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Shaken baby syndrome in Switzerland: results of a prospective follow-up study, 2002–2007

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Abstract Since the incidence of shaken baby syndrome in Switzerland was not known, we conducted a nationwide prospective follow-up study for a 5-year period (from 2002 to 2007). The data were collected through the Swiss Pediatric Surveillance Unit. Inclusion criteria were the presence, in a child ≤6 years of age, of 1) more than or equal to two clinical symptoms (altered consciousness, convulsions, respiratory irregularities, and bulging fontanel), 2) one eye finding (retinal hemorrhages, vitreous hemorrhages), 3) one MRI/CT finding (subdural hematoma, subarachnoid hematoma, and parenchymatous lesions), or 4) history of shaking. Exclusion criteria were age >6 years or documented accident/disease explaining symptoms/findings. To describe outcome, we used the King's Outcome Scale for Childhood Head Injury (KOSCHI). 56 cases were reported from 13 of 26 Swiss cantons, representing 80% of the Swiss population; 49 cases met the inclusion criteria. Preponderance of male infants was high (31 male and 18 female); median age at admission was 4 months (1–58). Clinical symptoms were present in 42 infants, retinal/vitreous hemorrhages in 39 infants, and pathological brain/head imaging in 46 infants. In 13 cases, the caregivers admitted shaking the child. Outcomes (KOSCHI 1–5; n=47 patients) were death (KOSCHI 1) 8 (17.7%), vegetative state (KOSCHI 2) 0, severe disability (KOSCHI 3) 11 (22.2%), moderate disability (KOSCHI 4) 14 (31.1%), and good recovery (KOSCHI 5) 14 (28.8%). Based on these data, the incidence of shaken baby syndrome in Switzerland is 14 per 100 000 live births, which corresponds to the incidence in other Western countries. Demographic characteristics and outcomes of Swiss patients were comparable to published studies.

Keywords Shaken baby syndrome · Non-accidental head injury · Traumatic brain hemorrhage · Incidence · Outcome

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Introduction

In the late 1960s, Ommaya and colleagues [25, 26] reported that high-speed rotational displacements of the head on the neck at the sagittal level alone, without direct head impact, could produce cerebral concussion and hemorrhages over the surface of the brain of subhuman primates. Attachments between the outer surface of the brain and the inner surface of the skull are subjected to powerful tensile and shearing stresses. The cortical veins, particularly where they enter the more fixed portions of the dural sinuses, can tear under such conditions and result in subdural hematoma.

In 1971, Guthkelch [8], a British neurosurgeon, reported details on two infants with subdural hematoma and no external marks of injury on the head. Extrapolating from the previous animal work of Ommaya et al. [25, 27], Guthkelch suggested that in some cases, repeated acceleration/deceleration rather than direct violence was the cause of the hemorrhage. Guthkelch was therefore the first to suggest that babies were injured by shaking.

Shaken baby syndrome (SBS) is a common and preventable cause of infant mortality and severe permanent disability [24]. The term SBS, or non-accidental head injury (NAHI), is used for a constellation of clinical symptoms and findings such as retinal hemorrhages and subdural and/or subarachnoid hematomas. Patients present with a variety of symptoms and injuries that may include poor feeding, vomiting, neurological

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findings such as irritability, lethargy, and convulsions, posterior rib fractures, and metaphyseal fractures. Other "red flags" that may indicate SBS include parents' accounts of the incident that differ over time or differ between the parents/caregivers [7]. When no history of head trauma or only minor trauma is reported to treating physicians, the positive predicative value for abuse is 0.92 [11].

In the USA, the incidence of NAHI in children 2 years old or younger is estimated to be 17/100,000 person years, compared with 15.3/100,000 person years for accidental traumatic brain injury [16]. The incidence is higher in male children and in children aged 12 months and younger.

In Switzerland, there have been no previous studies on SBS to date. The aim of the present study was to evaluate the incidence of SBS in Switzerland and to investigate the main clinical symptoms present at admission and during the clinical course and to ascertain the outcome of the victims.

Methods

Children's hospitals in Switzerland were invited to report all cases of confirmed or suspected SBS to the Swiss Pediatric Surveillance Unit (SPSU). The SPSU is an established system for monitoring rare pediatric conditions. All Swiss children's hospitals (n=36) receive a monthly card asking them to report cases of the conditions being surveyed. The hospitals' replies go to the SPSU, which notifies the relevant investigator.

