SPECIAL ISSUE: ABUSIVE HEAD TRAUMA

Epidemiological data on shaken baby syndrome in France using judicial sources

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Abstract

Background The frequency of and risk factors for shaken baby syndrome remain poorly documented for several reasons: the real number of "benign" cases of shaken baby syndrome are not known; information sources used are diverse, there have been changes over time in the definition of this pathology and few population-based epidemiological studies are available.

Objective Estimate the frequency of fatal shaken baby syndrome and determine its risk factors through research carried out on fatal cases in three regions of France while comparing them to data from international publications.

Materials and methods A retrospective epidemiological study of all cases of fatal shaken baby syndrome affecting infants younger than 1 year of age who were examined by the courts during a 5-year period in a defined geographical area. Shaken baby syndrome cases were compared with infanticide cases for risk factors and a comparison was made of family characteristics with those of the general population.

Results Thirty-seven cases of shaken baby syndrome were recorded (a rate of 2.9 for 100,000 live births). As in other studies, we found a strong predominance of male victims (78%), young age (median age: 4 months) and a high rate of prematurity (22%). Conversely, results on family educational and socioeconomical levels differ from those reported in numerous studies. Parent perpetrators of shaken baby syndrome belong to higher social classes than those of other types of homicide and socially reflect the population they come from.

Conclusion Our study suggests 1) that epidemiological studies on shaken baby syndrome should include both medical and judicial information sources and 2) that primary prevention strategies (especially in maternity wards) should target all social classes.

Introduction

While there are abundant publications of a clinical nature on shaken baby syndrome, few population-based epidemiological studies exist that enable calculating the frequency of this pathology and identifying its risk factors.

This lack of an epidemiological approach has several explanations. First, the number of unknown "benign" cases makes it difficult to measure the global frequency of the syndrome. These cases, which are no doubt frequent, are not always diagnosed when they occur and their sequelae are probably rarely linked to their initial cause. In addition, the difficulty of comparing frequencies calculated from several different studies is related to the nature and number of information sources used as well as to changes over time in nosologies applied to the pathological condition. Thus, shaken baby syndrome, abusive head trauma, nonaccidental head injury and inflicted traumatic brain injury are not synonymous terms and do not necessarily encompass the same anatomical lesions. Recent publications have attempted to address the question of terminology [1], with the American Academy of Pediatrics adopting the term abusive head trauma to include any mechanism (shaking, impact, etc.) resulting in inflicted injury to the head and its contents [2].

In France, in spite of the recognition starting in the 1960s that a substantial part of subdural hematomas are of intentional origin, the concept of shaking as a causal mechanism was not evoked until the late 1980s [3]. This terminology is still in use

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and is the one employed in diagnostic recommendations issued in 2011 by the High Authority on Health and by SOFMER (a French society of physical medicine and rehabilitation) [4].

Data presented below are from a population-based study on cases of fatal shaken baby syndrome recorded during a 5-year period by the justice system.

Materials and methods

Cases included in the study and documents used

A retrospective study was carried out in 26 of 27 courts (one small court refused to participate) in 3 regions of France: Brittany, Île de France (Paris region) and Nord-Pas-de-Calais. These regions have very different socioeconomical characteristics and rural/urban composition. During the study period, there were 1,286,253 live births, comprising 34.6% of all births from the 22 regions of continental France [5]. Included in the study were all cases of infants dying before the age of 1 year during a 5-year period (1996–2000) that were submitted to the courts by the state prosecutors' offices in the three regions. This period was chosen so judicial proceedings would be concluded at the time of data analysis. Complete court case data cover the period from 1996 to 2008, due to often lengthy investigations.

The study was carried out at all levels of the judicial system, including criminal and appellate courts. Following identification and selection of cases using computer tools available in each court, judicial files were obtained and data entered into individual anonymous questionnaires by the research team (a field coordinator and two trained investigators). Each file was exhaustively examined and several documents were analyzed: transcripts of interviews by police and interrogation by the investigating judge of the author and witnesses (parents, family members, friends, work colleagues, neighbors and first responders at the scene of the death), prosecutor's charges and the report of the forensic examination of the infant. Therefore, each case makes available medicolegal data, information on death circumstances, family characteristics and judicial management of the case.

