Disability and Visual Outcomes following suspected

Abusive Head Trauma (AHT) in children under 2 years

Corresponding Author
Dr Juliana Wright
[1]Department of Paediatrics, Oxford University Hospitals NHS Foundation Trust, Headley Way, Oxford OX3 9DU
juliana.wright@ouh.nhs.uk
07736933701

Co-authors
Miss Sally L. Painter[2], Dr Sheethal Kodagali[1], Dr Nicholas R. Jones[3], Mrs Andrea Roalfe[3], Dr Sandeep Jayawant[4], Mr John Elston[5], Dr Geetha Anand[1]

[1]Department of Paediatrics, Oxford University Hospitals NHS Foundation Trust, Headley Way, Oxford OX3 9DU
[2]Paediatric Ophthalmology, Birmingham Children's Hospital, Steelhouse Lane, Birmingham, B4 6NH
[3]Nuffield Department of Primary Care Health Sciences, University of Oxford
[4]Paediatric Neurology, Oxford University Hospitals NHS Foundation Trust, Headley Way, Oxford OX3 9DU
[5]Paediatric Ophthalmology, Oxford University Hospitals NHS Foundation Trust, Headley Way, Oxford OX3 9DU

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ABSTRACT

Aim: To report disability and visual outcomes following suspected AHT in children under 2 years.

Methods: We present a retrospective case series (1995 to 2017) of children with suspected AHT aged ≤24 months. KOSCHI (King's Outcome Score of Childhood Head Injury) was used to assess disability outcomes at hospital discharge and at follow up. The study used a retinal haemorrhage score (RHS) to record findings at presentation and a visual outcome score at follow up.

Results: We included 44 children (median age 16 weeks). At presentation, 98% had a subdural haemorrhage and 93% retinal haemorrhage. At discharge, 61% had moderate to severe disability, and 34% a good recovery. A higher RHS was observed in those with more disability (r=-0.54, p=0.0002).

At follow up, 14% had a worse KOSCHI score (p=0.055). 35% children had visual impairment, including 9% with no functional vision. Those with poorer visual function had a higher RHS (r=0.53, p=0.003). 28% attended mainstream school without support; 50% were in foster care or had been adopted, 32% lived with birth mother and 18% with extended family.

Conclusion: It is known that injuries from suspected AHT result in high levels of morbidity; our cohort showed significant rates of disability and visual impairment. Those with higher disability at discharge and poorer visual function showed more significant retinal changes. The extent of disability was not always apparent at hospital discharge, impacting on provision of prognostic information and targeted follow-up.

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INTRODUCTION

Abusive Head Trauma (AHT) describes an inflicted injury to the head. It typically presents as an encephalopathic infant with intracranial and retinal haemorrhages (1), although a range of signs and symptoms are recognised. Incidence in the United Kingdom (UK) ranges between 21 to 24 per 100,000 children under 1 year (2) and it is associated with significant morbidity and mortality. The intracranial injury is commonly a subdural haemorrhage (SDH); characteristically bilateral, multiple, with interhemispheric fissure and posterior fossa involvement when compared to unintentional injury (3). Retinal haemorrhages in AHT are typically bilateral, multi-layered, with extension to the periphery of the retina(4). In a child with head trauma and retinal haemorrhages, the odds ratio of AHT is 14.7 (95% confidence interval 6.39 to 33.62) and the probability of abuse 91%(5), as against non-abusive head trauma.

Studies of long-term outcomes associated with AHT are limited but suggest two thirds of survivors suffer long-term neurological, behavioural or cognitive effects. Additional difficulties such as language, socio-behavioural, visual, hearing and sleep problems are reported(6), but are thought to be under-recognized. Literature on visual outcomes is limited, with visual impairment varying from 40% (7) – 66% (8).

The primary objective of this study was to determine the immediate and long-term disability outcomes in children under 2 years of age following suspected AHT. The secondary objective was to report visual outcomes.

METHODS

The NHS Health Research Authority approved this retrospective cohort study, with Ethical approval granted from the London-Hampstead Research Ethics Committee (reference 16/LO/2133).

