Autopsy findings of co-sleeping-associated sudden unexpected deaths in infancy: Relationship between pathological features and asphyxial mode of death

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Aim: Co-sleeping is associated with increased risk of sudden unexpected death in infancy (SUDI)/sudden infant death syndrome (SIDS). The aim of this study is to examine autopsy findings from a single UK specialist centre to determine the relationship between co-sleeping and cause of death.

Methods: Retrospective analysis of >1500 paediatric autopsies carried out by paediatric pathologists over a 10-year period. SUDI was defined as sudden unexpected death of an infant aged 7–365 days; deaths were categorised into explained SUDI (cause of death was determined) and unexplained SUDI (equivalent to SIDS).

Results: There were 546 SUDI; sleeping arrangements were specifically recorded in 314; of these, 174 (55%) were co-sleeping-associated deaths. Almost two thirds (59%) of unexplained SUDI were co-sleeping compared to 44% explained SUDI (95% confidence interval (CI) 1.0–27.2%, P = 0.03); however, this difference remained statistically significant only for the first 5 months of life (95% CI 3.5–33.2%, P = 0.01). In unexplained SUDI aged <6 months, there were no significant differences between co-sleeping and non-co-sleeping deaths with respect to ante-mortem symptoms, intrathoracic petechiae, macroscopic lung appearances, pulmonary haemosiderin-laden macrophages, and isolation of specific bacterial pathogens; however, fresh intra-alveolar haemorrhage was reported more commonly in co-sleeping (54%) than in those that were not (38%; 95% CI 1.4–30.5%, P = 0.03).

Conclusions: Co-sleeping is associated with unexplained SUDI/SIDS in infants aged <6 months, suggesting that co-sleeping is related to the pathogenesis of sudden death in younger infants. The finding that intra-alveolar haemorrhage is more common in co-sleeping suggests that a minority of co-sleeping-associated deaths may be related to an asphyxial process.

Key words: autopsy; co-sleeping; infancy; SIDS; sudden unexpected death; SUDI.

Introduction

Sudden unexpected death in infancy (SUDI), defined as the sudden and unexpected death of an infant aged less than 1 year, comprises a heterogeneous group of infant deaths, including those in which a review of the death scene and complete post-mortem examination will disclose a cause of death (explained
SUDI), and deaths that will remain unexplained even after such investigations. The latter cases may also be classified as sudden infant death syndrome (SIDS) if the infant died during sleep.

Many risk factors have been identified for unexplained SUDI/SIDS, including prone sleeping,4–7 co-sleeping,8–14 and maternal smoking,8,15–17 as well as high ambient room temperatures, excessive clothing and/or bedding, head covering, preterm birth and/or intra-uterine growth restriction, multiple pregnancy, high parity, young maternal age and low socio-economic class.8,18–22 Previous studies have also consistently shown that use of pacifiers (soothers or dummies), room sharing without co-sleeping, and breastfeeding significantly reduce the risk of SIDS.21,23

Co-sleeping or bed sharing remains a controversial risk factor for SIDS; it is normal practice in many cultures, and despite recommendations to the contrary, it is still commonly practiced in the UK.24,25 However, there is strong epidemiological evidence that co-sleeping is associated with an increased risk of SIDS when there is a history of maternal smoking or where parents have consumed alcohol or taken drugs in the preceding 24 h.8,26 Although the actual number of co-sleeping-associated SIDS deaths has decreased in the UK, the proportion of unexplained SUDI/SIDS deaths associated with reported co-sleeping has increased from around 12% in the 1980s to almost 50% in 1999–200320 to 54% in 2003–2006.26 While there are many epidemiological studies on co-sleeping, autopsy-based data relating to such deaths remain limited. The aim of this study is to determine the prevalence and significance of co-sleeping in relation to cause of death in a large series of SUDI infants referred for autopsy to a single specialist centre in the UK and investigated to a common protocol.

Methods

This study is a retrospective review of 546 SUDI autopsies, reviewed as part of a larger series of >1500 consecutive paediatric autopsies performed over a 10-year period, 1996 to 2005, at a single specialist centre. SUDI was defined as the sudden and unexpected death of an infant aged 7 to 365 days. Local research ethics committee approval was obtained prior to the start of this project.

