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Extended follow-up of neurological, cognitive, behavioral and academic outcomes after severe abusive head trauma

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ABSTRACT

Studies about long-term outcome following abusive head trauma (AHT) are scarce. The aims of this study were to report long-term neurological, cognitive, behavioral and academic outcomes, ongoing treatments and/or rehabilitation, several years after AHT diagnosis, and factors associated with outcome. **In this retrospective study, all patients admitted to a single rehabilitation unit following AHT between 1996 and 2005, with subsequent follow-up exceeding 3 years, were included. Medical files were reviewed and a medical interview was performed with parents on the phone when possible.** The primary outcome measure was the Glasgow Outcome Scale (GOS). **Forty-seven children (out of 66) met the inclusion criteria (mean age at injury 5.7 months; SD = 3.2). After a median length of follow-up of 8 years (range 3.7–12), only seven children (15%) had “good outcome” (normal life – GOS I) and 19 children (40%) presented with severe neurological impairment (GOS III and IV). Children sustained epilepsy (38%), motor deficits (45%), visual deficit (45%), sleep disorders (17%), language abnormalities (49%), attention deficits (79%) and behavioral disorders (53%). Most children (83%) had ongoing rehabilitation. Only 30% followed a normal curriculum, whereas 30% required special education services.** Children with better overall outcome (GOS I and II) had significantly higher educated mothers than those with worse outcomes (GOS III and IV): graduation from high school 59% and 21% respectively ($p = 0.006$). This study highlights the high rate of severe sequelae and health care needs several years post-AHT, and emphasizes the need for extended follow-up of medical, cognitive and academic outcomes.

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Introduction

Abusive head trauma (AHT), also labeled various ways in the literature, such as *shaken baby syndrome*, *shaken impact syndrome*, *whiplash-shaking injury*, *inflicted head trauma*, *non-accidental head injury*, is an inflicted brain injury defined by an acute brain injury (often associated with subdural or subarachnoid hemorrhage) where no history or no compatible history

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with the clinical presentation is reported. Diagnostic criteria have recently been published (Laurent-Vannier et al., 2011). AHT occurs most often under 2 years of age, at the average age of 6.2 months (Makaroff & Putnam, 2003), with a male:female ratio of around 3:2 (Fanconi & Lips, 2010; Talvik et al., 2006). The annual incidence rate is estimated at 14 to 40/100,000 infants (Fanconi & Lips, 2010; Niederkrotenthaler, Xu, Parks, & Sugerman, 2013; Talvik et al., 2006) and is certainly underestimated (Chadwick, Chin, Salerno, Landsverk, & Kitchen, 1991; Williams, 1991). Mortality rates range from 11% to 36%, with a median of 20–25% (Barlow, Thomson, Johnson, & Minns, 2005; Chevignard & Lind, 2014). Initial clinical severity, brain lesions on imaging findings and outcome are more severe than following accidental traumatic brain injury (TBI; e.g., injury following clearly accidental mechanisms, such as road traffic accidents or witnessed falls) occurring at the same age (Chevignard & Lind, 2014; Ewing-Cobbs, Prasad, Kramer, & Landry, 1999; Hymel, Makoroff, Laskey, Conaway, & Blackman, 2007; Niederkrotenthaler et al., 2013).

Studies reporting outcomes following AHT with follow-up exceeding 5 years, when children have reached school age, are scarce (Chevignard & Lind, 2014). Furthermore, of the available studies, samples are small and attrition rates are high (48–57%). Overall, the studies report high rates of visual, neurological (motor deficits, post-traumatic epilepsy, etc.), cognitive, behavioral and sleep impairments, and special education needs, with only 8–36% of children achieving a “good outcome” (see Table 1). As environmental demands increase and the child’s abilities are expected to develop (Barlow et al., 2005), a delayed presentation of sequelae is very frequent in infants whom are thought to have recovered after the acute phase. This can result in an apparent *sign-free interval* in some children with deficits emerging a few years later, leading to severe consequences on independence for daily life activities and academic achievement (Barlow et al., 2005; Bonnier, Nassogne, & Evrard, 1995; Duhaime, Christian, Moss, & Seidl, 1996; Karandikar, Coles, Jayawant, & Kemp, 2004).

Factors predicting a worse outcome include: (1) demographic and environmental factors (such as family instability, low parental socio-economic status, younger age at injury); (2) initial clinical and radiological markers of injury severity (initial severe presentation, such as lower Glasgow Coma Scale (GCS) scores, presence, depth and duration of impaired consciousness, necessity for cardio-pulmonary resuscitation or intubation, raised intra-cranial pressure, occurrence of seizures at any time, presence and extent of retinal and vitreous hemorrhages; extent and severity of brain lesions on initial or delayed imaging); and (3) post-injury factors (cranial growth deceleration, persistent adverse environmental influences, uncontrolled seizures) (Chevignard & Lind, 2014).

The aims of this study were to assess the long-term neurological, cognitive, behavioral and academic outcomes; assess the amount of treatments and rehabilitation still needed several years after AHT; and identify the factors that influence these outcomes.

Methods

Study Design and Procedure

The study sample for this retrospective study consisted of all infants who had been consecutively admitted to one single rehabilitation unit, dedicated to children with acquired brain injury in the Paris area (France), following a diagnosis of AHT (initially treated and diagnosed at the same regional pediatric neurosurgical unit), between January 1996 and December 2005.

