The Challenges of Assessing the Incidence of Inflicted Traumatic Brain Injury

A World Perspective

Desmond K. Runyan, MD, DrPH

Objective: Commentary on the methods available for ascertaining the incidence of inflicted traumatic brain injury (inflicted TBI) and the difficulties involved in defining and measuring this condition in young children.

Design: Review of published and unpublished international data regarding parental shaking of infants compared to studies assessing incidence.

Results: Review of parental report data reveal that the shaking of young children is a surprisingly common act in a wide variety of countries and cultures. While 2.6% of parents of children aged under 2 years in the U.S. report shaking their child as an act of "discipline," survey data from lesser-developed countries on four continents indicate that shaking, as a form of discipline, may be many times more common among infants in their countries and that the consequences, short of hospitalization or death, are inadequately studied. Methodologic challenges to epidemiologic work to develop better estimates are discussed.

Conclusions: These data highlight the challenges faced in ascertaining the epidemiology of inflicted TBI in young children. While there is scientific evidence that the shaking of young children can produce profound destruction of children's brains and lives, these data reveal that there are many other children who are shaken by their caregivers but escape the acute clinical presentation of "shaken baby syndrome" or for whom the injuries are not recognized as due to inflicted TBI. The impact of these private acts must be further studied as there may be other long-lasting and serious intracranial impacts that have not been characterized.
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Introduction

More than 60 years ago, John Caffey noted an association between long-bone fractures and subdural hematomas and reported that some children in his series had retinal hemorrhages.[1] In 1971, A. Norman Guthkelch, an English neurosurgeon, described two children with subdural hematomas with no external signs of trauma and postulated the role of rotational forces as the mechanism of injury.[2] John Caffey coined the term "shaken baby syndrome" in 1972 for children with intracranial hemorrhage without external signs of trauma inflicted by shaking the child.[3] He described a combination of intracranial and intraocular bleeding and metaphyseal "chip" fractures of the long bones linked to vigorous shaking. The mechanism of injury is postulated to be the abrupt acceleration–deceleration injury with rotational forces.[4,5] These movements cause motion of the brain within the skull and dura, tearing bridging vessels passing through the dural membrane and leading to intracranial hemorrhage. A variety of other intracranial injuries are described including intraparenchymal shear injuries in the brain, subarachnoid hemorrhage, diffuse axonal injury, and hypoxic–ischemic encephalopathy.[6] Terms applied to the condition by investigators include abusive head trauma (AHT), inflicted traumatic brain injury (inflicted TBI), shaken baby syndrome (SBS), whiplash shaken baby syndrome and shaken-impact syndrome.[5]

Despite the volume of case reports, reviews, and cohort studies, there remain significant questions related to the amount of force, duration, and even the nature of the shaking event. It is hypothesized that traumatic brain injury (TBI) in young children, inflicted by shaking, may be an occult but leading cause of infant mortality and mental retardation in the developing world. Studies of the incidence of these behaviors and studies assessing the consequences are needed to establish the extent and impact of the shaking on children. These data will be useful in establishing the extent of the problem, allowing the development of

From the Department of Social Medicine, The University of North Carolina at Chapel Hill, Chapel Hill, North Carolina

Address correspondence and reprint requests to: Desmond K. Runyan, MD, DrPH, Campus Box 7240, Department of Social Medicine, The University of North Carolina, Chapel Hill NC 27599-7240. E-mail: drunyan@unc.edu.

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careful epidemiologic studies of the consequences, and assessing progress in prevention.

Review of Parental Self-Report Data

In a 1995 Gallup poll survey, 4.4% of parents of children aged <2 years in the U.S. reported having shaken their child as a means of discipline.[7] This survey asked just a single question among a variety of other questions regarding discipline strategies. More information is needed to establish the importance of this observation. However, it was not an isolated observation. A recent survey of parental discipline practices in North and South Carolina revealed that in 2002, 2.6% of parents of children aged <2 years surveyed by an anonymous telephone survey reported using shaking to discipline the child.[8] Interestingly, older children were shaken at about the same rate; 2.5% of parents of children aged >2 years also reported using shaking as discipline. These data are limited as the parents were responding to a single question on the Conflict Tactics Scale–Parent-Child version (CTS-PC).[9] Information about the vigor or nature of the shaking that the parents describe is not available. Regardless, the prospect that 2.6% of parents report shaking young infants is a distressing discovery.

In surveys parallel to that completed in the Carolinas, clinical epidemiologists in the International Clinical Epidemiology Network (INCLEN) completed population-based analyses of parents in specific communities in India, Chile, the Philippines, Egypt, and Brazil (manuscript in preparation). This project, entitled WorldSAFE, has been described in the WHO's *World Report on Violence and Health*.[10] In population-based samples of mothers that ranged from 400 to over 1000 in size, the investigators asked mothers what disciplinary practices they had used in the past year with one child, selected randomly from among that mother's children and identified by age and gender. Thus, it was possible to examine discipline patterns by age and gender. The frequency of shaking reported by mothers of children aged <3 years ranged from 6.6% of mothers in the Manila, Philippines, neighborhood of Paco to 42% of mothers in the urban slums of India. The rate of admitted shaking for children aged <3 years was 19% in a community in Egypt and 23% in nonslum portions of India. In addition to the proportion of mothers reporting shaking, the frequency with which shaking was used by a mother was highly variable.

