



ORIGINAL ARTICLE

Circularity bias in abusive head trauma studies could be diminished with a new ranking scale



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Abstract: Causality in abusive head trauma has never been fully established and hence no gold standard exists for the diagnosis. Implications hereof include bias introduced by circular reasoning and a shift from a trustful doctor patient relationship to a distrustful one when the caregiver statement is questioned. In this paper we examine seven recent abusive head trauma studies including 476 diagnosed abuse cases for circular reasoning as well as the role of the caregiver statement in the diagnosis. Secondly, we present a novel ranking scale for the diagnosis of abusive head trauma designed to minimize circular reasoning. We found circularity to be a potential source of bias in all seven studies. The caregiver statement (lack of trauma mechanism or trauma mechanism considered incompatible with clinical findings) was listed as a diagnostic item in 329 (69%) of 476 cases. Applying our ranking scale to the abuse cases showed that the demands of our ranking scale were not fulfilled in 440 (92%) cases. We conclude that most abuse cases in the studies were, to some extent, diagnosed on criteria based on circular reasoning. The caregiver statement was one of the most frequently used diagnostic items. Hypothetically, caregivers offer no or inadequate explanation to the clinical findings in assumed abuse cases. Thus, when this feature is encountered, it is regarded as indicative of abuse adding further to the risk of circularity bias. We propose the use of our novel ranking scale in abusive head trauma research in an effort to minimize circular reasoning.

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1. Introduction

In 1971 pediatric neurosurgeon Gutkelch hypothesized, that intracranial and intraocular bleeding in children with no external signs of injury was the result of shaking.¹ Radiologist Cafey subsequently published on the subject in a similar vein.² This hypothesis has been subjected to debate because the causal connection between exposure and clinical findings has never been established. It is unclear if shaking on its own is forceful enough to produce the clinical findings or if there is a lucid interval between exposure and onset of symptoms. Additionally, re-bleeding of a chronic subdural hematoma and strokes has been suggested as the cause of clinical findings in suspected abuse cases.³⁻⁶ Despite these controversies it is widely accepted that the triad findings of subdural hemorrhage (SDH), retinal hemorrhage (RH) and encephalopathy are pathognomonic or highly specific for the syndrome known today as “Abusive Head Trauma” (AHT), so named by the AAP committee COCAN (American Academy of Pediatrics and Committee on Child Abuse and Neglect).⁷

Because the causal connection between exposure and clinical findings has never been established, no gold standard or standard case definition for AHT exists. Often clinical findings are occult and only investigated and disclosed when they are suspected and since the background population prevalence of these findings is unknown, the same applies to their predictive value. When there is no standard definition of abuse, it is often defined by the same variables that are subsequently analyzed as variables of abuse. Or, as stated by Piteau et al., “As there are no standardized criteria for the definition of abuse, most authors developed their own criteria, and many of these are fraught with circular reasoning”.⁸

Recent attempts have been made to combine data from several observational studies to identify common diagnostic ground which has resulted in a substantial increase in clinical findings considered indicative of AHT.⁸⁻¹⁰ These findings now include subdural hemorrhage, cerebral ischemia, retinal hemorrhages, skull fractures, intracranial injury, metaphyseal fractures, long bone fracture, rib fracture, seizure, apnea, bruising of the head, neck, ear and torso and no adequate history of trauma. The predictive value of this increasing list of findings is unknown and as long as research is fraught with circularity bias, the attempt to identify common diagnostic ground is more or less futile.

Circularity bias in AHT studies has received increased attention by several authors and different approaches have been suggested with the specific aim of avoiding circularity.^{8,11,12} Among these is the ranking scale developed by Maguire et al.⁹ (Box 1). However, “...for features that have been traditionally associated with abuse (such as subdural hemorrhage and retinal hemorrhage), this ranking scale does not compensate well for circularity” as suggested by Piteau et al.⁸

When the causative mechanisms in AHT are unclear, the evaluation of the proposed injury mechanism offered by the caregiver becomes important. Hypothetically, caregivers offer no or inadequate explanation to the clinical findings in assumed abuse cases. Thus, when this feature is encountered, it is regarded as indicative of abuse adding further to the risk of circular reasoning as well as creating an atmosphere of distrust in the doctor-patient relationship.

Box 1 Ranking scale for the diagnosis of AHT suggested by Maguire et al.⁹

Abuse ranking	Criteria used to define abuse
1	Abuse confirmed at case conference or civil, family, or criminal court proceedings or admitted by perpetrator or independently witnessed
2	Abuse confirmed by stated criteria, including multidisciplinary assessment
3	Abuse diagnosis defined by stated criteria
4	Abuse stated as occurring, but no supporting detail given as to how it was determined
5	Abuse stated simply as “suspected”; no details on whether it was confirmed

The objective of this short paper falls in two parts; firstly we examine recent AHT studies for possible bias caused by circular reasoning as well as the role of the caregiver statement in the abuse diagnosis. Secondly, we present a novel ranking scale designed specifically with the intent of avoiding circularity in AHT studies.

2. Methods and results

Two recent AHT studies were selected based on the study design and number of abuse cases. The studies’ inclusion criteria were examined for circularity and checked against our own suggested ranking scale for abuse. The first study was conducted in 2011 by Maguire et al.⁹ who selected 14 AHT studies identified as “high quality comparative studies”. Of these, six entered the study based on data availability. Only cases of confirmed abuse as defined by the ranking scale previously presented by the authors were included. The other study was conducted in 2013 by Hymel et al.¹⁰ who aimed at deriving a clinical prediction rule that, if validated, could be used as a tool for *excluding* AHT by identifying predictive clinical variables. They conducted their study from 14 pediatric intensive care units across the USA and the study population consisted of children less than three years old with acute head-injury admitted for intensive care. Children were categorized as abused by the following six definitional criteria which, according to the authors, were selected specifically to avoid circularity; admission by caregiver, independently witnessed abuse, caregiver denying head trauma, inconsistencies in the account by caregiver over time, caregiver account inconsistent with the developmental state of the child and presence of extracranial injuries considered suspicious of abuse. The following clinical variables were identified as predictive; acute respiratory compromise, seizures, bruising of ear, neck or torso, subdural hemorrhage and skull fracture.

The ranking scale we developed has three levels; ranking 1, 2 and 3. Ranking 1 is regarded as first grade evidence followed by ranking 2 and 3 in declining order.

Ranking 1: *Recorded*. With the introduction of smart cell-phones follows a potential for recording everyday events in spontaneous home-videos. We consider it likely that the number of recorded cases of abuse (as well as accidents) of small children and infants will increase and we regard

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