For this study, the inclusion criteria was a diagnosis of AHT that had led to referral to the rehabilitation department, with subsequent follow-up by the treating clinician after the hospital phase for at least 3 years (so that the children were all at least of pre-school age at the time of the study). The diagnosis of AHT was based on (Committee on Child Abuse and Neglect, 2001): (1) the presence of an intracranial hemorrhage, including subdural hemorrhages, with a history that was inconsistent with the observed injuries and/or that changed over time; (2) with or without associated retinal hemorrhages or bones fractures; (3) exclusion of any hematological or genetic condition capable of causing spontaneous intracranial hemorrhages. Referral and admission to the rehabilitation unit usually occur when, after the acute phase, the infants are medically stable, but require specialized medical care and rehabilitation following their brain injury (Chevignard, Toure, Brugel, Poirier, & Laurent-Vannier, 2010), for hemi- or quadriplegia, truncal hypotonia, swallowing difficulties, etc. Those whose injuries are not sufficiently severe to require inpatient rehabilitation care can receive outpatient follow-up in clinics, with care (when needed) organized in the community (those children were *not* included in the study). On the other hand, a few children with very severe brain injury and neurological impairment are sometimes discharged directly from the neurosurgery department to a specialized long-term medical unit, and would not be included here either.

The study was approved by the local Research Ethics Committee. The study sample was identified using the department’s admission records. Parents were contacted by mail, and asked if they wished to participate in the study. If they agreed, a telephone medical interview was performed. If the families could not be reached by phone, follow-up data was collected from the medical files. In the context of their routine medical follow-up in the department, when needed (e.g., in order to determine rehabilitation goals, to implement school adaptations or to organize orientation towards special education), and when appropriate (e.g., patients able to perform cognitive testing), a number of patients had undergone comprehensive neuropsychological assessment at some point.

Table 1
Summary of the main studies reporting long-term outcome following AHT.

	Follow-up period	Number of patients	Methods	Assessment	Good outcome	Epilepsy	Motor deficits	Visual impairment	Language/cognitive deficits	Behavior disorders	Special education needs
Karandikar et al. (2004)	1.8–8.5 y	45	Retrospective	King's Outcome Scale for Childhood Head Injury	55%	11%	–	24.4%	37%	28.8%	41%
Tanoue, Matsui, Nozawa, and Aida (2012)	2.25–9 y (mean 4.6 y)	24	Prospective	GOS	50%	–	–	–	–	–	–
Talvik et al. (2007)	4.67 y	22	Prospective	KABC; GOS; Rankin	9%	32%	22% severe; 59% light to moderate	23.5%	77% delayed	–	–
Barlow et al. (2005)	4.9 y	25	Prospective and cross-sectional	Neurological examination; GOS; Sheshia Outcome Scale; Neuropsychological and adaptive behavior assessment	32%	20% (60% intractable)	60%	48%	64%	52%	–
Ilves, Lintrop, Talvik, Sisko, and Talvik (2010)	2.3–9 y (mean 5.2 y)	22	Retrospective + prospective	Neurological examination; GOS; Rankin Disability Score	9%	–	–	–	–	–	–
Oliver (1975)	4.2, 5.2 and 7 y	3	Case reports	Neurological examination; developmental status	–	2 out of 3	1 hemiparesis; 2 bilateral spasticity	All	All: Profound mental retardation	2: impaired	–
Bonnier et al. (2003)	2.5–13 y (mean 6 y)	23	Retrospective/cross-sectional follow-up for children over 3 y	Neurological examination; GOS; neuropsychological assessment	1 (4%)	39%, refractory in 86%	65%	39% (2/3 blind)	30% selective – 61% multiple neuropsychological impairments; 21% IQ ≥ 85	–	–
Laurent-Vannier, Toure, Vieux, Brugel, and Chevignard (2009)	6.7 y	1	Case report	Neurological examination, neuropsychological assessment	–	Yes	Hemiplegia, scoliosis	Yes	Severe; VIQ = 44; PIQ = 45	Severe	Special education institution
Bonnier et al. (1995)	4–14 y (mean 7.2 y)	12	Prospective with control group	Serial neurological, psychological & social assessments; standardized developmental tests	1 (7%)	33%	42%	33%	92% mental retardation (IQ < 80) and learning disabilities	50%	38.5%
Stipanovic et al. (2008)	7.3 y	11	Case control	Neuropsychological assessment	54%	–	–	–	Mean FSIQ = 86; attention and executive function deficits	–	–
Rhine, Wade, Makoroff, Cassedy, and Michaud (2012)	3–11.5 y (mean 7.4 y)	8	Retrospective	Neurological examination, chart review, GOS	1 (11%)	–	–	–	–	–	–
Duhaime et al. (1996)	5.5–15.5 y (mean 9 y)	14	Retrospective	Medical telephone interview; GOS	5 (36%)	21%	57%	36% (4 blind)	71% language and/or cognitive deficits	21.4%	36%
Fischer and Allasio (1994)	8–15 y (mean 10.1 y)	10	Retrospective	Telephone interview and chart review	3 (30%)	20%	70%	30% (2 blind)	60% cognitive deficits	30%	40%

Note: y: years; GOS: Glasgow Outcome Scale; KABC: Kaufman Assessment Battery for Children; IQ: Intellectual Quotient; FSIQ: full-scale IQ; –: data not reported.

Table 2

Demographic and medical characteristics of the study sample, compared to patients lost to follow-up.

	Patients included (<i>n</i> = 47)	Patients lost to follow-up (<i>n</i> = 19)	<i>p</i> ^a
Sex-ratio (boys/girls)	3.7	1.1	NS
Mean age at injury [months; mean (SD)]	5.7 (3.2)	6 (3.2)	NS
Mothers' education level equal or superior to graduation from high school [<i>n</i> (%)]	34 (72)	8 (42)	0.03
Mean length of stay in ICU [days; mean (SD)]	24.6 (9.6)	25 (4)	NS
Initial intubation [<i>n</i> (%)]	39 (83)	13 (68)	NS
Seizures at diagnosis [<i>n</i> (%)]	40 (85)	16 (84)	NS
Subdural hemorrhages [<i>n</i> (%)]	46 (98)	18 (95)	NS
Fractures [<i>n</i> (%)]	8 (17)	3 (16)	NS
Skin lesions [<i>n</i> (%)]	9 (19)	7 (37)	NS
Retinal hemorrhages [<i>n</i> (%)]	41 (87)	18 (95)	NS
Hemiplegia upon admission to rehabilitation department [<i>n</i> (%)]	29 (62)	9 (47)	NS
Length of stay in rehabilitation department [days; mean (SD)]	145 (265)	150 (134)	NS

NS: non-significant ($p > 0.05$).^a Fisher exact test (2-tailed).

