

## Simultaneous sudden infant death syndrome: case report

### 同步嬰兒猝死綜合徵：個案報告

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Simultaneous sudden infant death syndrome (SSIDS) is death of both twins within 24 hours which is compatible with the sudden infant death syndrome criteria and death is independent of each other. A pair of twin babies, approximately two months old, was found dead in the supine position in bed by their mother. After scene, autopsy, toxicological and histopathological investigations, specific cause of death could not be found. There were 41 pairs of twin babies with SSIDS reported in the literature between 1900 and 1998. This is the second case report from Turkey. (*Hong Kong j.emerg.med.* 2010;17:297-302)

同步嬰兒猝死綜合徵為兩孿生兒於24小時內在不受對方影響下死亡，而符合嬰兒猝死綜合徵的標準。一對大約兩月大的孿生兒被母親發現在床上仰臥死亡。經現場、驗屍、毒理學及組織病理學調查後，仍未能找出明確的死因。在1900至1998年間的文獻中有41對孿生兒的同步嬰兒猝死綜合徵報告。這是土耳其的第二個個案報告。

**Keywords:** Sudden infant death, twins

**關鍵詞：**嬰兒猝死、孿生兒

### Introduction

Simultaneous sudden infant death syndrome (SSIDS) is the death of a pair of twins occurring at the same time. Its cause cannot be explained with the clinical history, case assessment, crime scene investigation or autopsy.<sup>1-4</sup> Sudden infant death syndrome (SIDS) is

the leading cause of death in babies and the incidence is between 0.5 and 2 per thousand live births in many different European regions.<sup>2-5</sup> According to epidemiological studies, the risk factors of SIDS are: black race, male gender, prematurity, low birth weight, the 2-5 months babyhood period, illegitimate babies, low maternal age, winter season, low socioeconomic class, low maternal education, alcohol, cigarette, and illegal drug abuse in antenatal and postnatal periods.<sup>2,6,7</sup> Sleeping in the prone position, sleeping out of the baby bedstead and sleeping with a person in the same bed have also been suggested as risk factors for sudden baby death related to sleep.<sup>7,8</sup>

SSIDS is a kind of twin babies contemporary death and the death is independent of each other.<sup>6</sup> SSIDS, in twin babies, is very rare in the literature. It was reported between 1900 and 1998 in the literature that 41 pairs of twin babies died because of SSIDS.<sup>1,9,10</sup> In the past years, only one case was found in Turkey.<sup>1</sup>

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## Case presentation

In February 2007, a pair of 2 months and 8 days old twin male babies was found dead in the supine position in their bed by their mother in Erzurum, Turkey.

The mother was 22 years of age and graduated from high school. She was a nurse and her blood group is O Rh(+). The twins were her first pregnancy. The mother had no history of alcohol, cigarette or illegal drug abuse. The father was 30 years of age and he was a petty-officer. His blood group is A Rh(+). The family belonged to the middle class in the society. The grandmother of the twins had history of five sudden infant deaths in early childhood.

The twin babies were delivered by Caesarean section and were premature (approximately eight months). Their birth weights were about 2500 grams. The twins were closely observed in the neonatal intensive care unit and they were incubated due to apnoea problem (the first baby was incubated for five days, and the second one was incubated for ten days). Finally, both babies were discharged. The babies were breast-fed for about 45 days and followed with formula feeds (Milupa Preaptamil).

Both babies had been vaccinated the second doses of oral polio vaccine, diphtheria-pertussis-tetanus and hepatitis B three days before death. The first baby had high fever and was given a full teaspoon (~40 mg) of ibuprofen syrup by the mother. Later, the fever settled according to the mother's description.

### *Progress*

According to the history from the mother, the babies were sleeping in bed in the supine position after feeding at around 4:00 pm. At around 5:30 pm, she saw the babies were not breathing, their eyes were closed and they were motionless. When she saw their babies in that condition, she tried to perform mouth to mouth ventilation and she called for his husband. According to the mother's information, the room temperature was approximately 23-24 degrees Celsius.

When the babies arrived at the emergency department at about 6:15 pm, both babies' pupils were fixed and dilated. No heart beat or respiration was detected.

Asystole was shown in the electrocardiogram, and cardiopulmonary resuscitation with intubation was performed for 30 minutes. However, they showed no response to resuscitation and finally succumbed.

### *Postmortem examination*

The babies' heights and weights were 54 cm/3900 grams and 53 cm/3750 grams respectively. The weights of the internal organs of both babies (brain-cerebellum, heart, lung, liver, spleen, and kidneys) were close to each other, and the internal organs were of normal size according to their age.

