TECHNICAL NOTE

Roger W. Byard,¹ M.B.B.S., M.D. and Lisbeth L. Jensen,^{1,2}* M.D.

How Reliable is Reported Sleeping Position in Cases of Unexpected Infant Death?

ABSTRACT: Examination of sudden infant death syndrome (SIDS) deaths in South Australia over a 7-year period from 2000 to 2006 was undertaken. There were 32 out of 35 cases where details of position when found were known. The data confirmed a marked decline in deaths in the prone position over the past decade, but showed no significant decline in cases reportedly found dead in the supine position. Posterior lividity was present in most cases (n = 30), 10 of whom also had anterior lividity. Posterior lividity was attributable either to the position of the body after death or to the effect of supine postmortem storage. In six cases, however, fixed anterior lividity indicated that death had occurred in the prone position in the side (n = 1) and in the supine position (n = 5). This contradiction indicates that caregivers' descriptions of terminal sleeping positions may not be supported by autopsy findings. The numbers of SIDS deaths reported in the supine position in South Australia may not, therefore, represent a genuine tally, but instead may be a function of inaccurate reporting. This may act as a confounding factor in studies attempting to link sleeping position with other risk factors.

KEYWORDS: forensic science, sudden infant death syndrome, sleeping position, prone, risk factors

There is a well-established link between an increased risk of sudden infant death syndrome (SIDS) and prone sleeping (1,2). Recently in South Australia the number of SIDS deaths has fallen dramatically, largely due to falls in the number of deaths in the prone (face down) position (3). Numbers of deaths in the supine (face up) position have remained relatively unchanged. To examine this trend and to evaluate the location of livor mortis, or lividity, as an autopsy marker of the position of the body after death the following study was undertaken.

Materials and Methods

Autopsy files at Forensic Science SA over a 7-year period from 2000 to 2006 were reviewed for all cases where death had been attributed to SIDS. Diagnoses were made using the NICHD and San Diego definitions (4,5) with full clinical history reviews, death scene, and autopsy examinations. Case details were reviewed and the position of the body when found and the distribution of lividity on skin surfaces were recorded. Autopsy photographs were examined in cases where there was a contradiction between the pattern of lividity and the reported position of the infant when found. No control group was utilized as the current study was evaluating the correlation between caregivers' histories and autopsy findings in a specific group of infants who had died of SIDS.

Background

Forensic Science SA provides autopsy services for the entire state population of approximately 1.5 million people. Over 95% of

¹Discipline of Pathology, The University of Adelaide, Frome Rd, Adelaide, SA 5005, Australia. ²University of Aarhus, Peter Sabroes Gade 15, DK-8000 Århus C,

²University of Aarhus, Peter Sabroes Gade 15, DK-8000 Århus C, Denmark.

Received 9 Sept. 2007; and in revised form 29 Dec. 2007; accepted 27 Jan. 2008.

the state's coronial autopsies are performed at the center, including all cases of unexpected infant death that proceed to postmortem. Once an infant has been found deceased at a home address full police and coronial investigations are initiated. Scene examinations are performed by Forensic Response Section police officers who liaise with pathologists. Trained police officers interview family members using a standard unexpected infant death proforma based on international guidelines. The scene, including the house, rooms, and bedrooms, with a particular focus on the bed or cot where the infant was found, are videotaped and photographed. On occasion a doll is used for re-enactments. Full autopsies are performed once a skeletal survey has been reported by a pediatric radiologist. Ancillary investigations including bacteriology, virology, and toxicology are performed, as well as vitreous humor sodium measurements and metabolic evaluation. Brains are retained for formal examination by a neuropathologist.

Results

All 35 cases underwent full police, coronial, and pathological investigations as detailed above. Out of 35 cases, caregivers were able to specify the position of the body when found in 32. Two cases where this information was not available were co-sleeping situations where uncertainty existed as to how the infants had been lying when found. The third case was an isolated rural family where information was incomplete. Infants were found on their sides in four cases, supine in 14 (44%), and prone in 14. Thirty infants had documented posterior dependent lividity, 10 with additional anterior lividity. One case each had only lateral and anterior lividity, respectively.

