bilateral skull fractures, femoral and tibial fractures (case 18); skull and rib fractures (case 36) [8]. It would be frankly disingenuous to suggest that the one infant who was 'thrown' and who also had a fractured femur, had been subjected to anything less than violent handling.

There is considerable data to signal that these infants were actually victims of violent physical abuse. Geddes *et al.* [8] describe the principal details of the 37 babies studied ([8], see table 2). The data show the following:

- 15 out of 37 (41%) babies had serious injuries to other body parts (fractured ribs, fractured long bones, intra-abdominal injuries);
- 16 out of 37 (43%) babies had skull fractures and in six of these cases the skull fractures were bilateral; and
- 27 out of 37 (73%) babies had sub-scalp bruising.

These other injuries were all ones that equate very strongly with violent handling and, in respect of the skull fractures and sub-scalp bruising, indicate unequivocal evidence of forceful impact injuries. Four infants who had cerebral contusional tears, a lesion that is highly characteristic of inflicted head injury [24] also had rib fractures (case 1); bilateral skull fractures (case 4); bilateral skull and lower limb fractures (case 18) and rib and skull fractures (case 27).

In the light of this catalogue of other injuries, which can only have resulted from violence, is it logical or reasonable to surmise that the injury that actually led to the death of each of the babies was the consequence of anything but a violent act?

If fractures, soft tissue injuries and contusional tears are taken to be markers of trauma then the data show the following:

- 11 of 22 (50%) evaluable babies with no neck injury had one or more such markers; and
- 10 of 13 (77%) evaluable babies with neck injury had one or more such markers.

Geddes *et al.* do not indicate the cases with sub-scalp bruising in the individual patient data [8], but it is understood that only eight of the 37 had no markers of any trauma (G. Adams, personal communication 18 February 2004); the figures derived above may underestimate the proportion of children with neck injury and a marker of trauma. Accepting that the num-

bers are small, these figures suggest that those with neck injury had, as a group, suffered more physical abuse than those who did not have the neck injury and, intuitively, this would go against any suggestion that the neck injury indicated less than violent handling.

On the basis of the data contained within the learned article, it is very difficult to see how the authors' contention that less than violent handling could have been the cause of the deaths of the babies studied can be logically sustained.

Geddes herself has acknowledged that the manner of handling that she envisaged was such that an onlooker witnessing the act would think 'My God, what are they doing to that child? Stop' ([25], p. 41).

It is on this critical background that the more recent article [9] should now be addressed, rather than upon the implicit presumption that less than violent handling has been established as causative of fatal inflicted head injury.

GEDDES *ET AL.* [9]

In this paper, initially submitted in 2001 but published in revised form in 2003, Geddes *et al*. [9] correctly indicate that there are non-traumatic causes of subdural haemorrhage in foetuses, neonates and infants. It is noteworthy that the causes referenced are identifiable conditions, such as infections and disorders of coagulation. No cases due to unidentifiable or idiopathic pathological processes are referenced from the literature.

It is understood from remarks made by Geddes at an international meeting at which the work was presented that the index case that led to the research described in Geddes *et al*. [9] was that of a baby born at 25 weeks gestational age, who developed a severe infection secondary to chorioamnionitis and who died 1 week after birth. At post-mortem, witnessed by chance by Geddes, there was a large, unilateral, subdural haematoma.

As Geddes *et al*. [9] themselves state and as is well known to paediatric neurosurgeons and neuroradiologists, the subdural haemorrhages typically seen in infants suspected of having suffered severe inflicted head injury are, in the authors' own words, 'Characteristically spread over both hemispheres, often no more than a thin film . . . it does not require neurosurgical intervention; SDHs acting as mass lesions tend to be seen in older age groups' (p. 19).

