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TITLE PAGE

RECURRENT SUDDEN UNEXPECTED DEATH IN INFANCY: A CASE SERIES OF SIBLING DEATHS

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ABSTRACT

Objectives

To determine the Sudden Unexpected Death in Infancy (SUDI) rate for infants born after a previous SUDI in the same family; and to establish the causes of death and frequency of child protection concerns in families with recurrent SUDI.

Design

Observational Study using clinical case records

Setting

The United Kingdom's Care of Next Infant (CONI) programme which provides additional care to families who have experienced a (SUDI) with their subsequent children.

Patients

Infants registered on CONI between January 2000 and December 2015

Main outcome measures

Cause of death, presence of modifiable risk factors for SUDI and child protection concerns.

Results

There were 6608 live-born infants registered on CONI with 29 deaths. 26 families had 2 deaths, 3 families had 3 deaths.

The SUDI rate for infants born after one SUDI is 3.93 (95% CI 2.7 to 5.8) per 1000 live births.

Cause of death was unexplained for 19 first and 15 CONI deaths. Accidental asphyxia accounted for 2 first and 6 CONI deaths; medical causes for 3 first and 4 CONI deaths; homicide for 2 first and 4 CONI deaths.

10 families had child protection concerns

Conclusions

The SUDI rate for siblings is ten times higher than the current UK SUDI rate. Homicide presenting as recurrent SUDI is very rare. Many parents continued to smoke and exposed infants to hazardous co-sleeping situations with these directly leading to or contributing to the death of 6 siblings. SUDI parents need support to improve parenting skills and reduce risk to subsequent infants.

Keywords

Sudden Unexpected Death in Infancy

Sudden Infant Death Syndrome

Cause of death

Child Protection

INTRODUCTION

Each year in England and Wales 300-400 infants die suddenly and unexpectedly¹. These deaths are referred to as Sudden Unexpected Death in Infancy (SUDI) which is defined as the death of an infant that was not predicted as a possibility in the 48 hours prior to the death or the collapse that led to death². Following investigation many SUDI remain unexplained and labelled as Sudden Infant Death Syndrome (SIDS)³. There were 183 unexplained infant deaths in England and Wales in 2017⁴. There is considerable variation internationally in the classification of these deaths, which may be categorised as SIDS, unascertained or accidental asphyxia⁵. Current SIDS understanding suggests a complex interplay between intrinsic vulnerability, a critical period of homeostatic development, and exogenous stressors^{6,7} including infection^{8,9}. Modifiable risk factors for SUDI include non-supine sleeping, parental smoking, and co-sleeping on a sofa or bed with parents who smoke or have consumed alcohol¹⁰.

After SUDI, parents are understandably anxious about having further children. The Care of Next Infant programme (CONI), was established in 1988 to assist families with infants born after SUDI, by supporting an enhanced health visitor service. CONI is co-ordinated nationally by CONI Headquarters (CONI HQ) currently at the Lullaby Trust. Families receive regular home visits by their health visitor until the infant is at least 6 months and are provided with apnoea monitors, Basic Life Support Training, symptom diaries and weight charts. CONI is supported by a steering group of specialist paediatricians, paediatric pathologists and medical statisticians. CONI is an optional programme; although exceptionally families have been required to register as part of child protection plans. CONI is widely available in England, Wales, Northern Ireland, the Crown Dependencies and British Forces overseas.

CONI HQ maintains a database of all registered infants. Mothers complete registration documents antenatally detailing maternal age, parity, estimated date of delivery and smoking status;

registration is for individual infants not families, mothers need to re-register for CONI with each new child. Clinical details about the cause and circumstances of death for the child that led to the registration on CONI are not requested unless there is a subsequent infant death. In the event of a CONI SUDI the family is offered a detailed case review after all local investigations are complete including the Coroner's Inquest and any legal cases. It is typically at least one year after the death before CONI HQ makes contact with bereaved parents.

The detailed case review by CONI specialist clinicians may include meeting the health visitor and GP, and a family visit to obtain a detailed medical history and account of events for all deaths. Parents are offered a second opinion on any pathology slides from the CONI steering group pathologist. If parents do not wish to have the clinical service from CONI HQ, the cause of death as detailed by the local clinical team is recorded and no further action taken. As a result of this CONI HQ has one of the most complete collections of case records relating to cases of repeat SUDI worldwide.

