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SPECIAL ISSUE: ABUSIVE HEAD TRAUMA

Long-term outcome of abusive head trauma

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Abstract Abusive head trauma is a severe inflicted traumatic brain injury, occurring under the age of 2 years, defined by an acute brain injury (mostly subdural or subarachnoidal haemorrhage), where no history or no compatible history with the clinical presentation is given. The mortality rate is estimated at 20-25% and outcome is extremely poor. High rates of impairments are reported in a number of domains, such as delayed psychomotor development; motor deficits (spastic hemiplegia or quadriplegia in 15–64%); epilepsy, often intractable (11–32%); microcephaly with corticosubcortical atrophy (61–100%); visual impairment (18–48%); language disorders (37–64%), and cognitive, behavioral and sleep disorders, including intellectual deficits, agitation, aggression, tantrums, attention deficits, memory, inhibition or initiation deficits (23–59%). Those combined deficits have obvious consequences on academic achievement, with high rates of special education in the long term. Factors associated with worse outcome include demographic factors (lower parental socioeconomic status), initial severe presentation (e.g., presence of a coma, seizures, extent of retinal hemorrhages, presence of an associated cranial fracture, extent of brain lesions, cerebral oedema and atrophy). Given the high risk of severe outcome, long-term comprehensive follow-up should be systematically performed to monitor development, detect any problem and implement timely adequate rehabilitation interventions, special education and/or support when necessary. Interventions should focus on children as well as families, providing help in dealing with the child's impairment and support with psychosocial issues. Unfortunately, follow-up of children with abusive head trauma has repeatedly been reported to be challenging, with very high attrition rates.

Keywords Abusive head trauma · Shaken baby syndrome · Traumatic brain injury · Long-term outcome · Cognitive impairment · Children

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Introduction

The period from infancy to the toddler stage is a time of rapid social, cognitive and behavioral development. The infant brain is immature and vulnerable to injury [1]. Very young children may be more vulnerable to the deleterious effects of acquired brain injury than older children [2, 3]. Further, the infant brain is more susceptible to shaking or impact injuries, due to poor head control and the relatively large size and weight of the infant head compared with the body, leading to significant diffuse brain injury [4, 5]. In infants, violence is the most frequent cause of mortality and morbidity following traumatic brain injury [6]. Abusive head trauma is an extremely serious form of inflicted brain injury, secondary to violent shaking, with or without impact, of an infant by an adult, sufficient to cause brain injury [7, 8]. Abusive head trauma,

also labeled in the literature as shaken baby syndrome, shaken impact syndrome, whiplash-shaking injury, inflicted head trauma and non-accidental head injury [9], most often occurs in children younger than 2 years old, at a mean age of 6.2 months (standard deviation [SD]=2.7) [10], with a male: female ratio of 3:2 to 3:1, without a clear explanation of this difference [11, 12]. It can occur in any socioeconomic level [13], although a predominance has been reported in those with lower socioeconomic status, unmarried mothers, low maternal age, low birth weight, a history of perinatal illness, previous injury to the children and the presence of environmental stress factors in the family [14–16] .

Incidence rate is estimated at 14 to 29.7 infants per 100,000 person-years [17–20], and is certainly underestimated [21], as abusive head trauma is not always detected and diagnosed. It could especially be misdiagnosed [21–23]. Abusive head trauma could represent 17–56% of severe traumatic brain injury cases in infants [16].

Abusive head trauma is characterized by an acute brain injury (most often focal or multifocal subdural and/or subarachnoid hemorrhages, in 69–95% of cases), where no history or no compatible history with the clinical presentation is reported. It is associated with uni- or bilateral retinal hemorrhages in 75–90% of cases [6, 24], skull (shaken impact) or other fractures of the skeletal system, rib fractures and/or metaphysal fractures, possibly in different stages of healing, and bruises. Diagnostic criteria have recently been published by the French National Health Authority [7, 8]. Abusive head trauma can lead to a number of initial brain lesions, with several possible mechanisms in one child: Those include space-occupying effect of a large subdural haematoma, cerebral oedema, hypoxic-ischaemic changes, contusion of the brain, tears of the brain and diffuse axonal injury [9]. Diffuse cerebral hypoxia can result from intracranial hypertension, status epilepticus or respiratory arrest caused by cervical or brainstem lesions.

Clinical presentation can be immediately typical of severe traumatic brain injury, with inaugural or early seizures, which are extremely frequent (65–74%) [25, 26], including up to 40% clinical or subclinical status epilepticus, apnoea, impaired consciousness, hypotonia, motor deficit, rapid cranial circumference increase, vomiting or even sudden death in the most severe cases [22, 23]. However, clinical presentation can also be mild or nonspecific (e.g., poor feeding, isolated vomiting, irritability, fussiness, lethargia).