In six of the cases although the position of the bodies when found was recorded as "side" (n = 1) and "back" (n = 5), this did not accord with the autopsy findings. Specifically, in addition to posterior lividity in five of these cases, there was also fixed anterior lividity in all six. Fixed anterior lividity indicated that these infants had been lying face down for some time after death and had most

^{*}Dr. Lisbeth Jensen was a Visiting Research Fellow at The University of Adelaide sponsored by the Sudden Infant Death Research Foundation of South Australia.

likely died in that position, despite the claims of back and side positions when found.

These infants had been sleeping in their own cots (n = 2), on a mattress on the floor (n = 1), or co-sleeping with parents (n = 3). There were equal numbers of male and female infants (M:F = 1:1), with an age range from 4 to 28 weeks. All infants were in the care of their parents at the times of their deaths. Resuscitation had been attempted in three of the six cases. Postmortem intervals ranged from 7 to 71 h. There was no evidence of airway obstruction on scene examination or autopsy examination in any case. In addition there was no evidence of trauma or underlying organic diseases that could have caused or contributed to death. Ancillary testing (radiologic, microbiologic, biochemical, and metabolic) was unremarkable. Details of the cases are summarized in Table 1.

Discussion

Sudden infant death syndrome (SIDS) refers to the unexpected death of a previously well infant where death scene and autopsy investigations fail to reveal a cause of death (6–8). A variety of definitions have been proposed for SIDS, with the most recent being formulated in San Diego, U.S.A., in 2004 (5), based on the previous NICHD definition (4). According to the San Diego definition, SIDS is "the sudden and unexpected death of an infant under 1 year of age, with onset of the lethal 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" (5).

Numerous possible causes of SIDS have been proposed over the years, ranging from failure of autonomic control of respiration, to occult infections, gastroesophageal reflux, cardiac arrhythmias, anaphylaxis, and trace metal deficiencies; however, none have been proven (7,9). Research into central nervous system abnormalities has implicated reduced brainstem receptors and neurotransmitters, brainstem gliosis, and lowered levels of decosohexanoic acid (10–12). One of the major difficulties with SIDS is that there are no pathognomonic autopsy findings to prove the diagnosis; i.e., although thymic petechiae, pulmonary edema, and fluid blood are all encountered, they are in no way specific (7,13).

Another issue that arises is the difficulty in distinguishing between suffocation and SIDS. Infants may die in their beds and cots from a variety of causes that include wedging or hanging in dangerous cots, or being overlain by a sleeping adult (14,15). Infants being breastfed in bed are sometimes also at risk of smothering (16). The importance of the sleeping environment has been well recognized and formal death scene examination is now a required part of standard definitions (4,5). Guidelines for scene examination have been established by authorities such as the Centers for Disease Control and Prevention in the United States (17). A result of instituting formal death scene examinations has been an increase in diagnoses of cases of accidental asphyxiation (18,19).

 TABLE 1—Details of six infants who were allegedly found dead lying on their backs and sides.

Case	Age (weeks)	Sex	Reported Position	Expected Lividity	Actual Lividity
1	4	М	Supine	Posterior	Posterior + anterior
2	5	F	Supine	Posterior	Posterior + anterior
3	8	F	Supine	Posterior	Posterior + anterior
4	11	F	Supine	Posterior	Posterior + anterior
5	20	Μ	Side	Lateral	Anterior
6	28	Μ	Supine	Posterior	Posterior + anterior

Despite the lack of clarification of possible etiological pathways, epidemiological studies in the 1990s revealed a series of factors that increased the risk of SIDS including prone and side sleeping, cigarette smoke exposure, soft bedding, and overheating (2,6). It is now apparent that SIDS is not a single disease with one cause, but is instead a heterogeneous amalgam of underlying predispositions and external factors occurring at a vulnerable period in life, best conceptualized in the "fatal triangle" or "triple risk" models (20,21).

The recognition that prone sleeping position was associated with a significantly increased risk of SIDS has been one of the major achievements in the field of sudden infant death in the past 20 years. While this association was reported by Abramson in 1944 (22) it was Beal in South Australia (23) and de Jonge in the Netherlands (24) who were responsible for drawing attention to this link in more recent times. An increased incidence of SIDS was noted in the Netherlands after policies recommending prone sleeping position for infants were introduced, and once prone infant sleeping was discouraged the incidence of SIDS fell by 40% (24). Fleming et al. in the United Kingdom found an increased risk of SIDS of 8.8 times in infants who slept prone (1). Although one of the criticisms of the earlier studies was that retrospective analysis may have been skewed by recall bias (25), other studies have shown this not to be a major confounding factor (26,27).

