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CASE REPORT

Sudden twin infant death on the same day: a case report and review of the literature

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Abstract Sudden infant death syndrome (SIDS) is a major contributor to infant mortality. The cause of death is unknown: suggested possibilities include cardiovascular disease, anaphylactic shock, and suffocation. The occurrence of simultaneous sudden infant death syndrome is uncommon, such cases being extremely rare in forensic pathologic practice. We report two 10-week-old male twins who appeared well at the time of their evening feeding, yet died while sleeping on their backs. Both infants had petechial hemorrhages on the visceral pleura, epicardial surface of the heart, and thymus gland. Microscopic examination revealed pulmonary edema, intra-alveolar hemorrhage, and minor lymphocytic infiltration, again in both infants. In this report, we discuss the risk factors for SIDS, which should be considered individually or in combination as possible causes of death.

Keywords Autopsy · Forensic pathology · Petechial hemorrhage · Risk factors · Simultaneous sudden infant death syndrome

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Introduction

Sudden infant death syndrome (SIDS) occurs within the first 365 days of life [1]. Unexpected infant death and SIDS remain major public health concerns; investigation of unexpected infant deaths is a significant component of forensic pathology [2, 3]. Differentiation between traumatic and natural deaths is necessary. However, a careful clinical history, thorough autopsy, and examination of the site of death often fail to identify the cause of death. Recently, studies involving the pathophysiology of unexpected infant deaths and SIDS have focused on the cardiorespiratory nuclei of the brainstem and the cardiac conduction system [4, 5].

The incidence of SIDS in twins is higher than in singletons [6, 7]. In part, this phenomenon reflects twins' higher incidence of premature births and low birth weights [8]. Previous reports have stated that the death of both twins on the same day is extremely uncommon [9–11]. Some forensic pathologists consider simultaneous death in twins to be suggestive of homicide; however, homicide cannot always be confirmed based on case histories, thorough autopsies, investigation of the scene, and toxicologic analysis [12]. We report a case of unexpected deaths of twin male infants on the same day. We also present a review of relevant published reports with a focus on site of death, risk factors, anatomic and histopathologic findings, and cause of death.

Case report

At midnight the mother of two 10-week-old male twins found them both lying on their backs in separate beds in their home, lifeless. She took them to the emergency unit of

a hospital where death was confirmed. She had fed them cow's milk at about 10:00 p.m. and put them to bed; they appeared normal at that time. A police officer provided us with comprehensive health records and information pertaining to their medical and family histories. Their records and histories included no evidence of potentially fatal diseases. Their parents were both over 30 years old, had been married for 5 years, and had no other children. The twins had been the mother's first pregnancy and they had been born vaginally. Neither parent smoked cigarettes. The parents had only been educated to primary level and had a low incomes and low social status. Because their mother had failed to produce breast milk, the twins had been fed cow's milk since birth. They had received their first doses of hepatitis B and bacillus Calmette-Guérin vaccines at the time of birth and their first doses of oral polio and diphtheria, pertussis, and tetanus (DPT) vaccines 60 days after birth.

An investigation of the place where the twins had died revealed no obvious risk factors. There was a round table, a television, two beds, and a coal stove in the twins' bedroom. The house was of poor quality; in fact, the police officer described their living conditions as deplorable. The twins were dressed in identical thick cotton garments and disposable diapers. White stomach contents were noted in and around the twins' mouths and on their faces. There were no stains, vomitus, or suspicious findings on their clothing. One infant had been sleeping with his mother in her bed and the other infant had been sleeping in another single bed. Both infants were in supine positions with their faces uncovered.

Autopsies were performed approximately 24 h after death. The babies both had the same autopsy findings. External examination showed two well-developed, well-nourished male twins with no apparent physical anomalies. They had dark purple postmortem hypostasis involving their backs and postmortem rigidity had fully developed. Their faces were pale and mildly cyanotic. Their conjunctivae were not congested and they had no petechial hemorrhages of their faces or eyes. There was no evidence of trauma. Internal examination showed dark red cardiac blood that had not clotted. There were petechial hemorrhages on the epicardial surfaces of their hearts (twin A: 23 g; twin B: 27 g), especially at the atrioventricular junctions posteriorly (Fig. 1). Their thymuses were studded with petechiae and larger ecchymoses. Their lungs (twin A: left, 41.8 g and right, 47.6 g; twin B: left, 35.8 g and right, 45.7 g) were markedly congested and edematous, with numerous petechiae and larger hemorrhages (Fig. 2). There was no abnormal material in the upper and lower airways. Their brains (twin A: 534 g; twin B: 597 g) were slightly swollen, but normal in size. The anterior and posterior