The selection of the index case is, therefore, particularly strange, considering the statement that 'traumatic rupture of one or more bridging veins would be a more likely explanation of significant unilateral bleeds' (p. 20). The pattern of subdural haemorrhage in the only non-traumatic case in the series to have a macroscopic subdural haemorrhage is, therefore, not the pattern that the authors hypothesize would result from the mechanism they propose.

The choice of subjects without head injury comprised 17 intrauterine deaths, three spontaneous abortions, 21 perinatal or neonatal deaths and nine infant deaths. Few paediatricians would regard this cohort as being a good model for the hypothesis under investigation.

Apart from the aforementioned index case, of the 50 cases reported who had not suffered head injury, none had any subdural haemorrhage that was visible with the naked eye. In their hypothesis, Geddes *et al*. rely heavily upon the immaturity of the structures in displaying vulnerability to hypoxia, such that there is bleeding into the dura, which they postulate could reach the subdural space. It is, therefore, paradoxical that despite the immaturity of most of the cases without head injury that they studied, they have only observed one case of subdural haemorrhage that was visible with the naked eye. If the hypothesis was correct, then the most immature children would have been at greatest risk of experiencing actual subdural haemorrhage. This was clearly not the case as, the index case excepted, none had macroscopic subdural haemorrhage.

Geddes *et al*. [9] recorded the weight of the brains in their series at post-mortem. In 12 cases, death occurred *in utero* and the brains were macerated. These were excluded from consideration. Of the remaining 38 cases, 37 brains (97%) were either normal or below normal expected weight. Only one brain (3%) was increased in weight to a level that the authors' regarded as causing raised intracranial pressure. In this case the cause of death was not ascertained. Thus, in the vast majority of their cases, severe brain swelling was absent and, therefore, could not be implicated as the cause of the intradural haemorrhages. This fact undermines the hypothesis that apnoea leads to severe hypoxia, brain swelling, raised intracranial pressure and then subdural and retinal bleeding. It serves to emphasize the difference between the cases in this study and those in the earlier study in which brain weight was increased in 82% of cases in which it was measured [7].

The hypothesis regarding the causation of subdural haemorrhage is further contradicted by the very large, albeit much older, series of Towbin [26] encompassing autopsy material from over 600 infants, including stillborn and live born infants. The pattern of intracranial haemorrhage was quite clearly related to the cause. Subdural, brainstem and spinal cord haemorrhage was

caused by trauma, whereas intraventricular and subarachnoid haemorrhage was caused by hypoxia. No relationship was found between subdural haemorrhage and hypoxia or between spinal haemorrhage and hypoxia.

The chapter in an international textbook of foetal and neonatal neurology and neurosurgery concerning the brain pathology of hypoxic-ischaemic injury [27] makes no reference to subdural haemorrhage as part of the pattern of intracranial response to hypoxic-ischaemic insult at any age from 0–40 weeks gestational age.

In the extensive experience of the neuroradiology co-authors of neonatal non-traumatic hypoxic-ischaemic insults, multiple supratentorial and infratentorial subdural haematomas are extremely rare. Furthermore, of those infants referred for neuroimaging following near miss or fatal cot deaths in which a traumatic aetiology has been excluded, subdural haematomas are notably absent (figure 1).

Geddes *et al*. [9] cite three case reports of intrauterine subdural haemorrhage [28–30]. In none of these cases was hypoxia thought to be implicated and, in extensive reviews of the existing literature, none of the three reports cited a case of foetal subdural haemorrhage due to hypoxia, although one alluded to hypoxia as a proposed mechanism.