In South Australia there has been a marked decline in numbers of SIDS deaths, falling from a peak in 1987 of 52 cases per year to less than five in recent years (3). While the decrease in numbers has been partly due to increased accuracy in the diagnosis of sleeping accidents and probable accidental asphyxia (19), with increases in numbers of cases deemed "undetermined," there is no doubt that there has been a genuine reduction in the incidence of SIDS related directly to the publicizing of risk factors by the "Reduce the Risks" campaign (7) with marked reduction in deaths in the prone position (28).

There are no pathognomonic pathological findings to assist with the diagnosis of SIDS, however, certain postmortem changes such as livor mortis may provide useful information. Livor mortis, or lividity, refers to pooling of blood in dependent parts of the body after death. It commences soon after circulation ceases and may change in location for a number of hours after death if a body is moved. After some time, usually hours, lividity becomes "fixed" and does not alter despite changes in body position. This may be useful in showing the position that a body was in after death, although lividity in most infants who die of SIDS (as in the present study) is usually found along the posterior surfaces of the body at autopsy as a result of the bodies having been placed on their backs soon after death (29).

The finding of fixed anterior lividity in the six cases investigated who were reportedly found on their side or back, however, indicated that they had been lying prone for some time after death, and not in the positions stated by caregivers. There may be a number of reasons for the disparity between the autopsy findings of fixed anterior lividity in these cases and the caregiver's history, including inaccurate recall or inadvertent movement of the body after death. This may occur during attempted resuscitation by both family members and ambulance officers. Another possibility may be an attempt to obscure the fact that an infant had been placed to sleep prone, as this may suggest that health warnings had not been heeded. If the desire to avoid such an accusation is sufficiently strong, caregivers may maintain that infants had been sleeping on their backs or sides, despite having been found prone. Certainly, back and side sleeping, which was claimed in six of the reported cases could not be supported by the autopsy findings. This was also noted in a recent study, where subsequent information in 12.4% of

cases contradicted the initial report of position at the time of discovery. It was considered that caregivers may not provide accurate histories because of feelings of guilt and embarrassment, and also because of concerns of possible child protection or legal issues (30).

It should also be noted that agreement between the caregiver's story and the autopsy findings does not necessarily validate the history, as patterns of lividity often change with body handling, i.e., anterior lividity from lying prone may disappear if the body position is changed soon after death (17). This also explains the presence of posterior, in addition to anterior, lividity in five of the six reported cases (Table 1), as posterior settling of blood had occurred once they had been placed on their backs after discovery and during their time awaiting autopsy.

Given the possibility of inaccurate reporting of infant positions, it appears that caregivers' descriptions of terminal sleeping position cannot always be accepted as completely reliable. This trend may have increased in recent years with the wide publicizing of the risks associated with prone sleeping. One result of this may be that deaths in the prone position may now be reported as supine. If this is so, then epidemiological studies relating reported infant positions to other factors may be faced with a significant confounding factor that may make subsequent interpretation of results difficult.

References

- Fleming PJ, Gilbert R, Azaz Y, Berry PJ, Rudd PT, Stewart A, et al. Interaction between bedding and sleeping position in the sudden infant death syndrome: a population based case-control study. Br Med J 1990;301:85–9.
- Mitchell EA, Tuohy PG, Brunt JM, Thompson JMD, Clements MS, Stewart AW, et al. Risk factors for sudden infant death syndrome following the prevention campaign in New Zealand: a prospective study. Pediatrics 1997;100:835–40.
- Tursan d'Espaignet E, Bulsara M, Wolfenden L, Byard RW, Stanley FJ. Trends in sudden infant death syndrome in Australia from 1980–2002. Forensic Sci Med Pathol 2008;4:83–90.
- Willinger M, James LS, Catz C. Defining the sudden infant death syndrome (SIDS): deliberations of an expert panel convened by the National Institute of Child Health and Human Development. Pediatr Pathol 1991;11:677–84.
- Krous HF, Beckwith JB, Byard RW, Rognum TO, Bajanowski T, Corey T, et al. Sudden infant death syndrome (SIDS) and unclassified sudden infant deaths (USID): a definitional and diagnostic approach. Pediatrics 2004;114:234–8.
- Moon RY, Horne RS, Hauck FR. Sudden infant death syndrome. Lancet 2007;370:1578–87.
- Byard RW, Krous HF. Sudden infant death syndrome—overview and update. Pediatr Dev Pathol 2003;6:112–27.
- Byard RW. Possible mechanisms responsible for the sudden infant death syndrome. J Paediatr Child Health 1991;27:147–57.
- Byard RW, Moore L, Bourne AJ, Lawrence AJ, Goldwater PN. *Clostridium botulinum* and sudden infant death syndrome: a 10 year prospective study. J Paediatr Child Health 1992;28:156–7.
- Kinney HC, Filiano JJ. Brain research in SIDS. In: Byard RW, Krous HF, editors. Sudden infant death syndrome, progress and possibilities. London: Arnold, 2001;118–37.