We performed a case note review which identified children aged 2 years or less, from the Ophthalmology Department's database of children examined for suspected AHT. These cases were tertiary referrals from the local area and surrounding District General Hospitals, between 1995 and 2017. Each case had undergone a comprehensive medical and social care assessment in line with the departmental safeguarding policy for suspected non-accidental injury. We included only those cases where there was an explicit clinical conclusion of AHT. The assessment included evidence of encephalopathy, intracranial injury, ophthalmic assessment and skeletal survey in varying combinations to form the overall clinical picture. Naturally occurring causes of the presentation were excluded by appropriate investigation. Clinicians' follow up documentation was also used but court proceedings were not, due to inconsistent filing in the clinical notes. The specifics of departmental safeguarding policies evolved over the study period but the distinguishing clinical features and principles of a full Multi-Disciplinary Team (MDT) assessment remained constant. The term 'suspected AHT' has been used throughout the manuscript in acknowledgement that the full causative circumstances may never fully be known.

Children were excluded if there was not enough evidence for a diagnosis of AHT, there was a pre-existing developmental or visual problem, or insufficient medical information to complete a proforma.

A standardized data collection proforma was used to record anonymised data. King's Outcome Scale of Childhood Head Injury (KOSCHI) was used as a measure of disability(9). This was chosen as it was devised specifically for children with a head trauma, with the advantages are that it is easy to apply retrospectively, providing functional information, and is applicable to both short and long-term outcomes. The score was allocated from the detailed clinical notes of two Senior Paediatric Neurologists describing the child's developmental and functional profile. Additional information from standardised neurodevelopmental or neuropsychological assessment was included where available. Wilcoxon signed rank test was used to compare KOSCHI scores at discharge and follow-up. McNemar's test was used to compare the proportion of children making a good or full recovery (KOSCHI 5) at discharge and follow-up.

Ophthalmic data

All children had been assessed at presentation by the same Senior Ophthalmologist (JE) including pupil dilated indirect ophthalmoscopy, with RetCam and Optos imaging when available. Retinal findings were graded with a Retinal Haemorrhage Score (RHS) adapted from Binembaum et al(10) to include structural retinal changes (retinoschisis and peri-macular folds). These are described in shaking injuries (11) and are relevant as features of poor visual outcomes. Damage to the development of normal binocular vision was assessed at follow-up using a Visual Function Score (VFS), scored between 0 (normal vision) and 7 (no useful visual function). Associations between ordinal scales were measured by Spearman's rank correlation. Statistical analysis was performed with Stata v15.0.

RESULTS

In total, 44 children were included from an original cohort of 64. 25 (57%) were male, and 19 (43%) female with an age range from 19 days to 13 months (median=16 weeks). Those that were excluded did not fulfil our inclusion criteria for various reasons (n=2 aged more than 2 years, n=3 possible alternative diagnosis, n=15 lack of follow-up data). Subdural haemorrhage was present in all but one child; additional intracranial and skeletal injuries were frequent (Table 1). Two thirds were admitted to Paediatric Intensive Care (29/44 [65%]), of whom 26 required ventilation, 10 cardiovascular support, and 29 neurosurgical intervention. The mean duration of Intensive Care stay was 6.3 days (range 1 to 16 days).

Table 1: Type, severity and patterns of physical injury

Injury	**Number (%)**
Subdural haemorrhage	43 (98)
• Bilateral	32 (74)
• Unilateral	11 (26)
Subarachnoid haemorrhage	7 (15.9)
Intra-ventricular haemorrhage	4 (9.1)
Para-falcine haemorrhage	2 (4.5)
Parenchymal infarct	3 (6.8)
Cerebellar infarct	1 (2.3)
Hydrocephalus	2 (4.5)
Midline shift	2 (4.5)
Hypoxic ischaemic encephalopathy	12 (27.2)
Upper thoracic spinal subdural haematoma	1 (2.3)
Neurosurgical Intervention	29 (66)
• Intracranial pressure monitoring	12 (27)
• Subdural aspirations	13 (30)
• Peritoneal shunt	8 (18)
• Craniotomy	4 (9)
• Extra-ventricular drain	1 (2)
Skeletal Survey	32 (72.7)
• Abnormal	18 (56)
○ Long bone fractures	10 (22.7)
○ Rib fractures	10 (22.7)
○ Skull fractures	8 (25)
○ Vertebral fractures	1 (3.1)

• Multiple abnormalities	7 (38)

Disability

At hospital discharge, 17/44 (38%) had severe disability (KOSCHI 3), 10 (23%) moderate disability (KOSCHI 4) and 15 (34%) had made a good recovery (KOSCHI 5). Two of the cohort died (KOSCHI 1) before discharge, and no patients were in a vegetative state (KOSCHI 2).

