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ORIGINAL ARTICLE

Sudden infant death syndrome in the Federal District, Brazil: is there evidence?

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Abstract

Objectives: The syndrome of sudden infant death (SIDS) affects children aged between one month and one year of age. Although it has long been reported, there is still no known cause. Although it has long been reported, there is still no known cause. The present work aims to raise the profile and prevalence of SIDS in the Federal District, Brazil. **Methods:** Descriptive, transversal and retrospective study relating to deaths, beyond autopsies and the demographics of children between 28 days and one year of age was performed at the Institute of Forensic Medicine of the Federal District between 2003 and 2009. **Results:** The number of infant deaths registered during the study period was 431. Of these, seven (1.62% of total) died while sleeping and four met the conceptual criteria. The mean age was 102.28 (±11.28) days. However, none was diagnosed as SIDS and theirs causes were analyzed. **Conclusion:** There was no report of SIDS in the Federal District between 2003-2009, contrary it was reported in South/South-Eastern of Brazil whose incidence varied between 0.13 and 1/1,000 live births. This finding suggests underdiagnosis and indicates the lack of a SIDS specific protocol for post-mortem identification. This study shows the need to implement an investigative protocol for SIDS, knowledge, and dissemination of the syndrome among health professionals and the community and shows the importance of awareness campaigns and prevention of this phenomenon.

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INTRODUCTION

Since 1969 three internationally accepted definitions of sudden infant death syndrome (SIDS) have been used[1-3]. The latest definition for SIDS established in San Diego, California, by a work-group in 2004 states that it is "the sudden and unexpected death of an infant less than 1 year of age, with onset of the lethal episode apparently occurring during sleep that remains unexplained after a thorough investigation, including performance of a complete autopsy, and review of the circumstances of death and the clinical history"[3]. Additionally, its subclassification divides this condition into categories I and II, according to the presence of classical features of SIDS and complete documentation of the case. There is another condition established as "unclassified sudden infant death (USID) category", which includes deaths that do not meet the criteria for categories I and II for SIDS, but for which alternative diagnoses of natural or unnatural conditions are equivocal, including cases where autopsies were not performed[3].

SIDS affects children under one year old and 90% of cases occur up to six months of life. The highest mortality frequency takes place between the second and fourth months of life, being more frequent in males (1.4:1) and occurring predominantly in the winter[4,5]. The incidence varies significantly between countries, according to the methodology used for its diagnosis. The use of different definitions of SIDS makes it difficult to compare studies on the subject[6]. In Ireland, which has an organized record containing all cases of unexplained deaths, the incidence of SIDS in the period 2006-2008 was 0.34/1,000 live births[7]. Unfortunately, in many less highly developed countries, the incidence is not yet known. While the Buenos Aires Metropolitan Area in Argentina presented a 1995 SIDS mortality rate of 1.4/1,000 live births[8], in Brazil the incidence is not clear because there are no official statistics and the few studies on the topic are restricted to small regions and the protocols for SIDS diagnosis are not always followed or obeyed[9,10]. A study involving 72 cases of SIDS based on information from parents and medical records, found an incidence of 1/1,000 live births whilst no case was diagnosed as SIDS on death certificates[5]. Another study based in autopsies of babies less than one year of age at Ribeirão Preto, South-Eastern Brazil, reported an incidence of 0.13/1,000 live births[10]. A case-control study among the population of Porto Alegre, Southern Brazil, found an incidence of 0.55/1000 live births[11].

It is noteworthy that Brazilian reports show a discrepancy with the worldwide incidence of SIDS. The possible explanations for under-diagnosis are that at autopsy the medical examiner generally does not have information to hand about the association of risk factors for SIDS, such as: death in previously healthy infants, autopsy that did not justify the cause of death and negative death scene analysis. A review of the place of death is also rarely performed, making the final diagnosis unclear. In addition, the pathologist is often not an expert in child pathology or aware of research into SIDS[9]. Two studies showed no case of SIDS in official mortality record in southern Brazil until 2000 and only 2 cases between 2001 and 2003, indicating that SIDS probably had been misdiagnosed[11,12]. In these cases, the autopsy reports do not justify the immediate cause of death and usually these indicate infections or broncho-aspiration, although these are not enough to explain death, especially as aspiration may have occurred soon after death[9,10].

In the Federal District, Brazil, all sudden unexpected and unexplained deaths of children are examined by the Institute of Forensic Medicine, and these reports are not shared with an official notification system for SIDS. Hence, there are no official statistics about SIDS incidence in this area of the country. The objectives of this study were to verify if any SIDS definitions were used by coroners in their autopsy reports, the incidence and prevalence of SIDS among children who died in their sleep, and if there is evidence for SIDS as cause of death among them.

