Int J Legal Med (2010) 124:631–635
DOI 10.1007/s00414-009-0407-y

CASE REPORT

Simultaneous sudden unexpected death in infancy of twins: case report

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Received: 24 November 2009 / Accepted: 16 December 2009 / Published online: 16 January 2010

Abstract Simultaneous sudden infant death syndrome (SIDS) in twins is an extremely rare event. Some believe these are natural deaths, whereas others suggest they are unnatural. We report monozygotic twins dying at 12 weeks of age. Extensive investigation concluded that the cause of death was natural. Our case fulfilled the criteria of simultaneous SIDS in twins. However, thermal stress due to excessive thermal insulation, use of a pillow and placing twins in the same cot all represent a potentially unsafe sleeping environment. We recommend the term "simultaneous sudden unexpected death in infancy of twins" to describe such cases.

Keywords SUDI · Simultaneous death · Twins · Thermal environment · Unsafe sleeping environment

Introduction

Twins are at higher risk of sudden infant death syndrome (SIDS) than singletons [1]. However, this largely appears to be due to lower birthweight of twins. After the "Back to Sleep" campaign in the US, twins continued to have a higher SIDS rate (1.3/1,000 live births) compared with singletons (0.7/1,000) [2]. The risk factors for SIDS (male, small for gestational age, black, unmarried and maternal smoking) were similar for both singleton and twins. The risk of SIDS in twins is similar for both monozygous and dizygous twins [3, 4].

Simultaneous SIDS in twins has been reported but is extremely rare. The literature mainly deals with single case reports [5, 6]. The deaths are classified as either simultaneous sudden infant death syndrome in twins [7, 8], or due to injuries associated with environmental hazards [9] or "unascertained" [10]. The literature is limited and generates strong opinions. To quote one author: "The likelihood of twin infants dying suddenly and simultaneously of SIDS, a natural disorder, defies credibility" [9].

The aim of this paper is to describe a further case of simultaneous unexpected deaths of twins and to discuss the possible causes of death in this case.

Case report

There had been one previous child born to young Maori parents. That child was aged 3 at the time of the birth of the monozygotic twins and she was well. This pregnancy was complicated by pregnancy-induced hypertension. The mother smoked one to two cigarettes a day during pregnancy. Labour was induced at 36 weeks gestation. Both infants were delivered vaginally without complication. Twin 1 weighed 2,100 g (10th–15th percentile) [11], length was 43.1 cm (5th–10th) and head circumference 32 cm (25th). Twin 2 weighted 2,700 g (50th percentile), length was 46 cm (25th) and head circumference 33 cm (50th). The infant girls were bottle fed from birth. They were discharged home at 7 days of age.

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At 20 days of age both infants were admitted to hospital for 2 days due to an upper respiratory tract infection (URTI). Two weeks prior to death both infants had minor URTI which was treated in the community with antibiotics. Five days prior to death they had the 3 months immunisation (OPV, DTaP and HibHepB). The 6 weeks immunisation had also been given on time. Both parents smoked.

On the evening prior to the death each infant was given a bottle between 9:30 and 10:00 pm and a dose of paracetamol for mild fever. Both infants were placed to sleep on their backs alongside each other in the same cot, which was in the parental bedroom. The infants went to sleep at 10:30 pm. Both infants were wearing a Stretch N Grow and cardigan and wrapped in separate blankets, and then a folded fleece blanket and quilt covered both infants. There was a sheepskin underlay. The infants' heads rested on an adult sized pillow which was used for the first time. A portable gas heater was used to heat the bedroom and this was on from 4:00 to 5:00 pm.

The infants did not wake during the night as was their usual practice. Both the infants were found dead at approximately 6:50 am. They were lying on their backs and their heads were not covered by bedding. Their clothing was very damp. Cardiopulmonary resuscitation was attempted by ambulance officers when they arrived at 7:14 am, but this was unsuccessful. The infants were aged 12 weeks.

