

Munchausen syndrome by proxy and sudden infant death

A W Craft, D M B Hall

Media vilification of paediatricians acting in cases of alleged child abuse has resulted in widespread confusion about research data and threatens the systems to protect vulnerable children. How should we move forward?

Everyone can empathise with the grief of parents who have lost a baby and even more so with the nightmare scenario of being wrongly held responsible for the child's death. It is therefore not surprising that the media showed intense interest in three recent cases in which mothers were accused of murdering their babies.¹ Nothing in this article should be construed as personal opinion on any individual case, but we are concerned that everything we have learnt over the past 40 years about sudden infant death syndrome and the spectrum of child abuse seems to have been forgotten.

Attacks on paediatricians who give evidence in child protection cases^{w1} and journalists' use of phrases like "Meadow's discredited theory of Munchausen syndrome by proxy" have left the public and the health professions anxious and bewildered. In this paper we outline the evolving understanding of the relation between unexplained death in infancy and child abuse, draw lessons from recent events, and offer proposals for the future.

History

Caffey described what was undoubtedly child abuse in 1946^{w2} but was reluctant to draw the obvious conclusions. Another 16 years passed before Kempe and colleagues published their seminal paper on the battered child syndrome in 1962.^{w3} At about the same time, the problem of "cot death" began to attract interest. The phenomenon of unexplained death in previously well babies had been known for centuries and was blamed on overlying, "status thymolymphaticus," witchcraft, and many other causes, including infanticide.

The first major conference on cot death was held in Seattle in 1963, and the possibility that some such deaths might be due to foul play was raised but not pursued.² A second conference in 1969 adopted the term sudden infant death syndrome. The hypothesis that episodes of prolonged apnoeic pauses, possibly of genetic origin, might be the basis for sudden infant death syndrome was investigated by Steinschneider^{w4} and many others. This resulted in the widespread adoption of apnoea alarms across the United States and subsequently in the United Kingdom.

Over the next decade doubts emerged about the apnoea hypothesis.² At the Baltimore conference in

1982 Southall et al reported, firstly, that after monitoring almost 10 000 babies they were unable to show any relation between apnoeic pauses and sudden infant death^{w5-7} and, secondly, that they had found no case of a second cot death in the same family after following the siblings of over 200 babies who had died from sudden infant death syndrome.^{w8}

It is now known that sudden infant deaths do not follow a random pattern.^{w9 w10} Substantial relations exist between unexplained death and social background, social chaos, poverty, parental mental illness, prematurity, parental smoking and substance misuse, and pre-existing symptoms of illness in the baby.^{w11} The relation with prone sleeping position is still not fully explained, but the United Kingdom's Back to Sleep campaign was associated with a large reduction in incidence.³ However, a completely satisfactory explanation is still elusive in many infant deaths.

Link between child abuse and sudden infant death

The evolving interest in unexplained death and child abuse converged when several families who had

Sir James Spence
Department of
Child Health, Royal
Victoria Infirmary,
Newcastle upon
Tyne NE1 4LP
A W Craft
*professor of child
health*

Institute of General
Practice, Northern
General Hospital,
Sheffield, S6 6GY
D M B Hall
*professor of
community
paediatrics*

Correspondence to:
D M B Hall
d.hall@sheffield.ac.uk

BMJ 2004;328:1309-12

References w1-36 are on bmj.com



Such scenes have left many paediatricians reluctant to take on child protection cases

apparently lost several babies from sudden infant death syndrome were subsequently found to have murdered their children.^{2 w12} In 1982, Taylor and Emery introduced the concept of gentle battering to describe cases in which suffocation by an object or a hand led to the infant's death.^{w13} Several strands of evidence support this. Some parents of babies thought to have died from sudden infant death syndrome have subsequently admitted suffocating their infant. Other unequivocal evidence of physical abuse is sometimes found. The siblings of suspected cases (sometimes including genetically unrelated adopted siblings) are more likely to have died than would occur by chance.

Southall et al used covert video surveillance to investigate apparent life threatening events and showed that parents do indeed cause suffocation by deliberate airway obstruction. They emphasised that the aim of covert video surveillance was to protect the child rather than to secure convictions.^{4 w14} Covert video surveillance is now an accepted procedure when there is no other way of confirming a diagnosis of suffocation.