A common concern of parents after sudden infant death is 'Will it happen again?' Any increased risk of SUDI in siblings is likely to reflect genetic vulnerability as well as similar environmental risk factors. The only systematic review of recurrence risk in SIDS was based on data published between 1970 and 2005 suggesting an increased but unquantified risk¹¹. Previous research by CONI¹² showed most repeat deaths are due to natural causes but some may be unnatural, raising concerns for professionals managing such families. However, there is no consensus on which proportion of repeat deaths may be unnatural^{13 14} and much of the increased recurrence risk relates to parental smoking, maternal age and social deprivation¹⁵. Since this last research the standard of UK SUDI investigation has improved considerably¹⁶ enabling a cause for death to be determined in more cases. The incidence of SUDI has declined dramatically with remaining cases occurring more frequently in socially deprived families¹⁷; so we aimed to readdress the issue of recurrence risk to better inform parents and professional practice.

The objectives for this study were:

1. To determine the SUDI rate for infants born after a previous SUDI in the same family
2. To establish the causes of death in families with recurrent SUDI
3. To determine the frequency of child protection concerns in families with recurrent SUDI

METHODS

This is an observational study of deaths of infants registered on CONI analysing routinely collected clinical data, reported using STROBE guidelines¹⁸.

Setting

The UK CONI programme between 01 January 2000 and 31 December 2015.

Participants

All infants born and enrolled in CONI between 01 January 2000 and 31 December 2015.

All CONI registered infants dying unexpectedly in the first year of life, with deaths occurring between 01 January 2000 and 31 December 2015.

'Index death' refers to the first infant death in the family that led to the enrolment on CONI

'CONI death' refers to the unexpected death of an infant in the first year of life enrolled on CONI

Data sources

For CONI deaths, the data sources were the CONI clinical records. These included CONI registration forms, correspondence from local clinical teams, any detailed case reviews if conducted by CONI specialist clinicians, and press reports of court cases. Registration forms were completed by mothers antenatally and included brief details of the index death and smoking habits. For surviving infants the only data source was the CONI registration form.

Each CONI clinical record was assessed for the quality of information available for index and CONI deaths. The records were reviewed by a pair of researchers to determine the cause of death and relevant risk factors using a standard template. The file and completed template was then reviewed

by a second pair of researchers to ensure consistency (JG /AM or CDW /AW). As inconsistency in categorisation of causes of death in SUDI is common¹⁹ with coroners often solely relying on pathologists findings for the cause of death²⁰ and pathologists reticence to use the term SIDS²¹, we determined the cause of death based on information in the CONI clinical record. The definitions we used for each cause of death are detailed in table 1; we required detailed information on the circumstances of death to categorise deaths as due to accidental asphyxia.

Details of risk factors are shown in table 2.

Due to inadequate information we did not consider intrinsic risk factors such as low birth weight or prematurity.

Statistical methods

Data on the quality of clinical information, risk factors and cause of death were entered into an Excel 2010 spreadsheet, and descriptive statistics used for analysis. The 95% confidence intervals used the Wilson score method in Open-Epi²².

Ethics

This is a long-standing service evaluation so does not require ethical approval. All mothers gave consent to share information with CONI at registration. Following a death, parents are asked by their local healthcare team to consent to share information with CONI.

RESULTS

There were 6608 live-born infants registered on CONI in the years 2000-15, 171 were first born infants to mothers whose male partners had previously had an unexplained infant death. There were no data available on the number of eligible families whose chose not to register with CONI.

29 unexpected infant deaths following the index death occurred in 26 families, 23 with two deaths and three with three deaths. All CONI deaths occurred in families where index deaths had the same mother. This gives a SUDI rate for infants born in families with one previous SUDI of 3.93 (26/6608, 95% CI 2.7 to 5.8) per 1000 live births and the risk of a third death in a family with two previous SUDI of 115 (3/26, 95% CI 40 to 290) per 1000 live births.

If only unexplained CONI deaths (SIDS or unascertained) are included this gives an unexplained infant death rate for infants born in families with a previous SUDI of 2.27 (15/6608, 95% CI 1.4 to 3.7) per 1000 live births.

Detailed clinical information was available for 18/26 (69%) index cases and 25/29 (86%) CONI cases, as shown in table 3.

Causes for death

The causes for death are shown in figure 1. All unascertained deaths were classified as such due to inadequate information. In 12 families, index deaths and CONI deaths were both classified as unexplained (SIDS or unascertained).

Three index deaths had medical causes identified after the CONI death, one as a result of CONI specialist clinician review. Causes included metabolic, neuro-muscular and respiratory conditions.

Three CONI infants died of medical causes after unexplained index deaths.

