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The Pitfalls of Diagnosing Sudden Infant Death Syndrome: Case Studies

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Abstrakt The autopsy diagnostics of children generally include specific features, however no clear guidelines are available. Most discussions centre around the diagnosis of SIDS. In practice, some cases may be falsely diagnosed as SIDS whereas they actually involve intentionally violent acts. The aim of this paper is to describe the pitfalls of diagnosing the causes of the sudden and unexpected deaths of infants using three actual case reports from the Czech Republic. The common feature of these cases was the assumption that the death had been caused by SIDS or was diagnosed as such, while child abuse and neglect syndrome or other maltreatment was suspected. The research method of choice was a retrospective content analysis of all the medical and forensic autopsy reports of three infants who died within the 1st year of life. Two of these cases were judged as the result of SIDS, the third one as a violent death. Forensic-medical diagnoses affect the subsequent procedures of law enforcement authorities.

Key words: death, infant, violence, forensic medicine, autopsy, guidelines, SIDS, CAN

1. INTRODUCTION

Ever-improving medical procedures and the increasing medical success rate make the death of a child seem senseless and futile to the parents as well as to the social environment. Society views a child's death as "unnatural", "abnormal", "untimely" (Kubíčková 2001). Despite decreasing occurrences, extraordinary attention continues to be devoted to infant mortality in both medicine and medical policy (Gladkij and Koldová 2005). Infant mortality rate is an important indicator in evaluating the state of public health, i.e. the statistic is associated with the quality and availability of health care as well as the general living standard of a country (CDC, ÚZIS [Institute of Health Information and Statistics of the Czech Republic]).

Statistics enumerate the infant death rate as well as the diagnoses that led to each of these mortalities. Nevertheless, the investigation of circumstances surrounding a death is not easy. The autopsy diagnostics of children includes specific features such as the failure of vital functions, which can also occur when macroscopically detected changes have not yet developed in the child as they would have in adults (Hirt et al. 2015, p. 92). No guidelines exist for autopsy diagnostics, a situation which causes discrepancies in the conclusions of diagnostic procedures at forensic medicine workplaces (Olecká 2018). In order to investigate death circumstances, multidisciplinary teams have been created in many countries. Hymel (2006) argues that in an ideal case the multidisciplinary team for a death investigation should include a child-care social worker, a police investigator, a nurse, a forensic doctor, a paediatrician with child abuse experience, a coroner, a representative of the emergency medical services system, a child pathologist and a local prosecutor. Such circumstances would help make the data valid.

The most frequent discussions on data validity and reliability concern the diagnosis R95 SIDS, i.e. Sudden Infant Death Syndrome (Sheehan et al. 2005). This diagnosis accounts for the largest unified group of deaths within infant mortality in the age range of one month to one year in all developed countries (Houšťková and Bouška 2002, p. 2). The incidence of SIDS is approximately 0.5 per one thousand of children born alive (Zdraví 21, p. 17). The syndrome is defined as a sudden, unexpected death of a child under one year of age, with the onset of death episode clearly happening during sleep, and not otherwise explicable after a thorough investigation, including a complete autopsy, investigation of death circumstances and a clinical history of the infant (Krous et al. 2004). The origin if SIDS is multifactorial (Busuttil 2009, Ottaviani 2007, Enid Gilbert-Barness 2013), and the impact of outer risk factors, including social factors, is taken into consideration in the diagnosis. The causes of SIDS are not univocally known, however, hypoxia, vegetative nervous system malfunction, impact of infections, hereditary metabolism defects and immunological deviations have all been mentioned (Houšťková and Bouška 2002, p. 2).

External influences have also been linked to higher incidences of SIDS (Byard and Krous 2003, p. 113):

Infants of mothers with a low level of education and low socio-economic status;
Boys, especially in the age range of two to four months, mostly in winter months;
Hyperthermia, soft pillows and mattresses in inclining positions;
The child sleeping on its stomach;

The child exposed to cigarette smoke during pregnancy and infancy;
Poor prenatal care and drug abuse (mainly opiates) by mothers;
Infants with higher levels of prematurity and children of mothers with higher birth rate with short intervals between pregnancies;
SIDS is a rarity in Asian communities.

There generally exists no univocal agreement for the diagnostics of sudden deaths within individual countries, let alone across national borders. Numerous authors have documented erroneous diagnoses in cases that in reality were caused by deliberate acts of violence (Krugman 1994, Meadow 1999, Le Fanu 2005, Stanton 2001, Nunes 2001). Infanticide can sometimes be indicated by examining the death circumstances or by an external body examination, but in cases in which no signs are obviously detectable, the murder can only be exposed by a thorough post-mortem body examination including a whole range of instrumental tests (Levene and Bacon 2004). Nevertheless, these tests are quite specific and, due to the inconsistent standards of validity, the procedures are often not conducted. The decision of whether an expert assessment of the causes of the death of the infant is to be ordered is taken by the police authority or other bodies involved in criminal proceedings.