At follow-up, 19 (45%) children were severely disabled (KOSCHI 3), 11 (26%) moderately disabled (KOSCHI 4) and 12 (29%) had made a good recovery (KOSCHI 5). The average length of follow-up was 4 years 5 months (0.2-13.8 years; interquartile range 8.8 years). There were no later deaths.

Six (14%) children had a worse KOSCHI score at follow-up than at discharge, representing increasing level of disability (p=0.055). The children originally classified as making a good or full recovery (KOSCHI 5a & 5b), dropped from 34% to 29% at follow-up (p=0.08). Two children dropped from KOSCHI 5a at discharge to 4b at follow-up, and one child dropped from KOSCHI 5a to 4a. The additional challenges for these cases were poor concentration, behavioural challenges, or social and emotional differences. One child showed a small improvement, from KOSCHI 4a to 4b, but functionally remained within a moderate disability category. These data are shown in Table 2.

Table 2: Number (%) by KOSCHI score at discharge and at follow-up

KOSCHI Score		Discharge (n=44)	Follow-up (n=42)
1	Death	2 (5)	-
2	Vegetative	0	0
3 a	Severe Disability	9 (20)	11 (26)

b		8 (18)	8 (19)
4 a b	Moderate Disability	7 (16) 3 (7)	6 (14) 5 (12)
5 a b	Good Recovery Full recovery	5 (11) 10 (23)	4 (10) 8 (19)

Neuropsychology evaluation was available for a sub-group of 7 children with moderate to severe disability at follow-up. These children had Learning Disability (n=4), visuo-spatial difficulties (n=3), Autism Spectrum Disorder (n=2), Attention Deficit Hyperactivity Disorder (n=1), epilepsy (n=2), tics (n=1) and sleep difficulties (n=1). At school age, data available for 25 children showed that 12 (48%) were in a specialised educational setting, 6 (24%) were in mainstream school with support, and 7 (28%) in mainstream school with no support. At follow-up, care arrangements included placement with a grandparent (n=5), fostered or adopted (n=14) or placed with single parent (mother) (n=9). None were in their father's care.

Visual Outcomes

Retinal haemorrhages (RH) were present in 93%, bilateral in 88%. 1 case had a sub-conjunctival haemorrhage and 2 had no ophthalmic signs of AHT. The retinal haemorrhage grading score (n=43) ranged from 0 to 16 (median=8). A negative association was found between RHS and KOSCHI scores at discharge ($r=-0.54$, $p=0.0002$), with higher retinal haemorrhage score observed with lower level of recovery.

Follow-up data were available for 34 children. The mean age was 1.0 years (range 1 month – 11.8 years), with 13 children having follow-up beyond one year. Normal vision (VFS 0) was found in 65% children, visual impairment (VFS 1 – 6) in 26%, and no functional vision (VFS 7) in 9%. Whilst most children had a visual function score of 0 (indicating normal visual function), 6 children had a score of 5 or more, indicating severe visual impairment. Of the children who had a VFS score of 5 or more, the underlying cause for poor vision was ocular damage (permanent retinal damage or optic atrophy n=2), cortical visual impairment with no ocular damage (n=1), and mixed ocular and cortical visual impairment (n=3). Two children had jerky pursuit eye movements at the ages of 1 months and 11.1 years respectively, suggesting that the fine control mechanisms of eye movements had been affected. Data was available for 29 children with both RHS and VFS; analysis showed that those with a higher retinal haemorrhage score tended to have worse visual outcomes ($r=0.53$, $p=0.003$), see figure 1.