All post-mortem examinations were performed by specialist paediatric pathologists at a single centre. Clinical information regarding the circumstances of death was provided by Her Majesty’s Coroner (HMC) or the investigating police. A local autopsy protocol was followed that included the use of detailed ancillary investigations, which was modified slightly during the 10-year period, and comprised a full macroscopic post-mortem examination in addition to histological examination, post-mortem radiology, bacteriological and virological investigations, and metabolic studies, as well as other analyses, such as biochemical or toxicological investigations, in selected cases. The autopsies were entered into a specially designed Microsoft Access database; to ensure consistency for data analysis and interpretation, all data entry and data extractions were carried out by a single paediatric pathologist (MAW). Using strictly defined criteria, the final cause of death was classified as either explained SUDI or unexplained SUDI following review of the post-mortem findings (which included the macroscopic and histological findings, and the results of all ancillary investigations) and of the clinical history available to the pathologist at the time of the autopsy (including any given details of the death scene investigation). For the purposes of this study, co-sleeping was defined as the sharing of a sleeping surface between the infant and at least one adult or child during the infant’s last sleep. Statistical methods used included the comparison of proportion test and the Mann–Whitney U-test.

Results

During the 10-year study period (1996 to 2005 inclusive), a total of 1516 paediatric post-mortem examinations were performed in the centre; of these, 965 were infants under the age of 1 year, of which 546 met the criteria for SUDI (sudden unexpected death of an infant aged 7 to 365 days). In 202 (37%) infants, an identifiable cause of death was diagnosed following a detailed post-mortem examination (‘explained SUDI’); the other 344 (63%) deaths remained unexplained despite extensive post-mortem investigations (‘unexplained SUDI’).1 In 36 of the 546 SUDI, there was no information provided by HMC or the police about whether the infant had died during sleep. Of the remaining 510 SUDI, 361 (71%) were reported as having died during sleep, but in 47 of these, no specific information was provided regarding their precise sleeping arrangements during the last sleep. Of the remaining 314 infants, 174 (55%) were co-sleeping-associated deaths, while 140 (45%) were not.

There were significantly more deaths associated with sleep in the unexplained SUDI group (279 of 329 unexplained SUDI in whom the sleep status was recorded, 85%) compared to the explained SUDI group (82 of 181 infants in whom the sleep status was recorded, 45%; difference 39.5%, 95% confidence interval (CI) 31.1% to 47.5%, P < 0.0001; Fig. 1). In the unexplained SUDI group, in more than half of all sleep-associated deaths, the infant had been co-sleeping, usually with one or both parents (144 of 246, 59%), compared to only 44% in explained SUDI (30 of 68, 44%; difference 14.4%, 95% CI 1.0% to 27.2%, P = 0.03; Fig. 2).

Co-sleeping-associated deaths occurred at a significantly younger age in both explained and unexplained SUDI, likely reflecting that this sleeping practice is more common in younger infants (Fig. 3). However, co-sleeping-associated deaths were significantly more common in unexplained SUDI than explained SUDI in the first 5 months of life (132 of 201 (66%) unexplained SUDI vs. 24 of 51 (47%) explained SUDI; difference 18.6%, 95% CI 3.5% to 33.2%, P = 0.01), but not in older infants aged 6–12 months (12 of 45 (27%) unexplained SUDI vs. 6 of 17 (35%) explained SUDI; difference 8.6%, 95% CI –15.1% to 35.3%, P = 0.38; Fig. 4), suggesting that co-sleeping may play a role in the pathogenesis of unexplained SUDI in a proportion of deaths in the younger age group. Overall, 32 of all 174 (18%) co-sleeping associated deaths were reported to have co-slept on a sofa, 26 of which were in the unexplained SUDI group (26 of 144, 18%); 10 of the 26 (38%) were reported to show intra-alveolar haemorrhage at post-mortem.

