CURRENT TOPIC

Cot death after CESDI

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The three year study of sudden unexpected death in infancy (SUDI), recently completed as part of the programme of the Confidential Enquiry into Stillbirths and Deaths in Infancy (CESDI), includes the largest and most comprehensive study of cot death yet conducted in the UK. Over 450 sudden deaths, of which 80% were classified as sudden infant death syndrome (SIDS) were investigated in five (former) NHS regions between 1993 and 1996, both by confidential inquiry and by case-control technique. Outline results for the first two years were given in the third report of the National Advisory Body for CESDI¹ while more detailed reports on various aspects of the case-control study are being published in professional journals.^{2,3} The national intervention on sleeping position in 1991 was followed by a marked and most welcome fall in the incidence of cot death, the national rate declining from 1.61/1000 live births in 1990 to 0.6 in 1993. However SIDS is still the largest category of deaths in the postperinatal period, and the SUDI study has shown that several major problems remain to be addressed. Some of these I should like to discuss.

Definition of SIDS

Definition by exclusion is sometimes unavoidable but never really satisfactory. It is particularly unsatisfactory in the case of SIDS because some specific causes of unexpected infant death, such as suffocation or metabolic disorder, may leave little or no obvious sign. There are those who would argue that SIDS is not a proper diagnosis anyway but just a cloak for our diagnostic penury: there is always a specific cause of death if only we knew how to find it. In addition there remains the problem of what degree of necropsy findings constitutes an adequate cause of death. In the SUDI study there were several instances in which the expert regional panel, guided by their paediatric pathologist who had seen the slides, disagreed with the interpretation of the reporting pathologist, upon whose opinion the cause of death had been registered. Usually the reporting pathologist thought his findings constituted an adequate cause of death while the panel did not; this occurred in at least one in 10 of unexpected deaths not registered as SIDS (the exact proportion is uncertain because panels did not always record this point). Consistent errors of this sort will falsely lower the SIDS rates

recorded by the Office of Population Censuses and Surveys (OPCS, now the Office for National Statistics), the distortion being greater now that deaths are fewer. The subjectivity inherent in the definition of SIDS means that some disagreement is inevitable, but it could be kept to a minimum if necropsies on infant dying unexpectedly were done only by specialist pathologists. Meanwhile all pathologists who do infant necropsies should at least try and achieve better consensus.

Briefing the pathologist

When a person dies unexpectedly the coroner's officer is required to look into the circumstances and write a report. This report may be the only information available to the pathologist before he does the necropsy. Coroner's officers are usually police officers and their experience of sudden deaths in infancy is variable. Some are well informed and write comprehensive reports. Others write more limited reports that mainly address the possibility of foul play. The SUDI study noted several instances where the pathologist was not made aware of significant details in the history or circumstances of the death which might have prompted him to look for particular features at necropsy. For example, a history of non-accidental injury in siblings would have suggested a skeletal survey, previous unexplained deaths in the family would have justified metabolic studies, while a report of an old gas fire burning in the baby's bedroom would have called for an assay of carboxyhaemoglobin. The necropsy is of the greatest importance on several counts: it is the essential basis for the diagnosis, it is traumatic for the family, it is time consuming for the pathologist, and it is quite expensive. It does not make sense for the pathologist to embark on it without adequate briefing. In some areas this problem is tackled by furnishing the coroner's officer with a checklist of questions to ask after a sudden infant death. However this is unlikely always to elicit the varied and subtle features in the history of the baby and the family and in the circumstances of the death that may give clues about its cause. The ideal arrangement, which already happens in a few areas, would seem to be a joint investigation by the coroner's officer and a paediatrician, the former concentrating on forensic aspects, the latter considering the

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multiplicity of possible causes of death in infancy. I return to this theme later.