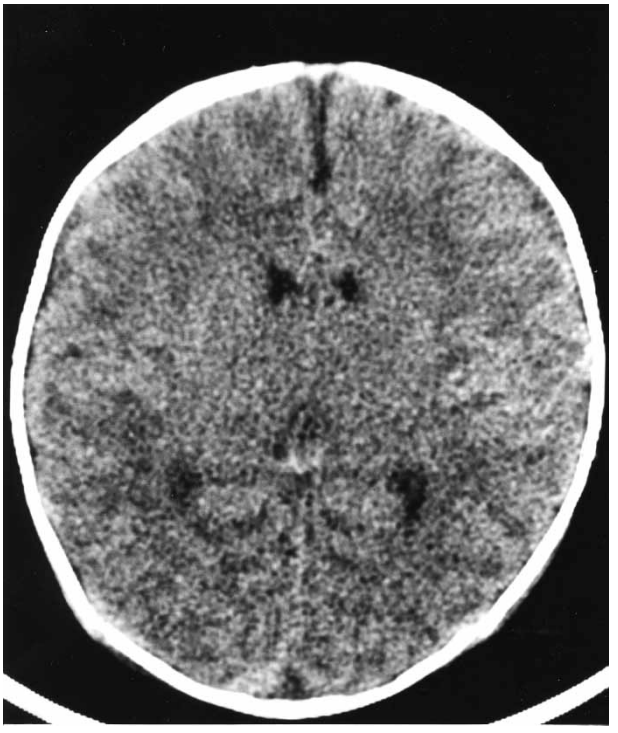

Figure 1 34 day-old male infant admitted following an acute life threatening event. Axial CT scan demonstrates loss of definition and hypodensity of the deep gray matter nuclei and subtle global hypodensity but no extra-axial haemorrhage or focal parenchymal injury. The infant died shortly after the scan. Post-mortem revealed global hypoxic-ischaemic change but no evidence of trauma.

It is, therefore, apparent that not only is there a paucity of evidence from their own series to support actual subdural haemorrhage as a consequence of hypoxic-ischaemic insult in babies and infants, there is substantial established experience to indicate that macroscopic subdural haemorrhage may not occur at all under the circumstances suggested by Geddes *et al*. [9].

It is the common experience of paediatric neurosurgeons and neuroradiologists examining live children who may have suffered inflicted head injury that subdural haemorrhage is frequently apparent on imaging before the appearance of any hypoxic-ischaemic changes in the brain tissue. By contrast, subdural haemorrhages are not a feature of cases of severe brain swelling of non-traumatic origin in the absence of coagulopathy or, possibly, severe hypernatraemia. Subdural haemorrhage is not a feature of even the most severely raised intracranial pressure due to non-traumatic intracranial mass lesions or severe hydrocephalus, even though at open operation such infants may have massively enlarged venous drainage systems. This provides further evidence that there is no causative link between brain swelling, raised intracranial pressure and subdural haemorrhage in infants.

Furthermore, cases of subdural haemorrhage in multiple sites due to undoubted inflicted head injury are frequently encountered in which there has been no or no major clinical or radiological evidence of hypoxic-ischaemic encephalopathy at any stage in the history. Brain swelling is not universal and raised intracranial pressure was absent in seven out of 37 fatal cases (19%) studied by Geddes *et al*. [8]. Those who care for such children in life encounter a very broad clinical spectrum from the mild and deceptively non-specific transient upset, to the child who requires full multi-organ support on an intensive care unit. In a study of 63 infants aged less than 2 years who had subdural haemorrhages, 85% being cases of inflicted head injury, only 16 had brain swelling on CT or MR. Furthermore, of 21 cases which displayed apnoea, only nine had swelling on CT or MR, compared to seven out of 42 cases who had not displayed apnoea [31]. This suggests that hypoxic-ischaemic encephalopathy is simply one feature in a variable spectrum that includes subdural and retinal haemorrhages, rather than a fundamental causative factor. If the whole range

of the condition is considered, those cases presenting with multiple subdural haematomas but no hypoxic-ischaemic encephalopathy are actually much more common than those presenting with hypoxic-ischaemic encephalopathy.

The aforementioned observations provide strong evidence that hypoxic-ischaemic encephalopathy is not necessary for the production of subdural haemorrhage and that in cases in which there is hypoxic-ischaemic encephalopathy of traumatic origin, subdural haemorrhage actually precedes the hypoxic-ischaemic changes.