Autopsy findings

Both infants had no dysmorphic features and there was no bruising. The skin of both infants was in good condition and there was no napkin dermatitis. Twin 1 had a left occipital plagiocephaly. Her body weight was 4,490 g (3rd) [12], crown-to-heel length 57.5 cm (15th) and head circumference 39.2 cm (50th). There were multiple petechial haemorrhages over the epicardium, visceral pleura and thymus. Twin 2 had a right occipital plagiocephaly. In addition there was a Mongolian blue spot on the left buttock and a 1.5-cm diameter strawberry naevus on the left shoulder. Body weight was 4,950 g (15th percentile), crown-to-heel length was 57 cm (15th) and head circumference 40 cm (50th−85th). There were some petechial haemorrhages on the epicardium, visceral pleural surfaces and thymus. The internal organs were normal and neither infant had macroscopic or microscopic evidence of infection. Routine histology was also normal. This includes H&E staining of the lungs (four to five sections), trachea and larynx, heart (right and left ventricles and septum including coronary arteries), diaphragm, both kidneys, spleen, liver, pancreas, brain (multiple sections from cervical spinal cord, brain stem, midbrain, cerebellum, cerebral hemispheres to include deep grey nuclei, cortex, white matter and hippocampus), dura, adrenals and thyroid.

Other investigations

Toxicology screen, including paracetamol, was negative for both infants. Carbon monoxide levels at a saturation of 5% (twin 1) and 6% (twin 2) were found. The portable gas heater and cylinder were less than 1 year old and were in good working order and without leakage. Furthermore, it had a shut-off system, such that if there were not enough oxygen in the air whilst the heater was running, the gas supply to the heater would automatically be shut-off. The minimum air temperature at the closest weather station that night was 5.5°C.

Discussion

The investigation of the deaths of these twins was thorough. It included a complete autopsy, examination of the death scene and review of the clinical history. The investigation is similar to that currently recommended [13, 14]. Unnatural deaths have to be considered in this case of simultaneous unexpected death in twins. However, there was no evidence of bruising or previous inflicted trauma and the infants appeared well cared for. Furthermore, various reports to the coroner emphasised that the children were loved and well cared for.

Poisoning by the gas heater was unlikely as it was in good condition. The normal level of carboxyhaemoglobin for non-smokers is <2% and for smokers 5–9% [15]. The level of carbon monoxide in both infants' blood was considered to be due to environmental tobacco smoke [16]. Although parental smoking is a risk factor for SIDS and it has been argued as a cause of SIDS, it cannot be proven to be the cause of SIDS in an individual case [17].

The infants were immunised 5 days prior to death. Although some claim that immunisation causes SIDS [18, 19] various studies indicate that immunisation is associated with a lower risk of SIDS [20–22].

On the night of the death an adult pillow was used for the first time. Pillows have been implicated in SIDS in some studies [23, 24] but not all [25]. The usual postulated mechanism is due to rebreathing expired gases if the infant is face down in the pillow [26]; however, in this case the infants were found supine. Possibly airway obstruction could be produced by the pillow flexing the head forwards, as has been demonstrated in car seats when the head falls forwards [27].

Bed sharing is a well-established risk factor for SIDS; however, this usually refers to bed sharing with an adult [28–30]. Bed sharing with an older sibling is associated with an increased risk of SIDS [23]; however, there are no data on the risk of SIDS when cobedding with a twin, and this is an area that requires urgent research [31]. Further-

more, it is not known how frequently twins share the same cot, and their position within the cot.

Although the thermal properties of the clothing and bedding are not completely known, it seems likely that the thermal insulation was in excess of that required for the environmental temperature. In addition, the thermal effect of co-sleeping with a twin is uncertain, although each twin would act as a heat source for the other twin. Endogenous fever needs to be considered but is unlikely. Although the infants had an URTI 2 weeks prior to death, they appeared to have recovered completely at the time of death. Immunisation is also an unlikely cause of fever in this case. Fever, if it occurs, is usually seen in the 48 h following the immunisation, and is not present at 5 days. The parental observation that the infants were found very damp suggests the infants were thermally stressed. Although there are numerous causes of being found very sweaty, we have reported that duvet use and head covering were associated with being found very sweaty, suggesting that sweating is related to thermal stress [32]. Thermal factors were a risk factor for SIDS but only appeared to be important when the infants were sleeping prone [33]. Now that few babies sleep prone, thermal factors are no longer risk factors for SIDS [34].

