Analysis and comment

Special report

Was message of sudden infant death study misleading?

Jonathan Gornall

An investigation by the *BMJ* has uncovered serious concerns about an important paper on sudden infant deaths recurring within a family. The paper has featured in the appeal case at several recent high profile murder appeals and has also influenced international practice

In January 2005 the *Lancet* published an analysis of data on babies born to parents who had previously had a baby who died suddenly and unexpectedly.¹ John Emery, a highly respected paediatric pathologist and expert on sudden infant death, was listed as the paper's seventh author, although he died in May 2000, more than two years before the first draft was completed and three years before the paper was submitted to the *Lancet*. The six other authors acknowledged in the paper that Professor Emery was "largely responsible for the setting up of this study and for investigation of the earlier cases" but played no part in drafting it. However, they did not make clear that after Professor Emery's death they recategorised deaths that he had classed as unnatural or of indeterminate cause as natural deaths. Furthermore, evidence of Professor Emery's views shortly before his death in May 2000 suggests that his name has been used to support a conclusion with which he would not have agreed.

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Provenance

The paper analysed deaths of babies in the Foundation for the Study of Infant Death's Care of the Next Infant scheme, which was set up to support parents with a history of sudden infant death. The lead author was the statistician Robert Carpenter. An honorary professor in the Medical Statistics Unit at the London School of Hygiene and Tropical Medicine, Professor Carpenter had worked alongside Professor Emery for 30 years. Another of the authors was Alison Waite, a health visitor and the national head of the scheme, who had been coauthor on several of Professor Emery's earlier papers.²⁻⁴

The *Lancet* paper was sponsored by the Foundation for the Study of Infant Death. Although the authors stated that the sponsors "had no role in study design, collection, analysis, or interpretation of data, or the writing of this report," they did not mention that Professor Carpenter had been a trustee of the foundation for more than 30 years.

The only previous report on the progress of the Care of the Next Infant scheme had been prepared for the foundation by Professor Emery in 1998. At this stage 5000 babies had been through the scheme,

among whom there were 35 unexpected deaths. Emery and his coauthors, including Carpenter and Waite, had concluded that at least 14 of these deaths (40%) were unnatural.⁴ By the time Carpenter and colleagues came to write the 2005 paper, the number of babies studied had increased to 6373 and the number of unexpected deaths had risen to 46. This time, however, the authors concluded that only six of the 46 deaths (13%) were unnatural. (Two families in the scheme had two sudden infant deaths, which did not contribute to these figures.)

Response to paper

The Carpenter paper was written at the height of the public debate over the statistical evidence given by paediatrician Roy Meadow at the trial of Sally Clark, who was convicted of murdering her two sons, and addressed his opinion that suspicion of foul play should increase with each subsequent sudden infant death within a family.⁵ The paper's finding that 87% of the unexpected deaths in a second child were natural seemed to vindicate Professor Meadow's critics and was

seized on by the media. The study, reported one newspaper, showed that: "The vast majority of second infant deaths in families who have already suffered the sudden loss of a baby are down to natural causes."⁶ Another said the study proved Professor Meadow's "murder theory" was wrong and reported lead author Professor Carpenter as saying that "Our data suggest that second deaths are not rare and that the majority—80 per cent to 90 per cent—are natural."⁷ The paper formed part of the evidence at the hearing that led to Professor Meadow being struck off the General Medical Council register for serious professional misconduct.

The American Academy of Pediatrics announced a policy change on the basis of the paper's findings: "Carpenter et al calculated an 87% probability that a second SIDS death within a family would be of natural cause ... Therefore, the task force supports the position that the vast majority of either initial or second sudden unexpected infant deaths within a family seem to be natural rather than attributable to abuse, neglect, or homicide."⁸

Questions over interpretation

The *BMJ* has learnt that before its publication the Carpenter team's interpretation of the data was questioned internally at the foundation by Chris Bacon, then the foundation's medical adviser. Dr Bacon was credited on the paper for his "encouragement and helpful comments" but was clearly unhappy with its conclusions. In a letter to the *Lancet* in March 2005, a month after he left the foundation, he said: "The investigators seem to have taken the benign view that a death should be classified as natural unless there was compelling evidence to the contrary—an approach that is appropriate in the courts but not in scientific debate ... this paper will be influential, but those quoting it should be aware that its data do not support such clear-cut conclusions."⁹