Analyses of the patterns in these countries is ongoing, but the high rate of 42% of infants shaken in the urban slums of India mirrors the rate of mothers reporting zero years of education. An examination of the difference in frequencies of shaking for older children and younger children reveals that, across samples in disparate countries, infants are reportedly shaken at a rate of 83% of the rate of older children. In some countries, however, the shaking of younger infants greatly exceeded that for older children, while in a few other countries the pattern was clearly that the older children were most at risk. Figure 1 presents the WorldSAFE data with the North and South Carolina data included, although the other countries examined rates for children aged <3 years and the Carolina data were calculated for children aged <2 years.

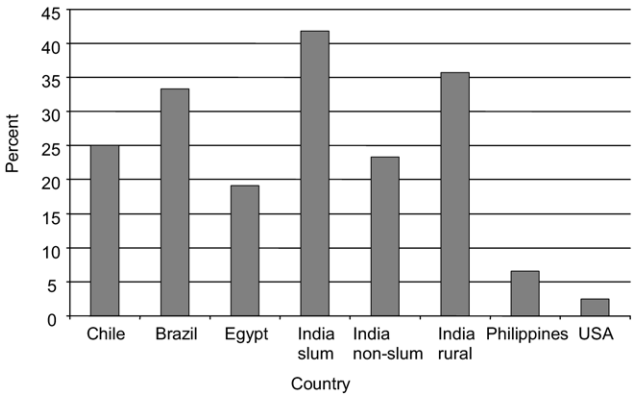

Figure 1. WorldSAFE reports of shaking of young children for discipline.

In 2004, UNICEF added a set of seven parental discipline questions to the third version of the Multiple Indicator Cluster Survey (MICS3),[11] a household survey used to assist countries in filling data gaps for monitoring the situation of children and women with a variety of measures referred to as indicators. It is intended to produce statistically sound and internationally comparable estimates of these indicators. The MICS was developed to measure progress toward international goals in child wellbeing. MICS1 was conducted around 1995 in more than 60 countries; MICS2 was conducted in 2000 and MICS3 is now in the field. One MICS3 question is nearly identical to the CTS-PC and WorldSAFE questions; that question specifically asks if the parent has shaken the child. The MICS3 instrument has been limited to families with children aged >2 years, and the time period for the question is specified to be within the past month. MICS3 survey data, anonymized as to the specific countries providing the data, have been shared with the author for two eastern European countries, three central Asian countries, and one West African country. With sample sizes of over 3000 parents in each country, self-reported shaking of children as discipline in the last month ranged from 18% to 36%. These data have not been stratified by child age or by family composition. Nevertheless, they suggest that shaking is a frequent disciplinary strategy. These preliminary MICS3 data, with the identity of the individual countries obscured, are presented in Table 1.

In 2005, the International Society for the Prevention of Child Abuse and Neglect (ISPCAN), as a contribution to the UN Secretary-General's ongoing study of

Table 1. UNICEF MICS3 data on frequency of shaking as child discipline[a]

Country	N	% shaken
West Africa	6019	36
Central Asia	5369	36
Central Asia 2	5064	30
Central Asia 3	3297	18
Eastern Europe	1004	18
Eastern Europe 2	2697	18

[a]Children aged >2 years. (Data from Edilbarto Loaiza, UNICEF, personal communication).
MICS, Multiple Indicator Cluster Survey.

children and violence decided to build on the work of the WorldSAFE consortium and develop a new set of instruments for completion by children, by parents, and by newly emancipated youth that might shed light on the frequency of harsh disciplinary practices and/or abuse in all parts of the world (manuscript in preparation).

In 2006, the ISPCAN supported a series of pilot studies of a new instrument that is similar in intent to the CTS-PC in that parents are asked about discipline behaviors. This instrument was developed with input from over 100 scientists in 40 countries in order to produce an instrument that would be useful across cultures and in developing countries. While the samples that were administered this instrument in the pilot study were small, the data again lead the reader to conclude that shaking as discipline is quite common (Figure 2).

Collectively, these data indicate that the shaking of young children is a common phenomenon with a frequency among countries often referred to as lesser-developed countries exceeding, in a dramatic fashion, that of the U.S. While the nature and duration of the shaking reported by these parents is not known, for most of the children it is likely to be brief and of relatively low severity. Using the calculated rate of inflicted TBI leading to hospitalization or death of 17 per 100,000 children in the first 2 years of life from North Carolina,[12] and 2.6% of parents admitting shaking, the ratio of children hospitalized or dying from inflicted neurotrauma compared to the numbers of reported shaken children may be estimated at 1 to 152. Apparently, many shaken children escape detection and perhaps even significant health consequences. It is unknown whether shaking of these young infants in a more mild fashion can be linked to the development of mental retardation, learning difficulties, or behavioral problems. Mental retardation and behavioral problems have been shown for children whose TBI does come to medical attention. If the ratio of shaken to injured infants is an accurate one, and the ratio holds for other countries, shaking may be a leading cause of infant mortality and morbidity throughout the world. More careful study is needed to assess the degree of harm.