Meanwhile, other more subtle forms of abuse were identified.⁵ In 1976, Money and Werlwas described a case of dwarfism caused by starvation in which the parents presented misleading accounts of the child's illness, suggesting an analogy with Munchausen's syndrome.^{w15 w16} The term Munchausen syndrome by proxy received wider publicity when in 1977 Meadow reported a case with deliberate fabrication of bizarre symptoms.⁶ Many manifestations of Munchausen syndrome by proxy are now recognised—for example, intentional poisoning by salt or medicines and fictitious accounts of seizures.⁷

It gradually became clear that parentally induced apnoeic attacks^{w17} and cases of deliberate suffocation should be regarded as a form of Munchausen syndrome by proxy. Contrary to the impression created by the recently announced review of 5000 child protection cases, non-accidental suffocation is not a common paediatric diagnosis. A UK-wide survey of Munchausen syndrome by proxy in 1992-4 identified just 128 cases in two years, of which only 32 were related to suffocation.⁸ However, a growing number of case reports suggest that many manifestations of Munchausen syndrome by proxy are underidentified because of lack of professional expertise and public awareness.

The most recent estimate is that around 10% of all otherwise unexplained sudden infant deaths may be caused by deliberate suffocation, though this probably varies over time and between populations.⁹⁻¹¹ Bleeding from the nose and mouth suggests deliberate suffocation,¹¹ and the presence of haemosiderin laden macrophages in the lungs might signify previous episodes of suffocation.^{w18 w19} However, in a single case neither the autopsy findings nor other background information are likely to provide unequivocal evidence.¹²

Recurrent sudden infant death syndrome

Since the Back to Sleep campaign, sudden infant death syndrome has become less common in the United Kingdom and the social class gradient has become

steeper. In low risk families, sudden infant death affects just over 1 in 8500 babies. The probability of any two randomly selected low risk babies dying of sudden infant death syndrome is that number squared (about 1 in 73 million), but this does not apply in an individual family that has already had one baby die from sudden infant death syndrome. Chance has no memory, so a low risk family would have a 1 in 8500 risk of a second death.

There is little evidence of familial clustering of sudden infant death syndrome, and more than one unexplained infant death in the same family suggests two other possibilities^{w20}: homicide or inherited conditions such as metabolic disorders (for example, medium chain acyl coenzyme A dehydrogenase deficiency), cardiac arrhythmias (including prolonged QT syndrome), immune deficits predisposing to infection, and abnormalities of ventilatory control.² These conditions probably account for a small fraction of cases,^{w21-23} but their existence and the possibility that some may remain to be discovered add a dimension of uncertainty in any death where autopsy findings are negative or equivocal.

Is Munchausen's the best label?

Only a minority of parents who deliberately harm their children have exhibited features of Munchausen's syndrome themselves,^{13 14} and little is known about the psychopathology underlying the fabrication of illness in older children. It probably differs from that described in parents who induce illness in infants by suffocation or other means. Some parents who induce illness in infants have a personality disorder^{w24} or (rarely) a psychotic illness.¹¹ In the postnatal period women have reported a variety of fantasies, obsessions, and anxieties regarding their babies,^{w25 w26} including concerns about their ability to care for the child and worries that they might cause them harm.^{w27} An association seems to exist between postnatal depression and sudden infant death.^{w28-30}

As there is no single psychological profile in Munchausen syndrome by proxy,^{10 w31-34} and the label makes unwarranted assumptions about the parent's mental state and motivation, many UK paediatricians feel that the term should be abandoned. The preferred term is now fabricated and induced illness.¹⁵ Fabricated and induced illness is seen as part of a spectrum of child abuse, and death caused by suffocation is at the severe end of that spectrum. The term keeps the focus on the presenting features of the child who needs to be protected, rather than on the supposed psychopathology of the parent.

Learning the lessons

Recent events have prompted some soul searching about the ways professionals respond to the unexpected death of an infant. We agree with Meadow's comment in his first paper on Munchausen syndrome by proxy: "doctors, although (occasionally) deceived, will rightly continue to believe what most parents say, most of the time."⁶ Stanton and Simpson reported a tragic case in which a mother who clearly loved her baby nevertheless smothered her. They commented on the "difficult cognitive processes involved for a paedia-

trician to shift from being the parent's ally in attempting to alleviate a child's suffering, to suspecting the parent of being the agent responsible for the child's suffering.^{17,26} This may explain why some professionals offer conflicting and sometimes improbable explanations for injuries to children, leading to difficulties for the judiciary and increased risks for children.

We need to rethink our approach to cases of unexplained infant death, particularly with families who have had more than one such tragedy. We stress the importance of being alert to inconsistencies in the history, scene of death inquiries,¹⁶ collection of background data,³⁵ and an autopsy by a paediatric or forensic pathologist with appropriate training, working to a standard protocol. In future this may sometimes need to include a search for genes that might be associated with conditions predisposing to sudden death. Parents have a right to expect that their baby's tragic unexplained death will be investigated as thoroughly as would be the case for any other citizen.