Clinical Details

Medical records were reviewed to ascertain demographic details such as age at injury, gender, mothers' education level (graduation from high school or not), the likely mechanisms of injury, acute admission details such as the initial or lowest GCS score, necessity of intubation, occurrence of seizures and/or status epilepticus, and details of other injuries.

Medical Structured Interview

Parents who agreed to participate were contacted by telephone for a semi-structured medical interview between July and September 2009. The medical interview consisted of questions on the following topics: current medical situation; current neurological condition (epilepsy, seizure control, past and ongoing anti-epileptic drugs and medications, motor deficit, spasticity, visual impairment); current cognitive impairments (namely language expressive and receptive impairment, visual-spatial and graphic/drawing difficulties, attentional disorders); current behavioral and sleep disorders; current academic situation (mainstream school with or without extra help; special education); ongoing rehabilitation therapy (number and type); social outcome (compensation from health care, judicial procedures). The interview lasted 10–15 min.

Qualitative Assessment of Outcome

The primary qualitative outcome measure, completed after the medical interview, was the Glasgow Outcome Scale (GOS) modified for children ("Circulaire DAS/DE/DSS n° 96-428 relative à la prise en charge médico-sociale et à la réinsertion sociale et professionnelle des personnes atteintes d'un traumatisme crânien", 1996; Miner & Houston Conference on Neurotrauma, 1986), where GOS I refers to *good outcome*; GOS II to *moderate disability*, including hemiparesis and/or cognitive impairments and/or child referral for outpatient rehabilitation therapy; GOS III to *severe disability*, including severe motor deficit and/or cognitive assessment in the deficient range and/or referral for inpatient rehabilitation; and GOS IV to *minimally responsive or vegetative state*.

Secondary qualitative outcomes were the presence of epilepsy, sleeping disorders, visual impairment, behavioral disorders, language difficulties, visual-spatial and graphic/drawing difficulties, the number of ongoing weekly rehabilitation therapies and current academic level and type of schooling.

Description of the Sample

During the inclusion period, 66 children were hospitalized in the rehabilitation department following a diagnosis of AHT. Among them, 47 met the inclusion criteria (follow-up exceeding 3 years) and 19 were lost to follow-up. The median length of follow-up was 8 years (range 3.7–12). The demographic and injury severity characteristics of both groups are summarized in Table 2. Children lost to follow-up did not significantly differ from the study group on demographic and initial medical/severity variables, except for mother education (lower in the lost to follow-up group).

In the study group, information was obtained over the phone for 12 families, and from medical files for 35 children. One family declined participation and 17 families had moved and could not be located. Most information was present in the medical files, allowing gathering the adequate information when a telephone interview was not possible. Data from the interview and from the medical files allowed determining the GOS score in all cases. Similarly, for most outcomes, such as epilepsy, motor deficits, language and communication deficits, the amount of ongoing treatments and rehabilitation,

and schooling modalities, the information was available in all cases, as those points are systematically addressed during the clinics. For some outcomes however (e.g., attention deficits, visual-spatial/drawing difficulties, and sleep disorders), the information was driven from either the telephone interview, the results of the neuropsychological assessment (when performed), or from a clear mention in the medical files that those outcomes were or were not a problem, at home and at school. This led us to identify a subgroup of 29 children for whom sufficiently detailed information was available for those specific outcomes, in addition to the outcomes available for the whole group. Those 29 patients did not differ statistically from the total group for the initial socio-demographic and medical characteristics. Thus, results are reported for the study group ($n = 47$) throughout the paper, unless otherwise stated.

Statistical Analysis

Demographic, clinical and severity data were collected for the whole sample: children who were included (follow-up exceeding 3 years – study group) as well as those who were lost to follow-up. Descriptive data were reported using mean (standard deviation – SD) or median (range) for continuous variables and n (%) for categorical variables. As a first step, children in the study group were compared to those lost to follow-up for demographic and initial severity variables, using Fisher's exact test (2-tailed).

In order to study factors influencing outcome, patients in the study group were separated into two groups according to their GOS score: a “good outcome” group (GOS I and II) and a “poor outcome” group (GOS III and IV). Fisher's exact test (2-tailed) was used to compare demographic and injury variables between the “good outcome” and the “poor outcome” groups. The alpha level for statistical significance was set at $p < .05$. Given the retrospective nature of the study and the heterogeneous data collection methods, no further multivariate analyses of predictors of outcome were undertaken.

Results

Overall Outcome: GOS Scores

In the study group ($n = 47$), only 7 children (15%) had returned to an apparently normal life (GOS I), 21 (45%) were rated GOS II, 17 (36%) GOS III and two children (4%) suffered extremely severe neurological impairment (GOS IV).

No significant differences were found in terms of age at injury, initial seizures, length of stay in ICU, or signs associated with the subdural bleeding (fractures, skin lesions or retinal hemorrhage) between the “good outcome” (GOS I and II; $n = 28$) and “poor outcome” (GOS III and IV; $n = 19$) groups. The “good outcome” group, however, had significantly lower initial rates of coma (68% vs. 95%; $p = 0.03$), required intubation less often (71% vs. 100%; $p = 0.01$), had lower rates of hemiplegia upon admission to the rehabilitation department (50% vs. 79%; $p = 0.02$), and their mothers had significantly higher levels of education (59% vs. 21% had graduated from high school; $p = 0.01$) relative to the “poor outcome” group. See Table 3 for a summary of results.