Geddes *et al*. [9] describe the findings on microscopic examination of three infants from their earlier series [8] who died following inflicted head injury. All three cases had retinal haemorrhage and bilateral subdural haemorrhage, the latter having been evident on pre-mortem scans and confirmed at post-mortem. None of the cases had any evidence of impact injury in the form of skull fracture; for reasons given above it is not known whether they did or did not have sub-scalp bruising.

Geddes *et al*. regard their finding of the microscopic appearances in the dura in these cases as supporting their hypothesis that hypoxia can cause bleeding from the dura and, thereby, the formation of the subdural haemorrhages.

Geddes *et al*. have not cross-referenced the cases from the previous study, but, from the details given, it would appear that the three cases reported were case numbers 24, 25 and 28 ([8], table 2). On the premise that this deduction is correct, it is appropriate to examine the details of those three cases as follows:

- *Case 24*: This baby was one of three (cases 3, 13, 24) in the series aged 7 weeks at the time of injury. It is noteworthy that the other two had features that were indicative of them having suffered extreme violence. Case 3 had allegedly been thrown and was found to have a recent fractured femur and bilateral skull fractures. Case 13 had old rib fractures and a skull fracture. Cases 3, 13 and 24 all had raised intracranial pressure, acute subdural haemorrhage, vascular axonal injury and global hypoxia-ischaemia. It would, therefore, appear that case 24 had a pattern of intracranial injury for which there was no apparent innocent explanation, but that was very similar or even identical to the two infants injured at the same age who had unequivocal evidence of major inflicted violent trauma. It would appear almost perverse to suggest that case 24 had died from an unidentifiable natural disease process or as a consequence of a less than violent act.
- *Case 25*: This baby, injured at the age of 5 weeks, had old rib fractures, a finding that correlates strongly with inflicted violence, and also had intracranial haemorrhage (ICH) separate from subdural haemorrhage. Not only can there be no reasonable doubt that this baby was the victim of an inflicted head injury, but the pattern of intracranial haemorrhage was not that of isolated subdural haemorrhage. Geddes *et al*. [9] provide no explanation for how the haemorrhage characterized as ICH could have arisen from the dura.
- *Case 28*: This baby, who was injured at the age of 8 months, had a subdural haemorrhage that was regarded as being of a size that it constituted an intracranial space-occupying lesion. It is, therefore, difficult to see how it was concluded that the baby died from the effect of hypoxic-ischaemic brain swelling, rather than from the effect of the space-occupying lesion. In any event, a subdural haematoma of a size that constituted a space-occupying lesion is clearly not the same as the thin layer of subdural blood frequently seen in multiple sites in babies suffering inflicted head injury. Geddes *et al*. [9] acknowledge that significant unilateral bleeds are 'more likely' to arise from traumatic rupture of bridging veins. Geddes *et al*. [9] make no observation that the dura examined was adjacent to the site of the subdural haemorrhage that constituted the mass lesion.

Crucially, none of the three aforementioned cases had the neck injury that formed the basis of the supposition that 'it may not be necessary to shake an infant very violently to produce stretch injury to its neuraxis' ([8], p. 1305). The absence of the neck injury must render the 'unified hypothesis' unsupportable, at least on the basis of any of the evidence provided.

Geddes *et al*. [9] propose a new, alternative, hypothesis that some cases might not have had a 'head injury at all' and that 'subdural and retinal bleeding might result from any event that initiated apnoea or significant hypoxia, with brain swelling' (p. 20). The absence of neck injury in the remaining two-thirds of cases might support the proposition. Only approximately one-third of the cases in the earlier series [8] had evidence of traumatic neural damage in the medulla or spinal cord. Geddes *et al*. do not seek to rely upon this observation and, indeed, the data are not supportive. If there was to be evidence in support of the sup-

position, it would be found in the cases who had no markers of trauma.