There is now convincing evidence that subtle pathological abnormalities, including gliosis, are present in the central nervous system of SIDS victims [35, 36]. These abnormalities may arise because of exposure to a suboptimal in utero environment including exposure to maternal smoking or because of premature birth [37, 38]. Future SIDS infants also show less body movement and arousal during sleep than controls and infants of smokers have an increased arousal threshold to an external stimulus as well as decreased spontaneous arousal [39]. Arousal is also less after obstructive apnoea in infants of smoking mothers [40]. Overheating may also contribute to decreased arousability [41]. Arousal is considered to be a critical component of the response to upper airway obstruction in infants and children.

The areas of the brain that are abnormal histologically in SIDS infants are areas containing serotonergic neurones and levels of serotonin have been shown to be decreased in these areas [33]. Serotonergic mechanisms can be disrupted because of exposure to smoking in pregnancy and also because of congenital disorders of serotonin metabolism [42–45]. It is likely that these genetic abnormalities explain some of the ethnic variation in prevalence of SIDS and also why some infants are at risk even when sleeping in the recommended sleep position [43, 45]. Given that these infants were monozygotic twins, a genetic abnormality is possible.

The first attempt at reviewing the world literature on simultaneous SIDS in twins was by Smialek in 1986 [7]. He identified nine cases. These cases were included in a later review by Koehler et al. [8]. This review identified 41 cases reported in the world literature from 1900 to 1998. They proposed three criteria for the definition of simultaneous infant death:

1. Both infants must independently meet the definition of SIDS from the National Institute of Child Health and Human Development [46].
2. The infants must be members of either a monozygotic or dizygotic pair.
3. The deaths must occur within 24 h of each other.

The panel defined SIDS as “the sudden death of an infant under 1 year of age, which remains unexpected after a thorough case investigation, including performance of a complete autopsy, examination of the death scene and review of the clinical history” [46]. Each of the infants would independently meet this criterion, as well as meeting the other two criteria.

In this review of 41 cases, the authors reported that 12 cases met all three criteria and thus could be classified as simultaneous SIDS in twins. Nine did not have a death scene investigation and therefore the authors could not eliminate environmental hazards. Fifteen cases had limited information and thus no judgement could be made. In five of the cases alternative diagnoses were offered (in pram together, sleeping in mother’s arms, hyperthermia, prone on a pillow and immunisation 3 h prior to death). These five cases illustrate the difficulty; each factor may be a risk factor for SIDS but does not necessarily indicate cause of death, and other reviewers might easily have labelled these deaths as SIDS.

The paper by Bass [9] reports on 13 pairs of simultaneous infant deaths in twins that originally had been labelled as simultaneous twin SIDS in ten, undetermined in two and sudden infant death (non-traumatic) in one. Further information was collected from health professionals and in six of the 13 cases by contacting the next of kin. Some of these deaths occurred over 20 years previously, and thus there may be serious problems of accurate recall. In each case an alternative diagnosis was offered, mainly hyperthermia or smothering. But again there is a problem of mixing risk factors with cause of death. For example, in two pairs deaths were reclassified as suffocation on a water mattress, apparently based solely on the fact that they were placed to sleep on a water mattress.

The single case report by Ramos et al. describes in detail the sudden and simultaneous death of twins [6]. The authors rejected the diagnosis of simultaneous SIDS, and instead conclude that a combination of several environmental factors (sub-lethal levels of carbon monoxide, over wrapping and mechanical obstruction of upper airways) resulted in death. Carter et al. challenged their findings and

felt that deliberate smothering was a possibility, and stated that although smothering could not be proven they would have alerted the relevant caring agencies and would have assigned the cause of death as "unascertained" [10].

We concluded that in our case the cause of death was natural. Our case fulfilled the criteria of simultaneous SIDS in twins. However, thermal stress, use of a pillow and placing twins in the same cot all represent a potentially unsafe sleeping environment and could be interpreted as a cause of death, either individually or in combination, or as possible risk factors, but not the cause of death. With the decreasing number of SIDS cases, identifying the cause of the remaining unexpected deaths in infancy has become more difficult. This case illustrates this difficulty. More cases of sudden unexpected death in infants are being defined as "unascertained" rather than SIDS [47]. It seems appropriate to define the cause of death in these twins as "simultaneous sudden unexpected death in infancy of twins", thus avoiding the term SIDS.

Acknowledgements EA Mitchell is supported by the Child Health Research Foundation.

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