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Age-related incidence curve of hospitalized Shaken Baby Syndrome cases: Convergent evidence for crying as a trigger to shaking[☆]

Ronald G. Barr^{a,b,*}, Roger B. Trent^c, Julie Cross^c

^a *Department of Pediatrics, University of British Columbia, Faculty of Medicine, Vancouver, BC, Canada*

^b *Centre for Community Child Health Research, BC Children's and Women's Hospital,
4480 Oak St, L408, Vancouver, BC, Canada V6H 3V4*

^c *Epidemiology and Prevention for Injury Control Branch, California Department of Health Services,
Sacramento, CA, USA*

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Abstract

Objective: To determine whether there is an age-specific incidence of hospitalized cases of Shaken Baby Syndrome (SBS) that has similar properties to the previously reported “normal crying curve,” as a form of indirect evidence that crying is an important stimulus for SBS.

Design and setting: The study analyzed cases of Shaken Baby Syndrome by age at hospitalization from hospital discharge data for California hospitals from October 1996 through December 2000.

Patients: All cases of children less than 18 months (78 weeks) of age for whom the diagnostic code for Shaken Baby Syndrome (995.55) in the International Classification of Disease, Ninth Edition, Clinical Modification was assigned.

Results: There were 273 hospitalizations for SBS. Like the “normal crying curve,” the curve of age-specific incidence starts at 2–3 weeks, has a clear peak, and declines to baseline by about 36 weeks of age. In contrast to the normal crying curve that peaks at 5–6 weeks, the peak of SBS hospitalizations occurs at 10–13 weeks.

Conclusions: The age-specific incidence curve of hospitalized SBS cases has a similar starting point and shape to the previously reported normal crying curve but the peak occurs about 4–6 weeks later. Of the likely predisposing

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* Corresponding authors.

causes, this pattern is only consistent with the properties of early crying. There are numerous explanations for the lag in the peaks between crying and SBS hospitalizations, including the possibility of repeat shakings prior to hospitalization. The importance of crying as a stimulus to SBS may provide an opportunity for preventive intervention.

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Keywords: Crying; Shaken Baby Syndrome; Child abuse; Prevention; Battering

Introduction

Shaken Baby Syndrome (SBS) is a form of intentional injury to infants and children inflicted by violent shaking with or without concomitant contact with a hard surface, resulting in head trauma including subdural hematomas, diffuse axonal injury, and retinal hemorrhages but also often fractures of the long bones or ribs, with little or no external evidence of trauma. Although usually attributed to John Caffey (Caffey, 1972, 1974) the first explicit report of shaking resulting in such lesions was published by the British pediatric neurosurgeon Dr. Norman Guthkelch (Guthkelch, 1971). In one of the case histories in Guthkelch's series and occasionally in Caffey's original reports (Caffey, 1974, 1972), crying as a proximal stimulus for the shaking is explicitly mentioned. In subsequent articles and reviews of Shaken Baby Syndrome, crying is often mentioned as a precipitant (Dykes, 1986; Levitt, Smith, & Alexander, 1996; Ludwig, 1984; Reijneveld, van der Wal, Brugman, Sing, & Verloove-Vanhorick, 2004) usually based on anecdotal reports. This is sometimes supported by the observation that the median age of the cases occurs in the first few months of life when crying is greatest. In the first report of incident factors limited to fatal cases garnered from investigative reports in the United States Air Force, Brewster, Nelson, and Hymel (1998) reported that perpetrators mentioned crying as a stimulus in 58% of the cases. The role of crying as a precipitating stimulus for shaking has also been incorporated in policy statements concerning Shaken Baby Syndrome. In the Policy Statement of the American Academy of Pediatrics on "Shaken Baby Syndrome: Rotational Cranial Injuries" (Committee on Child Abuse and Neglect of the American Academy of Pediatrics, 2001), "crying or irritability" is described as "often" the proximal cause of shaking, and pediatricians are encouraged to ask about "response to the crying infant" as part of anticipatory guidance to prevent Shaken Baby Syndrome. Similarly, in the Canadian "Joint Statement on Shaken Baby Syndrome" (2001) co-signed by eight organizations committed to its prevention, infant demands and "especially crying" are cited as triggers for shaking in exhausted or frustrated caregivers.