Neurological and Visual Outcome

Twenty-one patients (45%) had motor deficits, mostly hemiparesis (quadriplegia for three children), six had spasticity (13%), and one child suffered ataxia (2%).

Eighteen patients (38%), aged 4–9.7 years, still were on antiepileptic drugs and were seizure free, except for one child, aged 3.5 years, who required four anti-epileptic drugs. Epilepsy was significantly more frequent in children with a “poor outcome” (GOS III and IV) (63% vs. 18% for children with GOS I and II, $p = 0.002$) (see Table 3).

Twenty-one patients (45%) had visual impairment: cortical blindness ($n = 4$), heterotropia ($n = 9$), visual acuity deficits ($n = 2$), visual fields abnormalities ($n = 3$) and visual acuity deficits associated with visual agnosia ($n = 1$) (see Fig. 1).

Table 3

Comparison of clinical factors associated with overall outcome.

	‘Good outcome’ group (GOS I & II)	‘Poor outcome’ group (GOS III & IV)	p^a
Number of patients	28	19	
Age at injury [months; mean (SD)]	6.28 (3.6)	4.8 (2.7)	NS
Initial coma [n (%)]	19 (68)	18 (95)	0.03
Required initial intubation [n (%)]	20 (71.4)	19 (100)	0.01
Seizures at diagnosis [n (%)]	24 (86)	16 (84)	NS
Retinal hemorrhage [n (%)]	25 (89)	16 (84)	NS
Hemiplegia upon admission to rehabilitation [n (%)]	14 (50)	15 (79)	0.02
Mothers' education level > graduation from high school [n (%)]	16 (59)	4 (21)	0.006
Epilepsy at latest follow-up [n (%)]	5 (18)	12 (63)	0.002

NS: non-significant ($p > 0.05$); GOS: Glasgow Outcome Scale.

^a Fisher exact test (2-tailed).

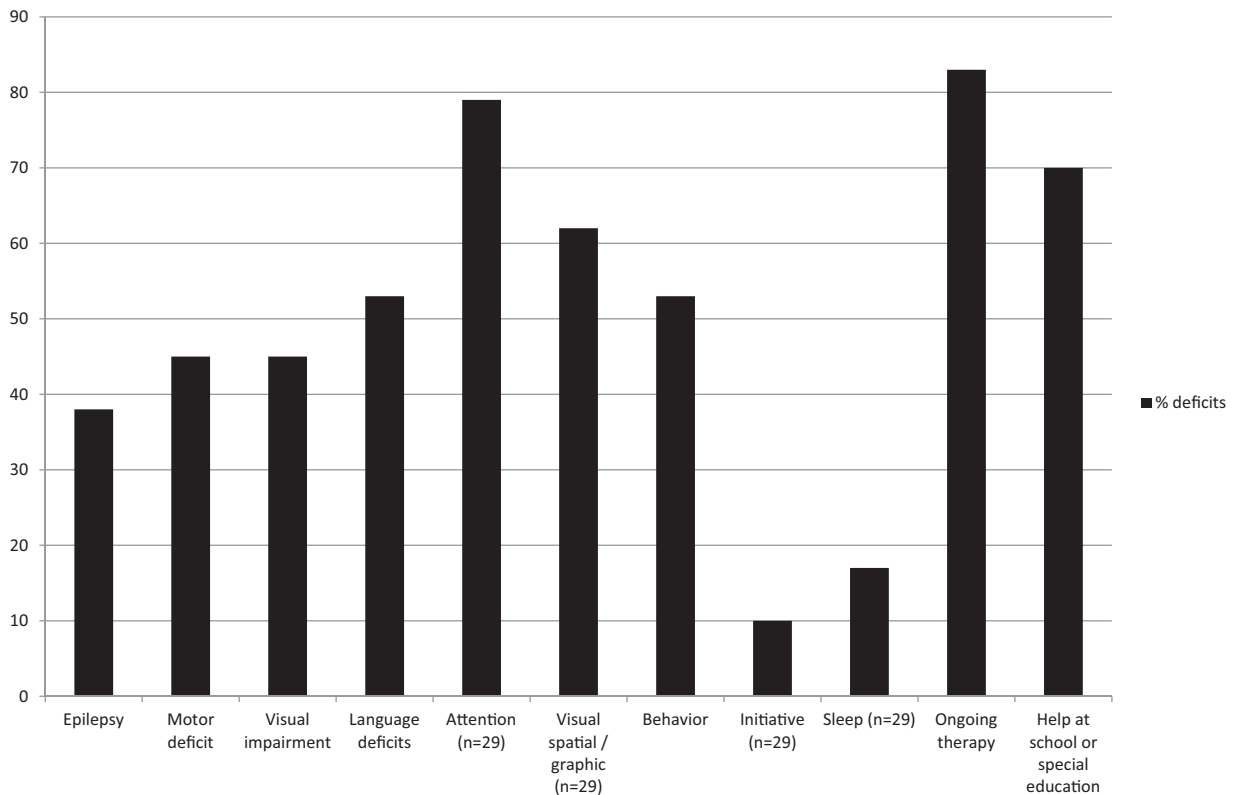


Fig. 1. Percentage of impaired children for the different clinical outcomes. *Note:* Percentages are reported for the whole group ($n = 47$), except otherwise stated. For language, 2 children with no communication abilities were counted as impaired. For schooling, all children who did not attend regular schools without specific help were combined in one group.