- Paterson DS, Trachtenberg FL, Thompson EG, Belliveau RA, Beggs AH, Darnall R, et al. Multiple serotonergic abnormalities in sudden infant death syndrome. JAMA 2006;296:2124–32.
- Byard RW, Makrides M, Need M, Neumann MA, Gibson RA. Sudden infant death syndrome: effect of breast and formula feeding on frontal cortex and brainstem lipid levels. J Paediatr Child Health 1995;31:14–6.
- Goldwater PN, Williams V, Bourne AJ, Byard RW. Sudden infant death syndrome—a possible clue to causation. Med J Aust 1990;153:59–60.
- Byard RW, Beal S, Bourne AJ. Potentially dangerous sleeping environments and accidental asphyxia in infancy and early childhood. Arch Dis Child 1994;71:497–500.
- Byard RW. Is co-sleeping in infancy a desirable or dangerous practice? J Paediatr Child Health 1994;30:198–9.
- Byard RW. Is breast feeding in bed always a safe practice? J Paediatr Child Health 1998;34:418–9.
- Centers for Disease Control, Prevention (CDC). Guidelines for death scene investigation of sudden unexplained infant deaths. Recommendations of the Interagency Panel on Sudden Infant Death Syndrome. Morbid Mort Week 1996;45:1–22.
- Byard RW, Beal SM. Has changing diagnostic preference been responsible for the recent fall in incidence of sudden infant death syndrome in South Australia? J Paediatr Child Health 1995;31:197–9.
- Mitchell E, Krous HF, Donald T, Byard RW. Changing trends in the diagnosis of sudden infant death. Am J Forensic Med Pathol 2000;21:311–4.
- Rognum TO, Saugstad OD. Biochemical and immunological studies in SIDS victims. Clues to understanding the death mechanism. Acta Paediatr 1993;389(Suppl.):82–5.
- Filiano JJ, Kinney HC. A perspective on neuropathologic findings in victims of the sudden infant death syndrome: the triple risk model. Biol Neonate 1994;65:194–7.
- Abramson H. Accidental mechanical suffocation in infants. J Pediatr 1944;25:404–13.
- Beal S. Sleeping position and sudden infant death syndrome. [letter] Med J Aust 1988;149:562.
- de Jonge GA, Engelberts AC. Cot deaths and sleeping position. [letter] Lancet 1989;ii:1149–50.
- 25. Southall D, Stebbens V, Samuels M. Bedding and sleeping position in the sudden infant death syndrome. [letter] Br Med J 1990;301:492.
- Drews CD, Kraus JF, Greenland S. Recall bias in a case-control study of sudden infant death syndrome. Int J Epidemiol 1990;19:405–11.
- Dwyer T, Ponsonby A-L, Newman NM, Gibbons LE. Prospective cohort study of prone sleeping position and sudden infant death syndrome. Lancet 1991;337:1244–7.
- Byard RW, Stewart WA, Beal SM. Pathological findings in SIDS infants found in the supine position compared to the prone. J SIDS Infant Mort 1996;1:45–50.
- Byard RW. Sudden infant death syndrome—a "diagnosis" in search of a disease. J Clin Forensic Med 1995;2:121–8.
- Pasquale-Styles MA, Tackitt PL, Schmidt CJ. Infant death scene investigation and the assessment of potential risk factors for asphyxia: a review of 209 sudden unexpected infant deaths. J Forensic Sci 2007;52:924–9.

Additional information and reprint requests: Roger W. Byard, M.B.B.S., M.D.

Discipline of Pathology

Level 3 Medical School North Building

The University of Adelaide, Frome Road

Adelaide, SA 5005

Australia

E-mail: byard.roger@saugov.sa.gov.au