Unrecognized murders have been estimated to represent from two to ten per cent of cases (Meadow 1999), with the children in such cases having died as a result of suffocation by blocking airways. This is the so-called suffocation method of smothering (covering the nose and mouth), which is also a form of child torture and a frequent death cause in childhood (Meadow 1990). Other forms of abuse can be present as well, e.g. the so-called Münchhausen syndrome by proxy. The most frequent perpetrator is the biological mother of the child. The death cause in such cases may not be clearly proven by autopsy, as the signs of mechanical obstruction of airways may not be apparent or no defensive wounds exist.

The aim of this contribution is to show the diagnostic pitfalls associated with investigating the sudden and unexpected deaths of infants. The analysis is based on chosen case interpretations in situations when no diagnostic guidelines existed at all.

2. FILE AND METHODS

The research method was a content analysis of autopsy files from medical and forensic autopsies of children under one year of age. Each autopsy file was analysed as a case study consisting of a descriptive and an exploratory section. The descriptive part presented the results of the doctor's examination and the autopsy of a child. The exploratory section was used as a source of information further detailing the circumstances of the child's death in the context of data from the descriptive part.

The presented cases were chosen from a research file containing the autopsy files of children who died suddenly and unexpectedly in the years 2007 – 2016 (Olecká et al. 2018). The autopsies of the deceased children from which the three below were selected were undertaken at four workplaces of forensic medicine in the Czech Republic. The reports also contained witness interrogations, police records, reports by doctors, experts and psychologists, and photographs of the crime scene. The data were analysed within the IGA_LF_2018_023 project "Retrospective analysis of mortality of children under five years of age who died suddenly, unexpectedly and violently." Three case studies from three different workplaces were chosen from the target file by the method of intentional choice. The common feature of all cases was a formulated assumption of death due to SIDS, or its diagnosis, while all three cases featured a suspicion of child abuse and neglect syndrome (CAN).

The results of each case study are summarized into three categories: autopsy report, family circumstances and death circumstances.

The research was approved the Ethics Committee of University Hospital and Faculty of Medicine and Dentistry Palacky University in Olomouc and also by 3 Head of Department of Forensic Medicine and Medical Law. Researchers were taught about the need to keep personal data confidental and unrecognisable.

3. CASE STUDIES

Case one: a girl (9 months), the cause of death: SIDS.
Autopsy report: The autopsy revealed significant venosity of all body organs, both lungs with lower airing, with isolated petechia under the pleura. A small amount of foamy fluid leaked out of a lung incision. Fontanelle mildly recessed. A discrete microscopic finding corresponding to the onset of bronchopneumonia, which could be one of many stress factors. Discreet abrasions in the right temporal headspace and between the left shoulder and the neck. On the left side of the forehead, a blood bruise 3x4 cm in size. Another small haemorrhage on the upper eyelid, the brain showing signs of swelling. The injuries could have originated by various mechanisms, by the act of blunt, low intensity violence, not a factor in the onset of death. The imminent cause of death has not been decidedly determined. A forensic expert claims the death fulfils the criteria for SIDS.

Family: The girl placed in an infant home in the early days of life but mother claims her back again. The child is properly vaccinated. She is, however, repeatedly hospitalized for failure to thrive due to poor feeding. Recommendation for regular weight checks at the paediatrician, devoting attention to the child, balancing its psychomotor development.

Death circumstances: The girl's weight stagnates one month after returning to mother care. She is hospitalized at the age of two months for failing to thrive. She drinks well and shows significant appetite, does not vomit during hospitalization. The problem of weight stagnation seems to be in the lack of feeding. The mother is instructed in the necessity of prevention of hypothermia. At the age of eight months another hospitalization is mandated for failure to thrive; an older haematoma is apparent on the girl's forehead after falling from bed. Results of sample tests are standard. Neurological examination claims the child's developmental stage is between the sixth and seventh month. Cow milk negative. The situation reported to the Social Services for investigation. According to their statement the child can be released and returned to its mother. According to the Police of the Czech Republic the girl found dead in a room, body bruised, brought to autopsy in a torn t-shirt.

Case two: a boy (4 months), cause of death: SIDS.
Autopsy report: non-specific, haemorrhages on the head, venosity of body organs, petechia under the pleura. The body bears signs of numerous blunt injuries of head, torso and lower extremities. The finding indicates repeated use of blunt violence of mild intensity by a smooth solid object such as a hand. The injuries are of various dates; the boy could not have caused them himself. The forensic expert found the repeated violence could not have endangered the vital functions due to its low intensity. The autopsy report did not reveal death in connection with the hostile act of another person.