After the autopsy, different organ samples (lung, heart, brain, cerebellum, liver, and kidney) were taken and sent to the Chemistry Specialization Bureau for histopathological and clinical toxicological analysis with blood (5 ml), gastric content, baby food (Milupa Preaptamil) and baby bottles.

### *Toxicological analysis*

For the analysis of organic compounds, the gas chromatography method was used. For the detection of insecticides, drugs and carboxyhaemoglobin levels, spectrophotometric analyses were used. Atomic absorption analysis was used for heavy metal detection. For the detection of systemic hypnotic/narcotic/stimulant drugs, bioassay methods were used. The results of toxicological analysis are shown in Table 1. Carbon monoxide and other toxic air particles could not be detected in the vicinity in the crime-scene investigation.

### *Autopsy report*

The brain and cerebellum were found to be normal. The kidneys, liver and spleen were hyperaemic. The lung surfaces were intensively haemorrhagic and oedematous. The first baby's heart surface had a green-like colour. The second baby's gastric mucosa was hyperaemic.

### *Microscopic study*

For both babies, it was reported that there was no abnormality in the heart, brain and cerebellum sections. There were marked congestion and haemorrhagic areas in the lungs; and there was also congestion in the liver, spleen, kidneys and stomach.

### *Summary of autopsy, toxicological and histopathological findings*

1. Hypnotic, narcotic and toxic compounds were not found in the blood, urine, internal organs, gastric content, feeding bottle and baby food.
2. The lungs were congested and haemorrhagic and the internal organs congested.
3. It was concluded that the cause of death could not be determined with the information and evidence from the clinical history, case assessment, crime scene investigation and autopsy.

### Discussion

Sudden infant death syndrome has been defined as the sudden death of an infant less than one year of age which remains unexplained after a thorough case assessment, including performance of a complete autopsy, investigation of the death scene, and review of the clinical history.<sup>1-4,7</sup> It is the leading cause of death in the postnatal period.<sup>1</sup> Simultaneous sudden infant

death syndrome is death of both twins within a period of 24 hours which meets with the SIDS criteria and fatality is independent of each other.<sup>1</sup> SSIDS is even rarer and only a limited number of cases have been reported in the literature.<sup>6</sup> The earliest reported case of SSIDS was cited by Hammar at the turn of the 20th century.<sup>11</sup> Between 1900-1998, 12 cases that met the criteria of SSIDS were reported.<sup>6</sup> Most of the SSIDS cases in the literature failed to describe even the most basic epidemiologic data such as age, sex, race, and place and time of death.

Although many pathophysiological theories for SIDS have been proposed, including respiratory dysfunction, cardiorespiratory instability, cardiac dysrhythmias, and inborn errors of metabolism, decisive pathogenic mechanisms triggering an infant's sudden death remain unclear.<sup>2,7,8</sup>

Many risk factors have been reported for SIDS (Table 2). One of the most important suggested risk factors for SIDS is the baby's sleeping position. It has been

**Table 1.** Results of toxicological analyses of the two babies

<b>Insecticide</b> (phosphate, carbamate, chlorine)	Not detected
<b>Drug</b> (acetaminophen, phenothiazines, pyrazolone derivatives, salicylates, strychnine)	Not detected
<b>Systemic hypnotic/narcotic/stimulant</b> (amphetamines and derivatives, benzodiazepines, barbiturates, cocaine, marijuana, tricyclic antidepressants)	Not detected
<b>Organic compound</b> (ethyl alcohol, methyl alcohol, formaldehyde, cyanide)	Not detected
<b>Plasma poison</b> (carboxyhaemoglobin, methaemoglobin)	Not detected
<b>Heavy metal</b> (copper, lead, arsenic, antimony)	Not detected

**Table 2.** Possible risk factors for SSIDS<sup>2,6-26</sup>

Baby's sleeping position (sleeping in prone position, sleeping in lateral position, sleeping out of the baby bedstead and sleeping with a person in the same bed)
Environmental risk factors (hypothermia, hyperthermia, carbon monoxide and other gases)
Vaccinations
Some familial metabolic or other genetic diseases (mitochondrial DNA, IL10, HLA DR, C4 gene mutations)
Mother's smoking, alcohol and illegal drug use (methadone, cocaine) before and during pregnancy and in postpartum period. Low social and economic status
Other factors related to the baby (male gender, 2-5 months babyhood period, prematurity, low birth weight, black race)

reported that sleeping in the prone position carries an increased risk for SIDS.<sup>8,12-14</sup> It has also been reported that sleeping in the prone position will cause diaphragmatic exhaustion, breathing air with a high concentration of carbon-dioxide and then asphyxiation.<sup>15,16</sup> Li et al also suggested that besides the prone position, sleeping in the lateral position is also a risk factor for SIDS.<sup>17</sup>

Low environmental temperature was described as a risk factor.<sup>15</sup> It has been suggested that SIDS is seen 2-3 times more in winter months than summer months.<sup>18,19</sup> Our cases also occurred in the winter months, which is compatible with the literature.