Lady Limerick, vice chairman of the foundation, in the course of a round table interview with her and three of the paper's authors (Carpenter, Waite, and Daman-Willems) told the *BMJ* that she had asked the authors for an explanation of why some deaths had been recategorised. In a document prepared by Ms Waite she explained that six of the deaths listed as unnatural in the 1998 paper and in a subsequent letter to the *BMJ*¹⁰ had been recategorised as natural deaths. In two of these cases coroners had recorded open verdicts. The other four were cases of overlying (infants had died while sleeping with their parents). Furthermore, Ms Waite said the authors had disregarded altogether four deaths that had taken place in children older than 1 year because they did not meet the internationally agreed definition of sudden infant death syndrome.

However, Ms Waite gave no explanation for the decision to categorise as natural all 13 deaths for which there was insufficient information to reach a conclusion. In five of these cases a police investigation was under way. In another seven parents or their doctors declined participation in the study's confidential inquiries. In the 13th case, the mother had been murdered.

The conclusion that 87% of repeat sudden infant deaths are natural does not even reflect the reservations about some of these cases that can be found within the narrative of the paper. The authors write: "We cannot exclude the possibility in our study that some of the 13 cases in which enquiries were not possible were cases of covert homicide." Furthermore, "We have counted as SIDS several cases in which asphyxia might have been a contributory factor."

Asked to justify the classification of these deaths categorically as natural, Professor Carpenter told the *BMJ*: "We've made clear that by unnatural we mean filicide ... everything else we've classified as natural." In other words, one category (unnatural) relies on incontrovertible evidence; the other (natural) relies on the absence of it. By excluding from their analysis the possibility of any grey area classification, the authors created the illogical corollary that all the deaths in the series that were not unnatural must be natural.

This would be the correct approach in a criminal court, where guilt must be proved beyond reasonable doubt, but Professor Emery recognised the importance of approaching such deaths differently when it came to scientific research. In a 1993 paper, Wolkind and Emery wrote: "These enquiries are not designed to produce legal evidence for use in court ... we are rarely completely certain but in all cases included in the 'probable filicide,' the conclusions of all members of the case discussion were that filicide was overwhelmingly the most probable cause."²

The three authors interviewed by the *BMJ* accepted that they could not be certain that Professor Emery would have agreed with what they had done. However, they thought that their approach had been within the spirit of Professor Emery's attitude towards parents and his last published words on the subject, in a letter to the *BMJ* just over three months before his death: "We do not need to create a pool of parents to which a label of unproved homicide is publicly attached."¹⁰

Yet Professor Emery's views on the probable causes of repeat sudden infant deaths are well documented, and there is plenty of evidence that he did not allow his compassion for individual parents to cloud his scientific objectivity. In three papers, a court report, and even that final letter to the *BMJ* his lowest estimate for the proportion of second sudden infant deaths that were unnatural was 34.5%³—a striking contrast with the 13% in the 2005 paper.

Further evidence of the divide between Professor Emery's apparent view and that credited to him after his death emerged during Professor Meadow's GMC hearing.¹¹ Professor Emery had been retained as an expert witness by Mrs Clark's defence team, and the two men had met in connection with the case before her trial in October 1999. Asked at the GMC hearing what he thought of the Carpenter paper, Professor Meadow described its analysis as "wild and amazing." Furthermore, he said, the paper "seems to me to be contrary to the declared position of Professor Emery shortly before he died ... one third of cases are, in his opinion, from natural causes ... one third roughly he says I think are child maltreatment and a third are, to his group, unknown."

Professor Meadow was referring to a report Emery had produced for Mrs Clark's defence team before her trial in November 1999, six months before his death.

The report was not used at the trial, but a fragment of it, pertaining to the rarity of two unexplained sudden infant deaths in one family, emerged in the judgment of Mrs Clark's first, unsuccessful appeal in October 2000.¹² (Mrs Clark's convictions were eventually quashed by the Court of Appeal in January 2003.) Emery wrote: "The occurrence of repeat unexpected deaths is a topic on which I have been doing research over the last few years and have been able to do a confidential study involving approximately 100 such deaths.

