

Simultaneous Sudden Infant Death Syndrome

A Case Report

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The first reported case of simultaneous sudden infant death syndrome (SSIDS) in Allegheny County, Pennsylvania, occurred on February 27, 1998. Two-month-old black fraternal twin girls were both found dead in their crib at the same time. After an in-depth death scene investigation, police investigation, toxicologic analysis, and complete autopsies, a specific cause of death could not be identified. The deaths of the two girls were therefore ruled simultaneous sudden infant death syndrome.

Key Words: Sudden infant death syndrome—Simultaneous sudden infant death syndrome.

Sudden infant death syndrome (SIDS) is the leading cause of death in the United States of infants during the postperinatal period (7–365 days), having caused 2,906 deaths in 1996 (1). In 1989, the National Institute of Child Health and Human Development defined infant deaths to be classified as SIDS if (1) the death occurs in an infant ≤ 1 year of age, (2) a thorough postmortem examination fails to demonstrate an adequate cause of death, (3) a death scene investigation is conducted and yields no evidence of unnatural cause of death, and (4) a review of the infant's and mother's medical records reveals no history of medical conditions that might have caused the death (2).

Hundreds of epidemiologic studies (3–6) have documented the death of a single infant, whereas twin SIDS deaths have received considerably less attention. Several studies have examined SIDS deaths wherein the twins' deaths were separated by >24 hours (3,7–12). However, the topic of simultaneous twins' deaths, known as simultaneous sudden infant death syndrome (SSIDS) (10), has been directly addressed in only a few articles (11,13,14).

The first attempt to locate, summarize, and evaluate all cited cases of SSIDS, in terms of a new proposed definition of SSIDS, was carried out by Koehler et al. in 1998. A search of the world medical literature resulted in locating 41 pairs of SSIDS (15).

CLINICAL SUMMARY OF CASES

On February 27, 1998, in Pittsburgh, Pennsylvania, 2-month-old black fraternal twin girls were found dead in their crib, both in the prone position. The mother, 22 years old and black, with B+ blood type, had five previous pregnancies; the first was a stillbirth at 24 weeks. The next two pregnancies ended in spontaneous abortion at 4 and 16 weeks, respectively, and the final two were carried to term and resulted in two girls, now 4 years old and 1 year old.

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The mother smoked at least half a pack of cigarettes per day during this twin pregnancy and continued smoking after delivery. Three or four months into the pregnancy, the mother had a urine infection; the culture was positive for *Staphylococcus saprophyticus*, which was treated with bacterin. Six days before delivery of the twins, the mother had a chlamydial infection, which was untreated at the time of delivery.

On December 3, 1997, she went into premature labor at 32+ weeks and delivered nonidentical fraternal twin girls. Both infants were delivered vaginally; however, the second infant was in a breech presentation. The first twin weighed 1,855 grams and had blood type B⁺. The second weighed 1,739 grams and had blood type O⁺. Both twins remained in the hospital nursery for 9 days, during which time they were treated for low bicarbonate levels and were given erythromycin for possible exposure to chlamydial infection. Both twins had hyperbilirubinemia, although only the first-born had a bilirubin level high enough to require ~48 hours of phototherapy. Before discharge, routine genetic screening tests were completed for both twins, and the results were unremarkable.

Both twins were transferred to a transitional care home before going home. The first twin stayed in the care home for 7 days with no problems. The second twin, however, stayed for 11 days, because initially she was being monitored for several apneic/bradycardic episodes. Use of the monitor was discontinued before her discharge home.

Both infants had well-baby visits at 1 and 2 months of age. On February 9, 1998, during the second well-baby visit, both twins were observed to have oral thrush and diaper rash, for which mycostatin and Desitin powder, respectively, were prescribed. During this visit, both twins received their first immunizations.

At 11:30 P.M. on February 26, 1998, the twins were each given a bottle of Enfamil and put to bed in the supine position. Both infants slept in a small wooden crib with wooden spokes, containing a mattress covered with a cotton sheet and a cotton comforter. The comforter was folded in half, and the infants were placed on top of it. At 1:00 A.M., both infants were crying, and their mother gave each a pacifier and left them to go back to sleep. The positions of the infants were unknown at this time. Typically, the twins slept through the night. When the mother woke up about 11:00 A.M., she immediately went to check on the twins, finding them in the prone position and unresponsive. She brought them into the living room and placed them on a sofa cushion to attempt resuscitation, which was unsuccessful.

A thorough scene investigation was conducted, which included the determination that very low carbon monoxide levels were present in the apartment and throughout the apartment building. Blood samples from each infant did not reveal the presence of carbon monoxide. When the police arrived at the scene, the apartment had a normal ambient air temperature. The gas stove in the apartment was not on. An electric space heater was located 48 inches from the infants. The apartment was filthy and roach infested, with empty beer cans in the trash. The twins' clothing, crib, and car seats, however, were all clean.