Compared to accidental traumatic brain injury, children who sustained abusive head trauma tend to have significantly less impact injury, lower initial Glasgow Coma Scale scores, more frequent signs of acute cardiorespiratory compromise, more frequent and prolonged impairments of consciousness, and more frequent bilateral hypoxic-ischaemic brain injury or swelling [27]. Signs of earlier brain damage/pre-existing brain abnormality have been reported in up to 79% of infants, such as cerebral atrophy, subdural hygroma and ex vacuo ventriculomegaly upon admission or after an earlier ultrasound investigation prior to the acute event [28].

Outcome after childhood traumatic brain injury in general

Childhood accidental traumatic brain injury often results in impairments in children's sensory-motor functioning [29] and cognitive, behavioral and emotional functioning, as well as poor social cognition [30]. Despite some improvement of cognitive function over time relative to the acute phase, children with severe traumatic brain injury show a slower rate of subsequent development, so that the gap between the children with severe traumatic brain injury and their peers tends to expand over time [31]. Further, some of these cognitive and behavioral problems may only become apparent after a delay, when developmental demands increase and cognitive processes are expected to be fully developed, especially in children injured at a young age [2, 32]. Together, these problems can have devastating long-term consequences on everyday functioning, social and academic areas, vocational outcome, and participation in society and community integration [30, 33]. Factors negatively influencing outcome following childhood accidental traumatic brain injury include (1) demographic factors, such as young age at injury, lower levels of pre-injury intellectual ability, academic achievement and behavioral status, lower socioeconomic status, parental education, family functioning and coping abilities; (2) injury-related factors, such as lower Glasgow Coma Scale score, longer duration of coma and extent of anatomical brain damage; (3) post-injury factors, such as neuropsychological deficits, behavioral disorders and poor school performance, which are related to long-term social functioning and quality of life and strongly predict parental stress and burden [34, 35]. Therefore, this group of young children with traumatic brain injury, in general, should be considered at high risk for poor development and preschool performance, secondary to environmental risk factors as well as their brain injury [36].

Mortality and short-term outcomes following abusive head trauma

Following knowledge of outcome and risk factors after childhood traumatic brain injury, and given that abusive head trauma often causes severe and diffuse brain lesions in very young children, children with abusive head trauma are at very high risk for poor outcome. Indeed, most short-term outcome studies report high mortality and morbidity rates. Mortality rates range from 11% to 36% [4], with a median of 20-25% [6, 10, 37], which is much higher than mortality following

accidental traumatic brain injury in infants, estimated at 2% for infants and children under age 3 and 4% for older children [15]. Initial severity and presentation are more severe in abusive head trauma than in accidental traumatic brain injury [27]. In a prospective series of 150 severe traumatic brain injuries in infants, Vinchon et al. [16] reported that although abusive head trauma represented 38% of all cases, it accounted for 71% of the mortality and 90% of severe morbidity at 6 months, as rated by the Glasgow Outcome Scale. The Glasgow Outcome Scale is an overall scale of outcome following brain injury, which has been modified for use with children. It is a five-point outcome score, in which a "good outcome" is assigned when the child has age-appropriate levels of functioning, and, if of school age, functions in school full-time without special education services; "moderate disability" is assigned on the basis of significant reduction in cognitive functioning, special education therapy, and/or motor deficits interfering with activities of daily living; "severe disability" is assigned when cognitive functioning is in the deficient range and/or severe motor deficits require substantial assistance with self-care skills; "persistent vegetative state" is assigned when there is no evidence of ability to communicate, verbally or nonverbally, or to respond to commands and, finally, "death" is the fifth outcome level [38].

In a large review [15, 39], overall outcome, as measured by the Glasgow Outcome Scale in 18 studies (n=837), indicated vegetative state in 5%, severe disability in 34%, moderate disability in 25% and good outcome in 13% (those children can attend regular classrooms but may require remedial help and may display behavioral disorders). Motor impairments were present in 38% (hemiparesis 19% or quadriparesis 35%; 15 studies; n=395), and cranial nerves were impaired in 20% (2 studies; n=45). Early post-traumatic seizures occurred in 32–79%, with an incidence of epilepsy in survivors of 30%, most often intractable (14 studies; n=312 children), (as opposed to only 5–15% in accidental traumatic brain injury). Blindness occurred in 15% of survivors (8 studies; n=135) and 45% had some degree of visual impairment (12 studies; n=581). Microcephaly was found in 50% (4 studies; n=67). Finally, intellectual/cognitive deficits were very frequent: intellectual deficiency (intellectual quotient <70) was reported in 54% (12 studies; n=275) and severe behavioral disorders in at least 38% (9 studies; n=208). Overall, 11% had normal outcome (9 studies; n=431). A large majority of survivors require multidisciplinary care [24].