"These have all been families who have presented as having two 'cot deaths.' My findings have been that approximately a third of the deaths have been due to a whole series of rare natural causes which had been missed by those doing the necropsy. A third of the deaths were associated with different forms of child abuse and having a whole number of features which indicated that they were unnatural deaths. In the final third no suspicion of unnatural death was found and no natural cause was found and these children constitute what may be termed at the moment as 'true idiopathic unexpected child deaths'."

His conclusion was stark: "The occurrence of two unexpected deaths in a family thus raises a definite suspicion of unnatural death which in my experience is confirmed ... in a third of such cases."

Clearly, Emery's interpretation of the data in 1999 is difficult to reconcile with the claim made by Joyce Epstein, director of the Foundation for the Study of Infant Deaths, for the 2005 paper that bore his name: "The study shows that any form of automatic suspicion, such as the so-called 'Meadow's Law', is unjustified."¹³

Use as evidence

A draft of the paper conveyed this message about the validity of "Meadow's law" to the three judges who in December 2003 heard and upheld the appeal of Angela Cannings. Mrs Cannings had been jailed in April 2002 for the murder of two of her children, and during the trial Professor Meadow had given evidence about the rarity of three unexplained infant deaths in one family (Mrs Cannings's first daughter had also died suddenly and unexpectedly, aged 13 weeks). The three unexplained deaths, said the appeal judges, was a fact to which "Professor Meadow attached huge importance." The evidence of the Carpenter paper helped to persuade them that he had been wrong to do so.

The three judges hearing the Cannings appeal were aware that the paper had not yet been peer reviewed, let alone accepted for publication. "It is anticipated," they noted in their judgment¹⁴ "that [the paper] will, subject to minor corrections, shortly be published." In fact, it was not published for another two years.

The judges went on to consider "the critical questions" that arose when multiple unexplained infant deaths occurred in the same family: "In addition to the concerns already noted about an excessively dogmatic approach, we must immediately note a substantial body of research, not before the jury, and received by us in evidence, suggesting that such deaths can and do occur naturally, even when they are unexplained." This body of research included the draft paper, which "claimed to be ... by far the largest

follow-up study of families who have had a cot death. It is sufficient to repeat the conclusion which was, 'that the occurrence of a second unexpected infant death within a family is not a rare event and is usually from natural causes.' Our inexpert analysis of the study does not suggest that the conclusion is unsupported by the research."

In the judges' final analysis, although they were "satisfied that there is a realistic, albeit as yet undefined, possibility of a genetic problem within this family, which may serve to explain these tragic events," it was plain that the Carpenter paper had played some part in the result of the appeal—namely, the quashing of Mrs Cannings's conviction. They wrote: "We have received significant and persuasive fresh evidence, which was not before the jury, some of it the result of further research, or research published post-trial, into the problem of SIDS generally, and some specific to Mrs Cannings and her extended family. The expert evidence was absolutely critical to these convictions. In our judgment the fundamental basis of the Crown's case, based on the extreme rarity of three separate infant deaths in the same family, and the pattern of events in this particular family is, for the reasons we have given, demonstrably undermined." The published paper also featured as evidence in the successful appeal last year of Donna Anthony, a mother convicted in November 1998 of the murder of her two children; both convictions were quashed. The judges noted with reference to the *Lancet* paper: "Recent research has wholly undermined the conclusion to be drawn from the coincidence of the deaths two of [sic] infant children in the same family."¹⁵ They added: "The conclusion that the deaths of these two children was unnatural and the consequent convictions depended substantially on the fact that there were two such deaths in the same family. Although there was evidence to support the conclusion that each death taken in isolation may have been the result of deliberate inference [sic] with the child ... the reality is that of itself the rarity of the two infant deaths in the same family underpinned to [sic] the views formed by and opinions expressed to the jury by the experts called by the Crown. It was critical to the evidence that they gave and must inevitably have permeated the way in which the jury would have approached this evidence. In the light of the decision in Cannings that approach would have been erroneous."