We classified six deaths as accidental asphyxia, in two cases this was the certified cause of death but four were certified as SIDS or unascertained. Two families each lost two infants from accidental asphyxia. Three asphyxial CONI deaths occurred following unexplained index deaths, and one after a medical death.

Homicides and probable homicides

There were six homicides or probable homicides, two index and four CONI deaths, in three families.

One mother was convicted of infanticide of both infants. There were suspicions of deliberate asphyxia of three infants who died in the care of the same parent. All three had marked pulmonary

haemorrhage on post-mortem examination with two infants dying in apparently safe-sleep environments. There were long-standing child protection concerns with this family all siblings were permanently removed from parental care by the Family Court, although no criminal charges were brought relating to the deaths. One father was convicted following a CONI death from non-accidental head injury but he was not involved in the family at the time of the index death from SIDS.

Families with three deaths

There were three families who lost three infants; in one family these were all probable homicides, in one they were all unascertained, and in one all were classified as SIDS; these infants all had similar hippocampal malformations potentially suggestive of a genetic syndrome.

Risk factors

There was a high prevalence of risk factors in all cases reflecting the extreme vulnerability of infants. Risk factors appeared more common in CONI cases but this was due to more information being available for CONI cases.

Table 4 compares risk factors in Index and CONI deaths.

Table 5 shows risk factors in each category of death.

3059/6579 (47%) of mothers who did not have a further infant death smoked antenatally.

DISCUSSION

This study found that repeat SUDI within families are rare, with only 29 deaths reported in 26 families over a 16 year period. However, the repeat SUDI rate of 3.93 per 1000 live births is more than ten times higher than the UK rate of unexplained infant deaths of 0.31 per 1000 in 2016, and more than nine times higher than the rate of 0.43 per 1000 in 2006²³. Covert homicide affected two families with suspicions only occurring after subsequent deaths. Concerns regarding child abuse or poor parenting occurred in more than half of CONI deaths. Accidental asphyxia accounted for six

CONI deaths, with index deaths from accidental asphyxia in two of these families. Most mothers smoked in pregnancy and a third of CONI cases died in hazardous co-sleeping situations.

CONI HQ has maintained a detailed case registry since its inception in 1988, and is accessible to the majority of families after SUDI. This is therefore one of the most complete case series of sibling SUDI deaths available. The CONI scheme is a voluntary programme so we would not be aware of further SUDI cases in unregistered families potentially leading to significant bias of the results. CONI recruitment is largely through maternity services and although open to paternal half-siblings these are probably under-represented thus deaths in these families may not have been included. Families with child protection concerns or following a concealed homicide, may be less likely to register with CONI to avoid professional scrutiny, leading to under-reporting of child protection risks or homicides. Some high risk families have been required to register with CONI as part of Child Protection Plans. Familial homicide cases are usually reported in the national press, and we are not aware of any sibling SUDI homicides that were not registered on CONI. It is possible that the repeat SUDI rate we determined is actually an under estimation due to higher risk families not enrolling in CONI but in the study period there was no alternative data source for unenrolled families for comparison. As detailed case review is only offered to families after a death on CONI, we had minimal information on risk factors in families with only one death; this was therefore an uncontrolled observational study. We were further limited by the lack of information on intrinsic risk factors such as low birth weight and pre-term birth for many deaths, so could not determine what role these played in repeat deaths. Given these significant limitations, we cannot make any conclusion about the effectiveness of the CONI programme in addressing modifiable risk factors for SUDI.

We were able to obtain detailed case information for the majority of deaths, although only a minority took part in detailed interviews with CONI specialist clinicians. Since the publication of the 2004 Kennedy SUDI Guidelines¹⁶, the quality of local clinical investigations has improved

considerably, with cases since 2008 subject to thorough review by Child Death Overview Panels. Some CONI deaths had joint home visits with death scene analysis by police and paediatricians and local case reviews, greatly improving the clinical information available compared with the previous CONI study¹³. Although few deaths that we categorised as accidental asphyxia were officially certified as such, we are confident of our classification. Accidental asphyxia is recognised much less often in the UK compared to other developed countries with similar child care practices such as New Zealand or the USA⁵, and cases are misdiagnosed despite detailed child death review²⁰. The cases we labelled as asphyxial had detailed descriptions of infants being found under parents or siblings, or co-sleeping on sofas with intoxicated adults. We acknowledge that this may over-diagnose accidental asphyxia as possibly an adult could overlay an infant after they have already died of natural causes; differentiating between SIDS and accidental asphyxia is challenging given this relies on parental accounts and scene examination as pathological findings are often insignificant²⁴ and not diagnostic²⁵.