As abusive head trauma occurs in very young children, impairments (or additional impairments) may only become obvious some years later [38], with severe consequences on everyday life independence and academic achievement [40–42]. Children who sustain abusive head trauma are at high risk for pervasive cognitive and adaptive behavior deficits. Cognitive deficits are global, including problems in motor, visual processing, and receptive and expressive language, with high rates of delays in language acquisition [43], as well as social competence and joint attention impairments [44].

Although greater depth of injury on neuroimaging in children with abusive head trauma when compared to children with accidental injury has been reported [27], impairments and overall outcomes are usually more severe after abusive head trauma than after accidental traumatic brain injury occurring at a young age [16, 36, 43, 45, 46], with more children exhibiting poor functional outcome, and more children suffering seizures and scoring in the deficient range for motor, cognitive abilities and adaptive behavior, even when controlling for injury severity and acute parenchymal injury. The high level of impairment found in this population could be explained, in part, by the initial delay in receiving care, because many children's conditions only become recognized when they have respiratory distress or seizures or are unconscious [4, 21].

Long-term outcomes

Outcome studies are very heterogeneous, with regards to follow-up periods (ranging from 1 month to 10 years, with most studies reporting relatively short-term outcomes), initial severity of the samples, source of cases, study design (often retrospective, without a control group) and outcome measures. Reports include telephone interviews with caregivers, retrospective chart review, as well as a few studies performing direct serial neurological examination and assessment of cognitive, adaptive and behavioral functioning, using standardized measures.

Studies reporting follow-up exceeding 5 years (when children reach school age) are scarce. They usually have small sample size (n=1–25) and high attrition rates, as the number of patients lost to follow-up is particularly high in this context (48–57%). Interestingly, follow-up is more likely to be accepted by caregivers when the perpetrator is known or if the child is in foster or adoptive care [37]. Further, there is no consensus on outcome measurement. Some studies report on neurological examination, caregiver report and overall level of impairment, using global scales, such as the Glasgow Outcome Scale, the Seschia's global outcome score or the Pediatric Overall Performance Category [4]. However, others report detailed information on medical and cognitive assessments.

Among studies reporting outcomes at least 5 years post-injury, high rates of long-term impairments are reported in a number of domains (e.g., visual, neurological, cognitive and behavioral) (Table 1). Indeed, only 8–36% of children are classified as having "good outcome." A delayed presentation of sequelae is very frequent in children who initially seemed to have a good outcome [40], with deficits emerging over time, when developmental delay becomes more apparent.

Table 1 Summary of the main studies on long-term outcome (at least 5 years) after abusive head trauma

	Follow-up period	Number of patients	Methods	Assessment	Good outcome	Poor outcome (disabled)	Epilepsy
Karandikar et al. 2004 [41]	1.8 to 8.5 y	45	Retrospective	King's Outcome Scale for Childhood Head Injury	55%		11%
Tanoue et al. 2012 [47]	2.25 to 9 y (mean 4.6)	24	Prospective	GOS	50%	4% mild; 25% moderate; 21% severe	–
Talvik et al. 2007 [48]	4.67 years	22	Prospective	KABC GOS Rankin Disability Scores	9%	Minor symptoms 14%; slight disability 64%; severe 13.6%	32%
Barlow et al. 2004 [37] and 2005 [38]	4.9 y	25	Prospective and cross-sectional	Neurological examination; Modified GOS; Seschia's Outcome Scale; Standardised tests of cognitive, behavioral and adaptive behavior tests	32%	68% abnormal: 16% mild; 16% moderate; 36% severe	20% (60% intractable)
Ilves et al. 2010 [28]	2.3 – 9 y (mean 5.2 y)	22	Retrospective+ prospective	Neurological examination; GOS; Rankin Disability Score	9%	13.6% severe: 77% mild impairment or moderate disability	–
Oliver 1975 [49]	1:7 y, 2:5.2 y, 3:4.2 y	3	Case reports	Neurological examination; developmental status	–	All 3 severe deficits	2: yes
Bonnier et al. 2003 [50]	2.5 – 13 y (mean 6 y)	23	Retrospective / cross-sectional follow-up for children older than 3 y	Neurological examination GOS; WPPSI; WISC; KABC	1 (4%)	8 (35%) moderate; 14 (61%) severe	39%, refractory in 86%
Vinchon et al. 2003 [51]	5 – 9.5 y (mean 6 y)	48 SDH, including 15 AHT	Prospective	Neurological examination; IQ tests; GOS	Normal: 81.2	10.4% moderate deficit; 6.2% severe deficit; 2.1% vegetative	–
Laurent-Vannier et al. 2009 [52]	6.7 y	1	Case report	Neurological examination, IQ tests	–	Severe outcome	Yes
Bonnier et al. 1995 [40]	4 – 14 years (mean 7.2 y)	12 (1/13 died)	Prospective; control group n=10 age-matched children admitted for abuse	Serial neurological, psychological & social assessments; Standardised developmental tests	1 (7%)	6/13: Vegetative 1 (7%) Severe 3 (23%) Moderate 4 (17%)	33%
Stipanicic et al. 2008 [53]	7.3 y	11 (GOS 2 and 3 excluded)	Case control	Standardised neuropsychological assessment	54%		–
Rhine et al. 2012 [54]	3 to 11.5 y (mean 7.4 y)	8 (out of 35)	Retrospective	Neurological examination, chart review, GOS	1 child (11%)	89%	–
Toure et al. 2007 [55]	7 to 10.2 y (mean 8.1)	23	Retrospective	Medical telephone interview GOS	22%	26% moderate; 35% severe disability; 17% vegetative	43.5% (20% intractable)