Analysis of the data shows that only seven out of 22 (32%) babies with no neck injury, who had thin acute subdural haematomas, but no markers of trauma, had evidence of raised intracranial pressure. As raised intracranial pressure is an indispensable link in the cascade of events proposed by Geddes *et al*., its presence in only one-third of those in whom it might be reasonable to consider a non-traumatic aetiology makes the additional hypothesis distinctly improbable.

The 'unified hypothesis' cannot explain the intraspinal, extradural or intradural haemorrhages that are not infrequently found in fatal cases of suspected inflicted head injury and which correlate strongly with optic nerve sheath haemorrhages [32].

Geddes *et al*. state that 'Most infants with inflicted head injury have severe hypoxic brain damage and rapidly develop grossly raised intracranial pressure from secondary brain swelling, which is documented on a scan taken on arrival at hospital and confirmed at post-mortem by markedly increased brain weight' (p. 19). Death occurring shortly after presentation occurs in 8–30% of cases [1, 3, 4]. The majority survive (figure 2) and, although most of those with subdural haemorrhages have significant neurodevelopmental disabilities, they do not suffer severe hypoxic-ischaemic damage or global cerebral swelling as judged by imaging in the acute phase. In those who do have parenchymal changes evident on CT scans obtained in the first few hours after the onset of encephalopathy or known time of injury, the cerebral swelling and hypoxic-ischaemic damage is often minimal or subtle. In many of these cases, fulminant changes develop on subsequent scans.

One of the commonest scenarios seen in life is the infant in whom imaging made soon after presentation demonstrates thin films of subdural haematomas and little or no brain swelling or hypoxic-ischaemic injury. Subsequent scanning shows little or no increase in size of the subdural haematomas, even in those who go on to develop hypoxic-ischaemic changes (figure 2). Retinal haemorrhages, when present, are usually unaccompanied by or associated with only minimal, optic disc swelling. Ophthalmologic follow-up in these cases almost invariably shows rapid or progressive resolution of the retinal haemorrhages.

Geddes *et al*. [9] includes co-authors from other disciplines: Adams (a paediatric ophthalmologist), and Tasker (a paediatric intensivist and respiratory physician). It is unclear from the paper what contribution they may have made, whether as the treating doctors