Our findings are similar to the previous CONI study¹³, showing an increased risk of SUDI in siblings with familial SUDI homicides being rare. Although there were detailed data available for the study it is still possible that some homicide cases were missed in unregistered families. Deaths may have been wrongly classified as unexplained due to inadequate information, or deaths due to accidental asphyxia actually being deliberately inflicted. In ten CONI deaths child abuse or neglect was a factor, four of these were homicides or probable homicides. In the other six there were significant concerns about parental care and parental decision making; these child protection concerns may or may not have contributed to deaths, but do show the vulnerability of CONI families.

This study highlights the need for comprehensive investigation of all SUDI, including detailed medical and social histories from parents, examination of the scene of death by an experienced healthcare professional, post-mortem examination and multi-disciplinary case discussion to determine the final cause of death. Some SUDI cases in this study appeared to be investigated inadequately; this issue is

a persistent problem in England despite the mandatory requirement for multi-agency investigation²⁶. Specialist review of deaths by the CONI team led to new medical diagnoses in three families. Other families appreciated the reassurance of thorough case reviews despite no new diagnoses being reached. Deaths from accidental asphyxia appear to be under-recognised and labelled as 'unascertained deaths' or SIDS instead. Although the term SIDS can include deaths where 'mechanical asphyxia or suffocation caused by overlaying has not been determined with certainty'³ we contend that it is unhelpful to include deaths with a very significant likelihood of asphyxia in this category. This lack of recognition of deaths that are likely due to accidental asphyxia limits learning and attempts at prevention both within families after a first SUDI, and in wider public health campaigns. An unascertained death by its lack of explanation is either viewed as unpreventable or the role of modifiable factors such as co-sleeping minimised.

Although our findings should provide reassurance to professionals dealing with a sibling SUDI case about the small possibility of homicide, it is important not to be complacent. The risk of repeat SUDI in a family is ten times that of the general population reflecting both inherent genetic risks as well as environmental factors such as maternal smoking and unsafe sleeping. CONI cannot address intrinsic risk factors, but these are very vulnerable families who need comprehensive care and support packages to help them understand safe-sleeping, address mental health problems and enhance their parenting capacity. Some families struggled to cope after the index death, using alcohol and illicit drugs putting subsequently born infants at greater risk. A second SUDI in a family is a tragedy, but a second accidental asphyxia death should be entirely avoidable and it is concerning that these occur despite CONI. Detailed multi-agency investigation of all SUDI, with recognition of accidental asphyxia deaths should enable CONI professionals to sensitively support families with subsequent infants and challenge parents when safe sleep practices are not followed. Unfortunately, cuts to health services have reduced the provision of CONI, and in some areas this is no longer available.

As SUDI rates continue to fall internationally, familial SUDI cases are becoming much rarer and it is more difficult to maintain a case registry such as CONI. A new National Child Mortality Database²⁷ has recently been set up in England collecting data following detailed Child Death Review procedures for all child deaths²⁸. This will allow accurate identification of all such cases in future, enabling greater understanding of risks within families.

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Contributors

JG, CDW, AM, and AW contributed to study design. JG, CDW, MC, RC, AM, AMK, and AW contributed to data collection and study implementation. JG, AM, CDW, MJC and AW did the analyses. JG, CDW, AM, and AW wrote the manuscript.

Competing Interests

All authors are members of the CONI national steering group managed by the Lullaby Trust. AW was funded by the Lullaby Trust as national CONI coordinator from 1988 to 2015 and employed by them from 2015-18. AMK was funded by The Foundation for the Study of Infant Deaths/The Lullaby Trust between 1988 and 2013. All other authors received expenses only from the Lullaby Trust towards their CONI work but no salary. JG has received research grants from the National Institute of Health Research (DRF 2010-0345)

All other authors declare no competing interests.

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What is already known on this topic?

Previous research has shown an increased risk of SUDI in siblings but it has not been possible to quantify this risk.

Some sibling SUDI cases may be homicides and child protection concerns in SUDI families are common.

No original data on sibling SUDI cases have been published since 2005.

What this study adds

This study shows a 10 fold increased risk of SUDI in siblings compared to the overall UK SUDI rate.

Homicide is a rare cause for sibling SUDI but child protection concerns are common and professionals may need to take action to safeguard infants.