Despite the reasonableness and acceptance of the assumption that crying is a trigger for shaking, the objective data supporting its role is limited at best. However, in the last 40 years, increasingly careful investigations of infant crying behavior have demonstrated specific and robust properties of crying that contribute to the frustration that caregivers experience in the first few months of life (Barr, Paterson, MacMartin, Lehtonen, & Young, 2005; Barr, St. James-Roberts, & Keefe, 2001). Furthermore, many of these properties have characteristics that provide an opportunity to acquire indirect convergent evidence of the importance of crying as a stimulus for shaking. The most important of these is a robust age-related pattern of crying. This pattern is manifest as increases in the average daily duration of crying in the first few weeks, a peak sometime in the second month of life, and then decreases to more stable levels by about the fifth month.

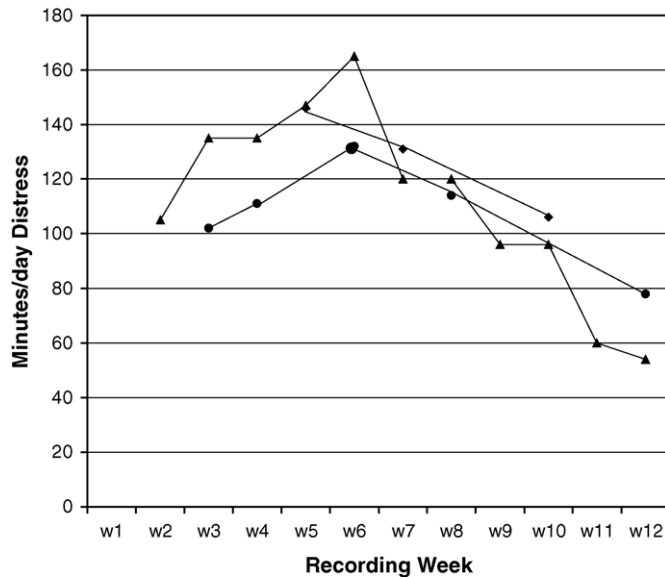


Figure 1. Crying amounts and patterns from three North American studies illustrating absence of secular trend. Triangles: data from Brazelton (1962); circles: data from Hunziker and Barr (1986); diamonds: data from Kramer et al. (2001).

This pattern was first described as typical of normally developing infants by Brazelton in 1962 who asked parents in his Cambridge, MA, practice to keep daily diaries of distress for the first 12 weeks of life (Brazelton, 1962). It has come to be known as the “normal crying curve.” Since then, a number of investigators have replicated this pattern (Alvarez & St. James-Roberts, 1996; Hunziker & Barr, 1986; St. James-Roberts & Halil, 1991). The replications have indicated how robust the pattern is, and that there has been very little secular change in either the pattern or the amounts of distress behavior since Brazelton’s original report. This is illustrated in Figure 1. The median amounts of distress behavior from Brazelton’s 1962 study (Brazelton, 1962) is represented along with the mean amounts of distress (crying and fussing) reported in Hunziker and Barr’s Montreal study in 1986 (Hunziker and Barr, 1986), and in a more recent Montreal study in 2001 using the same diary (Kramer et al., 2001). The important features of this pattern are that there is not simply “more” crying in the first weeks of life, nor is it just a generalized, linear decrease in crying from birth to older ages. Importantly, it manifests the properties of an “n-shaped” developmental curve, with a phase of increasing crying, a discernible peak or maximum, and a phase of decreasing crying. Although not illustrated by the data sets in Figure 1, this phase is followed by a period of relatively stable, low level crying throughout the remainder of the first year of life at less than half the overall amounts achieved in the first 3 months (St. James-Roberts & Halil, 1991).