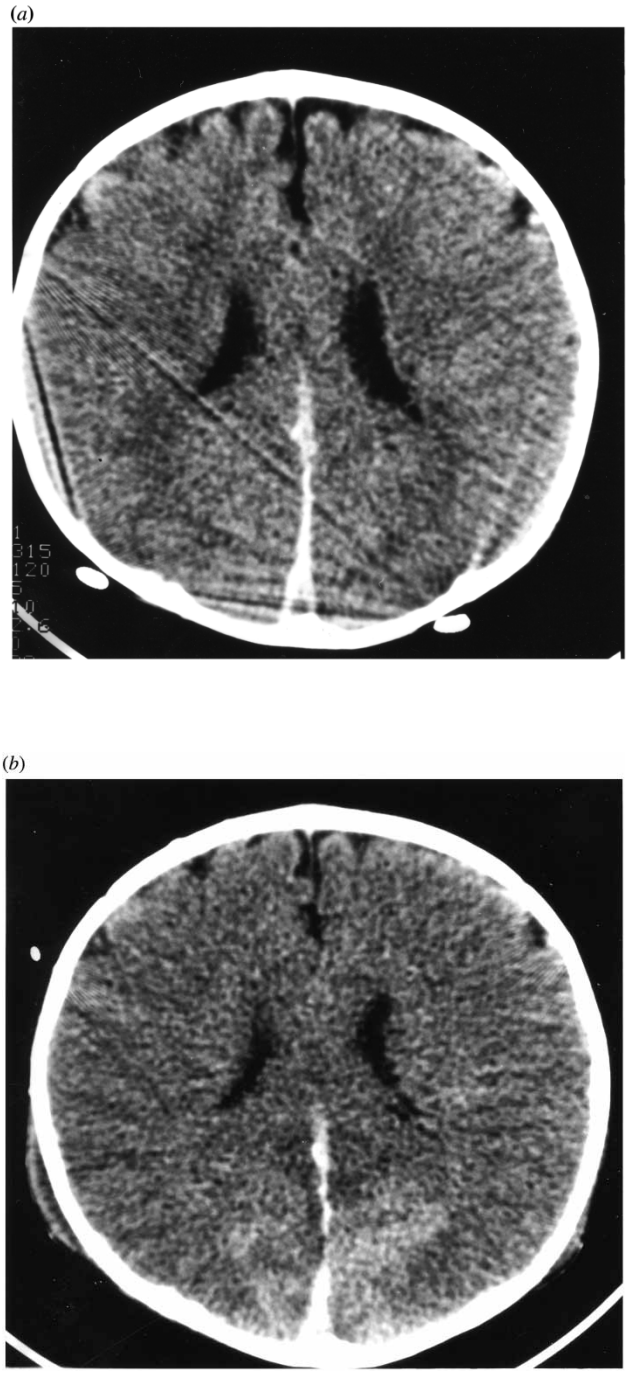

Figure 2 (*a*) 9 month old male admitted with acute encephalopathy and decerebrate posturing. CT scanning within 16 hours of presentation demonstrates acute bilateral posterior parafalcine subdural haematomas and early cerebral oedema. Note the preservation of cerebral sulci anteriorly. (*b*) CT scan 5 days later shows static size and appearance of the subdural haematomas. Global cerebral swelling and hypodensity has developed with effacement of the cerebral sulci. The right sided scalp swelling relates to a previously inserted scalp cannula. (*c*) (opposite page) Follow-up CT scan 2 months later demonstrates end stage hypoxic-ischaemic injury with development of large subdural collections secondary to cerebral atrophy.

(*c*)

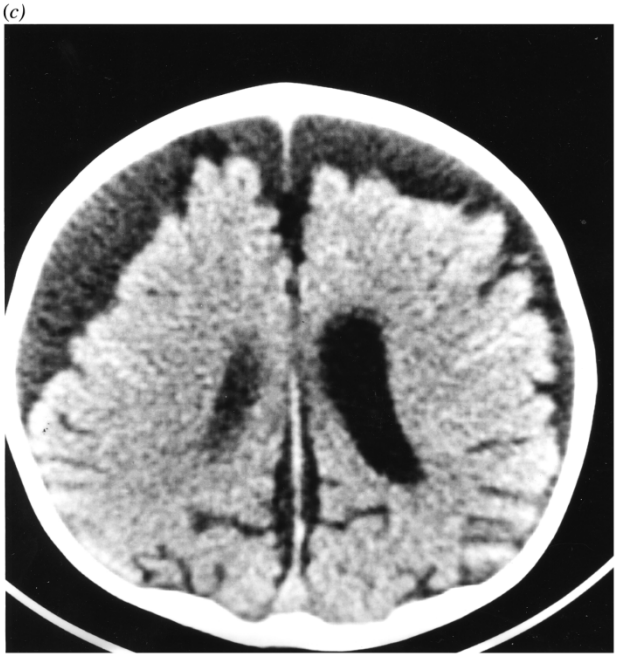

Figure 2 Continued.

of the children in any of the studies or by way of purely intellectual input. In neither of the authors' studies have the eyes been examined post-mortem and in the earlier study [7] the authors stated that 'the aetiology of retinal haemorrhages . . . is beyond the scope of this paper' (p. 1298). There is, therefore, no new ophthalmic pathology data from which conclusions can be drawn or that permits incorporation of retinal haemorrhages into the 'unified hypothesis'.