There are important lessons for the judiciary as well. In a criminal trial the standard of proof is "beyond reasonable doubt." In the United States, a Court of Appeals judgment stated that there might be insufficient evidence in any one child death to prove murder or infanticide, but evidence of repeated incidents was admissible as evidence of non-accidental deaths.¹² We agree, but at the same time feel that securing a conviction for murder is rarely the priority in cases where evidence rests on epidemiological data, probabilities, and clinical findings whose interpretation is subject to several opinions.^{17 18 36}

What is the alternative?

We believe that complex child deaths and child protection cases should normally be handled by civil proceedings, following the procedures introduced by the Wolff reforms. There should be a non-adversarial meeting of experts from each of the key agencies and disciplines to share and review all the evidence. More than one opinion should be obtained on difficult issues. The emphasis must be first on assessing and minimising the risk to any other children cared for by the parents and secondly on management of the parents' needs.^{7 14} Although the reasons why parents kill their babies are poorly understood, perhaps many deserve sympathy and treatment rather than imprisonment.

Some cases will nevertheless result in criminal proceedings. Professionals who offer expert evidence must not promote ideas that are unsupported by research but should present the facts as they currently see them. In an adversarial legal system, it is for the lawyers and the judge to make sure that experts explain to them, and to the jury, the limits of knowledge and the extent of uncertainty.

Expert evidence

Professionals expect to be accountable for, and challenged about, the reliability of their research, evidence, and interpretation. Nevertheless, they must be able to undertake their duties without fear of ill informed attacks on their competence and integrity by the media or parental organisations. Orchestrated

Summary points

Recent court cases regarding unexplained infant deaths have raised questions about the diagnosis of Munchausen syndrome by proxy and professional judgment

Many paediatricians are now reluctant to take part in child protection cases

A non-adversarial approach needs to be introduced to deal with sudden infant deaths

The public needs to be better informed of research on sudden infant death and child abuse

campaigns intended to discredit expert opinion¹⁹ have made professionals increasingly reluctant to participate in child protection cases. A recent survey undertaken by the Royal College of Paediatrics and Child Health found that 14% of paediatricians have had an official complaint made against them in respect of child protection issues arising in general paediatric practice. The often vexatious and malicious nature of these complaints is illustrated by the fact that although many resulted in unpleasant local or national publicity, only a tiny proportion were upheld.²⁰ Of 87 paediatricians referred to the General Medical Council whose cases have been completed, none was found guilty of serious professional misconduct.

Conclusions

We must restore the faith of the public and the professions in the child protection systems that are vital for a civilised society. Munchausen syndrome by proxy has captured the public imagination, but there is still much that we do not know about other aspects of child abuse. For example, questions remain unanswered about subdural bleeding and the interpretation of physical signs in child sexual abuse. We need urgently to review procedures and to fund more research into the causes, mechanisms, and diagnosis of child abuse. And we call on journalists, lawyers, and the judiciary to ensure that they are well informed about the mass of evidence and data gathered over the past 40 years in child protection.

We thank David Southall and R J Postlethwaite for helpful comments on an earlier draft of this paper.

Contributors and sources: AWC is president and DMBH is immediate past president of the Royal College of Paediatrics and Child Health. The idea for and preparation of this review resulted from our shared concerns about the state of child protection work both in the United Kingdom and in other developed countries. These concerns arose from the numerous attacks on the integrity and competence of paediatricians working in child protection brought to the attention of the college over the past four years. We have reviewed the literature and drawn on the extensive expertise of colleagues. DMBH is guarantor.

Competing interests: We and many members of the Royal College of Paediatrics and Child Health give, or have given, evidence as professional or expert witnesses in child protection cases.