As frequently reported in the literature, both babies were vaccinated three days before their death, there might be a relationship between vaccination and SIDS. Vennemann et al reported in their study that immunisation did not increase SIDS, on the contrary it could have a protective role.<sup>20</sup>

Opdal and Rognum reported that besides environmental factors, some genetic mutations may predispose to SIDS.<sup>21</sup> It has been claimed that all mutations which cause fatal metabolic diseases can also constitute a predisposition to SIDS. In particular, some mitochondrial DNA (mtDNA), IL10, HLA DR, and C4 gene mutations can increase the risk related to SIDS.<sup>21</sup> It has been suggested that paroxysmal supraventricular tachycardia and long QT syndrome-complex are seen more frequently in babies with genetic inclination and this inclination can cause unexpected baby death because of cardiac arrhythmias.<sup>22,23</sup> Our cases are thought to have genetic predisposition because in their grandmother's early obstetric history, there was a history of five infants' mortality with unknown reason. A simple upper respiratory tract infection can also be the reason for the deaths of babies with genetic predisposition.

Another leading risk factor for SIDS is maternal smoking. During the antenatal and postnatal periods, smoking is risky for both the baby and mother. It has been claimed that smoking in pregnancy will have a higher risk of early rupture of membranes, low birth weight and adversely affected foetal lung development.

It is also known that passive smoking can cause asthma, otitis media and SIDS in babies.<sup>24-28</sup> In the antenatal and postnatal periods, babies exposed to cigarette smoke are prone to viral infections. Unexpected baby death can be seen because of inefficiency in maintaining an open airway during the sleeping period.<sup>24</sup>

Another risk factor of SIDS is the male gender. It was shown that male infants appeared to show decreased arousability or decreased response to pain compared to female infants.<sup>14</sup> The age range that is at higher risk for SIDS is from 2 to 5 months of age.<sup>1,2</sup> The explanation for the age distribution of SIDS, particularly in partial sparing of the first month of life, could be an early protective mechanism that is limited in duration. For example, gasping is capable of restoring regular respiration in neonates, but the efficacy of the gasp depends on the glycogen store in the cardiac muscle, as improved oxygenation of the pulmonary capillaries will not help unless the oxygen is delivered to the brain. The notable ability of the foetus for anaerobic metabolism obviously still exists in the neonate, but gradually diminishes in the first few weeks.<sup>27</sup>

Although SSIDS is a diagnosis of exclusion, there were certain repetitive features common to the majority of SSIDS cases such as low birth weight, male gender, and death during sleep. Koehler et al<sup>6</sup> defined the diagnostic criteria for SSIDS in his study as follows:

1. The infants must be 7-365 days old and no apparent reason of death could be determined from autopsy, toxicological screening, crime-scene investigation or clinical history of the babies and mother.
2. The babies must be monozygotic or dizygotic twins.
3. Both babies' death must be within 24 hours.

When the 41 SSIDS cases reported in 1900-1998 were examined carefully, it was found that 12 pairs fulfilled three and 9 pairs fulfilled two of the aforementioned criteria respectively. Demographic data were not clearly mentioned in 15 pairs and diagnoses other than SSIDS should be considered in 5 pairs.<sup>6</sup> Most of the SSIDS cases reported in the past had inadequate toxicological and histopathological screening analyses, which are mandatory in making the diagnosis. It was reported

that there was extensive congestion and fresh bleeding areas in the lungs; and congestion in the liver, spleen, kidneys and stomach were also noted in the histopathological examination of our patients. The cause of congestion might be resuscitative attempts or crush asphyxia. Petechial bleeding and pulmonary oedema in lungs are seen frequently in reported SIDS cases but these are not diagnostic for SIDS.<sup>1,28</sup>

In summary, the cause of sudden infant death is unclear. Babies should be under the supervision of paediatricians especially between the 2nd and 5th months which is the period of highest risk for babies. Metabolic analysis and cultures should be taken, if necessary. Mothers should not smoke cigarette and should avoid drugs during pregnancy. Babies should be kept in bed in the supine position and single. Babies should be fed with mother's milk for at least four months after birth. Society should be aware of SIDS and the predisposing factors.

## Conclusion

Simultaneous sudden infant death of both twins has rarely been reported in the literature and most of the reports do not fulfil the criteria exactly. Our cases were defined and reported as SSIDS with the support of the clinical history, case evaluation, crime-scene investigation, autopsy, toxicological and histopathological analyses.

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