Others have found little correlation between raised intracranial pressure and retinal haemorrhage in children suffering inflicted head injury [33]. Tasker [34–36] has considerable experience in the fields of paediatric respiratory medicine, intensive care and the physiology of raised intracranial pressure.

Geddes *et al*. put considerable emphasis on the role of raised intracranial pressure in the evolution of the events that constitute their 'unified hypothesis'. The methodology employed for identifying the presence of raised intracranial pressure is, however, questionable. There is no reference to any intracranial pressure monitoring that may have been made in life in any of the cases in their first two papers [7, 8] and intracranial hypertension or its absence seems to have been assumed on the basis of the presence or absence of increased brain weight. This is hardly robust evidence. Tasker has previously commented, correctly, that 'invasive measurement may be the only means of recognizing this complication [raised intracranial pressure]' [36: p. 47]. Tasker [35] has also observed that there is an inconstant relationship between brain swelling, as judged by obliteration of subarachnoid spaces, and intracranial pressure in non-traumatic coma.

It is suggested that bleeding that might arise from dura and from dural veins damaged by the proposed hypoxia could be aggravated by 'systemic arterial hypertension, both sustained and episodic, that is commonly documented in children with raised intracranial pressure' ([9], p. 19). This proposal is flawed.

Geddes *et al*. [7–9] frequently allude to severe hypoxia as the consequence of apnoea and the causative agent for subsequent brain swelling, raised intracranial pressure and, thence, subdural and retinal bleeding. It is highly unlikely that the diffuse hypodensity seen on CT scans in severely injured infants represents the consequence of a pure hypoxic insult. Experience from imaging infants in life indicates that brain injury is more readily explained on the basis of ischaemia than hypoxia. One series of 14 children [37] confirmed as suffering from inflicted head injury showed either diffuse or focal cerebral hypodensity on computerized tomography (CT). Those children showing diffuse changes were significantly younger than those showing focal changes (mean age 5 months vs mean age 19 months). However, the appearances in both groups were of ischaemia rather than hypoxia. The authors commented upon the possible role of age-related pathophysiological responses and that the imaging in the very young infant probably reflects a global ischaemic insult (i.e. global hypoperfusion).

It is almost uniformly the case that those infants who have the apnoea upon which the 'unified hypothesis' relies also have circulatory collapse. This would be expected, as the regions of the central nervous system that are involved in the control of breathing are also involved in the control of circulation and cardiac function. Following admission to hospital, the most seriously injured children frequently require not only respiratory support, but also cardiac resuscitation and inotropic support to maintain a reasonable blood pressure. They are frequently submitted to repeat imaging and, despite the restitution of an adequate circulation and even a pre-terminal period of hypertension, no further subdural bleeding is seen on imaging or found at autopsy. The suggestion that systemic hypertension may be causative in the formation of the subdural haemorrhages is, therefore, incorrect.

Extensive experience gained from fatal and non-fatal cases indicates that the subdural haemorrhages are well formed very soon after admission to hospital,

as demonstrated on imaging. In particular, the acute subdural haemorrhages do not increase in size, even though the hypoxic-ischaemic changes appear and evolve. They may, in the future, go on to produce chronic subdural collections in some surviving cases through different mechanisms. The subdural bleeding, therefore, occurs very early in the condition, whilst the infant is still in a state of cardiovascular collapse, indicating a temporal sequence that is contrary to the proposed causative role of the hypoxia.

In the minority of infants with subdural haematomas that require surgical evacuation at open operation due to their space occupying effect, neurosurgical experience is that ruptured or avulsed bridging veins are encountered in these collections at craniotomy. This physical evidence of traumatic disruption of these veins is in contrast to the theory of Geddes *et al.* that 'severe brain swelling with venous congestion would produce widespread ''oozing'' from leaky hypoxic dural veins, possibly with a contribution from similarly leaky bridging veins, and that this is responsible for the typical thin film or patchy collections of subdural blood' ([9], p. 19).