Cognitive Outcome

Communication assessment data were not available for the two patients rated GOS IV. Poor vocabulary and/or comprehension difficulties were reported for 23/47 patients (49%). Attention disorders were reported for 23/29 patients (79%). Eighteen out of 29 patients (62%) had visual-spatial and graphic/drawing impairment (see Fig. 1).

Behavioral Outcome

Behavioral disorders were clearly reported by parents or in the medical files in 25 cases out of 47 (53%), including agitation, irritability, impulsivity, intolerance to frustration and temper tantrums. In the sub-group of patients with more detailed information ($n = 29$), lack of initiative was reported for three children (10%) and sleep disorders were present for five children (17%) (see Fig. 1).

Ongoing Rehabilitation and Psychiatric/Psychological Follow-up

Only nine children (19%) did not have any rehabilitation therapy, including the seven children rated GOS I. Overall 38 patients (83%) still needed rehabilitation: 28 children (60%) needed one or two different weekly rehabilitation therapies, and 10 children (21%) needed three or four different types of therapy. The most frequently prescribed rehabilitation therapy was speech and language therapy for language and cognitive disorders ($n = 27$; 57.5%), then physiotherapy ($n = 19$, 40.5%), psychomotricity ($n = 16$, 31%) and occupational therapy ($n = 10$, 21%). Psychomotricity is a healthcare profession well established in France. Psychomotricians receive training in psychological and cognitive domains, as well as in motor control (overall motor function and coordination, fine motor control, writing, bimanual coordination).

In the subgroup of 29 children with detailed records, three also required psychiatric follow-up and six had ongoing psychotherapy (31%) (see Fig. 1).

Type of Schooling

Only 14 children (30%) followed a mainstream curriculum with no additional support. Sixteen children (34%) were in a mainstream school, but required a special individualized schedule ($n = 16$), and/or the presence of a teaching assistant ($n = 9$). Fourteen children (30%) received special education services (specialized classes or medico-social institutions). Three children (aged 8 and 9 years) were at home and could not attend any educational institution (see Fig. 1).

Social Outcome

Few patients (10.6%) had received compensation for their prejudice. Three patients (aged 6, 8 and 10 years) were in foster families. Fourteen patients (30%) had moved since the accident.

Discussion

In this retrospective study, we report important clinical and schooling data characterizing outcomes several years after a diagnosis of AHT in a large group of children consecutively admitted to a rehabilitation department following injury. Results highlight long-lasting impairments after severe AHT. Indeed, less than 10% of the sample had returned to normal life according to the GOS. Moreover, after a median delay of 8 years post-injury (3.6–12 years), a large proportion of children suffered medical, visual, neurological, cognitive (including language difficulties), and behavioral impairments. Children still had high levels of rehabilitation needs and a majority required extra help at school or special education services. Only few patients had received compensation for prejudice.

In general a higher proportion of difficulties were reported by the sample included in the current study, relative to previous studies reported in the literature (Chevignard & Lind, 2014). Indeed, previous studies have reported 4–55% of good outcomes, and at the most, 39% of epilepsy, 48% of visual impairments, 64% of language impairments and 52% of behavior disorders. This could be partially explained by the severity of the AHT in our sample, as children had initially been referred for rehabilitation after their hospitalization in the ICU. In another study (Stipanivic, Nolin, Fortin, & Gobeil, 2008), however, with the opposite selection bias (children with the most severe deficits were excluded, as children had to be able to undergo comprehensive neuropsychological testing), high rates of cognitive deficits were also reported (relative to a group of age-matched controls). Despite these children potentially having less severe AHT than our sample, after an average delay of 7 years post-injury, children suffered slow speed of processing, intellectual ability in the low average range, comprehension and verbal fluency deficits, as well as significant deficits in working memory, divided attention, and various aspects of executive functioning.

Another explanation for the high rate of sequelae in our sample could be the length of follow-up, one the longest compared to previous studies. Indeed, as previously reported (Barlow et al., 2005; Bonnier et al., 1995; Duhaime et al., 1996), deficits might only become evident some years later, as environmental expectations increase. This can impact on language and cognitive development, praxis and graphic/drawing performance, and behavior. Indeed, behavioral disorders following AHT often only become apparent between the second and third years of life in previously calm infants. They are thought to be related to a combination of frontal lobe injury, speech and language deficits, and environmental factors (Barlow et al., 2005; Chevignard & Lind, 2014). The full extent of these deficits remains to be studied, as consequences of frontal lobe injury only become fully apparent after puberty in children with early frontal lobe lesions (Eslinger, Flaherty-Craig, & Benton, 2004; Eslinger, Grattan, Damasio, & Damasio, 1992). Further, in accidental TBI, young age at injury has been shown to lead to high rates of behavioral disorders (Chapman et al., 2010). Children with AHT are particularly young and suffer diffuse brain injury, and could therefore be at particularly high risk of subsequent behavioral impairments (Chevignard & Lind, 2014).

Regarding the effect of environmental and demographic factors associated with outcome, as in most studies following childhood accidental TBI, children with better outcomes (GOS I and II) had mothers who had significantly higher levels of education (i.e., SES) than those children with poorer outcomes. This could be related to an environmental protective factor (Anderson, Godfrey, Rosenfeld, & Catroppa, 2012; Keenan, Hooper, Wetherington, Nocera, & Runyan, 2007; Taylor et al., 2001; Yeates, Taylor, Walz, Stancin, & Wade, 2010), as higher educated mothers could be able to stimulate cognitive development more, seek adequate support and resources for their child. It could also involve genetic factors, higher educated mothers reflecting higher intellectual ability, with children benefitting from higher cognitive reserve, or perhaps a combination of both. The context of AHT may contribute to increasing family dysfunction because of the additional justice-related aspects, and the negative impact of poor family functioning on cognitive and behavioral outcomes have been well described in accidental TBI (Anderson et al., 2006; Taylor et al., 2001; Yeates et al., 2004). In AHT, family instability, low parental SES, and previous quality of child care have also been reported to be associated with worse outcomes (Chevignard & Lind, 2014; Keenan et al., 2007). In any case, the cognitive and behavioral outcomes are certainly explained by a combination of the consequences of the early brain injury, psychological factors and the overall family environment in which the children are raised. In some cases, the persistence of deleterious features such as family violence, absence of parental coping skills or support structures could lead to worse outcomes. Future research is required to tease out the effect of wider indicators of socio-economic status, such as parental education (used here as a proxy for SES), but also family functioning, coping skills, etc.

