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Case report

Simultaneous sudden infant death syndrome ☆

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Abstract

The simultaneous sudden deaths of twins rarely occur and therefore it has received limited attention in the medical literature. When the deaths of the twins meet the defined criteria for sudden infant death syndrome (SIDS) independently and take place within the same 24 h range it can be called as *simultaneous SIDS* (SSIDS).

The case(s): Twin girls (3.5-month-old) were found dead by their mother in their crib, both in supine position. The infants were identical twins and delivered at a hospital by cesarean section. Both infants were healthy and did not have any serious medical history. Two days prior to the incident, the twins had received the second dose of oral polio, DPT and the first dose of hepatitis B vaccines and they had fever on the first day of the vaccination and been given teaspoonful of acetaminophen.

Death scene investigation, judicial investigation, parental assessment, macroscopic and microscopic autopsy findings and the toxicological analysis did not yield any specific cause of death. The case(s) were referred to a supreme board composed of multidisciplinary medical professionals at the Institute of Forensic Medicine, Ministry of Justice, in Istanbul. The Board decided that the available data was consistent with SIDS.

These SIDS case(s) are presented because twin SIDS are rare and this is the first time that a *simultaneous* twin SIDS have been reported in Turkey. *Simultaneous* SIDS cases have many implications regarding definition, diagnosis and medico-legal approach.

Keywords: SIDS; SSIDS; Twins

1. Introduction

Sudden infant death syndrome (SIDS) continues to be a phenomenon of unknown cause. Although there is ongoing discussion about changing the definition, the current generally accepted definition of SIDS remains as follows:

The sudden death of an infant under 1 year of age, which remains unexplained after a thorough case investigation, including performance of a complete autopsy, examination of the death scene, and review of the clinical history.[1–10]

A number of different definitions of SIDS have been promulgated each emphasizing a different aspect: for example, an association with sleep; a requirement for extensive ancillary postmortem investigations (e.g., microbiological and toxicological testing); sub classifications based on the presence or absence of minor pathological findings; and specified upper and lower age limits.[11] According to the newly proposed definition,

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sudden infant-death syndrome (SIDS) is, "The sudden unexpected death of an infant <1 year of age, *with onset of the fatal 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".[1,2]

The diagnosis of causes of sudden infant death is an often complex and difficult process. Variable standards of autopsy practice and the use of different definitions for entities such as sudden infant death syndrome (SIDS) have also contributed to confusion and discrepancies.[12]

The *simultaneous* SIDS (SSIDS), as expected due to the rare occurrence, has received limited attention. A proposed definition for SSIDS suggests that in order to meet the criteria the infants should be twins, meet the defined criteria for SIDS independently and the term simultaneous implies that the deaths should take place within the same 24 h range.[13,14] The SIDS case is presented because the infants were twins and found dead concurrently and the twin SIDS occurs rarely.

2. The case(s)

Twin girls (3.5-month-old) were found dead by their mother in their crib, both in supine position in June in 2002 at a Central Anatolian city.

2.1. The parents

The mother was 19 and the father was 25 years old. They had been married for two years and were unsoited. They had not other children. The mother had given birth to twins at her first pregnancy. She was primary school graduate and did not work. Blood group was B Rh(+). She had not allergy or significant medical history. She did not smoke. Her grandmother gave birth to 3 sets of twins and 5 of the 6 off-springs had died after birth. Her uncle had twin off-springs and both died after birth due to unknown causes.

The father was also a primary school graduate and has no regular job. He had been smoking one package of cigarettes per day for 10 years. His blood group was AB(+). The family were of a low income and social status.

2.2. The gestation period

There was no documented or suggested pathology in control visits during the pregnancy. Delivery was at 39th gestational week by Cesarean section. Blood chemistry of the mother was within normal limits.

2.3. Medical records of the infants

Twin A: 1900 g, 44 cm prolapsed umbilical cord present, APGAR scores were 7 (1st min) and 9 (5th min), front fontanel measures 3×3 cm^2, circumference of the head: 32 cm, liver 1 cm palpable, no premature membrane rupture present, meconium (−). Blood type: A (+).

Twin B: 2110 g and 45 cm at delivery, APGAR score 5 at 1st minute, 7 at the 5th minute, front fontanel is 3×3 cm^2, circumference of the head is: 33 cm, liver palpable 1 cm, premature membrane rupture(+), meconium(+), 01.03.2002: 1/6 systolic murmur (no foci stated). Blood type: B (−).

Both babies were healthy and did not face any serious health problems.

2.4. Nutritional status

The twins were breast fed for 40 days. Then, they were fed by formula plus baby biscuits and cow milk.

2.5. Vaccination

The twins received the second dose of oral polio, DPT and the first dose of hepatitis B vaccines on June 3, 2002, two days prior to the incident. Both twins had been given 2 teaspoonful of acetaminophen due to fever on the first day of the vaccination. On subsequent two days they had no complaints of fever.

