Uncertainty in classification of repeat sudden unexpected infant deaths in Care of the Next Infant programme

It is misleading to classify every unexplained infant death as natural if no unnatural cause has been established, argue **C J Bacon** and **E N Hey**

Sudden unexpected infant death (cot death) has become much less common in recent years, and it is rare for a family to experience two such deaths. Carpenter and colleagues recently published valuable data on the repeat deaths that occurred among 5229 families in the Care of the Next Infant programme.¹ This voluntary scheme funded by the Foundation for the Study of Infant Deaths provides extra support to families in England and Wales who have had a cot death and now have a new baby. There were 48 sudden unexpected deaths among babies on the programme between 1988 and 1999, including two third deaths. After examining all the circumstances and postmortem findings of the 46 second deaths, the authors concluded that all but six of the babies died from natural causes. This contrasts with earlier studies by Emery² and Wolkind and colleagues,³ which concluded that a much higher proportion of repeat cot deaths were probably homicides.

Apart from two short letters,^{4 5} Carpenter and colleagues' report was initially unchallenged. It has proved very influential, being accepted by bodies such as the American Academy of Pediatrics.⁶ More recently, however, Gornall published a wide ranging critique of how the authors presented their data.⁷ The authors defended their classification,⁸ but we believe their dichotomy of the deaths into natural or unnatural is particularly open to criticism. Experience in child protection teaches that it is often impossible to determine whether the parents have been in some way and to some degree responsible for the unexplained death of their baby.

In this article we show how many of the second deaths might be more appropriately categorised as "undetermined" rather than "natural." This approach is now favoured by many pathologists when reporting on unexplained infant deaths.⁹

Analysis

Carpenter and colleagues divided families into groups with similar patterns of first and second deaths (table). We have not attempted to examine the first deaths because the published details are insufficient. From the information given in the paper we have reassessed the 46 second deaths to see how many might reasonably be regarded as undetermined. We take "natural

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deaths" to be deaths arising from disease or a wholly accidental event, "unnatural deaths" to be deaths due to homicide (murder, infanticide, or manslaughter), and "covert homicide" to be an unnatural death that was not initially recognised as such.

Classification

Second death homicide

In six families the authors classified the second death as unnatural. This was obvious in four cases: in one there was a serious head injury and a criminal conviction; in the other three there was inflicted injury but nobody was prosecuted, either because it was unclear who was responsible or because the perpetrator was a juvenile. In the other two cases homicide was not initially recognised, despite the presence of fractured ribs. A family court later decided one death was homicide. The other was initially attributed to sudden infant death syndrome but was reclassified as homicide after the father confessed to suffocating another child and was convicted of the murder of all three babies. Thus in all the six deaths that the authors classified as unnatural, either a court pronounced the death to be homicide or the evidence of homicide was extremely strong.

Original and alternative assessment of 46 second sudden unexpected infant deaths

Group of cases	Original assessment		Alternative assessment		
	Natural	Unnatural	Probably natural	Undetermined	Probably unnatural
Second death homicide		6			6
Both deaths explained	3		2	1	
One explained and one unexplained death	6		6*		
Both deaths unexplained	18		12†	6†	
Information incomplete because:					
Further review was declined	7			7	
Mother was murdered	1			1	
Case was under legal review	5			5	
Total (%)	40 (87%)	6 (13%)	20 (43%)	20 (43%)	6 (13%)

*Re-assessment not possible because the original paper provides no information on these cases.

†Numbers allotted to each category are arbitrary (for explanation see text).

Both deaths explained

In three families both deaths were attributed to specific natural causes: a familial metabolic defect in both babies, long QT syndrome in both babies, and pneumonia in the first baby followed by endomyocardial fibroelastosis in the second. Postmortem examination of the second baby whose death was ascribed to metabolic defect showed rib fractures, which were attributed to resuscitation. This was one of four cases in the study in which, despite the presence of fractured ribs, death was initially thought to be natural (although two of these, not including the metabolic defect case, were later reclassified as unnatural). A recent literature review shows that resuscitation very rarely breaks children's ribs and when it does the site is always anterior.¹⁰ Most rib fractures in babies are posterior and indicate abuse, as may some anterior fractures. This suggests that when rib fractures are found in addition to a natural disorder, the natural disorder cannot be assumed to be the cause of death. However, in the absence of details of the site of the fractures this particular case must remain categorised as natural. As regards the second baby whose death was attributed to long QT syndrome, at the inquest into the child's death the coroner recorded an open verdict, which indicates that he did not consider the evidence sufficient to attribute the death to natural causes.¹¹ On this basis, we have recategorised this death as undetermined.