Definition

The definition of shaken baby syndrome is the one recorded by the experts in forensic medicine who performed the autopsies and physical and histological examinations, and then used by judicial professionals. Shaken baby syndrome was defined as the association of one or more subdural hematomas and retinal hemorrhages, with the possible presence of axonal injury or cerebral edema.

Scientific steering committee for the research

An expert committee monitored the research project, bringing together representatives of the judicial system, pediatrics, forensic medicine, pathology, maternal and child health, psychology and epidemiology.

Study population

Two hundred forty-seven cases were recorded, 80 of which were considered by judicial professionals as intentional violent deaths. After reviewing all cases, the expert committee added 12 cases considered by the justice system to be accidental or natural deaths, but for which there were discrepancies between the nature of the lesions, the mechanisms evoked, and the age and level of development of the infant. These 92 cases of homicide of infants younger than 1 year of age are divided into three categories: 37 cases of shaken baby syndrome, 32 neonaticides and 23 other filicides.

Analysis

Frequency was calculated by relating the number of cases to the 1,286,253 live births during the study period. Risk factors were determined using two types of comparisons: 1) the comparison between the 37 cases of shaken baby syndrome and the 55 homicides (32 neonaticides and 23 filicides) and 2) the comparison of the characteristics of parents whose children died from shaken baby syndrome with those from the general population of the same age and regions, using data from the 1999 general population census [6].

With the authorization of the French Data Protection Authority, judicial data were crosschecked with official mortality statistics, using the date of birth and death, sex, administrative district of residence and death, as well as cause and place of death. World Health Organization International Classification of Diseases (ICD) cause of death codes assigned to children in the mortality statistics were matched to causes of death assigned to cases examined in the courts study.

Statistical analysis was carried out using the SAS software (SAS Institute Inc, Cary, North Carolina, USA). Bivariate analysis was done using the chi-square test and Fisher exact test for small numbers. Differences between groups were considered as statistically significant for P-values of 0.05 or less.

Results

Frequency

The rate of fatal shaken baby syndrome was 2.9 for 100,000 live births. This rate is a good deal higher that the one

Table 1 Distribution of the 37 cases of shaken baby syndrome according to diagnoses given in official mortality statistics *

World Health Organization International Classification of Diseases (ICD) causes	
Homicide	12
Nontraumatic cerebral hemorrhages	10
Accidental cerebral hemorrhages	7
Injury unspecified	4
Various medical pathologies	3
Unknown cause	1

*Codes of the 9th revision of the ICD for the research period 1996–1999 - Codes of the 10th revision of the ICD for 2000

calculated from official mortality statistics (Table 1). Indeed, at the time of death and certification of its cause, only one-third of cases were considered as being of intentional origin and nearly another third were attributed to nontraumatic cerebral hemorrhages.

Infant risk factors

In comparison with infant victims of homicide, those who died from shaking were characterized by a clear predominance of males (78% boys; Table 2), but there was no difference in the age distribution. The median age for fatal shaken baby syndrome cases was 4 months.

The percentage of premature infants was high in the two groups, especially in cases of shaken baby syndrome (22%) but is not statistically different.

On the other hand, there were significantly more histories of abuse among infant victims of shaken baby syndrome than among those dying from filicide (Table 2). More than half of infants who died from shaking presented with prior subdural hematomas and/or long-bone fractures, visceral lesions and skin lesions. Multiple shaking incidents were found in 13 cases.

Family risk factors

Among the 37 cases, the suspected perpetrator of the shaking incident was the mother, the father or both in 31 cases, and a baby-sitter in 6 cases. Perpetrators of shaking confessed in 24 of the 37 cases (no baby-sitters confessed to shaking the child).

The families of infants dying from shaken baby syndrome differed significantly from those of the other group of infants: parents lived as a couple more frequently (only 6% of mothers lived alone and 14% did not live with the biological father of the infant); the mother was more educated (nearly half had studied at a university) and was more often employed, in a white-collar job in a third of cases (Table 3).