In addition to the “peak” pattern, crying has other properties that are also specific to this age period that contribute to caregiver frustration. Most of these are included in clinical descriptions of “colic syndrome,” a syndrome that is now recognized as the upper end of a spectrum of crying phenomenology in otherwise normal infants (Barr, 1989, 1999, 2000; Ghosh & Barr, 2000; St. James-Roberts, 2001; St. James-Roberts, Conroy, & Wilsher, 1995, 1996). These properties include prolonged, unsoothable crying bouts that occur unexpectedly, seemingly unrelated to anything in the environment, during which the infant manifests a

facial grimace, increased motor tone, and curling of its legs up over its abdomen that raise concerns about gastrointestinal pain in many caregivers. These bouts do not occur randomly throughout the day, but tend to cluster in the late afternoon and evening hours. Although the prolonged, unsoothable crying bouts comprise less than 10% of the overall crying of infants with colic, they are specific to the first few months of life, and occur rarely thereafter (Barr et al., 2005).

Consequently, if crying is a stimulus behavior for shaking injuries as the anecdotal evidence and common sense suggests, the prediction would be that both the timing and the shape of the age-specific incidence of Shaken Baby Syndrome should be similar. However, a test of this hypothesis requires a different reporting of childhood injuries than is typical in order to test this hypothesis. In most reports, injury estimates are based on conventional age groupings of 1–4 years of age or 1 year age groupings (Agran et al., 2003). These are clearly too broad and too late to capture a developmentally based stimulant condition such as crying behavior. In a recent study designed to capture developmentally related risks of injury specific to young children, Agran et al. utilized E-codes (external cause of injury codes) that would be reflective of age-related developmental characteristics and 3-month age categories. Their study demonstrated that there was marked variability in both rates and specific causes of injury by 3-month age groupings. Although the leading cause of injury falls for the 0–3 years age group, battering (E-codes 967.0–967.9) had the highest rates specifically between 0 and 5 months, three to four times higher than any rate following 9 months of age.

While the early “battering” cluster in the Agran study included Shaken Baby Syndrome cases, both the codes and the 3-month age grouping are too broad for a specific indirect test of the hypothesis that the properties of early infant crying could be a stimulus for Shaken Baby Syndrome. In our study, we take advantage of a specific code for Shaken Baby Syndrome introduced into the California version of the ICD-9CM and analyze age-specific incidences of hospitalizations of SBS to determine whether SBS incidence reflects the temporal properties of early infant crying behavior.

Methods

We used data from the California hospital discharges from October 1996 through December 2000 to identify the day of age and cause of injury to California children less than 18 months (78 weeks) of age. A new code for Shaken Baby Syndrome was implemented in California on October 1, 1996. We examined data for 3 and 1/4 years because this produces larger and more stable numbers of hospitalizations per age. Hospital discharge data for California hospitals in 1996–2000 were obtained from the Office of Statewide Health Planning and Development (OSHPD), California Health and Human Services Agency. By law, each civilian hospital in California must report data to OSHPD on each hospital discharge, including up to 25 diagnosis codes, based on the *International Classification of Disease, Ninth Edition, Clinical Modification* (1996) for each initial hospitalization for injury. Subsequent hospitalizations for the same injury are excluded. However, hospitalizations that result in the patients dying in the hospital are included. OSHPD edits each discharge report for accuracy. We used the version of the publicly available data that includes age at admission reported in years and days for children up to their fourth birthday. We selected records of California residents who were younger than 18 months and had a diagnosis of 995.55 (Shaken Baby Syndrome). We analyzed the data in 2-week age brackets (0 and 1 week of age, 2 and 3 weeks of age, etc.) to be sure that we could identify the beginning of Shaken Baby Syndrome hospitalizations. However, the data are graphed in 4-week age brackets, as 2–5, 6–9, 10–13 weeks of age, and so on in