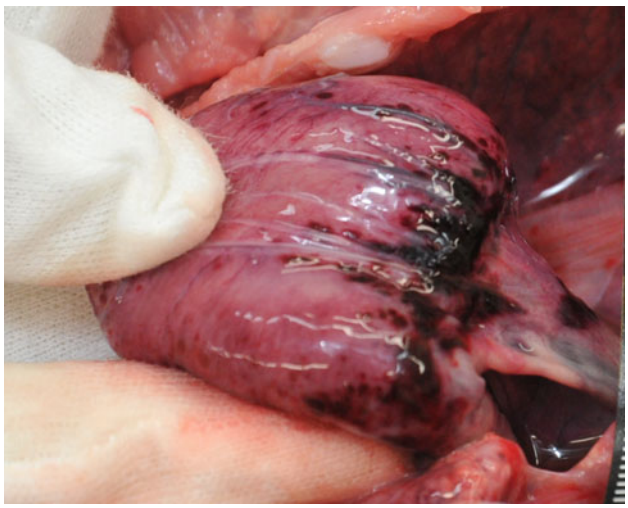

Fig. 1 Petechial hemorrhages on the epicardial surfaces of the hearts (posterior view)

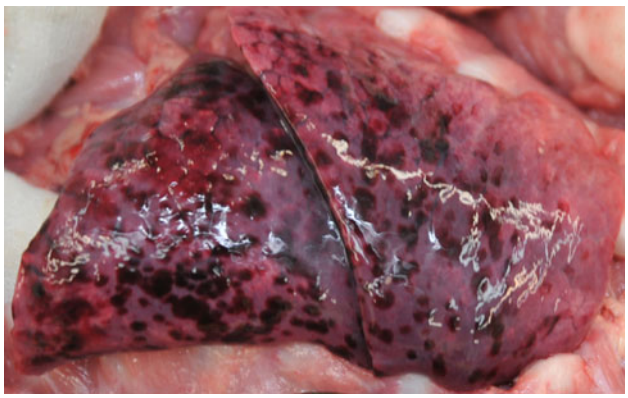

Fig. 2 Numerous subpleural hemorrhages

fontanelles were open in both babies. Their stomachs were partially full of undigested milk curds and the gastric mucosa was unremarkable. The other organs were unremarkable on gross examination.

Microscopically, both babies had extensive myocardial interstitial and focal subendocardial hemorrhages (Fig. 3). Their lungs had focal edema, hyperemia, intra-alveolar fresh hemorrhages (Fig. 4), spilled alveolar epithelial cells, and a small number of mononuclear cells. The other organs were immature for their age with severe hyperemia.

Systematic toxicologic analyses were performed using routine methods on blood, urine, stomach contents, livers, and cow's milk for ethanol and drugs of abuse (acidic, basic, and neutral organic drugs). They had 8 % (twin A) and 10.6 % (twin B) carboxyhemoglobin (COHb) in their blood. We did not detect any insecticides or drugs that are commonly used in the region in their samples.

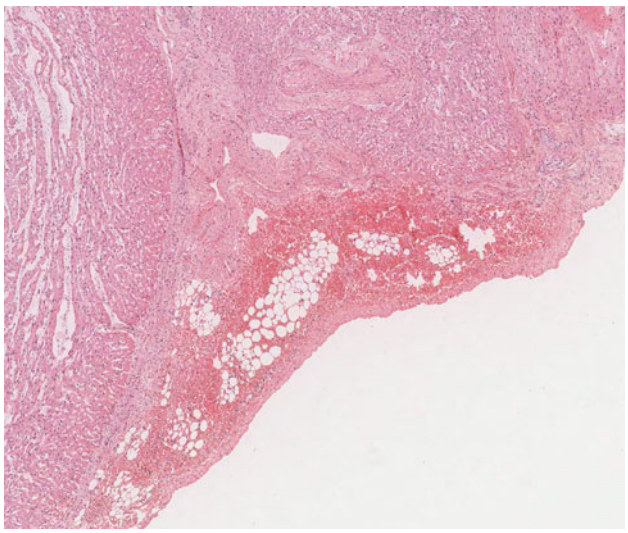

Fig. 3 Subendocardial fresh hemorrhages (*low* power; hematoxylin and eosin [H&E] stain)