Limiting further analysis to unexplained SUDI infants aged < 6 months (n = 201), there were no significant differences
between co-sleeping-associated deaths and non-co-sleeping deaths with respect to ante-mortem symptoms (as reported by parents), intrathoracic petechiae and macroscopic appearances of the lungs (as described by the pathologist at the time of the post-mortem examination), or pulmonary haemosiderin-laden macrophages and isolation of specific bacterial pathogens (Table 1). However, intra-alveolar haemorrhage was reported more commonly in unexplained SUDI that were co-sleeping (54%) than in unexplained SUDI that were not (38%; difference 16.5%, 95% CI –19.5% to 0.9%, \(P = 0.08\)).

**Discussion**

The results of this study show that co-sleeping is common, with more than half of all SUDI infants referred to our centre for autopsy having shared a sleeping surface with one or both parents during their last sleep. For both unexplained SUDI and explained SUDI deaths, co-sleeping was commoner in young infants; this notwithstanding, co-sleeping was found to be significantly more prevalent in unexplained SUDI compared to explained SUDI for infants under 6 months of age. These findings suggest that co-sleeping may be related to the pathogenesis of sudden unexpected death in around 20% of younger SUDI deaths. Significantly, of the unexplained SUDI deaths occurring during sleep in infants aged < 6 months, intra-alveolar haemorrhage was more prevalent in co-sleeping-associated deaths than in infants that were not co-sleeping, suggesting that a small proportion of co-sleeping-associated deaths may be related to asphyxia.

Co-sleeping remains a contentious risk factor for SIDS, despite epidemiological studies having shown an apparent association between co-sleeping and SIDS. The Confidential Enquiry into Stillbirths and Deaths in Infancy (CESDI) SUDI Studies demonstrated a significantly increased risk of SIDS for co-sleeping infants if at least one parent smoked (OR 12.35, 95% CI 7.41–20.59) or the mother had consumed more than two units of alcohol in the previous 24 h (OR 14.37 95% CI 4.66–44.29), but not for non-smoking parents (OR 1.08, 95% CI 0.45–2.58); these associations were recently confirmed in a subsequent UK-based study, which showed that co-sleeping significantly raised the risk of SIDS if parents had recently
consumed alcohol or drugs (odds ratio (OR) 14.34; 95% CI 3.78–78.76) but not if parents had neither used alcohol nor drugs (OR 2.27; 95% CI 0.93–5.57), suggesting that the increased risk of SIDS is not conferred by bed sharing per se but that the risk is modified by the circumstances in which co-sleeping occurs.26

The results of the current autopsy-based study are supported by several published epidemiological reports. A recent systematic review of 40 studies reporting on the association between SIDS and co-sleeping, including 30 case control studies and 10 prospective cohort studies conducted in 10 different countries, found an association between bed sharing and SIDS for younger infants, the strength of the association decreasing with increasing age.13 In a recent large multicentre German study published subsequent to the systematic review, bed sharing was associated with an increased risk of SIDS (multivariate OR 2.73, 95% CI 1.34–5.55), although the risk of SIDS remained significant only in younger infants less than 13 weeks old (multivariate OR 19.86, 95% CI 2.33–169.54).28 Similarly, in a study from Scotland, bed sharing was associated with an increased risk of SIDS in infants less than 11 weeks old (OR 10.20, 95% CI 2.99–34.80), and this association remained significant even when mothers did not smoke (OR 8.01, 95% CI 1.20–53.3).11 In a recent Dutch study of 138 SIDS deaths less than 6 months of age, bed sharing was found to be a significant risk factor for infants less than 4 months of age after adjusting for breastfeeding and parental smoking, but not for older infants.14 The consistent finding of an association between unexplained SUDI/SIDS and co-sleeping in younger infants is supported by the current and previous studies, suggesting that co-sleeping increases the risk of SIDS, particularly in younger infants.
infants regardless of smoking status,11,13,14,28 as also demonstrated in the present study, suggests that young infants may be intrinsically vulnerable to the risks of co-sleeping, even if the mechanism(s) involved remain unclear.