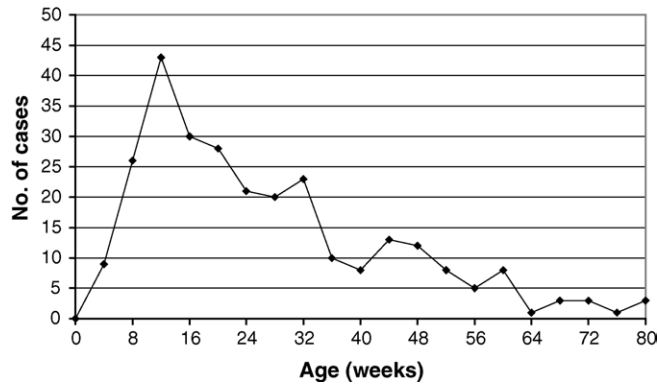


Figure 2. Age-specific number of hospitalized cases of Shaken Baby Syndrome in California hospitals from October 1996 through December 2000, in 4-week brackets.

order to illustrate better the shape of the age-specific incidence curve (Figure 2). To determine whether there was a risk of a “reporting epidemic” with the introduction of the new code, we performed a linear least-squares regression on quarterly reports of cases. Since only two cases were reported during the first quarter (the last quarter of October through December 1996) and this was clearly an outlier, only the subsequent quarters (years 1997 through 2000) are included (see Figure 3). Since all data are from a public use file with no identifiers, no IRB approval was required in order to carry out these analyses.

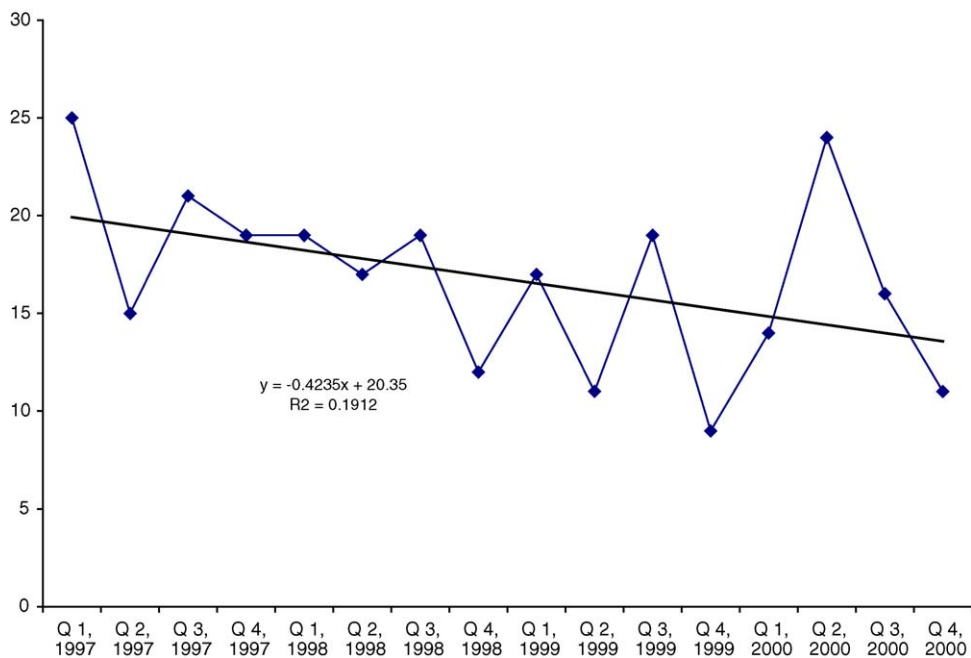


Figure 3. Trend in reported SBS hospitalizations by quarter year, California, 1997–2000.

Results

There were 273 hospitalizations for Shaken Baby Syndrome between October 1, 1996 and December 31, 2000. The youngest cases ($n = 3$) are reported for infants of 2–3 weeks of age. After that, the number of cases rises rapidly, reaching a peak at 10–13 weeks of age, and then shows a more gradual decline (Figure 2). At about 36 weeks of age, the number of cases remains around 10 per 4-week period for the remainder of the first year, and then consistently less than 10 after the first year.

To determine whether there was evidence for a “reporting epidemic” or a secular trend in the reporting of SBS hospitalizations, the data were reanalyzed to reflect the number of cases of infants under 18 months of age who were reported in successive 3-month quartiles from January 1, 1997 through December 31, 2000 (Figure 3). The regression is significant ($r = .44$, $p < .05$) and shows a consistent downward trend in the face of considerable quarter to quarter variability.