Table 2 Characteristics of the deceased children according to the cause of violent death (shaken baby syndrome or infant homicide)

Characteristics of the deceased children	Shaken baby syndrome (n=37) %	Homicides* (n=55) %
Gender		
Male	78	46
Female	22	54
	P=0.003	
Age at death**		
0–1 month	30	30
2–5 months	46	48
≥ 6 months	24	22
	NS	
Prematurity/low birth weight		
No	78	85
Yes	22	15
	NS	
Prior abuse**		
No	46	73
Yes	54	27
	P=0.05	

NS not significant

* Homicides include neonaticides (32 cases) and infant homicides (23 cases)

** Neonaticides excluded

The comparison of the distribution of professional categories of mothers who shook their infants (24, with 20 for whom information is available) shows it to be identical to that of professions in the general population to which they belong (Table 4).

Discussion

The frequency of fatal shaken baby syndrome calculated from this French study is 2.9 per 100,000 live births. Most recent epidemiological studies on shaken baby syndrome (or the different syndromes with new names [abusive head trauma, inflicted traumatic brain injury and nonaccidental head injury]) [7–14] were carried out on children younger than 2 years of age, rates by year of age are not always available and fatal cases are not always counted separately. The only two known studies comparable to ours for these characteristics are a Swiss study on shaken baby syndrome that gives a rate of fatal cases of 2.3 per 100,000 for infants younger than 1 year of age [7] and an Estonian study on inflicted traumatic brain injury that proposes a rate of 4.4 per 100,000 [14]. This last rate should be viewed with caution given the small number of fatal cases

Table 3 Characteristics of the deceased children's families according to the cause of violent death (shaken baby syndrome or infant homicide)

Characteristics of the families	Shaken baby syndrome (n=37) %	Homicides* (n=55) %
Mother's age**		
<25 years	30	36
25-29 years	35	29
≥30 years	35	36
	NS	
Mother's educational level**		
University level		
No	54	74
Yes	46	26
	P=0.05	
Mother's occupation**		
White collar	33	10
Employee or worker	27	34
No professional activity	39	56
	P=0.05	
Mother living alone**		
Yes	6	33
No	94	67
	P=0.003	
Biological father present at home		
Yes	86	60
No	14	40
	P=0.02	
Father's occupation		
White collar	37	22
Employee or worker	50	56
No professional activity	13	22
	NS	
Number of siblings		
0–1	69	70
≥2	31	30
	NS	
Abuse in the sibship***		
No	85	88
Yes	15	12
	NS	

NS not significant

* Homicides include neonaticides (32 cases) and infant homicides (n=23)

** 10 cases with unidentified mothers are excluded

*** If any other sibling present (n=47)

Table 4 Comparison of socioprofessional categories of mothers implicated in cases of shaken baby syndrome (n=24) with expected categories, based on those of the general population. There was no significant difference among the groups

	Distribution of socioprofessional categories of mothers perpetrators of shaken baby syndrome *		
	Observed numbers	Expected numbers**	Significance
White collar	4	5.3	
Employee or worker	6	7.5	NS
Unemployed	10	7.2	

* Socioprofessional category is known for 20 mothers out of the 24 implicated

** The expected numbers for each professional category if national population census data by Insee for 1999 are applied (in the same departments of the three regions, for women ages 15 to 44 years).

(four). In addition, the definition of nonaccidental head injury is clinically broader than that of shaken baby syndrome. Information sources for these two studies are solely medical (the Swiss Pediatric Surveillance Unit, and the principle neurosurgery departments and pediatric intensive care units in Estonia). The two studies state they are nationwide and thus have a geographical base, as does our study, which enables calculating frequencies and producing representative results.

Reported rates for infants younger than 1 year of age and for all cases (fatal, serious and benign) vary widely, going from 13 to 14 per 100,000 (Canadian study by Fujiwara [8]; study of nonaccidental head injury based on the British Paediatric Surveillance Unit [10]; the Swiss study [7]) to nearly 30 for 100,000 (28.7 in the Estonian study of inflicted traumatic brain injury [14]; 29.7 in the study of inflicted traumatic brain injury carried out in pediatric intensive care units in North Carolina hospitals [11]) to even more than 30 (33.8 in a study carried out in the Lothian region of Scotland [12]; 34 in a study of abusive head trauma carried out on a national level among American military families [9]; 39.8 in another national American study of abusive head trauma [13] carried out using the "Kids Inpatient Database" [15]). The lowest rate in all of these studies was obtained through passive surveillance using ICD codes for hospital discharges [8]. As we saw in our study on shaken baby syndrome and more generally for all the infanticides [16, 17], results from crosschecking judicial data with those from vital statistics clearly show that these routine data are not satisfactory as regards the accuracy of diagnoses. All of these recent epidemiological studies cited above use purely medical sources, but of very different kinds, a fact that of course influences the rates recorded: clinical data from hospital departments, specific surveillance systems and routine collection of nonspecific data.