Further and more worrying, in this study, children who were lost to follow-up had significantly less educated mothers than children in the study group, who benefitted from adequate follow-up. Of course, given the lack of data for children in

the former group, no conclusion can be drawn. One can fear, however, that with similar demographic, clinical and initial severity criteria, those children suffer severe impairments as well. One could hypothesize that the combination of lower SES with the lack of follow-up could negatively influence long-term outcome in those children.

In the current study, age at injury was not significantly related to outcome. This is consistent with the literature, however, mixed findings have been reported, with younger age at injury associated with a worse outcome following AHT in some, but not all studies (Chevignard & Lind, 2014). However, young age at injury is consistently reported to negatively influence outcome in childhood accidental TBI, and more widely in early brain lesions (Anderson et al., 2009; Johnson, DeMatt, & Salorio, 2009). The very high rate of impairment reported in this study is probably at least in part related to the impact of severe diffuse brain injury in very young children.

Regarding the effect of initial injury severity factors, our results confirm the effect on outcome of initial severity markers already reported in the literature, such as coma at diagnosis and necessity for intubation (Bonnier et al., 2003; Greiner, Lawrence, Horn, Newmeyer, & Makoroff, 2012; Keenan et al., 2007). We also found that hemiplegia upon admission to the rehabilitation department was correlated with a worse outcome. This is probably related to the initial brain injury severity, with deficits seen immediately after the child emerges from coma, in line with Bonnier et al.'s findings (Bonnier et al., 1995). Indeed, in that study, some children had no *sign-free interval*, with severe and permanent neurological abnormalities from the time of injury (including hemi- or quadriplegia), whereas almost half of the group had a *sign-free interval*. Among the children who apparently achieved *full recovery* after the injury, all (but one) became disabled after a delay ranging from 6 months to 5 years.

Surprisingly, we did not find initial seizures (Bonnier et al., 2003; Greiner et al., 2012) or retinal hemorrhage (Bonnier et al., 2003) at diagnosis to be related to outcome, as reported in other studies (Chevignard & Lind, 2014). This could be related to the very high rate of those features in our sample (84–89%). In the literature, occurrence of seizures at any time following AHT has been reported as a strong predictor of poor outcome (Chevignard & Lind, 2014; Keenan et al., 2007).

The burden of sequelae for families and society following AHT is extremely high. To our knowledge, so far, the need for on-going rehabilitation in this population has never been reported in the literature and few studies described academic outcome following AHT (Chevignard & Lind, 2014). Despite obvious differences in local academic systems, the rates of special education needs in our study were quite similar to those previously reported by Karandikar et al. in Great Britain (Karandikar et al., 2004), Fischer and Allasio in the USA (Fischer & Allasio, 1994), and Bonnier et al. in Belgium (Bonnier et al., 1995) (41%, 40% and 38.5% of children in special education respectively).

Given the severe consequences of AHT, affecting everyday life and academic achievement, we recommend that patients should receive specific care, starting as soon as possible post-injury. Care could include multidisciplinary rehabilitation when needed, and very long-term follow-up with adequate timely assessments allowing to implement and/or modify the intervention plan and school program in line with maturational needs (Chevignard et al., 2010; Limond, Adlam, & Cormack, 2014).

This study has a number of limitations. Apart from the recruitment bias and the retrospective nature of the study, the results are based on a parental interview and/or on retrospective data from the medical files. In spite of these methodological limitations, the medical files were very detailed, often including reports from the multidisciplinary team of neuropsychologists, speech and language therapists, and physiotherapists allowing precise information to be gathered qualitatively. Unfortunately, little was known about the conclusions of the justice and social enquiries, or this was not available in the medical files. No conclusion could be drawn, therefore, regarding the impact on psychosocial outcomes of potential further abuse. One could also question the reliability of the parents' reports if they were the suspected perpetrator. However if impairments were underestimated by some parents, it would only lead to even more severe outcomes. Further, there was no control group. Control cases are difficult to recruit because of the specificity of cerebral lesions of AHT and of the child's environment in case of inflicted injuries (Barlow et al., 2005). Despite these limitations, we report qualitative outcomes for a large sample of children following severe AHT, with one of the longest follow-up periods reported in the literature.

Conclusion

This retrospective study, performed in a relatively large sample of children following AHT, after a median follow-up of 8 years, confirms previous findings in the literature. It highlights severe outcome at school age following AHT, with significant proportions of persistent deficits, including epilepsy, motor deficits, cognitive and behavioral impairments, and significant consequences on academic achievement. Only one third of the sample followed a mainstream curriculum, while another third required special education services. This study also highlights the importance of parental SES and initial severity as influential factors on outcome, and the high level of needs of this population regarding ongoing medical and rehabilitation care. Systematic organized follow-up services should be established for those children and families, allowing for adequate care until referral to adult services when needed.