METHODS

This descriptive, cross-sectional and retrospective study was based upon the record system of the official data bank of the Institute of Forensic Medicine of the Federal District, Brazil. The research used the San Diego SIDS definition in its categories IA, IB, II and USID[3]. The autopsy reports of all deaths of children less than one year of age found to have died while asleep were analyzed for the period between January 2003 and August 2009. None of the parents or caregivers were interviewed nor was the child's clinical history reviewed. Sociodemographic variables were recovered, covering gender, age in days, ethnic background, season, if the infant was sleeping in the parents' bed, the position in the bed, autopsy reports and contents of the death certificates. In the case of misdiagnosis, broncho-aspiration, petechiae on the lung surface and cerebral edema were considered as possibilities for suspected cases of SIDS diagnosis[10]. The exclusion criteria were congenital malformations, infectious diseases, and external causes.

Sample

From the first analysis of these six and a half years of death reports, 431 deaths were registered for ages from zero to 365 days, and of these seven (1.62%) died in their sleep (four males and three females). The features of these seven deaths were analyzed and the results are described below. This study was approved by the Health Sciences Ethics Committee of the University of Brasilia, Brazil ID-017/2010.

RESULTS

The data analysis was run considering the investigation of seven suspected cases of SIDS with the incidence found to be of 0.027/1,000 live births and prevalence of 1.62%. The final

sample consists of four males (57%) and three females (43%), all previously healthy; six of them were white-skinned. The mean age was 102.28 (±11.28) days. As regards the season, two died in the winter, three in the summer and two in the fall. Co-sleeping was found in two cases and one was unknown. Only one infant was sleeping in prone position. The remaining cases were also unknown. The description of the autopsy findings and death certificates showed that no case of SIDS was reported by coroners in the studied period (Table 1). After data analysis, all deaths were considered in the USID category of San Diego's SIDS definition.

DISCUSSION

This study provides a description of the incidence, prevalence, epidemiological aspects, and autopsy findings of seven infants found dead in bed during a sleep period. The prevalence of SIDS was 1.62% and the incidence was of 0.027/1,000 live births considering all deaths in infants of less than one year of age examined in the Institute Forensic Medicine of the Federal District, Brazil. These rates are lower than in developed countries and some developing areas of Brazil, probably due to lack of protocols and to misconceptions about SIDS on the part of pediatricians, pathologists and coroners[5,7,11]. The mean age of 102±11.3 days of the affected infants was similar to that found in other studies about SIDS, which occurs mostly between the second and fourth months[7,12].

Regarding the most affected gender, the data are consistent with international reports, since they indicate a higher frequency of males (four cases, 57%) compared to females (three cases, 43%) in a ratio of 1.32 male deaths to one female, also similar to that found in the city of Porto Alegre, Brazil[11,12]. The predominance of males has been found in most published papers, leading some authors to suggest a sex-linked genetic hypothesis as the etiology of SIDS[6,9,13].

This study indicated a predominance of white (75%) individuals in relation to brown (25%). However, this analysis is geographically peculiar, due to the large mixture of origins and races among Brazilians, making accurate analysis of racial distinctions prone to the influence of the evaluator's subjective criteria. Data on seasonality indicate that there were two cases in the summer (40%), one in fall (25%), and one in winter (25%). In the spring, none was found. These values agree with some studies in Brazilian literature and diverge greatly from the study run in Pelotas, Southern Brazil and from international literature relating to developed countries, since both show a higher incidence in winter[4,5,10,11]. The higher prevalence of SIDS in summer in some Brazilian studies may result from an increase in body temperature, which leads to greater loss of water, changing the electrolyte concentration and reducing secretion of mucus in the respiratory tract[4,11]. A greater likelihood of exposure to viral and other infectious diseases in developing countries must also be taken into account[11]. The fact that the data from Pelotas, Southern Brazil are consistent with the literature of developed countries may be related to this region's more temperate climate, which is closer to that of many developed regions. These climatic and economic characteristics as previously cited did not occur in our study, whose prevalence of SIDS was 1.6%, lower than those found in Pelotas, of 4%, but higher than those found in Japan and the Netherlands, which are respectively 0.9% and 1%[12].

The death certificates reported by the coroners and under investigation in this study were mainly based on descriptions of respiratory failure (25%) and accidental aspiration of gastric contents (75%). The identification of respiratory failure secondary to accidental aspiration of gastric contents, as a cause of death in three of seven cases with strong suspicion of SIDS, shows a lack of knowledge by medical examiners in the performance of the autopsy, because this aspiration may have occurred after death and does not necessarily represent the cause of death[6]. This aspiration can be defined as a *post-mortem* event due to generalized muscle relaxation, which would allow passage of fluid into the upper airways, and therefore the secretion is seen only in the lumen of the trachea and main bronchi. On the other hand, when the cause of death is aspiration itself, the discharge is found in the lower airways such as bronchioles and alveoli[6].