The study period for SBS was from 1 July 2002 to 30 June 2007. The study was approved by the Ethical Committee of the University Children's Hospital of Zurich.

A first questionnaire was sent to the reporting physician immediately after the reporting. This questionnaire requested details pertaining to history, clinical presentation, diagnosis, and management. Details about perpetrators and crying habits were not asked. A second questionnaire was sent to the attending physician 9 months after hospital admission. Clinical features, radiological findings, legal proceedings, and subsequent outcome were recorded.

The data were handled strictly anonymously. Each child was identified by month and year of birth and age at admission. We had no communication with the reporting physician, and no investigations were requested. Live birth rate was obtained from the Swiss Federal Statistical Office.

The inclusion criteria were age <6 years, two or more of the clinical symptoms, and/or one eye finding, and/or one MRI/CT finding, or shaking history (Table 1). The exclusion criteria were age >6 years or documented accident or disease explaining the symptoms/findings.

To characterize global neurobehavioral outcome we used the King's Outcome Scale for Children Head Injury (KOSCHI), which is the specific pediatric adaptation of the original adult Glasgow Outcome Scale (GOS) [5]. The KOSCHI "provides a practical scale for pediatric head injury that will enable clinicians to describe rate and extent of recovery [5]". In this five-category scale, good outcome (KOSCHI 5) indicates to a return to age-appropriate or pre-injury level of functioning; moderate disability (KOSCHI 4) is assigned if the child 1) has a significant reduction in cognitive functioning, 2) has motor deficiencies, or 3) was referred to outpatient rehabilitation therapy. Severe disability (KOSCHI 3) is assigned if 1) cognitive scores are in the deficient range, 2) severe motor deficits are present, or 3) the child was referred to inpatient rehabilitation. The last two categories are KOSCHI 2, defined as vegetative state, and KOSCHI 1, death.

To make sure not to miss victims of SBS that died and were referred directly to institutes of pathology and forensic medicine, we asked these institutes to report these children to our study.

Table 1 Inclusion criteria

Age	<6years
Two or more clinical symptoms	Bulging fontanel
	Convulsions
	Respiration irregularities
	Altered consciousness
And/or one eye finding	Retinal hemorrhages
	Vitreous hemorrhages
And/or MRI/CT findings	Subdural hematoma
	Subarachnoidal hematoma
	Parenchymatous lesions
Or shaking history	Person admitted shaking child

Results

Of 56 reported cases, 49 cases of SBS met the inclusion criteria. These 49 cases came from 13 Swiss cantons, which is half the total number of the cantons in Switzerland. The total number of live births in Switzerland during the study period was 436,970; the 13 cantons from which there were no cases reported represent only 20% of this total number (Fig. 1).

Based on the total of reported cases, the incidence of SBS in Switzerland is 14/100,000 live births (0.14‰).

In Switzerland 21.1% of the population do not have Swiss citizenship (Swiss Federal Statistical Office). Of the 49 children with SBS here, 32 (65%) were Swiss citizens. Five (10%) were from Portugal, and four (8%) were from countries in the Balkan region. Three (6%) were immigrants from other EU countries, two (4%) were from Sri Lanka and India, one (2%) child was from Iraq, one (2%) was from Romania, and one (2%) was from Ethiopia.

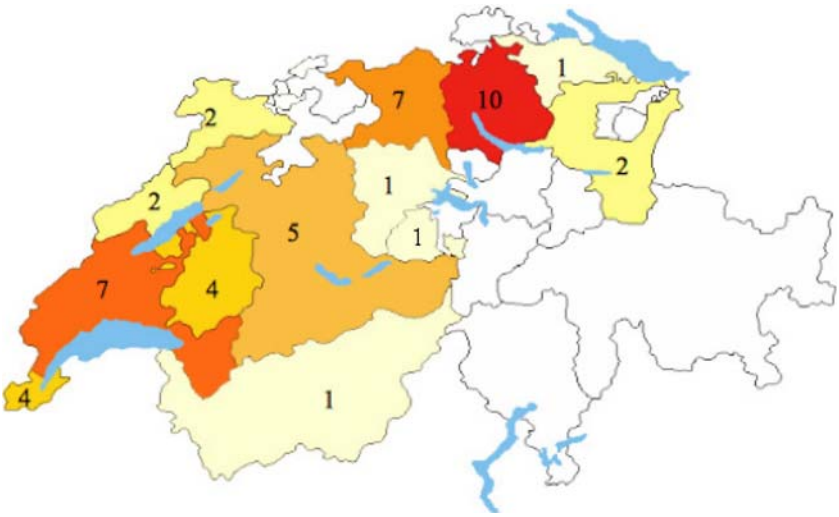

Fig. 1 Map of Switzerland with the 26 cantons. The numbers indicate the number of SBS cases declared during the study period (n=49)