Table 1 (continued)

	Follow-up period	Number of patients	Methods	Assessment	Good outcome	Poor outcome (disabled)	Epilepsy
Duhaime et al. 1996 [42]	5.5 – 15.5 (mean 9 y)	14 (of 62 survivors)	Retrospective	Medical telephone interview GOS	5 (36%) but 3 repeated grades or have tutors; 2 have behavior problems	6 (43%) severe disability or vegetative; 2 (14%) moderate	21%
Fischer and Allasio 1994 [56]	8 – 15 y (mean: 10.1 y)	10	Retrospective	Telephone interview and chart review	3 (30%)	7/10; 6 (60%) moderate or severe	20%

	Motor deficits	Visual impairment	Speech and language / cognitive deficits	Praxis / visual-spatial deficits	behavioral disorders	Special education needs
Karandikar et al. 2004 [41]	–	24.4%	37%	–	28.8%	41%
Tanoue et al. 2012 [47]	–	–	–	–	–	–
Talvik et al. 2007 [48]	22% severe; 59% light to moderate	23.5%	77% delayed mental development	–	–	–
Barlow et al. 2004 [37] and 2005 [38]	60%	48%	64%	44% normal; 16% mild; 20% moderate; 20% severe or profound	52%; sleep abnormalities 24%	–
Ilves et al. 2010 [28]	–	–	–	–	–	–
Oliver 1975 [49]	1: bilateral spasticity 2: right hemiparesis 3: bilateral spasticity	1: Limited vision and hearing 2: right homonymous hemianopia, impaired hearing 3: seriously impaired vision	1: Profound mental retardation 2: global DQ 26%; language DQ 17% 3: profound mental retardation	2: eye-hand coordination DQ 26%	2: impaired behavior	–
Bonnier et al. 2003 [50]	65%	39% (2/3 blindness)	30% selective – 61% multiple neuropsychological impairments; 21% IQ > =85	–	–	–
Vinchon et al. 2003 [51]	14.7%	6%	Mean VIQ (n=34) 105.9 (SD= 24.1), but “lower in AHT”	Mean PIQ (n=34) 105.5 (SD= 26.2), but “lower in AHT”	61.4%	85% normal school (16.5% with help); 15% special education
Laurent-Vannier et al. 2009 [52]	Hemiplegia, scoliosis	Yes	Severe; VIQ=44	Severe; PIQ=45	Severe	Special education institution
Bonnier et al. 1995 [40]	42%	33% (2 blind)	38.5%; 92% mental retardation (IQ <80) and learning disabilities		50% psychiatric problem	38.5%
Stipanicic et al. 2008 [53]		–	Reduced verbal fluency; mean FSIQ=86		Attention and inhibition disorders	–
Rhine et al. 2012 [54]	–	–	–	–	–	–

Table 1 (continued)

	Motor deficits	Visual impairment	Speech and language / cognitive deficits	Praxis / visual-spatial deficits	behavioral disorders	Special education needs
Toure et al. 2007 [55]	39% hemiparesis; 17% quadriparesis; 34% can't walk independently	34% (2 blind)	78% cognitive deficits		35%	17% school impossible; 35% special education; 13% special needs in regular education; 78% ongoing rehabilitation
Duhaime et al. 1996 [42]	57%	36% (4 blind)	71.4% language and/or cognitive deficits (memory, attention)		21.4%	36% special education; 28% normal education
Fischer and Allasio 1994 [56]	70%	30% (2 blind)	60% cognitive deficits	–	30%	40%