The mother and her boyfriend, the biologic father of all four girls, were very forthcoming with information. According to police at the scene, there was no indication of intoxication of either parents although the father did indicate that he had been drinking a day before the twins' deaths.

PATHOLOGIC EXAMINATION

The twins were transported to the Allegheny County Coroner's Office (ACCO), where at 2:00 P.M. that same day, autopsies were conducted. Twin A was dressed in a sleeper and a diaper. Twin B was dressed in a shirt and a diaper. The internal examinations found moderate pulmonary edema and cerebral edema in both infants. The examination failed to find any signs of physical violence, significant natural disease, or congenital abnormalities. The results of the autopsies are shown in Table 1. The organ weights were within normal limits, except for the lungs, which were heavy be-

TABLE 1. *Body measurements, internal organ weights, and biologic features of the twins*

	Twin A	Twin B
Birth weight (g)	1,855	1,739
APGAR		
1 min	8	2
5 min	9	9
Death weight (g)	4,080	3,645
Body measurement (cm)		
Crown-heel	49	52
Crown-rump	36	34
Head circumference	36.5	37.5
Chest circumference	34.0	34.5
Abdominal circumference	32.0	32.0
Organ weight (g)		
Thymus	18	14
Heart	28	26
Lungs	86	95
Liver	154	142
Spleen	8	6
Kidneys	37	29
Brain	532	489
Blood type	B+	O+

cause of pulmonary edema. Opening of the inner ear to check for otitis media was not conducted and is not a normal component of the autopsy procedure.

The toxicologic analysis was conducted on blood and eye fluid, and all results were negative on both twins. Table 2 lists the drugs that were screened and the results of that analysis. The results were identical for both twins. The bladders were empty, thereby preventing an analysis of the urine. Analysis for glucose in the blood was not conducted; however, testing was done for ketones, and the results were negative. Complete microscopic examinations were also conducted. Each internal organ was sectioned, and several slides (one section from each lobe of the lungs) were made and stained with hematoxylin and eosin. The microscopic review of the tissues was unremarkable, consistent with the gross findings. Microbiologic studies, including bacterial and viral cultures of blood, heart, and lung tissue, with specific lung cultures for *Chlamydia* spp., were all negative.

Both parents agreed to submit to a lie detector test, which showed no evidence of involvement in or knowledge of how the twins died. The twins had two sisters, one 4 years old and the other 1 year old. The 4-year-old was put to sleep in her own bedroom the night of the deaths. She was questioned by police several times. The first time was on the day of the twins' deaths; however, the inter-

view was brief because the child was very distraught and upset. She was again questioned on March 20, 1998, by the sex assault squad after the results of all the scientific examinations had come back negative. Although no sexual assault was ever suspected, the 4-year-old was questioned by the sexual assault squad because of their experience interviewing minors. Based on the interview, the police department determined that the 4-year-old played no role in, or had no further insight into, the deaths. The investigation also found no evidence to support the theory of sibling jealousy playing a role in the deaths. The twins' 1-year-old sister was at the grandmothers' residence at the time of the deaths.

It was the official conclusion of both the ACCO investigation and the police investigation that the parents had played no role in causing the deaths of these infants and that their deaths should be ruled natural in nature.

DISCUSSION

Simultaneous twin sudden infant death has received little recognition in the world literature and, when reported, leads the general public to consider them to be unnatural deaths. This perception was strengthened by the recent admission that a Philadelphia mother, who had eight infants die with their deaths attributed to SIDS, recently admitted to murdering her infants (16). And in Chicago, a grand jury is reexamining the four infants of Deborah Gedzius that were initially ruled as SIDS (16). Bass (17) and others (18,19) within the forensic field consider listing a cause of death as SSIDS to be due to a lack of proper death scene investigation. A worldwide search of the medical literature by the authors of this case summary resulted in the identification of 41 pairs of twins who died of SSIDS (15). It became apparent from the review that many of the reported cases of simultaneous sudden twin deaths failed to describe an adequate death scene investigation, review material, infants' clinical history, or results of the autopsy, or to outline additional studies carried out on materials taken during autopsy. In light of this review, it is clear that a general outline is needed for future presentations of SSIDS cases within the medical literature.