Boys were more frequently affected than girls (31 boys and 18 girls, resp. 62% and 38%). Median age at admission was 4 months. Boys were younger (4 months; range, 1–15 months) than girls (5 months; range, 1–58 months) (Fig. 2).

One or more clinical symptoms were found in 42 of 49 (86%) children with SBS (Table 2).

The Glasgow Coma Scale (GCS) [9] score on admission was documented and available for 38 of 49 (82%) children (26 boys and 14 girls) (Table 3).

Of 49 (98%) children, 48 had an ophthalmologic examination: 46 of 49 (94%) examinations were conducted by an ophthalmologist, one by a pediatrician and one by a neuropediatrician. In 39 of 49 (80%) patients, hemorrhages were detected: 28 retinal and eight vitreous hemorrhages; 12 were bilateral. Nine children had both retinal and vitreous hemorrhages; two of them were bilateral (Table 4).

In 46 (94%) of 49 children, MRI and/or CT showed brain/head injuries. Subdural hematomas were present in 41 of 46 (89%) cases, subarachnoidal hematomas in 11 of 46 (24%) cases. In 15 of 46 (37%) cases, subdural hematomas were associated with parenchymatous lesions. Of the 41 cases with subdural hematomas, 30 (73%) were bilateral and 11 (27%) unilateral. In one child (2%), there were parenchymatous lesions without hemorrhages. Five of 46 children (11%) had hygromas, and 5 of 46 children (11%) had brain edema. Intracerebral hemorrhages were present in 2 of 46 children (4%) and intraventricular hemorrhages in one of 46 (2%) (Table 4).

No additional symptoms such as bone fractures or bruises were found in 45% (22) of the children. Of the remaining 55% (27 children), 17 had bruises (mostly cranial and facial), and 16 had one or more fractures (Table 5).

Injury mechanism and perpetrator were known in only 18 of 49 (37%) cases. In 5 (28%) of these 18 cases, a fall was given as the reason for the injury. In 13 of the 18 (72%) cases, the person admitted shaking the child: Seven of these 13 persons (54%) did not give any reason for doing so, 5 of 13 (38%) stated that they shook the child in order to resuscitate it, and one of 13 (8%) stated that he/she shook the child in order to calm it. These confessions and statements were made during admission to the hospital and medical history taking and not during a legal investigation.

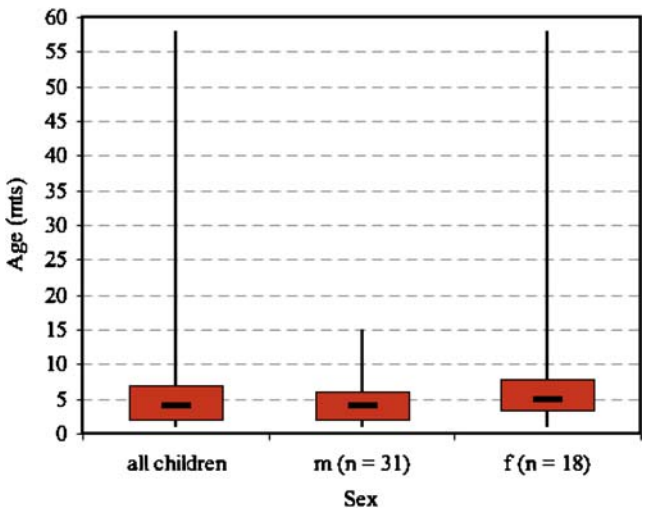

Fig. 2 Sex and age (n=49)

Table 2 Clinical symptoms (n=49)

	Boys (n=31) n	Girls (n=18) n
Altered consciousness	22	14
Respiratory irregularities	19	8
Convulsions	17	9
Bulging fontanel	12	5
No symptoms	5	2

Table 3 Glasgow Coma Scale at admission and sex (n=40)

	GCS ≤3 *n*	GCS 4–7 *n*	GSC 8–15 *n*	Median value
Boys	5	5	16	10
Girls	3	4	7	7.5

In total, 8 of 49 children died within days after the shaking (mortality, 16%): four girls and four boys. No children who died were referred directly to institutes of pathology and forensic medicine.