AHT busive head trauma, *GOS* Glasgow Outcome Scale, *§ NEPSY* Developmental NEuroPSYchological Assessment, *KABC* Kaufman Assessment Battery for Children, *WPPSI* Wechsler Preschool and Primary Scale of Intelligence, *WISC* Wechsler Intelligence Scale for Children, *IQ* intellectual quotient, *FSIQ* full-scale IQ, *VIQ* verbal IQ, *PIQ* performance IQ, *SD* standard deviation; *DQ* Developmental Quotient, *SDH* subdural hematoma, – data not reported

Neurological deficits and impairments

Microcephaly, related to cortical-subcortical atrophy, is frequently reported, 32% to 100%, with mean losses of 2 to 4 cranial circumference SD [38, 40, 57]. This worsening microcephaly over time indicates failure to maintain a normal head growth pattern and reflects cranial growth deceleration, or even cessation, observed shortly post-injury [40, 49]. Further, cranial growth deceleration and intraparenchymal brain abnormalities are significantly associated with poor short- and long-term neurodevelopmental outcome [40, 46, 50]. In Bonnier's series [40], cranial growth deceleration was the most severe and was a constant predictive factor of long-term neurological sequelae. In our series, in a group of 28 patients consecutively hospitalized in a rehabilitation department following abusive head trauma, mean cranial growth deceleration was 2.2 SD after a mean delay post-injury of 20 months. It was −0.5 SD in the group with good outcomes, contrasting with −4.4 for those with poor outcomes, with an extreme of −8 SD [13].

Delayed psychomotor development and motor deficits include central hypotonia, spastic hemiplegia or quadriplegia (15–70%), ataxia, dystonia, cranial nerve abnormalities or mixed patterns. Walking and gross and fine motor function are often impaired [38]. Overall, a high proportion of patients (36%) suffers severe neurological disabilities requiring significant long-term nursing and caregiver support in the community [37]. Patients often present with hydrocephalus and still have high rates of post-traumatic epilepsy several years post-injury (11–32%) [50], much more frequently than in accidental traumatic brain injury [4], often intractable (60%) [37], or even evolving towards infantile spasms/West syndrome [25, 38]. Non-accidental mechanism of injury has even been reported as an independent risk factor for the occurrence of early post-traumatic seizures [58].

Sensory deficits are also frequently reported, including sensorineural deafness [37] and a high incidence of visual impairment: Long-term visual impairment and blindness following abusive head trauma is frequent and disabling (18–48%). It is thought to be predominantly due to cortical injury, rather than to retinal haemorrhage. Visual impairment can result from a variety of causes, including occipital lobe injury/atrophy, optic radiation injury, optic atrophy, retinal fibrosis, retinal scarring and consequences of vitreous hemorrhages [10]. Visual dysfunction includes cortical blindness, visual acuity loss, visual field defects, visual agnosia, and abnormal extraocular movements with heterotropia [4, 37]. Those visual deficits can, in turn, increase developmental delays, learning difficulties and social integration.

Cognitive deficits — the major domain of impairments in the long term

Speech and language difficulties (37–64%) [38, 40, 41] including dysarthria, speech and language delay are often associated with marked broader cognitive impairment and behavioral abnormalities, paucity of speech or profound language problems [38]. In a prospective study where sequential assessment of language development was performed, the development quotient in speech and language decreased in five of the 11 patients, and it was even associated with developmental arrest in two children [38]. Stipanicic [53] reported impaired comprehension of instructions and verbal fluency in children more mildly impaired, when compared to matched controls, especially when more complex cognitive functions were involved simultaneously, such as working memory. In the latter study, although the most severely impaired children were excluded, so as to allow comprehensive neuropsychological testing to be performed, intellectual performance of children with abusive head trauma was in the low-average range and significantly lower than matched controls.