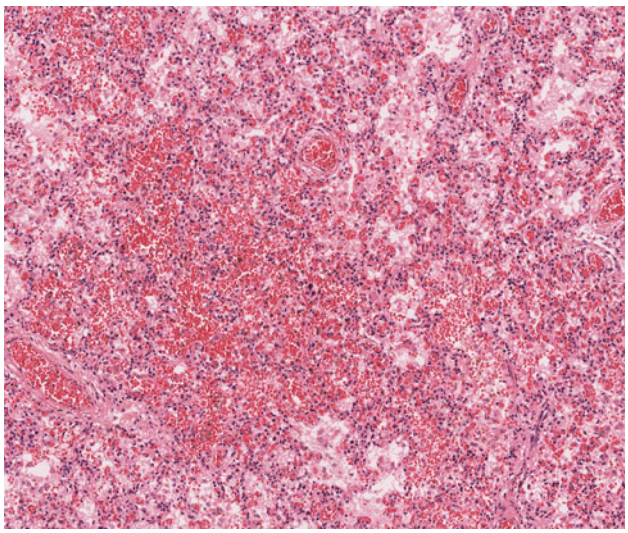

Fig. 4 Intra-alveolar fresh hemorrhages (*low* power; hematoxylin and eosin [H&E] stain)

Discussion

Worldwide, the main cause of death in infants <365 days of age is reportedly SIDS. The sudden death of twin infants on the same day without evidence of injury or disease is commonly known as SSIDS (simultaneous sudden infant death syndrome) [13, 14]; this phenomenon has also been reported in Europe and America. Forensic pathologists are often unwilling to make the diagnosis of SSIDS, especially in cases of simultaneous sudden infant death. A diagnosis of SSIDS evokes great bewilderment and suspicion of the family by others in general and criminal investigators in particular. SSIDS cases reported within large epidemiologic studies lack adequate information about the details of each case [6, 7, 14, 15]. The simultaneous death of both members of a pair of twins has received limited attention in published reports.

A paper by Ladham et al. [9] describes a case of 2-month-old black fraternal twin girls who were both found dead in their cribs, both in prone positions. Their mother had smoked during pregnancy and continued smoking after delivery. Three or 4 months into the pregnancy, the mother had had a urinary tract infection. She went into premature labor. The twins had each been given a bottle of Enfamil (Mead Johnson, Evansville, IN, USA) and put to bed in supine positions at midnight. Both infants had been crying and their mother had given each of them a pacifier and left them to go back to sleep about 2:00 a.m. The twins were found dead in prone positions the next day at noon. Balci et al. [10] reported a case of twin girls (3.5 months old) who were found dead in their crib, both in supine positions. The infants had been delivered by Cesarean section and were healthy. Two days prior to their deaths, the twins had received their second doses of oral polio and DPT and their first doses of hepatitis B vaccines. They had had fever on the first day of the vaccination, for which they had been given acetaminophen. Mitchell et al. [11] reported a case of monozygotic twin girls who died at 12 weeks of age in supine positions. Their mother had smoked during pregnancy. The infant girls had been bottle-fed from birth. They had received standard immunizations on time. On the evening prior to their deaths, each infant had been given a bottle of milk and a dose of acetaminophen for mild fever. The infants had gone to sleep at 10:30 p.m., had not woken during the night and were found dead at approximately 6:50 a.m.

Twins reportedly have a higher mortality rate than singletons because of their higher incidence of low birth weights and prematurity [16]. Environmental factors and sleeping position may play a greater role in SIDS deaths among twins than hereditary factors, especially in the case of infants who were sleeping in the prone position [17]. Virtually all epidemiologic studies have reported an association between the prone sleeping position and SIDS [18]. A number of researchers have reported that in infants who die in the prone position, the causes of death are likely asphyxia, re-breathing of gases, upper airway obstruction, and backwards pressure on the mandible [11, 19]. Furthermore, carbon dioxide can be re-inspired when infants are in the straight or near straight face-down position. Bedding that covers the face or entire head is a potential cause of asphyxia [20]. According to police investigators,

in the present case both infants had been sleeping in supine positions.

In the case reported here, we observed petechial hemorrhages on the twins' epicardia, visceral pleural surfaces, and thymuses at autopsy. The microscopic findings included extensive myocardial interstitial and focal subendocardial hemorrhages and focal edema, hyperemia, and fresh intra-alveolar hemorrhages. Yukawa et al. [21] noted that intra-alveolar hemorrhage is prominent and extensive in cases in which there are worrisome circumstantial features. Because petechial hemorrhages under the pleural surface and intra-alveolar hemorrhage are common in deaths caused by asphyxia, these abnormalities suggest that the child might have been smothered or covered. Intra-alveolar hemorrhage may be an indicator of a significant period of airway obstruction caused by being covered or smothered; however, these autopsy findings are not specific and can occur both in infants who die of natural and unnatural causes. Randall et al. [22] suggested that asphyxia is a possible cause of death in cases of SIDS and classified SIDS into the following five categories [22]: SIDS; possibly asphyxia-related; non-asphyxia-related, cause of death uncertain, and lacking risk factors for asphyxia; and known cause of death. Alm et al. [23] reported that the risk of SIDS during co-sleeping is further increased if the parents have consumed alcohol or drugs before co-sleeping. It has been theorized that these infants die of asphyxia because the parent with whom they are sleeping has rolled on them. Furthermore, investigation of the circumstances of death can reveal risk factors for asphyxia, which include sleeping in the prone or face-down position, soft bedding, bed sharing, covering, sofa deaths, and covering of the face. In our case, the mother had been sleeping with one of the infants, had consumed no alcohol or drugs, and claimed to have co-slept previously without rolling on her infants. The father was not at home that night. We observed a little white staining in and around the twins' mouths and on the faces. There were no stains, vomitus, or suspicious findings on their clothing. We considered that asphyxia caused by reflux of gastric contents was a possibility. However, there was no abnormal material in the upper and lower airways. Thus, there was no direct evidence that both infants had died of asphyxia.