Tasker has conducted research and published in the areas of the CT scan appearances in raised intracranial pressure of non-traumatic origin [35] and intracranial pressure monitoring in non-traumatic coma [34, 36]. None of these papers contains any reference to subdural haemorrhages or to retinal haemorrhages. It could reasonably be expected that, given his very extensive experience of non-traumatic coma, if any experience of subdural haemorrhages or retinal haemorrhages in this setting had come to light, it would have been alluded to in his contribution to Geddes *et al.* [9]. Such observations would have attained a new and greater significance which would have been supportive of the 'unified hypothesis'. None of Tasker's extensive and pertinent contribution to the literature appears in the reference list. As it stands, it would appear that Geddes *et al.* [9] fails to make full use of Tasker's published experiences.

The general absence of subdural haemorrhages in children with non-traumatic encephalopathy, even in the presence of severe changes on CT, has been noted by others. In a comparative retrospective study of 73 children (mean age of boys 12 months; mean age of girls 18 months) showing cerebral oedema or 'reversal sign' on CT, all of 46 children who had suffered inflicted head injury had subdural haemorrhages, but none of 26 who had identifiable non-traumatic pathologies had subdural haemorrhages [38].

A population-based study of non-fatal episodes of apnoea and lifelessness covering 38.5% of all live-born children in Sweden over a 2-year period did not report any cases of subdural haemorrhage or retinal haemorrhage [39].

Geddes *et al.* [9] are at pains to indicate that the 'unified hypothesis' could only apply to those 'isolated cases in which there is nothing apart from subdural and retinal bleeding to substantiate an allegation of inflicted trauma' (p. 20). The logical inference is that those with other evidence of trauma are not candidates, thus eliminating ~78% of cases. However, as only one-third of even the fatal cases have the cervicomedullary lesion demonstrated by Geddes *et al.*, the hypothetical population reduces further to ~7% at the very most.

For the 'unified hypothesis' to be tenable on the data presented, it would be expected that there would be some children who possessed all of the following attributes:

- the neck injury on microscopic examination,
- raised intracranial pressure,
- thin subdural haemorrhages,
- retinal haemorrhages, and
- absence of markers of trauma.

None of 13 (0%) babies displayed this combination. The authors have found no 'isolated cases in which there is nothing apart from subdural and retinal bleeding to substantiate an allegation of inflicted trauma' ([9], p. 20). By their own criteria, the 'unified hypothesis' is unsupportable on the basis of the authors' data.

Geddes *et al.* present no cogent argument for the secondary hypothesis that some children 'may not be cases of ''shaking'' or, indeed, of head injury at all'; or that this minority, who share the essential features of encephalopathy, subdural haemorrhages and retinal haemorrhages with the considerable majority have a different and unidentifiable natural disease process.

A spontaneously occurring natural disease might be expected to occur randomly; it is notable that those infants who develop 'infantile encephalopathy with subdural and retinal bleeding' only very exceptionally become unwell whilst being observed or handled by more than one carer, whether inside or outside the home. This must cast further doubt on the proposed non-traumatic aetiology.

Geddes *et al.* refer to 'genetic factors', but the work upon which they rely showed that the relationship between the genetic factor studied, apolipoprotein E polymorphism, and outcome was independent of focal or diffuse brain swelling [40]. It is, therefore, not correct to loosely implicate 'genetic factors' in the 'unified hypothesis' that relies upon brain swelling in its pathogenesis.

In conclusion, Geddes *et al*. advance two hypotheses that are not supported by the data contained within the studies performed. The effect is the production of a flawed hypothesis [9] for which there is no evidence, which relies upon another flawed hypothesis [8] for which there is also no evidence. The unbroken chain of events that would be required to comprise the 'unified hypothesis' has not been established by the data submitted.

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