These results are evidence of a lack of research protocol for *post-mortem* cases of suspected SIDS. Autopsy findings, such as lungs with petechiae in sub-pleural areas, white non-friable parenchyma and heart with petechiae were of a nonspecific nature and consistent with SIDS. These features are discussed by the literature and are often linked to asphyxia[14], although there is broad agreement today that what are now known as "Tardieu spots" are the result of intense lividity, leading to *post-mortem* rupture of dependently engorged blood vessels, entirely unrelated to asphyxia or any other mechanism of death. The occasional reference still made to them in the literature as *ante-mortem* petechiae or "asphyxial signs" betrays a misconception of their current meaning[15].

USID, an unclassified category of SIDS, includes deaths that do not meet the criteria for category I or II of SIDS, but for which alternative diagnoses of natural or unnatural conditions are equivocal, including cases for which autopsies were not performed[3]. Given the analysis of autopsies, it reaches the conclusion that the methodology used by the coroners differs from the protocols adopted in developed countries because bacteriological, virological, biochemical, cytogenetic, and radiological analyses were not performed. At no time was analysis of the brain stem, hippocampus, and cerebellum reported, structures closely related to SIDS. Hence, we prefer to maintain the hypothesis that the seven cases under analysis in this study might be better classified as USID than a SIDS, acknowledging the many uncontrolled variables in the data.

Table 1. Suspected cases of SIDS and related demographic and autopsy relevant information.

Patient	Gender	Age in days	Ethnic background	Season	Sleeping in the parents bed (Yes?)	Position in the bed	Death certificate	Review of necropsy
1	Female	25	White	Summer	No	Unknown	Asphyxia due to aspiration of gastric contents	• Presence of lacteal secretion in the nostrils, bronchi and lungs, without findings in trachea. • Remaining organs and structures of normal appearance.
2	Female	36	White	Fall	Yes	Unknown	Asphyxia due to aspiration of gastric contents	• Presence of yellowish pasty material in lower respiratory tract and stomach. • Tardieu petechiae on the lung surface. • Remaining organs and structures of normal appearance.
3	Female	235	Brown	Summer	No	Unknown	Acute respiratory failure	• Trachea and bronchia without aspiration contents. Lungs with whitish areas located in the upper lobes associated with diffuse Tardieu petechiae. Remaining organs and structures of normal appearance.
4	Male	48	White	Fall	No	Unknown	Respiratory failure secondary to aspiration of gastric contents	• Presence of lacteal secretion in the interior of the trachea and stomach. • Lungs hyper inflated with Tardieu petechiae on the lung surface. • Remaining organs and structures of normal appearance.
5	Male	68	White	Winter	Yes	Prone	Respiratory failure secondary to aspiration of gastric contents	• Presence of nasal discharge is thick. • Foamy secretion in the trachea without gastrointestinal tract components. • Lungs with whitish areas and sub-pleural petechiae.
6	Male	93	White	Summer	No	Unknown	Respiratory failure secondary to aspiration of gastric contents	• Petechiae on the basis of right and left ventricles; • Stomach with lacteal secretion. • Remaining organs and structures of normal appearance. • Presence of lacteal secretion in the trachea and stomach. • Lungs distended with Tardieu petechiae on the lung surface. • Remaining organs and structures of normal appearance.
7	Male	211	White	Winter	Unknown	Unknown	Unexplained death	• Excoriations on the lateral side of the right arm and left masseter region of the face. • Bruise on the left side of the face. • Lungs, liver, and spleen with diffuse petechiae. • Extensive subgaleal hematoma biparietal and occipital fracture on the inner surface of the occipital region the skull. • Separation of the lambdoid suture. • Subdural hemorrhage in the occipital region and cerebellum. • Normal bronchi and trachea. • Long bones intact. • Remaining organs and structures of normal appearance.

CONCLUSION

This study could, therefore, only discuss the data in terms of USID, as the death reports studied did not follow the SIDS protocol properly. The first step in preventing SIDS and USID is to diagnose existing cases to understand the context of its occurrence and make the professional class aware of its true incidence and clinical diagnosis, which seems to be underestimated in the Federal District of Brazil. The data found in the study corroborate those found in other Brazilian regions and developing countries. The profile of SIDS seen in a number of studies suggests that on a national level, there is a lack of clear policies to prepare pediatricians and medical examiners for the importance of the disease and then to develop prevention and awareness campaigns in relation to SIDS.

Worse than to feel the shadow of death over early lives, is the knowledge that despite years of national and international research, the country still hesitates to install a proper protocol for investigation and prevention of SIDS. Indeed, little is done to counter the trend to attribute false causes to these early deaths, e.g., accusations of infanticide. This study also prompts us to encourage research into causes and prevention strategies in different Brazilian states, as each can have its own regional risk factors.

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