Another major risk factor for infant SIDS is maternal smoking during pregnancy. After sleeping position, maternal smoking is the second most important modifiable risk factor for SIDS [24]. Avoiding smoking during pregnancy may contribute to prevention of SIDS deaths. A previous study has demonstrated that co-sleeping with a mother who smokes is associated with an increased risk of SIDS [25]. Infants who share a room with a smoker while in utero and after birth have an increased risk (the CO effect). Infants of smoking mothers are at exceedingly high risk of SIDS because they have heightened sensitivity to carbon monoxide and low thresholds for toxicity. Sturner [26] determined that >5 % serum COHb is consistent with acute intoxication. In the infants reported herein, COHb serum was 8 and 10.6 %. The twins' parents claimed that they had never smoked cigarettes or used illegal drugs. There was a coal stove for warming the room in the twins' bedroom. In infants with low percentages of serum COHb, as was present in our cases, SSIDS presumably results from the combined effects of various noxious stimuli, such as carbon monoxide, excessive wrapping, and bed covering, and a straight face-down position [13, 20]. Together such factors could have had additive or synergistic effects leading to death. Thus, environmental risk factors, including carbon monoxide, carbon dioxide, and other gases, should be considered possible causes of SIDS.

Because infants have inadequate thermoregulatory control, especially during the first 6 months of life, another important risk factor associated with SIDS is thermal stress, which can be caused by excessive clothing, multilayered covering, and high ambient air temperatures [27, 28]. Hyperthermia is difficult to diagnose posthumously because there are no characteristic autopsy findings or specific tests for it. In the present case, the twins were dressed in layers of cotton clothing and covered with blankets because it was winter. Furthermore, a coal stove was continuously heating the room. Therefore, the pathophysiologic cause of death was presumably hyperthermia.

Most studies have shown that vaccinations are not associated with an increased risk of SIDS [29].

However, there is an association between the timing of DPT vaccinations and the time of peak incidence of SIDS deaths. In the present case, the infants had received DPT vaccine about 2 weeks before their death. The twins had received their first doses of oral polio 45 days after birth; there was no evidence that they had had post-vaccine fevers. Therefore, there is no proven link between vaccination and death in the present case.

A few studies have investigated the possibility of familial genetic diseases [30]. One study of the molecular basis of SIDS reported the presence of long QT syndrome genes in this condition [4]. The family's medical history should be explored. Although genetic screening is not a routinely performed in forensic practice, blood, and other organs should be preserved for later diagnosis of hereditary diseases by genetic screening, especially in cases in which the organs have no obvious pathologic lesions.

Because SSIDS is exceptionally rare and is a diagnosis of exclusion, homicide must be ruled out first. We found no evidence of injury to the mouth, nose, or neck suggestive of compression in either infant. We found no fatal injuries in any other part of their bodies. Systematic toxicologic analysis was negative. No psychological disorders

associated with child abuse or neglect were identified. Therefore, there was no evidence for homicide.

In conclusion, SSIDS cases are rare in forensic practice. These cases should undergo complete autopsies, microscopic examination, toxicologic analyses, and investigations of the circumstances of death. Furthermore, a diagnosis of SIDS should be made only in cases that meet the criteria for this diagnosis. Various risk factors, including environmental conditions, sleeping position, and maternal smoking, must be considered individually or in combination as possible causes of death. The medical histories of the infants, including of immunizations and of their parents, should be elicited.

Key Points

1. For a diagnosis of SSIDS each infant should meet the criteria for SIDS individually and the deaths must occur within 24 h of each other.
2. When an apparent case of SSIDS occurs, thorough autopsies, microscopic examination, and toxicologic analyses must be performed to exclude the possibility of homicide.
3. Investigation of the site of death and known risk factors must be considered individually or in combination when determining the cause of death.
4. Blood and other organs should be preserved for later diagnosis of hereditary diseases by genetic screening and blood cultured for microbiological studies.

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