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References

- Anderson, V. A., Catroppa, C., Dudgeon, P., Morse, S. A., Haritou, F., & Rosenfeld, J. V. (2006). Understanding predictors of functional recovery and outcome 30 months following early childhood head injury. *Neuropsychology*, 20(1), 42–57. <http://dx.doi.org/10.1037/0894-4105.20.1.42>
- Anderson, V., Godfrey, C., Rosenfeld, J. V., & Catroppa, C. (2012). Predictors of cognitive function and recovery 10 years after traumatic brain injury in young children. *Pediatrics*, 129(2), e254–e261. <http://dx.doi.org/10.1542/peds.2011-0311>
- Anderson, V., Spencer-Smith, M., Leventer, R., Coleman, L., Anderson, P., Williams, J., Greenham, M., & Jacobs, R. (2009). Childhood brain insult: Can age at insult help us predict outcome? *Brain*, 132(Pt 1), 45–56. <http://dx.doi.org/10.1093/brain/awn293>
- Barlow, K. M., Thomson, E., Johnson, D., & Minns, R. A. (2005). Late neurologic and cognitive sequelae of inflicted traumatic brain injury in infancy. *Pediatrics*, 116(2), e174–e185. <http://dx.doi.org/10.1542/peds.2004-2739>
- Bonnier, C., Nassogne, M. C., & Evrard, P. (1995). Outcome and prognosis of whiplash shaken infant syndrome; late consequences after a symptom-free interval. *Developmental Medicine and Child Neurology*, 37(11), 943–956.
- Bonnier, C., Nassogne, M.-C., Saint-Martin, C., Mesples, B., Kadhim, H., & Sébire, G. (2003). Neuroimaging of intraparenchymal lesions predicts outcome in shaken baby syndrome. *Pediatrics*, 112(4), 808–814.
- Chadwick, D. L., Chin, S., Salerno, C., Landsverk, J., & Kitchen, L. (1991). Deaths from falls in children: How far is fatal? *The Journal of Trauma*, 31(10), 1353–1355.
- Chapman, L. A., Wade, S. L., Walz, N. C., Taylor, H. G., Stancin, T., & Yeates, K. O. (2010). Clinically significant behavior problems during the initial 18 months following early childhood traumatic brain injury. *Rehabilitation Psychology*, 55(1), 48–57. <http://dx.doi.org/10.1037/a0018418>
- Chevignard, M., & Lind, K. (2014). Long-term outcome of abusive head trauma. *Pediatric Radiology*, 44(Suppl. 4), S548–S558. <http://dx.doi.org/10.1007/s00247-014-3169-8>
- Chevignard, M., Toure, H., Brugel, D. G., Poirier, J., & Laurent-Vannier, A. (2010). A comprehensive model of care for rehabilitation of children with acquired brain injuries. *Child: Care, Health and Development*, 36(1), 31–43. <http://dx.doi.org/10.1111/j.1365-2214.2009.00949.x>
- Circulaire DAS/DE/DSS n° 96-428 relative à la prise en charge médico-sociale et à la réinsertion sociale et professionnelle des personnes atteintes d'un traumatisme crânien. (1996, juillet 4). (1996). *Bulletin officiel du ministère chargé des affaires sociales* n° 96/32.
- Committee on Child Abuse and Neglect. (2001). Shaken baby syndrome: Rotational cranial injuries technical report. *Pediatrics*, 108(1), 206–210. <http://dx.doi.org/10.1542/peds.108.1.206>
- Duhaime, A. C., Christian, C., Moss, E., & Seidl, T. (1996). Long-term outcome in infants with the shaking-impact syndrome. *Pediatric Neurosurgery*, 24(6), 292–298.
- Eslinger, P. J., Flaherty-Craig, C. V., & Benton, A. L. (2004). Developmental outcomes after early prefrontal cortex damage. *Brain and Cognition*, 55(1), 84–103. [http://dx.doi.org/10.1016/S0278-2626\(03\)00281-1](http://dx.doi.org/10.1016/S0278-2626(03)00281-1)
- Eslinger, P. J., Grattan, L. M., Damasio, H., & Damasio, A. R. (1992). Developmental consequences of childhood frontal lobe damage. *Archives of Neurology*, 49(7), 764–769.
- Ewing-Cobbs, L., Prasad, M., Kramer, L., & Landry, S. (1999). Inflicted traumatic brain injury: Relationship of developmental outcome to severity of injury. *Pediatric Neurosurgery*, 31(5), 251–258.
- Fanconi, M., & Lips, U. (2010). Shaken baby syndrome in Switzerland: Results of a prospective follow-up study, 2002–2007. *European Journal of Pediatrics*, 169(8), 1023–1028. <http://dx.doi.org/10.1007/s00431-010-1175-x>
- Fischer, H., & Allasio, D. (1994). Permanently damaged: Long-term follow-up of shaken babies. *Clinical Pediatrics*, 33(11), 696–698.
- Greiner, M. V., Lawrence, A. P., Horn, P., Newmeyer, A. J., & Makoroff, K. L. (2012). Early clinical indicators of developmental outcome in abusive head trauma. *Child Nervous System*, 28(6), 889–896. <http://dx.doi.org/10.1007/s00381-012-1714-z>
- Hymel, K. P., Makoroff, K. L., Laskey, A. L., Conaway, M. R., & Blackman, J. A. (2007). Mechanisms, clinical presentations, injuries, and outcomes from inflicted versus noninflicted head trauma during infancy: Results of a prospective, multicentered, comparative study. *Pediatrics*, 119(5), 922–929. <http://dx.doi.org/10.1542/peds.2006-3111>
- Ivles, P., Lintrop, M., Talvik, I., Sisko, A., & Talvik, T. (2010). Predictive value of clinical and radiological findings in inflicted traumatic brain injury. *Acta Paediatrica (Oslo, Norway: 1992)*, 99(9), 1329–1336. <http://dx.doi.org/10.1111/j.1651-2227.2010.01820.x>
- Johnson, A. R., DeMatt, E., & Salorio, C. F. (2009). Predictors of outcome following acquired brain injury in children. *Developmental Disabilities Research Reviews*, 15(2), 124–132. <http://dx.doi.org/10.1002/ddrr.63>
- Karandikar, S., Coles, L., Jayawant, S., & Kemp, A. M. (2004). The neurodevelopmental outcome in infants who have sustained a subdural haemorrhage from non-accidental head injury. *Child Abuse Review*, 13(3), 178–187. <http://dx.doi.org/10.1002/car.850>
- Keenan, H. T., Hooper, S. R., Wetherington, C. E., Nocera, M., & Runyan, D. K. (2007). Neurodevelopmental consequences of early traumatic brain injury in 3-year-old children. *Pediatrics*, 119(3), e616–e623. <http://dx.doi.org/10.1542/peds.2006-2313>
- Laurent-Vannier, A., Nathanson, M., Quiriau, F., Briand-Huchet, E., Cook, J., Billette de Villemeur, T., Chazal, J., Christophe, C., Defoort-Dhellemmes, S., Fortin, G., Rambaud, C., Raul, J. S., Rey-Salmon, C., Sottet, F., Vieux, E., Vinchon, M., & Willinger, R. (2011). A public hearing “Shaken baby syndrome: Guidelines on establishing a robust diagnosis and the procedures to be adopted by healthcare and social services staff”. Guidelines issued by the Hearing Commission. *Annals of Physical and Rehabilitation Medicine*, 54(9–10), 600–625. <http://dx.doi.org/10.1016/j.rehab.2011.10.002>
- Laurent-Vannier, A., Toure, H., Vieux, E., Brugel, D. G., & Chevignard, M. (2009). Long-term outcome of the Shaken baby syndrome and medicolegal consequences: A case report. *Annals of Physical and Rehabilitation Medicine*, 52(5), 436–447. <http://dx.doi.org/10.1016/j.rehab.2009.03.001>
- Limond, J., Adlam, A.-L. R., & Cormack, M. (2014). A model for pediatric neurocognitive interventions: Considering the role of development and maturation in rehabilitation planning. *The Clinical Neuropsychologist*, 28(2), 181–198. <http://dx.doi.org/10.1080/13854046.2013.873083>
- Makoroff, K. L., & Putnam, F. W. (2003). Outcomes of infants and children with inflicted traumatic brain injury. *Developmental Medicine and Child Neurology*, 45(7), 497–502.
- Miner, M., & Houston Conference on Neurotrauma. (1986). *Neurotrauma: Treatment, Rehabilitation, and Related Issues*. Boston: Butterworths.
- Niederkrotenthaler, T., Xu, L., Parks, S. E., & Sugerman, D. E. (2013). Descriptive factors of abusive head trauma in young children – United States, 2000–2009. *Child Abuse & Neglect*, 37(7), 446–455. <http://dx.doi.org/10.1016/j.chiabu.2013.02.002>
- Oliver, J. E. (1975). Microcephaly following baby battering and shaking. *British Medical Journal*, 2(5965), 262–264.
- Rhine, T., Wade, S. L., Makoroff, K. L., Cassidy, A., & Michaud, L. J. (2012). Clinical predictors of outcome following inflicted traumatic brain injury in children. *The Journal of Trauma and Acute Care Surgery*, 73(4 (Suppl. 3)), S248–S253. <http://dx.doi.org/10.1097/TA.0b013e31826b0062>
- Stipanich, A., Nolin, P., Fortin, G., & Gobeil, M.-F. (2008). Comparative study of the cognitive sequelae of school-aged victims of Shaken Baby Syndrome. *Child Abuse & Neglect*, 32(3), 415–428. <http://dx.doi.org/10.1016/j.chiabu.2007.07.008>
- Talvik, I., Männamaa, M., Jüri, P., Leito, K., Pöder, H., Hämarik, M., Kool, P., & Talvik, T. (2007). Outcome of infants with inflicted traumatic brain injury (shaken baby syndrome) in Estonia. *Acta Paediatrica (Oslo, Norway: 1992)*, 96(8), 1164–1168. <http://dx.doi.org/10.1111/j.1651-2227.2007.00362.x>