To make the diagnosis of simultaneous twin SIDS, the proposed definition for SSIDS and guidelines for a death scene investigation suggested by Koehler et al. should be followed (15). In brief, a SSIDS case is defined as follows: (1) both infants must meet the definition of SIDS individually, (2) infants must be either monozygotic or dizygotic

TABLE 2. Results of the toxicology analysis conducted on both infants

Blood
Plasma alcohol: not detected
Salicylate: not detected
ADX opiates-not detected
Carbon monoxide: not detected
Cyanide: not detected
ADX barbiturates: not detected
Acidic drugs: insufficient sample
Neutral drugs-insufficient sample
Benzodiazepines (GLC): not detected
Basic drugs (GLC): insufficient sample
ADX cocaine metabolites: not detected
Naproxen: not detected
Chlorpheniramine: not detected
Brompheniramine: not detected
Oxycodone: not detected
TDX acetaminophen: not detected
Acidic drugs (GC/MS): not detected
Acidic drugs (GLC): not detected
Liver
Base: not detected
Bile
Alcohol: not detected

ADX, drugs of abuse (immunofluorescence technique); TDX, therapeutic drugs (immunofluorescence technique); GLC, gas liquid chromatography; GC/MS, gas chromatography-mass spectrometry.

pairs, and (3) deaths must occur within 24 hours of each other (15). In many jurisdictions, a forensic pathologist is required to go to the scene of all infant deaths. At the ACCO, the forensic pathologists have educated the police and the investigating deputy coroners on how to conduct a thorough scene investigation. Based on the 1996 interagency panel guidelines for death scene investigations for sudden, unexplained infant deaths (20), the ACCO created a checklist of questions that must be answered (Table 3), which is provided to the investigating police officer in each case. In difficult cases or in those arousing suspicion, either the police or the deputy coroner will call for a pathologist to come to the scene, as was done in this case.

In all infant deaths, a photographer will fully document the scene, and photos should be available for viewing by the pathologist involved in the case as soon as possible. To improve our ability to have the pathologist view death scenes, this office is now using digital photography. This will enable the pathologist to view the death scene immediately upon the return of the deputy coroner from the scene. Recent or past exposure to gases, such as carbon monoxide and carbon dioxide, should always be tested for. Finally, genetic testing should also be considered in all infant deaths arousing suspicion. In this case, the death scene investigation, autopsy, toxicologic analysis, and additional tests did not reveal any direct cause of the simultaneous twin deaths.

TABLE 3. *Suspected SIDS questionnaire*

1. Mother's full name and age?
2. Father's full name and age?
3. Did mother have prenatal care: If yes, where?
4. Child's birthday?
5. Where was the child born?
6. Type of delivery (vaginal or cesarean)?
7. Any complications during birth?
8. Child's medical history?
9. Length of gestation?
10. Weight and length of child at birth?
11. APGAR scores at birth?
12. Does the child have any brothers or sisters? If yes, give the names and ages?
13. Were there any complications during delivery?
14. If any previous pregnancies, were there any complications during them?
15. When was the baby last seen alive, and by whom?
16. Description of how the child was found as to position, clothing, covered or not covered, and to what extent?
17. Description of crib/bassinet as to fixture, mattress, covering on mattress and bumper pads if any?
18. Room temperature and type of heat or air conditioner?
19. Type of formula the child fed on?
20. Name, address, and phone number of the child's pediatrician?
21. When was the child's last visit with a pediatrician?

Those investigating a sudden infant death must be aware and search for all known physical and environmental risk factors that have been associated with SIDS. Epidemiologic studies on SIDS have frequently reported that the mothers used illegal drugs such as cocaine (21) and methadone (22), as well as legal drugs such as tobacco (23) and alcohol (24), before, during, and after pregnancy. A recent study has reported that nicotine metabolites (nicotine and cotinine) were found in the pericardial fluid of infants who died suddenly (25). It was theorized that their metabolites around the heart muscle may affect cardiac function and trigger SIDS (25). Therefore, a thorough review of the infants' and mothers' medical histories must be conducted to identify the presence of known maternal alcohol and legal or illegal drug use before, during, and after the pregnancy. The relationship (correlation) between smoking and SIDS is well documented (23–29); however, none of the SSIDS cases presented within the authors' literature review made any mention of this modifiable risk factor. Environmental risk factors shown to be associated with SIDS include carbon monoxide (14), carbon dioxide (18,30,31), other gases, excessive bedding (32,33), hypothermia (34), hyperthermia (35–37), parental behavior such as bed-sharing (34,38) or cosleeping, and homicide (18).

One of the most important nonenvironmental modifiable risk factor associated with SIDS is the prone sleeping position (33). The association between the prone sleeping position and SIDS has been reported in virtually all epidemiologic studies (34–36,39,40). Studies from around the world (41–44) have reported a drop in the SIDS rate after a change in sleeping position from the prone to the supine. On the basis of these and other studies, the American Academy of Pediatrics in 1992 recommended that healthy infants be placed in the supine sleeping position (45).

The twins' deaths summarized herein demonstrated several known environmental risk factors for SIDS. The mother smoked throughout and after the pregnancy. The twins were both found in the prone position at the time of their death. These factors have been thoroughly documented in the worldwide literature as being associated with sudden infant death and may have played a role in these deaths.

Overall, with the findings we had and the subsequent negative polygraph tests of the parents, the ACCO and the local police department feel confident that the deaths were natural. This was the first documented case of simultaneous sudden infant twin deaths in Allegheny County, Pennsylvania.

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