Discussion of Methodologic Issues to Assess Inflicted TBI Incidence

Challenges Related to Changes in Risk Factors

Accurate assessment of the incidence of inflicted TBI has profound implications for child health. Public health evaluations of the frequency of this phenomenon can help in the design and assessment of prevention and intervention programs. Policy and programmatic efforts of public health agencies are developed, in large part, in response to data about threats to health. Current efforts at preventing inflicted TBI require careful evaluation of whether these efforts have any evidence of effect. With relatively low base occurrences in the population and risk factors such as first child, young parent, natural disasters, and even military parents that need to be considered, there are likely to be rather dramatic fluctuations in cases seen at tertiary care centers.[13,14] With low base occurrences, the confidence intervals will be large around estimates of frequency and comparisons around intervention programs will lack statistical power. Distinguishing natural variation and the impact of other events on any apparent reductions in caseload is needed in order to determine whether cases are less frequent after prevention efforts. Enthusiasm for reduced caseloads in hospitals after education programs must be tempered by recognition that the caseload in eastern North Carolina in 2001 was half the caseload in 2000, and the only difference was that the area did not sustain a devastating hurricane in the second year.

Determining a Case Definition

When is a case of inflicted TBI appropriate to be counted? As a part of assessing incidence, an important task is defining what constitutes a case. Do all children need to have evidence of intracranial injury? How should the presence or absence of skull fractures be

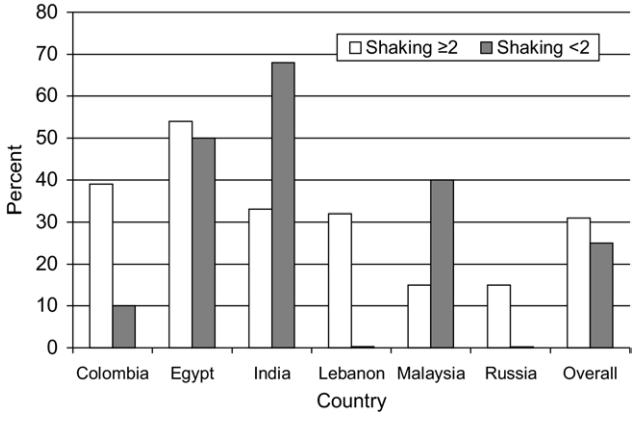

Figure 2. International Society for the Prevention of Child Abuse and Neglect (ISPCAN) Child Abuse Screening Tool (ICAST-P) pilot data for convenience samples of about 100 parents in each country.

included? What about children with retinal hemorrhages or metaphyseal chip fractures in the absence of intracranial injury? Should epidural hematomas ever be included? The inherent difficulty is that the injuring of young children by an adult shaking them is likely to be a private act, experienced in a childcare setting in which the caregiver and the child are alone. While this private act can have devastating consequences, the child cannot report and the caregiver may hesitate to admit for fear of family and/or legal ramifications. In a series of cases from North Carolina, nearly 60% of the cases of what was judged to be inflicted TBI had no history by a caregiver of what might have happened.

Data Quality and Recording

Low base-occurrence conditions require studies extending across multiple communities or even across states and countries. Collection of data by investigators or their staff in far-flung parts of a country complicates efforts to ensure uniform coding and definitions. Individual records may be accessed by only a single individual, and assuring that the individual's reading and recording of pertinent facts mirrors what is done in other locations involves extensive training and well-developed coding manuals. Any incidence study of TBI will require careful training and standard approaches to definitions.

Establishing Denominators at Risk

One of the hallmarks of public health research is the concept of population-based research in which the population to be studied is clearly defined and there is little confusion about calculating incidence and prevalence. Studies based on caseloads of individual hospitals or even collected hospitals are ultimately unsatisfactory for establishing incidence when the population that the hospital serves is uncounted and other hospitals may see some of the children that are of interest. Efforts to establish cooperative research among hospitals and using carefully defined population characteristics can help sort this threat to incidence estimates.

Summary

Incidence estimates for inflicted TBI are of more than academic interest. Issues of hypotheses related to learning disabilities, developing-world infant mortality, the usefulness of prevention strategies, and even the impact of natural disasters are all tied to our ability to ascertain whether the problem is increasing or decreasing in society. Improved data about the degree of force and the risk of other minor injuries mimicking inflicted TBI will be better addressed by clear and careful estimates of the incidence of inflicted TBI. Investigators will need to attend to sources of data, case definitions, coding of data, multisite collaborations, and defining the population at risk.

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