- Talvik, I., Metsvaht, T., Leito, K., Pöder, H., Kool, P., Väli, M., Lintrop, M., Kolk, A., & Talvik, T. (2006). Inflicted traumatic brain injury (ITBI) or shaken baby syndrome (SBS) in Estonia. *Acta Paediatrica (Oslo, Norway: 1992)*, 95(7), 799–804. <http://dx.doi.org/10.1080/08035250500464923>
- Tanoue, K., Matsui, K., Nozawa, K., & Aida, N. (2012). Predictive value of early radiological findings in inflicted traumatic brain injury. *Acta Paediatrica (Oslo, Norway: 1992)*, 101(6), 614–617. <http://dx.doi.org/10.1111/j.1651-2227.2012.02635.x>
- Taylor, H. G., Yeates, K. O., Wade, S. L., Drotar, D., Stancin, T., & Burant, C. (2001). Bidirectional child-family influences on outcomes of traumatic brain injury in children. *Journal of the International Neuropsychological Society*, 7(6), 755–767.
- Williams, R. A. (1991). Injuries in infants and small children resulting from witnessed and corroborated free falls. *The Journal of Trauma*, 31(10), 1350–1352.
- Yeates, K. O., Swift, E., Taylor, H. G., Wade, S. L., Drotar, D., Stancin, T., & Minich, N. (2004). Short- and long-term social outcomes following pediatric traumatic brain injury. *Journal of the International Neuropsychological Society*, 10(3), 412–426. <http://dx.doi.org/10.1017/S1355617704103093>
- Yeates, K. O., Taylor, H. G., Walz, N. C., Stancin, T., & Wade, S. L. (2010). The family environment as a moderator of psychosocial outcomes following traumatic brain injury in young children. *Neuropsychology*, 24(3), 345–356.