It is recognised that in a small proportion of co-sleeping infants, death may be caused by accidental asphyxia due to overlaying, but for the majority, the mechanism of death is poorly understood, with little direct evidence to support a diagnosis of asphyxia.29 That said, the diagnosis of asphyxia is notoriously difficult at post-mortem as there are no specific pathological findings.30 It has previously been suggested that moderate to severe degrees of pulmonary haemorrhage may be a marker of asphyxia due to either accidental or inflicted suffocation,13 although its recognised that intra-alveolar haemorrhage (IAH) per se is common in SIDS and reported in more than half of cases.31 One such study found that ‘significant’ IAH, which the authors arbitrarily defined as >5% of the total lung surface area assessed, was significantly more prevalent in SIDS infants that were co-sleeping/potentially overlain than in SIDS infants that were not.31 However, Berry33 observed that the majority of the co-sleeping deaths in that study occurred in younger infants, that younger age of death was associated with more extensive IAH, and that further work was required to ascertain whether young age or co-sleeping, or both, were independent variables resulting in increased IAH. The present study has also shown that IAH in infants < 6 months of age is more prevalent in co-sleeping than in infants that were not co-sleeping; while not specific, this further suggests that a proportion of otherwise non-suspicious co-sleeping-associated deaths in younger infants might be related to asphyxia, either as the cause of death, or occurring in combination with other factors.

The main reasons given by parents for bed sharing include the facilitation of breastfeeding, comforting an irritable infant and/or helping the infant or mother get better sleep.25,27 Previous studies have shown an increased association between co-sleeping and both the rate and the duration of breastfeeding, although this does not necessarily imply a causal relationship.13 In a recent longitudinal, population-based study in the UK, co-sleeping was associated with a significantly higher prevalence of breastfeeding in the first 15 months of life in the groups that bed shared in infancy and that bed shared constantly throughout the 4-year study period.27 It has been argued that co-sleeping represents a biologically and culturally evolved behaviour, as human infants, like other primate infants, require close physical contact between themselves and their mothers to regulate body temperature, breathing and other vital functions.14 However, the authors emphasise that this does not make co-sleeping inherently safe, and that it is the context and manner in which such practices are performed that make them safe or unsafe. Most epidemiological studies lack the complexity and/or statistical power to differentiate between these different (i.e. safe vs. unsafe) sleep practices, but the observation that there are many cultures where bed sharing is commonly practiced but which carry a low incidence of SIDS, such as Japan and Hong Kong, supports the notion that other co-factors may be important in mediating the risk of SIDS associated with co-sleeping.24

The high proportion of co-sleeping deaths on a sofa (18%) in the present study is especially worrisome, and highlights an ongoing need for further public awareness campaigns. Co-sleeping with an infant on a sofa has been shown to carry an almost 23 times greater risk of SIDS than infants not co-sleeping (OR 22.05, 95% CI 6.52–74.56, compared with an OR of 2.07, 95% CI 1.46–2.95, for infants found in the parental bed),6 similar data are reported in other studies.6,11,12,26 The reason for this association remains unclear, although it is possible that co-sleeping on a sofa increases the risk of accidental asphyxia.15

The main limitations in the present study relate to the absence of reliable information about other associated risk factors for
SID, such as parental smoking or alcohol use. This is an inherent issue with any retrospective autopsy-based study, since the clinical details are dependent on the information supplied by HMC and/or police at the time of the autopsy. The major strengths of this study relate to its large sample size, this being the largest single-centre pathology study to date in which all SUDI autopsies were carried out by a small number of specialist paediatric pathologists to a common autopsy protocol, and the use of an appropriate post-mortem comparison group (the explained SUDI group) to determine the significance of differences in the frequency of co-sleeping in relation to cause of death. Moreover, this is the first time that specific autopsy findings have been compared between co-sleeping and non-co-sleeping unexplained SUDI/SIDS infants, demonstrating that there are no specific pathological findings at post-mortem to determine mode of death in this setting.

In conclusion, while co-sleeping is common, it is associated with unexplained SUDI in infants aged < 6 months. While there is evidence to suggest that other co-factors are required while bed sharing to confer the increased risk of SIDS, our data support previous findings that co-sleeping may be related to the pathogenesis of sudden unexpected death in younger infants. The finding that IAH is more commonly reported in co-sleeping-associated deaths than in age-matched infants that were not co-sleeping suggests that a proportion of co-sleeping-associated deaths may be related to asphyxia.

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