2.6. Scene investigation, postmortem examination and autopsy

According to the mother's account; she had changed the diapers of the twins the previous night, fed them and let them sleep at about 23.30 p.m. The infants slept together in supine position in a small crib as usual. During the night she woke several times; she changed the diapers and fed both. In the morning, she realized that both were unresponsive. No clue as to the possibility of intentional or accidental death was present within the testimonies of the parents and the other witnesses during the preliminary investigation.

A thorough scene investigation was conducted and revealed no significant risk factors. The first medical examination was performed at the scene at 12 a.m. The twins were together at the same crib, both in supine position, in clothes and their heads were near the crown of the bed. Both were dressed with double pyjamas and homemade textile diapers covered by nylon commercial stuff. There were no stains or vomiting material. There were two settees, a stove and a television in the room and the parents slept in the same room. The stove was not burning. The clothes of the twins were examined. There were no suspicious stains or findings. The room was tidy. The twins were transferred to the Forensic Medicine Department of the City University.

Both twins had seborrhea on the scalp and diaper dermatitis at vulvae and genital regions with erosions and epidermal peeling. Anterior and posterior fontanels were open in both babies. Internal examination revealed that both babies had lungs with incomplete separated lobes, mild

edema and hyperemia. The thymus had petechial hemorrhages. Mucoid content at the lower pharynx and epiglottis of the 2nd baby was seen. Frothy appearance was seen in trachea and down to the bronchus. Both hearts had small petechial hemorrhages on the surface. The organ weights were within normal limits.

The postmortem examination failed to reveal any signs of physical violence, significant medical disease, or congenital abnormalities.

2.7. Histopathological examination results

Heart: Both twins showed immaturity consistent with the age, hyperemia. *Lungs*: Intensive edema, intraalveolar fresh hemorrhage, spilled alveolar epithelial cells, increased light-mild amount of mononuclear cells, hyperemia. *Liver*: Little mononuclear cell increase in the sinusoidal cells, hyperemia, natural appearance at the parenchyma cells. *Kidney*: Several mononuclear cell clusters intra capsular, hyperemia. *Brain*: Hyperemia.

The autopsies indicated no macroscopic or microscopic pathologic findings to which death can be attributed.

2.8. Laboratory analysis

Systematic toxicological analyses (STA) were performed within internal organs (lungs, liver, kidney, brain, stomach, and heart), other specimens (pericardial fluid, feeding bottle, water cups, baby clothes, towel, fine muslin, and bed sheets), blood and urine. Other analysis performed were insecticides from the specimens, psychoactive drugs in blood and urine, COHb levels in blood. Spot tests are used with ITK in CEDIA and CO-OXIMETER.

None of the items in the STA were found in the specimens, 5.3% COHB was found in the blood of the twin A and 5.2% COHB was the blood of twin B.

No pathogen organisms were isolated from the blood and tissue cultures in microbiological examinations.

2.9. Psychiatric evaluation of the parents

Psychiatric interviews were performed on the first week of the deaths. The information was obtained from the parents and the relatives. There were no findings of misconduct on behalf of the parents. The mother thought that the vaccines given the previous day might be responsible from their death.

Psychometric tests and assessments results revealed that both parents had depressive mood and anxiety. Both felt guilty about inadequacy in taking care of the children and felt regret.

As a result of the history, interviews, test results and psychiatric evaluation, within present findings, no psychological disorders suggestive of child abuse or neglect were found. No involvement on behalf of the parents in the death was concluded.

2.10. Forensic evaluation results

The death scene investigation, judicial investigation, parental assessment, macroscopic and microscopic autopsy findings and the toxicological analysis did not yield any specific cause of death. Likewise, the clinical history of the parents and the infants, the gestational follow up during the pregnancy did not imply any pathologic condition the death can be attributed to. The case was referred to a supreme board composed of multidisciplinary medical professionals at the Institute of Forensic Medicine, Ministry of Justice, in Istanbul. The Board decided that the available data was consistent with SIDS.

3. Discussion

Sudden infant death syndrome (SIDS) is the leading cause of death of infants during the post perinatal period 7–365 days.[6–9,13,15] Simultaneous sudden deaths of twins during infancy has received little recognition in the world literature. A worldwide search of the medical literature resulted in the identification of 41 pairs of twins who died of SSIDS from 1900 to 1998.[13,14] Many of the reported cases of simultaneous sudden twin deaths failed to describe an adequate death scene investigation, infants' clinical history, or results of the autopsy, or to outline additional studies carried out on materials taken during autopsy. 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. is adopted in this case report.[14] In brief, a SSIDS case is defined as follows: (a) each infant should meet the criteria of SIDS individually, (b) infants must be either monozygotic or dizygotic pairs, and (c) deaths must occur within 24 h of each other (9). Ladham et al. presented the death of two months old black twin girls as the first SSIDS case of the region.[13,14]