Figure 1 Visual function vs retinal haemorrhage score

DISCUSSION

Our retrospective study shows significant visual impairment and disability in children under 2 years with suspected AHT. Over 90% of cases had both SDH and retinal haemorrhages and other injuries typically seen in AHT, such as long-bone fractures and ischaemic encephalopathy, were also present.

Significant long-term injury was higher in our cohort compared to existing literature reports. Our study was conducted at a tertiary centre, which increases the likelihood of the most

severe cases of AHT. Although the cases analysed were from the Ophthalmology database, it included all suspected AHT referrals, a small number of which did not have retinal haemorrhage.

The AHT literature relies on a variety of different definitions, with variables including age range, and type of injury; comparing outcomes from different studies requires caution. Our findings support the existing research showing that long-term disability following AHT is common(1,6,7). There was some evidence ($p=0.055$) of a difference between the discharge and follow-up disability scores, with a tendency to worsen over time. We also highlight those originally classified as making a good or full recovery (KOSCHI 5a & 5b), dropping to a moderate disability score at follow-up ($p=0.08$). Although this did not show a statistically significant result in our dataset, it remains an important observation for the individual child and their family. Those in the higher functioning group are more likely to be discharged from services and this is relevant if co-morbidity is recognised later.

In addition, we describe co-morbidity from social, emotional and behavioural conditions, as well as epilepsy and sleep difficulties in a subsection of the cohort. Whilst there may be genetic and environmental influences that could contribute to these diagnoses, this data emphasises the negative implications of AHT for overall long-term functional outcome.

AHT typically occurs in infants, damaging an immature and developing brain. It is unsurprising therefore that disability rates are high, and additional impairment is being recognised at a later age when more complex, higher functioning skills are demanded. Of concern is the fact that assessment at discharge appears to under-estimate the long-term

risk of disability. Follow-up MRI, particularly with newer available technologies, may provide valuable information to support prognosis(3) and is an area of further research.

Many previous studies have sought to define the retinal findings in AHT(4). Our study has linked a score to include features seen in shaking injury with functional visual outcomes. We demonstrate a significant finding of poorer visual function in those with a higher retinal haemorrhage score. A second significant finding using this score was of higher levels of disability at discharge in those with a higher RHS. Further research on longer-term outcomes is required.

Strengths and Limitations

The strengths of this study are in the use of standardised assessments, independent senior review of both KOSCHI and Ophthalmic analysis, and the length of patient follow-up. Making a diagnosis of suspected AHT is challenging and over the time period of this study, the evidence base and thus individual departmental safeguarding guidelines have changed. We acknowledge this; at the time of injury the cases included were investigated by experienced Consultants across all relevant sub-specialist fields. We are confident that we have only included clear cases where there was no ambiguity in the clinical conclusion of AHT, based on the best available investigations and evidence at the time. Our follow-up data also supported this.

There are inherent difficulties in collecting and interpreting retrospective data, and we acknowledge that a standardised neuropsychology assessment would be a preferred way of detailing outcomes. The KOSCHI score has limitations of interpretation in younger children, particularly under 2 years. It has been used in other studies of suspected AHT in the

absence of a suitable alternative retrospective measure. Some data remained unobtainable, despite efforts to retrieve it from local hospitals.

CONCLUSION

This cohort suffered high rates of morbidity from significant disability (71%) and visual impairment (31%). Those with higher disability at discharge and poorer visual function showed more significant retinal changes. This study highlights that deficits may become more apparent over time which has implications for counselling families, and also in adequate provision of services for support. Long-term prospective studies would provide additional evidence for monitoring and support of neurodevelopmental and visual co-morbidity, to improve functional outcome for the affected children and their families.

What is already known on this topic?

1. Abusive head trauma in children under 2 years causes significant disability in survivors
2. Visual impairment following AHT is variable and not widely studied

What this study adds:

1. We add to the sparse literature showing that disability level may be underestimated at time of discharge from hospital

2. Children with poorer visual function at follow-up showed a higher retinal haemorrhage score

Contributorship Statement:

JE, NJ, SJ and GA planned and designed the study, including the ethics application. Ophthalmic data analysis and interpretation by SP and JE. Overall data collection by SK, JE and GA. GA and JE critically revised the manuscript. Statistical analysis by SP, JW and AR. Paper written by JW, with all authors approving the final paper.

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