Professional dissent

The *BMJ* has also learnt that David Hall, then immediate past president of the Royal College of Paediatrics and Child Health, wrote to the *Lancet's* editor, Richard Horton, in November 2005 to express his alarm at the Carpenter paper (personal communication). Professor Hall said the Care of Next Infant dataset had "offered an unparalleled opportunity to take a dispassionate look at this extremely important question. Unfortunately, the analysis was seriously flawed."

The *Lancet* suggested Professor Hall could write an online comment but rejected his suggestion that it should publish a "detailed review" of Carpenter's findings. Hall's letter highlighted concerns that "in every case where there might be reasonable doubt the authors have come down on the side of a natural death

rather than classifying it as indeterminate, which we believe would be the correct solution for a substantial number of these reported deaths. There is furthermore the interesting statement regarding the 18 ... cases for which data were available that 'all of them had high risk factors' and by implication the series contains no example of a repeat classic cot death in a low risk family."

Professor Hall's conclusion was that he and others regarded the paper's findings as "seriously misleading" and thought they could have "serious consequences" for child protection.

Competing interests: None declared.

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- 3 Waite A, Emery J. Repeat cot deaths in families enrolled onto a support programme [abstract]. Proceedings of fourth sudden infant death syndrome international conference, Bethesda, 23-26 June, 1996.
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- 5 Meadow R. Fatal abuse and smothering. In: *ABC of child abuse*. 3rd ed. London: BMJ Publishing, 1997:27-9.
- 6 Study casts doubt on 'Meadow's law' of sudden child death. *Independent* 2004 Dec 31.
- 7 "Murder" theory on infant deaths disproved. *Herald* 2004 Dec 31.
- 8 American Academy of Pediatrics Task Force on Sudden Infant Death Syndrome. Policy statement. *Pediatrics* 2005;116:1245-55.

Summary points

An analysis of sudden infant deaths suggested that almost 90% of second deaths in the same family are natural

Classification of deaths in the study was changed after the death of one of the senior authors

Deaths of indeterminate cause were counted as natural

The study was used as evidence in murder appeals

- 9 Bacon C. Repeat sudden unexpected infant deaths. *Lancet* 2005;365:1137.
 - 10 Emery JL, Waite AJ. These deaths must be prevented without victimising parents. *BMJ* 2000;320:310.
 - 11 GMC Fitness to Practise Panel. *Case of Meadow: day 10, 6 July, 2005*. London: GMC, 2005.
 - 12 R v Sally Clark [2000] EWCA Crim 54 at 115-6.
 - 13 Study casts doubt on 'Meadow's law' of sudden child death. *Independent* 2004 Dec 31.
 - 14 R v Angela Cannings [2004] EWCA Crim 1.
 - 15 R v Donna Anthony [2005] EWCA Crim 952.
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Primary care

Does practice based commissioning avoid the problems of fundholding?

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Current changes to primary care are meant to improve services for patients, but history suggests that they may not deliver and could even decrease satisfaction

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By the end of 2006 all general practices in England are expected to be responsible for a budget to purchase secondary care and community health services for their patients.¹ What is remarkable about the practice based commissioning initiative is its similarity to the general practitioner fundholding experiment of the previous decade, which Labour abolished after it came to power in 1997. There is therefore potential to take stock of the lessons from fundholding and to assess the prospects for commissioning in that light.^{2 3}

Fundholding

Fundholding was a cornerstone of the NHS reforms of the 1990s. It allowed general practices to hold budgets to purchase secondary care for their patients on the grounds that it would make general practitioners more aware of the financial implications of their prescriptions and referrals. It was also assumed that it would give general practitioners greater leverage in negotiating those referrals with other health providers, as a clear resource would be attached to their commissioning decisions, and

that services would become more responsive to patients as a result. Practices were also allowed to retain any budgetary surplus to invest in additional patient services or in enhancing practice premises.

There were three varieties of general practitioner commissioning in the 1990s that gradually extended the scope of the scheme: the fundholding scheme itself; total purchasing pilots, which extended the range of services general practitioners purchased with the rationale that their purchasing would be more responsive than that of health authorities; and general practice commissioning pilots, where groups of practices collaborated to exercise a greater collective influence over commissioning to make health authorities more sensitive to local needs.

Evidence on fundholding

Studies show that fundholding increased general practitioners' awareness of the resources involved in referring patients. Both fundholding and total purchasing pilots lowered referral rates for elective