Various degrees of intellectual disability and cognitive deficits are very prevalent, including intellectual deficits, visual information processing, visual and verbal memory, visuo-motor integration, attention/concentration, executive functioning and social skills deficits [38, 41, 48, 53]. Cognitive scores are in the deficient range (<1st percentile) in more than half of the assessed patients [38]. Further, some children whose intellectual quotient was within the normal range and who attended regular classrooms were found to suffer significant memory deficits that had not been identified in the classroom [38]. A sign-free interval has been reported by Bonnier [40], with learning disabilities becoming apparent several years post-injury in children initially thought to be normal on early follow-up. In this study, the group was divided in two subgroups: (1) children without sign-free interval (7/13), where children remained severely and permanently abnormal from the time of the shaking; and (2) children with sign-free interval (6/13), who had apparently fully recovered after the injury. All but one left this normal category and became disabled after a delay ranging between 6 months and 5 years. Delayed deficits include hemiparesis, detected around 18 months of age, psychomotor retardation (especially in language, adaptability and social behavior) around 24 months of age, major behavioral disorders, detectable 3 to 6 years post-injury, and mental retardation (intellectual quotient ranging from 53 to 80), becoming evident within 5 years of the event, all of them requiring special education. Rhine et al. [54] also reported similar evolution of the proportion of children with poor outcomes over time, with 63% of poor outcome at 6 months, 56% at 12 months, 80% at 36 months and 89% at 81 months, with a number of children demonstrating one or more new deficits over time, among the domains of locomotion, language, behavior and feeding. Similarly, Fisher and Allasio [56] reported that among three children rated normal upon discharge, only one was still normal on follow-up at 10 years of age.

Stipanicic [53] reported significant impairment in divided attention, working memory and various aspects of executive functioning, such as reasoning, mental organization/planning, and mental alternation and inhibition, with a negative impact on intellectual ability and memory (especially retrieval), as well as slower execution time, in comparison to matched controls. This highlights the need to monitor cognitive development closely, over prolonged periods of time, including executive skills, since the frontal regions only reach maturity at the end of adolescence.

Behavioral and sleep disorders have been less often studied. They include agitation, aggression, temper tantrums, rage outbursts, stereotypical behaviors, inhibition or initiation deficits or autism spectrum disorders (23–59%) [38]. Behavioral problems in particular have rarely been assessed directly, and seem to be present in more than half of affected children. They entail self-injurious and self-stimulatory behaviors (24%), such as head banging, eye poking and biting, occurring in children with neurological abnormalities; severe temper tantrums (36%) or rage reactions; evidence of frontal lobe dysfunction with hyperactivity; impulsivity, marked problems with attention and ritualistic behaviors, especially in older children. Many of these problems only become apparent between the second and third years of life, in previously quiet infants, and they are reported by caregivers to be the most difficult issues to deal with. They are attributable to a combination of frontal lobe injury, speech and language abnormalities and environmental factors. Therefore, their frequency may increase with age, as the consequences of frontal lobe injury may be underestimated unless follow-up is extended into adolescence and early adulthood. Sleep disturbances are also common (24%) [38].

Adaptive behavior has been reported to be severely impaired, with a wide range of difficulties across all domains (e.g., communication, daily living skills and socialization domains, with all mean scores ranging from 73 to 79, which is well below the normal mean of 100) [38].

Those combined deficits have obvious consequences on academic achievement, with high rates of special educational needs in the long term (15–41%; Table 1) [16, 40–42, 52].

Further, in those studies that performed sequential assessments, children who changed outcome category all did so to a more severe outcome category (e.g., normal to mild or moderate to severe) [37]. Many of the children with moderate to severe outcomes have complex disabilities with varying combinations of motor, language, cognitive and behavioral abnormalities, which all compound significant learning difficulties and require the support of a multidisciplinary team and high levels of care and needs for activities of daily living [38]. To

our knowledge, the need for adaptive housing and vocational training is currently unknown, in the absence of studies with sufficient follow-up. Barlow [38] predicted that 40% of a cohort of children with abusive head trauma would never live independently in the community.

Table 1 lists the studies reporting long-term outcomes following abusive head trauma and the principal findings in overall outcome, as well as in more specific domains such as epilepsy, cognitive, behavioral or academic outcomes when available.

Factors influencing outcome following abusive head trauma

Factors influencing outcome are of various types, and include pre-injury demographic and environmental factors, clinical and radiologic markers of injury severity, and post-injury factors.

Pre-injury demographic and environmental factors

Pre-injury factors, such as previous medical or developmental abnormalities, previous unrecognized abuse and trauma, family instability, low parental socioeconomic status and previous quality of child care are associated with death or worse cognitive and motor outcomes [15, 43, 59]. The effect of young age at injury (e.g., younger than 6 months) is an indicator of poor prognosis in some [42, 48], but not all studies [16, 38, 46, 54].

Clinical and radiologic markers of injury severity

Initial severe presentation has been related to worse overall outcomes. Lower initial Glasgow Coma Scale scores (8 or less, e.g., coma) are consistently related to worse outcomes [19, 36, 54]. Presence, depth and duration of coma or impaired consciousness [16, 28], necessity for cardiopulmonary resuscitation and/or intubation, apnoea or respiratory difficulties, hypotension, initial hyperglycemia, other indices of neurological injury severity, such as initial Pediatric Trauma Score, raised intracranial pressure, lower cerebral perfusion pressure, length of stay in the intensive care unit, occurrence of seizures at any time (early post-traumatic seizures, especially when status epileptic occurred, and onset of late post-traumatic epilepsy), presence and extent (e.g., bilateral) of associated retinal and vitreous hemorrhages, have all been related to worse outcomes [4, 25, 26, 50, 54, 60–62]. Cardiopulmonary resuscitation and seizures were reported to be the strongest predictors of poor outcome at 1 year [36].