One explained and one unexplained death

In six families one death was attributed to a specific natural cause and the other to sudden infant death syndrome. The authors gave no additional information so we cannot review the categorisation of these cases.

Both deaths unexplained

In 18 families both deaths were attributed to sudden infant death syndrome. The authors state that all these families were at high risk of cot death and that the second deaths exhibited many untoward features: violent family relationships (9 cases), pathology findings suggestive of asphyxia (9), parental mental health problems (6), concern about the welfare of a subsequent baby (4), imprisonment of the father (3), previous apparent life threatening events (3), deafness of both parents (2), open coroner's verdict (2), failure to thrive (1), and abuse of a previous child (1).

One of the difficulties in distinguishing between sudden infant death syndrome and covert homicide is that both tend to occur against a background of social disadvantage.^{12 13} The features reported in these families could have been associated with either. We do not suggest that violence in the family, for example, necessarily implies that a baby's death was unnatural; but we think that when the family has two unexplained deaths this possibility at least has to be considered and may sometimes be true. To most observers the number of untoward features in these 18 cases would seem disproportionate. Since the available information does not allow individual assessment of each case, we have

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resorted to the arbitrary estimate that a third of them might be classed as undetermined.

Many of the deaths in this group had features of asphyxia. This raises the questions of whether some should have been attributed to unintentional harm rather than to sudden infant death syndrome, and whether in some the circumstances were such that they might have been regarded as unnatural—as two of the authors had previously suggested.¹⁴

Another unusual finding was successive unexplained deaths of babies in two families in which both parents were profoundly deaf. Such a concurrence of extremely rare events is unlikely to have resulted from chance alone and raises the question of whether the deaths were in some way related to the deafness or an associated disorder.^{15 16} One of these, long QT syndrome, was considered but discounted by Carpenter and colleagues and is not supported by a recent genetic study.¹⁷ Whatever the explanation, this finding is important and suggests that babies of profoundly deaf parents should receive extra surveillance.

Information incomplete

Information on the second death was incomplete for 13 families. In one of these the mother was murdered. In five families further inquiry was not attempted because the police had become involved. In two of these a parent faced prosecution (though neither finally resulted in conviction), and in three the other children were taken into care. The authors classified these five deaths as natural, apparently because they regarded judicial decisions or decisions not to prosecute as equivalent to medical diagnoses. We do not think this is justified. In criminal trials the jury must be persuaded beyond reasonable doubt that the defendant is guilty, and the Crown Prosecution Service recommends prosecution only when it considers that a court "is more likely than not to convict" and that prosecution is in the public interest.¹⁸ Failure to reach the high level of proof required in criminal proceedings does not mean that a possible cause of death has been excluded by the normal standards of medical diagnosis. We therefore categorise these five cases as undetermined.

In another seven families further inquiry was declined. The second baby to die in one family had two fractured ribs and a healing fracture of the clavicle, and a third death in another family resulted in all the remaining children being taken into care. Four of the babies in these families died while sleeping with a parent on a sofa. In the absence of more complete information, classification of all these deaths as natural seems unwarranted. The authors did, however, acknowledge the information was insufficient to enable them to distinguish between sudden infant death syndrome and a specific natural cause. Since distinction between sudden infant death syndrome and covert homicide can be equally difficult, it seems illogical to exclude covert homicide when there is not enough information to exclude a specific natural cause.

Discussion

The table summarises how an alternative analysis of these second deaths might categorise 13% as probably unnatural, 43% as probably natural (although this includes the six cases we could not review), and 43% as undetermined. This contrasts with the assessment by Carpenter and colleagues that 87% of the deaths were natural. But it is closer to the findings of Emery² and Wolkind and colleagues,³ who thought that about two fifths of the unselected repeat deaths they studied probably resulted from homicide.

Our purpose is not to second guess the original authors, who, being directly involved in the cases and having access to additional information, were much better placed than we are to assess them. However, we hope we have shown how a comparatively small change of perspective can result in a large change in the conclusions reached. We think the perspective we have taken is no less reasonable than that of the paper's authors and would be shared by most paediatricians. Most, for example, would not designate as "natural" so-called accidents that are entirely foreseeable and preventable.

We acknowledge that our assessments may be wrong, which is why in the table we prefix the categories natural and unnatural with probably, as well as introducing the third category of undetermined. We think using a dichotomy of natural or unnatural is unhelpful. It glosses over complexities and uncertainties and fosters polarisation. It is also more likely to be erroneous, as shown by the case where two deaths that were classified natural had to be reclassified after a third baby was murdered. Uncertainty may be uncomfortable, but it is truer to reality, more conducive to scientific inquiry, and safer for children than a dogmatic stance at either pole.