All research carried out on shaken baby syndrome shows the large predominance of males and this fact merits further in-depth study. The role of crying has been shown as the

principle factor triggering shaking [18] and it would be appropriate to raise several questions. Is it less acceptable for a boy to cry? Do boys cry more than girls? Is their crying more annoying and difficult to bear?

The frequency of histories of prematurity is also noted in most studies. In our study, 22% of infants dying from shaken baby syndrome were born prematurely whereas the 1998 national perinatal study reported a rate of 6.8% (a highly significant difference) [19].

Repetition of shaking is also noted in several studies, one of which was carried out in France using judicial sources [20]. In fact, shaken baby syndrome rarely has the characteristics of a unique and accidental event that it has long been said to have. It is a form of abuse and often occurs in the context of several kinds of maltreatment [21, 22]. As we noted in our study, 54% of children who died from shaking had former subdural hematomas and/or long-bone fractures, visceral and skin lesions. This percentage reaches 81% in a recent Dutch study [22].

The role of family demographic and socioeconomical factors is unevenly assessed in the literature. In a review published in 2004, Guttierez et al. [23] cite several risk factors for shaken baby syndrome: poverty, unemployment, low education, single marital status and lack of social support. In the Scottish study published by Minns [12] in 2008, it is noted that "the incidence rates from this prospective study for NAHI (nonaccidential head injury) are considerably higher than other published UK surveys and are not considered to reflect a cluster effect. The perpetrators in this study fit a strongly skewed profile aggregating to the lowest socioeconomic groups in the community." And the American study carried out by the Kids Inpatient Database [13] concludes by affirming that "socioeconomically disadvantaged families with children under 1 year are an important focus for primary prevention." On the other hand, in a retrospective study using medical files on 101 children ages 0 to 4 years with shaken baby syndrome, Sinal [24] noted that "most victims (76%) lived with their mothers and biologic father or mother's boyfriend" – something we have also found – and that there was no difference between Caucasian and non-Caucasian populations in the rate of shaken baby syndrome nor in the rate of fatalities in such cases. In the United States, the analysis of socioeconomical factors has indeed crystallized around ethnicity in the case of shaken baby syndrome, as is the case with child abuse in general.

Overall, we can say a presupposition exists as concerns the association between child abuse (including shaken baby syndrome) and disadvantaged social classes. Yet, in an analysis of racial differences in the evaluation of pediatric fractures for physical abuse, Lane et al. [25] showed that "… minority children…were significantly more likely to have a skeletal survey performed compared with their white counterparts, even after controlling for insurance status, independent expert determination of likelihood of abuse, and appropriateness of performing a skeletal survey…. This group of children was also more likely to be reported to CPS compared with white toddlers, even after controlling for insurance status and likelihood of abuse… Minority children at least 12 months old with accidental injuries were more than 3 times more likely than their white counterparts to be reported for suspected abuse…"

Not only do social services have easier access to the poorest families, but the latter are the object of more marked suspicions than those from the upper social classes. Nonfatal cases of shaken baby syndrome are doubtless more easily concealed and less often the object of suspicion in the upper social classes. This demonstrates the importance of analyzing fatal cases from judicial sources, since all social classes are probably represented in this kind of sample. This was the choice we made, as well as that of carrying out a study in a geographically defined population. This allowed us to show that the distribution according to professional category of mothers who shook their children was identical to that of the regional population to which they belonged, as recorded at the time of the general population census. In their review of the literature on risk factors for shaken baby syndrome, Guttierez et al. [23] cite psychological factors such as unrealistic child-rearing expectations, rigid attitudes and impulsivity, all characteristics that can be found among management-level white-collar workers for whom the presence of a baby who cries night after night may be experienced as a career impediment.

Conclusion

Out study suggests 1) that epidemiological studies on shaken baby syndrome should involve several information sources (medical, judicial and psychological) and 2) that primary prevention strategies (in maternity wards especially) should target all social classes.

Conflicts of interest None

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