Radiographic findings have a strong association with outcome. The following are associated with poor short- and long-term neurodevelopmental outcomes: 1) In the early stage, presence of an associated skull fracture, evidence of preexisting brain injury, extent and severity of brain lesions (e.g. diffuse cerebral oedema, loss of grey-white matter differentiation, high intracranial pressure, transtentorial herniation, subarachnoid haemorrhage, parenchymal lesions [particularly if diffuse], oedema, injury to the brainstem, diffuse axonal injury, infarction [especially if present in different vascular territories in several lobes and on both sides of the brain], subdural haematoma and cerebral atrophy); 2) in the subacute phase, any focal changes in the basal ganglia, development of new intracerebral changes; and 3) in the 3 months following the injury, decrease in white matter, development of severe atrophy or any kind of new focal intracerebral changes (e.g. new acute subdural haematoma, diffuse gliotic changes or multicystic encephalomalacia) [24, 28, 36, 42, 46, 47, 50, 51, 54, 60–63]. In multivariate analyses, Rhine et al. [54] found that children with cerebral oedema within 72 h of injury are 27 times more likely to have a moderate or poor Glasgow Outcome Scale score at follow-up, and children who spent 10 days or more in the intensive care unit are approximately 21 times more likely to have a poor outcome as measured by the Glasgow Outcome Scale at follow-up. Similarly, Duhaime et al. [42] reported that all children with bilateral diffuse hypodensity and loss of grey-white matter differentiation on CT scan (the big black brain) remained blind, nonverbal and nonambulatory. Diffuse parenchymal lesions tend to cause high rates of motor and cognitive dysfunctions and higher frequency of blindness and epilepsy [50]. More sophisticated imaging techniques are being investigated, such as the apparent diffusion coefficient, which has been found to be strongly associated with poor neurodevelopmental outcome, when abnormal in the acute phase (<4 days) in any of the following regions: basal ganglia, thalamus, brainstem, corpus callosum, white matter, cerebral cortex, cerebellar vermis, cerebellar cortex and mean total brain. However, during the early phase (up to 1 month post-injury), only apparent diffusion coefficient in the basal ganglia, thalamus, brainstem and corpus callosum remained associated with a poor outcome [64]. In this latter study (n=14), worse outcome was associated with lower apparent diffusion coefficient values, with values in the various brain regions of interest in the acute phase (<4 days) ranging from 0.3 to 0.4 for patients with severe disability, contrasting with values ranging from 0.8 to 1.1 for patients with good recovery. However, authors did not propose an apparent diffusion coefficient cutoff value.

After 2 weeks, cerebral atrophy is frequent, either frontotemporal (68%) or diffuse and severe (32%), with a decreased amount of white matter in up to 73% of infants, with a significant relation to poor outcome [28]. This atrophy seems to be related to three etiopathogenic mechanisms: contusions, infarcts/stroke and white matter scars [50].

After the acute phase, cranial growth deceleration (measured by the extent of head circumference growth) is

significantly correlated to poor outcome [40, 46, 49, 57]. Further, the amount of cerebral atrophy on late imaging (>9 months post-injury) is predictive of poor neurodevelopmental outcome, as well as the size of the corpus callosum, of the hippocampus (especially when a decrease occurs) and the amount of white matter [28]. The extent of intraparenchymal lesions is related to the severity of motor and cognitive dysfunctions and the atrophic areas and white matter scars tend to match the sites of oedema seen on the initial imaging [50]. For instance, in imaging performed between 0.5 and 3 months post-injury, Bonnier et al. [50] found that stroke was associated with poor outcome in 78% of children, and white matter injuries in 82%, several years post-injury. However, a number of children with no lesions on imaging still develop residual disabilities, such as hemiparesis and mental deficiency, visual sensory defect, or visual-spatial impairment and attention deficit [50].

Post-injury factors

Post-injury factors influencing outcome include persistent adverse environmental influences, emotional abuse, neglect or reinjury (which could occur in 10-30% of cases), negative influence of uncontrolled seizures or medical complications [15]. Finally, outcome as measured at discharge with overall scales of functional performance does not tend to change much over 2 years post-injury, children's functional outcomes remaining relatively stable over time at early follow-up, with deficits noted at hospital discharge maintained in most cases [36].