Discussion

Although it is widely assumed that crying is a trigger for Shaken Baby Syndrome, the objective data supporting its role is limited at best. Indeed, in Brewster et al.’s 1998 report on 32 maltreatment deaths in the armed forces, they note that there were no previous studies at that time that examined factors surrounding the final, fatal incident. It is understandable that systematic evidence implicating crying (or any other behavior) as the stimulus is rare for a number of reasons. First, the syndrome itself, although all too common, remains a relatively infrequent diagnosis, and therefore difficult to study. In perhaps the best incidence study to date, Shaken Baby Syndrome was reported to occur in 24.6 per 100,000 children (95% confidence interval 14.9–38.5) under 1 year of age in and around Edinburgh, Scotland (Barlow & Minns, 2000). In a study of inflicted traumatic brain injury in North Carolina, the incidence in infants under 1 year of age was 29.7 per 100,000 person years (95% confidence intervals 22.9–36.7) (Keenan et al., 2003). Second, the incidents of shaking themselves are rarely witnessed by a third party. Third, records are often missing or incomplete. In Brewster et al.’s 1998 study, most of the records were missing for 8 of 40 (20%) substantiated cases. Fourth, perpetrators are usually not forthcoming about the actual incidents surrounding the shaking. When they do offer a description, it may not be an accurate one. A probable reason for the lack of an accurate description is that shaking an infant simply because it was crying would be unlikely to be accepted as a legal defense. Fifth, it is extremely difficult to know whether a history of crying or colic obtained at the time of diagnosis reflects crying secondary to the trauma inflicted by the shaking (including broken bones) or a cause of the shaking. The cause-effect relationship is especially difficult if there have been repeated episodes of shaking prior to the one that results in the diagnosis (Alexander, Crabbe, Sato, Smith, & Bennett, 1990; Brewster et al., 1998; Ewing-Cobbs et al., 1998; Jenny, Hymel, Ritzen, Reinert, & Hay, 1999). For these reasons, it is unlikely that there will ever be an accurate direct accounting of the role of crying as a stimulus for Shaken Baby Syndrome.

However, the very specific age-related “normal” crying pattern that is associated with prolonged and inconsolable crying bouts provides an opportunity to test indirectly the hypothesis that crying is a trigger for some cases of Shaken Baby Syndrome. As predicted, the age-specific incidence curve for hospitalized cases of Shaken Baby Syndrome in California hospitals has a number of similar, but not in all cases identical, properties compared with the “normal crying curve.” The first is that the curve begins its ascent when infants are 2–3 weeks of age. Although none of the published studies of diary recorded crying

includes data before the second week of age, most of the clinical descriptions of colic indicate that the syndrome begins to be expressed at about 2 weeks of age (Barr, 1991; Wessel, Cobb, Jackson, Harris, & Detwiler, 1954). The second is that the number of cases rises rapidly thereafter. The third is that there is a recognizable peak or maximum that occurs in the region of 10–13 weeks of age. Fourth, following the peak, there is an almost linear decline until about 36 weeks of age, after which the rate remains at about 10 cases per 4-week age block until the end of the first year of life. After 1 year, the age-specific incidence remains very low, if not nonexistent. In short, the age-specific incidence curve in hospitalized Shaken Baby Syndrome cases has a similar starting point and a similar shape to the normal crying curve.