In another study by Darios et al. the incidence of SIDS was 82% higher in twins as compared to singletons. The excess was especially prominent in term infants. The probability of a 2nd twin dying of SIDS, given that at least one twin had died of SIDS was 4.9 times higher than the overall risk of a twin dying of SIDS.[16]

Similar to the relevant studies non specific findings like multiple petechial hemorrhages of the thymus and lungs and pulmonary edema are determined at the autopsy in this cases.[6–9,14,17] These findings, individually or in combination, are not specific and can occur in infants who die of natural and unnatural causes. However, the location of the petechiae is significant as it suggests SIDS rather than undetermined infant death.[18]

Although, SIDS is a diagnosis of exclusion, there were certain repetitive features common to the majority of SIDS cases and this case is not an exception.[1,19] Twin pregnancies, low birth weight, the infants' age (months), young ages of the parents, low socioeconomic status, parental education, poor infant care, death during sleep and early

in the morning, lack of breast feeding are some of them in this case.[7–10,15,20–24]

Also common modifiable risk factors like maternal smoking or substance abuse and sleeping in prone position, sleeping on adult beds or bed sharing with adults were not present in this case.[5,10,11,20,23–32]. For infants less than 4 months of age the risk of SIDS while sleeping prone has been shown to be higher during winter than summer.[33] Prone sleeping has also been associated with excess thermal insulation. It is possible that these infants were too heavily wrapped, inducing overheating, which has the most serious implications for the smallest infants with the least mature regulation of temperature. In these cases the death took place in the summer season. Despite the season both were dressed with double pajamas and covered with blankets. Hyperthermia is considered as a risk factor in some studies.[23,34–36]

Whether passive smoking is a risk factor for SIDS is still controversial. Some studies have found secondary smoking to be a risk factor.[21,23,29,37,38]. Arnestad et al. could not find a significant relationship between SIDS and the father's smoking habit.[10] However, we think that sharing the same room with a smoker during the pregnancy and after birth presents an environmental risk factor of CO effect.[23,39] Toxicological analysis yielded 5% COHb for both twins. Some studies have shown an increased risk of SIDS associated with co-sleeping or sleeping together with a mother who smokes as being the greatest hazard[5,10,13,20,23,40–48] Although there is no bed sharing in this case, it still deserves discussion since they share the room.

In the past there was concern that vaccinations might cause SIDS as the peak ages for SIDS is 2–4 months, which coincides with the age for vaccinations[9] However studies have shown that vaccinations are not associated with an increased risk of SIDS.[42,49,50] The twins received the second dose of oral polio, DPT and the first dose of Hepatitis B vaccines two days prior to death. They had fever and been given teaspoonful of acetaminophen each the first day of the vaccination. On subsequent two days they had no complaints of fever. The pediatricians consulted about the possible connection between deaths and immunization also confirmed that there is no proven link between vaccination and SIDS. According to the pediatricians, hypersensitivity reaction was not an expected incident since the twins were vaccinated the second doses and a day has passed over. Moreover, the vaccination records of the health unit were examined with the instruction of the health directorate and there were no complications or complaints in other children given the same vaccines consecutively.

There is always the possibility of some familial metabolic or other genetic disease.[3,7] When the familial history of the mother is explored; the mother's mother had 3 twin births to 6 children, of whom 5 died sometime after the delivery. And the mother's own twin sibling and the twins of the uncle of the mother also died. In order to evaluate the possible hereditary metabolic disease; specimens from blood, brain and muscle tissues were also obtained and stored in deep freezer with the suggestion of the pediatricians. However at the point of conclusion the Council of Forensic Medicine, Ministry of Justice, did not find it essential in the diagnosis. Such analysis is not a part of routine analysis performed at the Council of Forensic Medicine laboratory. Besides it does not have facilities of its own to perform it. The weakness of this study with regard to the diagnosis seems to be the lack of analysis for inherited metabolic disease.

However, as the judicial authorities are interested in the cause of death in connection to any criminal activity, their reluctance to afford the costs of the further examinations as to the metabolic and genetic disorders is not a surprise. Furthermore, some of the examinations performed in this case out of routine procedures have been possible on behalf of the forensic medicine team involved.

4. Conclusion

SIDS is rarely diagnosed in Turkey probably because of the deficiencies in the course of differential diagnosis. There are numerous unexpected sudden deaths of infants in Turkey. However, these cases are usually not classified as SIDS because full autopsy, scene investigation records and sufficient laboratory analysis are not performed in most of the cases. Therefore, we cannot draw any conclusions about the epidemiology of SIDS in Turkey. The situation in Turkey calls for a standard procedure to be established and put into practice in order to diagnose SIDS cases.

This case is the second case that the supreme board has concluded as consistent with SIDS. And the first time that a *simultaneous* SIDS is to be reported in Turkey. In this case the infants are twins, meet the defined criteria for SIDS independently and the deaths took place concurrently. Cases of twin *simultaneous* SIDS reminds us that further studies are required both to illuminate the causes of SIDS and thus, try to prevent them.

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