Detailed follow-up was available for 39 of the surviving 41 babies; the mean follow-up period was 13 months (range 3–59 months). Of the 39 survivors, 25 (64%) were disabled: 11 severely (KOSCHI 3) and 14 moderately (KOSCHI 4). 14 of 39 (36%) children had a good outcome (KOSCHI 5) (Table 6).

The symptoms of the disabled children assigned KOSCHI scores 3, 4, and 5 are listed in Table 7.

In 38 of 49 cases, we had information on the GCS score at hospital admittance. Comparison of initial GCS scores with KOSCHI scores shows that lower GCS scores result in lower KOSCHI scores (Table 8).

Discussion

According to Kochanek et al. [19] non-accidental head injury (NAHI) is the leading cause of death from traumatic brain/head injury in children younger than 2 years of age.

Table 5 Additional symptoms (n=49)

	n
No additional symptoms	22
Additional symptoms	27
Bruises	17
Cranial/facial/neck	15
Extremity	6
Chest/abdomen	6
Fractures	16
Cranium	8
Rib	5
Tibia	5
Radius	3
Femur	2

Jayawant et al. [13] conducted a three-year retrospective study of all children under the age of 2 years with subdural hematoma in South Wales and south west England and found a subdural hematoma rate of 12.8/100,000 per year in children under 2 years of age; the incidence of subdural hematoma in children under 1 year of age was 21/100,000 per year. The researchers estimated that NAHI accounted for 82% of these cases.

Barlow and Minns [1] conducted an 18-month prospective study of NAHI in Scotland; they surveyed pediatric wards, pediatric intensive care units, neurosurgical units admitting children, the Death Register, and the Scottish Health Information System. There were no cases of NAHI older than 12 months. Shaken impact syndrome occurred with an annual incidence of 24.6/100,000 children younger than 12 months.

Table 4 Ocular and brain imaging findings

	n	Bilateral *n*	Unilateral *n*
No hemorrhages	9		
No examination	1		
Hemorrhages	39		
Retinal hemorrhages only	28	11	17
Vitreous hemorrhages only	8	1	7
Combination of retinal and vitreous hemorrhages	9	2	7
No brain/head injuries detected	3		
Brain/head injuries detected	46		
Subdural hematomas	41	30	11
Subarachnoid hematomas	11	7	4
Parenchymatous lesions with hemorrhages	17	14	3
Parenchymatous lesions without hemorrhages	1		
Hygroma	5		
Brain edema	5		
Intracerebral hemorrhages	2		
Intraventricular hemorrhages	1		

Table 6 KOSCHI[a] score (n=39)

	n (%)
KOSCHI[a] 3 (severely disabled)	11 (28)
KOSCHI[a] 4 (moderately disabled)	14 (36)
KOSCHI[a] 5 (good outcome)	14 (36)

[a] KOSCHI = King's Outcome Scale for Childhood Head Injury [5]

In Estonia, Talvik et al. [30] conducted a 6-year retrospective and prospective study surveying two tertiary centers for pediatric intensive care. The overall incidence of SBS was 28.7/100,000 infants up to 1 year of age. Looking at the prospective and retrospective groups separately, the incidence was 40.5 and 13.5/100,000 children under 1 year of age, respectively. Finally, the study by Hobbs et al. [12] found a rate of NAHI of 7.1/100,000 under age 2 and 14.2/100 000 under age 1.

According to our data, the incidence in Switzerland is 14/100,000 live births, which is similar to the incidence obtained by the Jayawant study and the Talvik study, higher than that found by Hobbs, and lower than that found in the study by Barlow and Minns.

SBS is largely restricted to children under 3 years of age, with the majority of cases occurring during the first year of life [6, 9, 21]. Our findings are consistent with previously published data on SBS [12, 15, 30] that highlighted the young age of the victims and the preponderance of boys. This suggests that SBS is a form of child abuse triggered by children's crying. In a study by Lee et al. [20] it emerges that crying is an important stimulus for SBS, especially in the first 4 months of age.