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Tables and figures

Table 1 Classification of causes for death

Category	Description
Homicide or probable homicide	Cases where parents had been found guilty in criminal courts of murder or infanticide, or cases where there had been strong suspicion of homicide and family courts had taken action to protect other siblings.
Accidental asphyxia	Asphyxia was considered probable in cases where both the autopsy findings and the circumstances of death were supportive based on reports of sleep scenes. Circumstances supportive of asphyxia included: infants found under parents, or at the bottom of parents' beds under bedding, or if there were other significant suffocation hazards, or evidence of entrapment or wedging. Autopsy findings may be inconclusive but can include facial petechiae or pulmonary haemorrhage ²⁰ .
SIDS	Cases meeting the San Diego definition of SIDS categories IA, IB and II (including those with the possibility of accidental asphyxia) ³ where the results of complete investigations were available in the case file, this must have included a death scene examination, post-mortem examination, medical history and case review.
Medical causes of death	Cases with full medical cause for death on post-mortem or coroners' reports.
Unascertained	Cases where there was inadequate information in the case file with which to make a judgement.

Table 2 Risk factors for SUDI

Risk Factor	Definition
Maternal smoking	Maternal smoking either antenatally or post-natally
Unsafe co-sleeping	Co-sleeping on sofa at time of death Co-sleeping in bed at time of death, if carer smoked, had consumed alcohol or illicit drugs in preceding 24 hours
Maternal mental health problems	Maternal mental health problems at time of index death or CONI death
Poor parenting	Documented concerns about parenting capacity by health or social care professionals at time of index or CONI death Research team concerns about parenting capacity based on case file information, including cases where parents co-slept with infants having

	consumed more than two units alcohol or used illicit drugs.
Child abuse or neglect	Death due to homicide or probable homicide Index case or CONI case on child protection plan at time of death Siblings placed on Child Protection Plan, or removed from maternal care after index or CONI death.

Table 3 Quality of clinical information for index and CONI deaths

Category	Index death	CONI death
1 CONI registration form only	7	2
2 CONI registration form plus press report of court case	1	2
3 CONI registration form plus clinical correspondence detailing registered cause of death	7	12
4 As category 3 plus detailed clinical summaries from health visitors, general practitioners and paediatricians	2	2
5 As category 4 plus interview with family	1	2
6 As category 5 plus specialist pathology review of tissue samples	8	9

Table 4 Comparison of risk factors for Index and CONI deaths

	Index death (n=26)	CONI death (n=29)		
Risk Factor				
Maternal smoking in pregnancy		Smoking CONI death	Non-smoking CONI death	Smoking unknown CONI death
	Smoking index death	17	0	0
	Non-smoking index death	2	5	0
	Smoking unknown index death	5	0	0
Hazardous co-sleeping		Co-sleeping CONI death	Solo sleeping CONI death	Sleep unknown CONI death
	Co-sleeping Index death	7	4	0
	Solo sleeping index death	2	3	1
	Sleep unknown index death	2	1	9
Maternal mental health (MH) problems		MH problems CONI death	No MH problems CONI death	MH problems unknown CONI death
	MH problems index death	6	0	0

antenatally or postnatally	No MH problems index death	3	6	0
	MH problems unknown index death	4	2	8
Parenting Concerns		Parenting concerns CONI death	No parenting concerns CONI death	Parenting concerns unknown CONI death
	Parenting concerns index death	5	0	0
	No parenting concerns index death	7	8	0
	Parenting concerns unknown index death	5	0	4
Child abuse or neglect		Abuse or neglect CONI death	No abuse or neglect CONI death	Abuse or neglect unknown CONI death
	Abuse or neglect index death	4	1	0
	No abuse or neglect index death	3	15	0
	Abuse or neglect unknown index death	3	0	3

Table 5 Risk factors for each category of death

		SIDS/ unascertained			Accidental asphyxia			Medical cause			Homicide/ probable homicide		
		Yes	No	Un- known	Yes	No	Un- known	Yes	No	Un- known	Yes	No	Un- known
Maternal smoking in pregnancy	Index death	12	4	3	2	0	0	0	1	2	1	0	1
	CONI death	11	4	0	6	0	0	3	1	0	4	0	0
Hazardous co-sleeping	Index death	6	5	8	2	0	0	1	0	2	1	0	1
	CONI death	11	4	0	6	0	0	1	1	2	0	3	1
Maternal	Index	5	3	11	0	2	0	0	1	2	1	1	0

mental health problems antenatally or postnatally	death												
	CONI death	4	4	7	5	1	0	2	1	1	2	2	0
Parenting concerns	Index death	3	10	6	1	0	1	0	1	2	1	1	0
	CONI death	5	7	3	6	0	0	2	1	1	3	1	0
Child abuse or neglect	Index death	2	13	4	0	2	0	0	1	2	1	1	0
	CONI death	3	10	2	1	5	0	2	1	1	3	1	0