Family: the mother of the boy is a 24-year old woman who grew up in a children's home. To run her household, she earns extra money by prostitution. A suspicion of neglect exists with her two older children. The boy is placed in an infant home after birth; the mother

takes him back after two weeks to be eligible for social security payments.

Death circumstances: The mother repeatedly beats the boy by hand all over his body (the first time approximately two months before the boy's death). The father of the boy knew of the beatings but never prevented the mother from the acts of violence. The mother uses higher intensity of violence on the day of death after having argued with the boy's father. She then places the boy in a cot where the boy dies. The mother never calls for help. She packs the dead boy in a plastic bag and buries him in a forest. Afterwards she reports a child kidnap. The mother is sentenced to three years in prison for torturing the child under her guardianship.

Case three: a boy (9 months), cause of death: intensive shaking of a baby (Shaken Baby Syndrome)
Autopsy report: Epidural bleeding in the whole spinal canal, brain swelling and bleeding into meninges detected during the autopsy; no further injurious changes. The boy of a small size and significantly low weight died at home as a result of brain swelling and bleeding between spinal membranes.

Family: the mother is a 27-year old woman who does not work; she collects a disability pension due to her epilepsy. The boy is from her fourth pregnancy (first pregnancy terminated in abortion, one child is in care of the mother's sister, second child – the older sister of the boy – lives with the mother). Every child has a different father; two of the fathers supposedly died of liver cirrhosis. The father of the boy was killed in a motorcycle accident when the boy was three weeks old. The mother lives with a man who is not in an intimate relationship with her; he can live in her home in an exchange for babysitting. The man is 27-years old, a smoker, with vocational training, unemployed, not registered with the Labour Office, on probation for criminal acts of theft and inflicting bodily harm. The landlord claims the mother is often late with the rent; the landlord does not want to prolong the lease agreement. The neighbours claim the family is obviously short of money; the mother often borrows things and food she never returns. She was also seen begging on the main square. The children are apparently neglected, dirty, smelly, they wear old clothes and are hungry. The apartment also smells bad. The mother gives an impression that she is not interested in her children. The paediatrician claims the mother does not attend the compulsory preventive examinations; the Child Protection Authority notified three times at approximately one-month intervals. The mother has not taken the child to attend the required hip sonography examination on time. The boy did not gain weight. The paediatrician discussed the matters with the mother several times. The boy was hospitalized at the age of five months for strangulation of two toes on the left foot. The toes were strangulated by hair, which the mother had not noticed. The neurologist who examined the boy at the age of seven months stated the boy was pale and weepy with hypotonic syndrome. A share of social deprivation was acknowledged and general stimulation recommended. The older sister of the boy was also hospitalized, taken to the hospital under the assistance of the Police of the Czech Republic with haematoma and pulled-out hair. No further details of her hospitalization are known.

Death circumstances: two days before the boy's death the mother departs to visit relatives and leaves her two children in care of the subtenant. She does that frequently. She gives the subtenant 40 Czech Crowns [approximately €1.50] and instructions to follow according to which the boy should be given an analgesic suppository if he cries due to teething. During the first day the man supposedly takes care of the children and feeds them. On the second day the boy cries since early morning and cannot be soothed. The man tries several times to soothe the boy and put the him to sleep. When the boy spits out the pacifier the man gets angry and shakes the boy intensively in different directions six to eight times. This calms the baby and the man puts him into the cot. Forty minutes later the man finds out the boy is not breathing; with help of a neighbour he calls the ambulance service and he himself tries to resuscitate the boy. The ambulance service doctor continues the resuscitation to no avail. The man confesses to shaking the boy only after several interrogations; he had never shared this fact with the medical staff of the ambulance service.

4. DISCUSSION

All the presented cases have the following characteristics in common: the child shows signs of neglect, the mother does not show real interest in the child (placement in an infant home as a part of case history, numerous instances of babysitting by a stranger), violence of various intensity committed on the child during its life, signs of inappropriate behaviour towards the child noticed by people in its surroundings (neighbours, doctors, social workers), unsatisfactory socio-economic situation of the family. The first two cases are closed with a finding of SIDS based on the diagnostic presupposition per exclusionem – the death was unexpected from the point of view of case history, nor explained by a thorough autopsy, further laboratory testing or by the circumstances at the place of death. In the third case the spinal canal was opened beyond the standard autopsy procedures. This step demonstrated epidural bleeding in the whole spinal canal and lead to the conclusion that the child had been shaken and died of Shaken Baby Syndrome.