The main difference between the two curves is the relatively later appearance of the maximum incidence for Shaken Baby Syndrome hospitalizations (10–13 weeks of age) compared to the peak of the crying curve (5–6 weeks of age). There are a number of possible explanations for this lag. One is, of course, that we have no indication in this data set as to what stimulated the shaking in these cases. Consequently, many of these violent acts may have other triggers or no triggers at all. A second is that there is some other developmental influence that is determining of SBS hospitalization incidence. Possible candidates could be difficulties with feeding or sleeping, two other early regulatory behaviors that can be of significant concern to caregivers. There are, however, no obvious reasons why this peak pattern should relate to feeding or sleeping. Concerns for these behaviors are not related to the crying pattern (Wolke, Meyer, Ohrt, & Riegel, 1995). Feeding problems are likely to occur earlier, and sleep usually tends to get better at 3–4 months, when “sleeping through the night” begins to be established (Anders, 1979; Anders, Halpern, & Hua, 1992). Third, there are a number of plausible reasons for the lag that are consistent with crying being the stimulus behavior. For example, there is increasing evidence that a significant number of diagnosed cases have suffered previous episodes of shaking and/or abuse (Alexander et al., 1990; Brewster et al., 1998; Ewing-Cobbs et al., 1998; Jenny et al., 1999). Jenny et al. (1999) reported that 31% of 173 cases were “missed” when presenting the first time to emergency services. In the series reported by Alexander et al. (1990), 50% of victims of shaking that resulted in intracranial injury had evidence of coexisting direct external trauma, and 71% had evidence of prior abuse, neglect, or both. In the series by Ewing-Cobbs et al. (1998), 45% of the children with inflicted head injury had signs of pre-existing brain injury. In short, the shaking episode that brings the child to the emergency room may only be the last in a series of shaking episodes that began days to weeks earlier. Another factor that may contribute to the later peak for SBS hospitalizations is the increasing recognition that there are a small number of infants who continue to have persistently high crying levels after the 2-month peak (sometimes referred to as “persistent caregiver-infant distress syndrome” [Barr, 1995, 2000; Papousek & von Hofacker, 1995]). There is also a small number of infants without prior colic at the typical time who become high criers later (Barr, 2002; Clifford, Campbell, Speechley, & Gorodzinsky, 2002). Both of these groups may provide cases of crying-stimulated shaking that contribute to the delay in the peak incidence of Shaken Baby Syndrome cases. While some or all of these factors may contribute to the time lag in the peak of the Shaken Baby Syndrome hospitalizations, there is no independent evidence within the study to assess their relative contribution.

The remarkably similar onsets and shapes, including the distinct peak patterns in the early months of life, of the age-related hospitalizations and crying serve as indirect evidence convergent with common sense and the anecdotal reports that early crying is a likely stimulus for Shaken Baby Syndrome. Indeed, in light of the early clustering of all forms of “battering” in the first 5 months of life (Agran et al., 2003), crying may have wider significance as a stimulus for other forms of abuse as well.

If crying is a stimulus for shaking and other forms of abuse, there are a number of implications for increasing the effectiveness of primary prevention programs for Shaken Baby Syndrome. An important one is that improving the understanding by caregivers of the unique properties of early crying and its ability to frustrate them (Donovan, Leavitt, & Walsh, 1997; Leavitt, 2001) should contribute to reducing the likelihood of shaking in response to crying. Unfortunately, parent advice about crying generally focuses on strategies for calming excessively crying infants or infants with colic. It rarely acknowledges that the prolonged unsoothable crying bouts and a peak pattern of crying are likely to occur regardless of soothing methods used and whether or not an infant has “colic.”

To address this need, prevention programs are being developed that are not limited to admonitions simply to “Never shake or hurt your infant.” Rather, Shaken Baby Syndrome is described as a serious negative consequence that occurs when frustrated caregivers lose control and shake their infants. In some of these programs, the frustrating properties of crying in normal infants are emphasized so that caregivers will be able to recognize them and be less frustrated by them. To illustrate, the National Center on Shaken Baby Syndrome has developed an educational program called the “Period of PURPLE Crying” to transmit this information in an effective, brief and memorable way (www.dontshake.com). The letters of the word “purple” each refer to one of six properties contributing to caregiver anxiety and frustration (P for the peak pattern, U for the unexpected timing of prolonged crying bouts, R for resistance to soothing, P for the pain-like grimace even though the infants are not in pain, L for the long crying bouts, and E for the later afternoon and evening clustering). Although the effectiveness of this or other interventions remains to be demonstrated, the likely relationship between the properties of early crying and shaking abuse incidents may provide improved opportunities for successful prevention efforts.

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Résumé

French language abstract not available at the time of publication.

Resumen

Spanish language abstract not available at the time of publication.