1 Marks N. An expert witness falls from grace. *BMJ* 2003;327:110.

- 2 Firstman R, Talan J. *The death of innocents*. New York: Bantam, 1998.
- 3 Fleming PJ, Bacon CJ, Blair PS, Berry PJ. *Sudden unexpected deaths in infancy—the CESDI SUDI studies 1993 to 1996*. London: Stationery Office, 2000.
- 4 Southall DP, Plunkett MC, Banks MW, Falkov AF, Samuels MP. Covert video recordings of life-threatening child abuse: lessons for child protection. *Pediatrics* 1997;100:735-60.
- 5 Rogers D, Tripp J, Bentovim A, Robinson A, Berry D, Golding R. Non-accidental poisoning: an extended syndrome of child abuse. *BMJ* 1976;ii:793-6.
- 6 Meadow R. Munchausen syndrome by proxy. The hinterland of child abuse. *Lancet* 1977;ii:343-5.
- 7 Eminson DM, Postlethwaite RJ. *Munchausen syndrome by proxy abuse: a practical approach*. Oxford: Butterworth Heinemann, 2000.
- 8 McClure R, Davis P, Meadow S. Epidemiology of Munchausen syndrome by proxy. *Arch Dis Child* 1996;75:57-61.
- 9 Emery JL. Child abuse, sudden infant death syndrome, and unexpected infant death. *Am J Dis Child* 1993;147:1097-100.
- 10 Wolkind SE, Taylor EM, Waite AJ, Dalton MF, Emery JL. Recurrence of unexpected infant death. *Acta Paediatr* 1993;82:873-6.
- 11 Levene S, Bacon C. Sudden unexpected death and covert homicide in infancy. *Arch Dis Child* 2004;89:443-7.
- 12 Meadow R. Unnatural sudden infant death. *Arch Dis Child* 1999;80:7-14.
- 13 Meadow R. What is, and what is not, 'Munchausen syndrome by proxy'? *Arch Dis Child* 1995;72:534-8.
- 14 Fisher G, Mitchell I. Is Munchausen syndrome by proxy really a syndrome? *Arch Dis Child* 1995;72:530-4.
- 15 Royal College of Paediatrics and Child Health. *Fabricated or induced illness by carers*. London: RCPCH, 2002.
- 16 Fleming P, Blair P, Sidebotham P, Hayler T. Investigating sudden unexpected deaths in infancy and childhood and caring for bereaved families: an integrated multiagency approach. *BMJ* 2004;328:331-4.
- 17 Southall DP, Samuels MP, Golden MH. Classification of child abuse by motive and degree rather than type of injury. *Arch Dis Child* 2003;88:101-4.
- 18 Golden MH, Samuels MP, Southall DP. How to distinguish between neglect and deprivational abuse. *Arch Dis Child* 2003;88:105-7.
- 19 Marcovitch H. GMC must recognise and deal with vexatious complaints fast. *BMJ* 2002;324:167.
- 20 Kmietowicz Z. Complaints against doctors in child protection work have increased fivefold. *BMJ* 2004;328:601.

(Accepted 7 May 2004)

Hands-on guide to questionnaire research

Selecting, designing, and developing your questionnaire

Petra M Boynton, Trisha Greenhalgh

Anybody can write down a list of questions and photocopy it, but producing worthwhile and generalisable data from questionnaires needs careful planning and imaginative design

This is the first in a series of three articles on questionnaire research

Department of Primary Care and Population Sciences, University College London, Archway Campus, London N19 5LW
 Petra M Boynton
lecturer in health services research
 Trisha Greenhalgh
professor of primary health care

Correspondence to: P M Boynton
 pboynton@pcps.ucl.ac.uk

BMJ 2004;328:1312-5

The great popularity with questionnaires is they provide a “quick fix” for research methodology. No single method has been so abused.¹

Questionnaires offer an objective means of collecting information about people’s knowledge, beliefs, attitudes, and behaviour.²⁻³ Do our patients like our opening hours? What do teenagers think of a local antidrugs campaign and has it changed their attitudes? Why don’t doctors use computers to their maximum potential? Questionnaires can be used as the sole research instrument (such as in a cross sectional survey) or within clinical trials or epidemiological studies.

Randomised trials are subject to strict reporting criteria,⁴ but there is no comparable framework for questionnaire research. Hence, despite a wealth of detailed guidance in the specialist literature,^{1-3 5 w1-w8} elementary methodological errors are common.¹

Inappropriate instruments and lack of rigour inevitably lead to poor quality data, misleading conclusions, and woolly recommendations.^{w8} In this series we aim to present a practical guide that will enable research teams to do questionnaire research that is well designed, well managed, and non-discriminatory and which contributes to a generalisable evidence base. We start with selecting and designing the questionnaire.

What information are you trying to collect?

You and your co-researchers may have different assumptions about precisely what information you would like your study to generate. A formal scoping exercise will ensure that you clarify goals and if necessary reach an agreed compromise. It will also flag up potential practical problems—for example, how long the questionnaire will be and how it might be administered.

As a rule of thumb, if you are not familiar enough with the research area or with a particular population subgroup to predict the range of possible responses, and especially if such details are not available in the literature, you should first use a qualitative approach (such as focus groups) to explore the territory and map key areas for further study.⁶

Is a questionnaire appropriate?

People often decide to use a questionnaire for research questions that need a different method. Sometimes, a



References w1-w17, further illustrative examples, and checklists are on bmj.com