Hymel (2006) claims it can be difficult or impossible to differentiate between the natural unexplained death of a child, an unintentional or accidental death, and the unnatural (intentional) death, e.g. of an infant (by covered airways with any soft object, plastic bag, sleeve, hand or the mother's breast). Cases in which the complex investigation of death circumstances has not taken place which show significant and adequate uncertainty about the causes or manner of death should be labelled as "unclear". Cases which Hymel (2006) classifies as uncertain contain a suspicion of infant death caused by an infection, metabolic disease, suffocation or child abuse.

Unless a criminal act is revealed and the offender is punished, another child born into the same family faces the same risk (DiMaio 1989). On the other hand, a situation in which innocent parents are accused is most certainly unwanted. The best solution is to execute a thorough, professional, disinterested and impartial investigation. The number of wrongly diagnosed cases decreases with the rising number of sufficiently investigated cases (Byard and Krous 2003). A thorough investigation of the causes of a child's death due to ill-treatment requires a careful and objective assessment by several specialists. Only the professional cooperation of doctors, police officers, lawyers and judges ensures justice and respect for the rights of children and entire families. To this end, the authorities in charge must provide appropriate support and resources for educating the specialists, who in turn must be equipped with tools and powers they need for proper diagnostics and case investigation (Jenny 2006).

No unequivocal agreement on autopsy procedures and diagnostics of sudden deaths exists within individual countries, let alone across the borders of nations (Sheeehan et al. 2005). Krous et al. (2004) divide SIDS into four categories. Category IA SIDS comprises cases when all the prototypical features of SIDS are present and all information has been thoroughly documented. Category IB SIDS comprises cases when all the prototypical features of SIDS are present, but the case has not been thoroughly documented (no investigation of the various scenes when death could have occurred has taken place,

and/or one of the following analyses has been not performed: toxicological, microbiologic, radiologic, chemical analysis of intraocular fluid or metabolic screening studies). Category II SIDS comprises the deaths of children which can be classified as category I with the following exceptions: age out of the IA or IB category (i.e. 0–21 days or 270 days/9 months from birth); the similar death of a sibling, close relative or a child in the care of the same caretaker; possible genetic disorders, neonatal or perinatal states (e.g. those relatable to premature birth); mechanical asphyxia or suffocation caused by covering nose and mouth are not excluded with certainty, abnormalities in growth or development which are not causally related to death are present; rather obvious inflammatory changes or other abnormalities are present that were clearly not the causes of death. The last category is Unclassified Sudden Infant Deaths (USID). These comprise deaths that do not fulfil the criteria for category I or II SIDS, but for which alternative diagnoses are not definite, including cases when an autopsy was not performed.

With regard to this classification the doctor has two options when performing the autopsy: to put forth the R95 diagnosis – SIDS (when strict diagnostic requirements are fulfilled), or the R99 diagnosis – other ill-defined and unspecified causes of mortality (when there is the lack of information that the definition requires). The presented cases demonstrate that a doctor's decision on a diagnosis has an impact on the follow-up steps of the authorities involved in criminal proceedings and thus the punishment of a potential offender. The R99 diagnosis does not preclude the fault of an unknown person, it presents a situation in which the possibility for further police investigation remains open. On the contrary, the R95 diagnosis relieves unknown persons from the responsibility for the death of an infant, and a caretaker of the child can be prosecuted at most for the abuse of a person in his / her care.

5. CONCLUSION

The expert community is currently working together to develop guidelines for data collection regarding the causes and circumstances involved in investigating the death of a child (Weber and Sebire 2011, Shapiro-Mendoza et al. 2010 and 2014, Devitt 2012, etc.). Since 1996, the US Centers for Disease Control and Prevention (CDC) have been developing a standardized form, protocol and guidelines to be used in these situations (Sudden, Unexplained Infant Death Investigation Guidelines). The general acceptance of these guidelines and their application in practice would undoubtedly lower the number of wrongly diagnosed deaths, which could as a result, lead to lower secondary criminality.

A recommended procedure for paediatricians in the case of a sudden death of an infant has recently been developed in the Czech Republic, along with a standardized questionnaire to be completed (Houšťková and Bouška 2002). Nevertheless, an analysis of autopsy files (Olecká 2018) has shown that this questionnaire is generally not used when the death circumstances of a child are being investigated. Standardized diagnostic guidelines for diagnostics regarding the sudden and unexpected deaths of children need to be developed. The development and updating of questionnaires on family anamnesis as well as on life and death circumstances of children are necessary. Greater attention needs to be devoted to risk factors on the mother's side (Ivanová et al. 2017). The primary emphasis must be placed on the cooperation and communication among all constituents dealing with the protection of the lives and rights of all children, a goal which can be facilitated even in investigations of the death of a child.

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