Direct costs of abusive head trauma

A large study looking at costs of traumatic brain injury in children younger than 3 years old compared the costs in accidental and abusive traumatic brain injury [65]. Patients with abusive head trauma were significantly younger and had higher initial severity and mortality rates than children with accidental traumatic brain injury, which usually results in higher costs. However, even after controlling for age and severity, children with abusive head trauma still stayed in the hospital 52% longer and had a mean total cost 89% higher than did patients with accidental traumatic brain injury. However this study focused only on initial inpatient costs, although given the severe outcomes following abusive head trauma, those children will be high resource users in the long term, including medical and rehabilitation care, but also a number of so-called indirect costs, such as reduced earning and productivity at the adult age, parents' lost wages from time spent caring for their child, and special education. The literature indicates that children's use of ancillary medical resources is high, as early as 1 and 2 years post-injury [63] [36], with up to one-third of children classified as high resource users. This was also found in our series (n=23) after 8 years follow-up, with only 35% of the group attending normal schools without special adaptations, and a large majority of children (78%) still requiring rehabilitation, often intensive, e.g., in two to four types (among physical therapy, speech and language therapy, occupational therapy, psychomotricity, psychological intervention) [55]. A recent study focused on all direct costs related to paediatric abusive head injury in New Zealand [66], including direct costs of hospital care, community rehabilitation, special education, investigation and child protection, punishment of offenders, and lifetime care for moderate or severe disability. Total direct costs averaged more than 1 million NZ Dollars per child, which is far higher than any other previous report. Further, the direct costs of acute hospitalization only represented 4% of those comprehensive lifetime direct costs.

Based on this information and the severity of long-term outcome, clinicians and politicians should be strongly urged to develop and implement widely applied primary prevention programs, which can only be cost-effective. Secondary prevention includes all the acute care interventions aimed at reducing secondary brain damage, such as limiting hypoxia, hypovolaemia, elevated intracranial pressure [5]. Tertiary prevention includes interventions provided to families when the children are sent home to help them cope with the deficits and management of care, and to prevent further abuse of the child.

Long-term care and follow-up after abusive head trauma

Given the potential long-term severe outcome, the risk of functional deficits emerging over time within multiple developmental domains [2, 40] and cognitive delays reported in children with initial mild injury as well as in children whose adaptive behavior was rated normal by their caregiver [43], comprehensive long-term follow-up, including systematic formal cognitive testing, should be systematically performed to monitor development, detect any problem and implement timely adequate rehabilitation interventions and special education and/or support when necessary [67].

Patients often require long-term multidisciplinary medical care, specialized education and the involvement of child welfare authorities [6]. Interventions focus on reducing impairments, enhancing independence in daily activities and quality of life, and minimizing caregiver burden. The overall goal is the child's successful integration in school and in the community. Areas targeted are tailored to each child, and interventions include physical therapy, occupational therapy, speech and language therapy and cognitive interventions, psychological support, behavior management, special education or tutoring, family information and support. Although these principles seem trivial, and are described in different models of care, no study to our knowledge has described and assessed

interventions focused on children who sustained abusive head trauma; most studies on interventions following traumatic brain injury are performed in older age groups who sustained accidental traumatic brain injury [5]. Given the important transitions occurring during the critical ages of 15 to 20, where the issues of professional training and individual living skills are raised, follow-up should be prolonged until early adulthood, when the child reaches a stable vocational outcome, before referral to adult services. Interventions should also focus on families, providing help in dealing with the child's impairment and support with psychosocial issues. However, unfortunately, follow-up of children with abusive head trauma has repeatedly been reported to be challenging and indeed the number of patients included in the studies with the longest follow-up periods is usually limited, with very high attrition rates.

Legal compensation

Legal compensation is a factor that can contribute to reducing the level of handicap. Indeed, it can allow funding for various interventions that would not otherwise be possible and for caregivers for the child if he/she does not achieve age-expected independence for basic and instrumental activities of daily living.

Conclusion

Overall, abusive head trauma is a very severe, avoidable condition, leading to high rates of mortality and long-term visual, neurological, cognitive, behavioral and developmental impairments, even when apparent initial early recovery is observed. Focus should be brought on systematic long-term follow-up of injured children, with adequate and timely developmental and cognitive assessments throughout the school years until transition into adulthood. These assessments should serve to implement adequate interventions and special education programs when needed. Research should focus on more systematic prospective very long-term outcome studies, as the outcome and autonomy of those patients in adulthood is not known at the moment. Of course, importantly, primary prevention programs should be developed and largely implemented.

Conflicts of interest None

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