In 80% of the children in our study, ocular hemorrhages were found, 70% of which were related to the retina. This is close to numbers (63.4–91%) previously reported [12, 15, 30].

Although non-ophthalmologists have a surprisingly good success rate in detecting the presence of these hemorrhages (up to 87%), they often fail to describe the hemorrhages in further detail [23]. For this reason, ophthalmology consultation remains mandatory in order to specify extent and details of retinal hemorrhages [3, 4].

Table 7 KOSCHI[a] score and symptoms (n=39)

	KOSCHI[a] 3 n	KOSCHI[a] 4 n	KOSCHI[a] 5 n
Visual problems	7	4	2
Epilepsy	6	2	0
Pathological tonicity	7	5	0
Hemiparesis	7	3	0
Pathological psychomotor development	11	6	0
Pathological development of head circumference	4	2	0

[a] KOSCHI = King's Outcome Scale for Childhood Head Injury [5]

Table 8 GCS[a] and KOSCHI[b] (n=38)

	GCS[a] ≤ 3 n	GCS[a] 4-7 n	GCS[a] 8-15 n
KOSCHI[b] 5	0	1	9
KOSCHI[b] 4	0	3	8
KOSCHI[b] 3	2	3	4
KOSCHI[b] 2	0	0	0
KOSCHI[b] 1	6	2	0

[a] GCS = Glasgow Coma Scale, [b] KOSCHI = King's Outcome Scale for Childhood Head Injury [5]

The most frequent neuroradiological finding in our study was subdural hematoma in 89% of cases, which is similar to the rate found in other studies: 81% of 22 SBS victims [30], 93% of 75 SBS victims [22], and 86% of SBS victims [17]. Similar results were reported in a postmortem study by Kivlin et al. [18] in a series of 111 patients (92.5%).

A study on outcomes showed that children with SBS have a significant mortality rate, and children who survive have serious morbidity and special education needs [15].

In our study, we report a mortality rate of 16%, moderate to significant disability in 28–36%, and good recovery at the time of follow-up in 36% of children with SBS. Our data correspond with other published studies that show a mortality rate ranging from 15 to 38% [16], moderate to significant disability in 30–50%, and full recovery in 30% of children with SBS [10]. However, there is a lack of long-term outcome data in survivors of SBS, particularly in less severely injured children.

Although the present study highlights the devastating effects of SBS, there are several limitations that should be mentioned. Underreporting and misdiagnosis remain a major problem, since the clinical setting and the symptoms are often non-specific. Vomiting, fever, irritability, and lethargy are common symptoms of a variety of conditions seen in children, including head trauma [14]. Another limitation of our study is the unexpected lack of reporting from half of the Swiss cantons, although they represent only 20% of total Swiss population and the children under 1 year of age living in these thirteen cantons represent only 21% of all children under 1 year of age living in Switzerland. Of the 13 cantons, which did not report, eight do not have a children's hospital. It is to presume that children living in these cantons were referred to a general practitioner or an adult clinic where symptoms of SBS may not be known. Lack of reporting may also be due to poor knowledge of SBS symptoms, making specific information for pediatricians important. The short follow-up period and the small number of victims limit our detailed knowledge of

the survivors, particularly in the less severely injured children. Finally, knowledge about injury mechanism and perpetrators in our study is limited.

Prognostic factors relating to poor outcomes emerging from our data are a lower score on the Glasgow Coma Scale at presentation, a higher number of lesions on imaging studies, and respiratory problems at presentation. This agrees with findings reported in a 2002 publication by Prasad et al. [28].

SBS is a frequent and serious public health problem in Switzerland. Between 1 in 3,000 and 1 in 4,000 children younger than 1 year of age are victims of significant or fatal NAHI, making SBS almost as common as cystic fibrosis and more common than acute lymphocytic leukemia [1, 19]. The outcome of SBS is devastating to the child. More work is needed to develop effective methods of prevention, for these are potentially healthy children prior to the injury. In a paper on infant crying as a trigger to shaking, Barr et al. [2] showed that the age-specific incidence curve in SBS cases has a similar starting point and a similar shape to the normal crying curve. The duration of crying at a given moment seems to be less relevant than the parents' perception of the crying in the long term [29]. Prevention should focus especially on the effect of crying on parents and on teaching parents how to cope adequately with crying.

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