It is unfortunate that the summary that headed the paper was much more categorical than the text, which is not easy to follow and will have been less widely read. We do not think the summary's unqualified assertion that 40 out of 46 repeat unexplained infant deaths were natural is justified by the data presented. Furthermore, quoting precise confidence intervals for numbers derived from subjective judgment imparts a false veneer of precision.

The paper will help avert unjustified suspicion of

SUMMARY POINTS

A recent study of families with two sudden unexpected infant deaths concluded only six (13%) second deaths were homicide

Review of the published material shows that this was a minimum estimate

The likely cause of many of the second deaths could not be confidently ascertained

parents, which is important, but we are concerned that it may also lead to mistakes in child protection. Following a recent decision in the Court of Appeal,¹⁹ the Crown Prosecution Service is less likely to embark on the prosecution of a mother suspected of causing the death of her baby. This will be generally welcomed. The process of child protection, however, has to be uncoupled from decisions about criminal proceedings. We would encourage professionals to keep an open mind in assessing unexplained infant deaths, to be aware of the difficulties in diagnosis, and to try to keep a balance between the need to support parents and the need to protect children.

Contributors and sources: CJB has long had an interest in cot death and has been a leading participant in regional and national studies, while ENH has conducted and reported on many studies relating to neonatal and infant deaths. This article, first submitted in October 2006, arose from discussions with other paediatricians who were concerned about the conclusions and possible effects of the paper in question. Both authors contributed equally to this article. CJB is guarantor.

Competing interests: Both authors have worked for the Foundation for the Study of Infant Deaths, CJB as trustee then as medical adviser, ENH as an honorary scientific adviser. CJB has written court reports in cases involving unexpected infant death, for which he received the standard fee.

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- 1 Carpenter RG, Waite A, Coombs RC, Daman-Willems C, McKenzie A, Huber J, et al. Repeat unexpected and unexplained infant deaths: natural or unnatural? *Lancet* 2005;365:29-35.
- 2 Emery JL. Families in which two or more cot deaths have occurred. *Lancet* 1986;1:313-5.
- 3 Wolkind S, Taylor EM, Waite AJ, Dalton M, Emery JL. Recurrence of unexpected infant death. *Acta Paediatr* 1993;82:873-6.
- 4 Bacon C. Repeat sudden unexpected infant deaths. *Lancet* 2005;365:1137.
- 5 Di Maio VJM. Repeat sudden unexpected deaths. *Lancet* 2005;365:1138.
- 6 American Academy of Pediatrics. Policy statement. The changing concept of sudden infant death syndrome: diagnostic coding shifts, controversies regarding the sleeping environment, and new variables to consider in reducing risk. *Pediatrics* 2005;116:1245-55.
- 7 Gornall J. Was message of sudden infant death study misleading? *BMJ* 2006;333:1165-8.
- 8 Carpenter RG, Waite A, Coombs RC, Daman-Willems C, McKenzie A, Huber J. Special report adds nothing new, say paper authors. *BMJ* 2007;334:7.
- 9 Limerick SR, Bacon CJ. Terminology used by pathologists in reporting on sudden infant deaths. *J Clin Path* 2004;57:309-11.
- 10 Maguire S, Mann M, John N, Ellaway B, Sibert JR, Kemp AM. Does cardiopulmonary resuscitation cause rib fractures in children? A systematic review. *Child Abuse Neglect* 2006;30:739-51.
- 11 Dorries C. *Coroners' courts: a guide to law and practice*, 2nd ed. Oxford University Press 2004: 272.
- 12 Emery JL. Child abuse, sudden infant death syndrome, and unexpected infant death. *Am J Dis Child* 1993;147:1097-100.
- 13 American Academy of Pediatrics, Committee on child abuse and neglect. Distinguishing sudden infant death syndrome from child abuse fatalities. *Pediatrics* 1994;94:124-6.
- 14 Emery JL, Waite AJ. These deaths must be prevented without victimising parents. *BMJ* 2000;320:310.
- 15 Jervell A, Lange-Nielsen F. Congenital deaf-mutism, functional heart disease with prolongation of the QT interval and sudden death. *Am Heart J* 1957;54:59-68.
- 16 Denmark JC. Mental illness and early profound deafness. *Br J Med Psychol* 1966;39:117-24.
- 17 Arnestad M, Vege A, Rognum TO, Isaksen CV. Sudden infant death syndrome not caused by Norwegian Jervell and Lange-Nielsen mutations. *Am J Med Gen* 2005;134A:459-60.
- 18 Crown Prosecution Service. *The Code for Crown Prosecutors*. London: CPS, 2004.
- 19 *Regina v Cannings; judgment. Case No 200201711 D3*. London: Royal Courts of Justice, 2004.