Quality of necropsy

Despite the guidelines for necropsies in infants who have died unexpectedly that were issued by the Royal College of Pathologists in 1992,⁴ the quality of necropsies in the SUDI study was found to be variable. Many were excellent—and not only those carried out by paediatric pathologists working in teaching centres. Others however were less adequate and omitted various items, including those that are always important such as lung histology, and those specifically indicated by the history, such as metabolic studies where there had been previous deaths in infancy. There appeared to be two main problems. First, if there were initial suspicions about the death, the coroner might instruct a forensic pathologist who, while very thorough in the gross examination and expert at detecting any evidence of foul play, once this was excluded might not regard it as his remit to pursue the minutiae of natural causes. Secondly, where a paediatric pathologist is not available the necropsy is often performed by a non-specialist who may not have the time or the inclination to follow the guidelines. The full protocol is indeed laborious and expensive—though in the SUDI study extra funding was available. Analysis of the pathology findings in the SUDI study is not yet completed; it would be very helpful if it could tell us which of the recommended investigations yielded most results, so that the busy pathologist could at least concentrate on areas most likely to be productive. Ultimately, however, it would be best if the increasing tendency to involve paediatric pathologists in infant deaths became universal; this goal should become achievable as the former increase and the latter decrease in number. Coroners are notoriously independent, but it is to be hoped that the Coroners' Society will take note of the National Advisory Body's recommendation that paediatric and forensic pathologists should join forces in investigating unexpected infant deaths that initially seem suspicious.

Deaths brought about by carers

The SUDI study excluded cases in which a criminal prosecution was being brought against the carers, which OPCS figures suggest comprise about 5% of unexpected deaths in infancy. The SUDI regional panels concluded that between 8% and 19% of the remaining sudden deaths may also have arisen, whether intentionally or not, from the action or behaviour of the carers, although the evidence was not strong enough to trigger prosecution and the death was likely to have been registered as SIDS. It has long been suggested that a small proportion of deaths registered as SIDS are in fact murder, manslaughter, or infanticide⁵—and this proportion may well have risen as the total number of deaths has decreased. As the National Advisory Body report points out, this poses a very difficult dilemma for all the professionals involved, not least the police. The majority of parents who lose a baby suddenly

and unexpectedly are innocent and in desperate need of compassionate support and informed advice. But it is vital to identify the minority of instances where the parents themselves have brought about the death, not for purposes of retribution (which may or may not be appropriate) but because all the present family members may need help and future babies need protection. When the cause of death is not obvious and parents deny liability, the experience that might lead to elucidation lies more with paediatricians than with policemen. Yet at present the police usually prefer to investigate on their own. This may sometimes result in a long and searching investigation, compounding the agony of the parents, of a death that a paediatrician might quickly have assessed as innocent. Conversely the police, if not alerted to the more subtle signs, might exonerate abusing parents, thereby making it harder to protect other children. Although it would at times be an uncomfortable partnership for either side, there could be advantages if paediatricians worked in liaison with police officers, both routinely with the coroner's officer, and with the CID when a death was being more fully investigated.

Local case discussion

Many districts routinely call a meeting after any unexpected infant death to discuss all relevant issues. Essential participants are the general practitioner, the health visitor, the pathologist, a paediatrician, and any other professional who has been involved. Agenda include the antecedents and circumstances of the death, the necropsy findings, any lessons for professionals, and arrangements for continuing support of the family, both in the short term and for any future pregnancy. Local case discussions were a requirement for the SUDI studies, and the National Advisory Body report recommends that they should become universal practice. In addition to their value in ensuring support for bereaved families and educating professionals, they also provide a forum in which the difficult but essential question of whether the carer might have caused the death can be addressed in confidence. The picture may only become clear when several people have fitted their pieces of the jigsaw together. The contribution of the primary care team is vital, so the meeting is best held in the general practitioner's surgery and at an hour of his choosing. The timing is dictated by the pathologist, who will usually want to see the histology before he gives an opinion. This means a delay of at least six weeks, which entails a loss of immediacy, and preliminary arrangements for supporting family must already be in place. Participation in a wider confidential inquiry does not reduce the need for a local case discussion, because the anonymity inherent in a confidential inquiry precludes intervention in individual cases. Responsibility for organising the local case discussion falls aptly within the remit of the designated paediatrician.

Support of parents

The SUDI study (in data not yet published) has confirmed previous findings that the support given to bereaved parents varies greatly from place to place. In some areas, particularly where there is a paediatrician with an interest in the subject, it may be prompt and effective. In other areas it is almost non-existent. A few years ago the British Paediatric Association and Foundation for the Study of Infant Deaths combined to compile a list of designated paediatricians in every district who would take a special interest in cot death, including the provision of support for the family. Some of these paediatricians are failing in this role. If adequate support is to be given, the first requirement is a network of informants—health visitors, general practitioners, other paediatricians, pathologists, casualty departments, mortuary attendants, coroner's officers—who know that they must get in touch with the designated paediatrician as soon as a death occurs. It is then not enough simply to write to the family offering them an appointment in hospital in three weeks' time; many will not take this up, especially if they have not met the paediatrician before. He must be proactive, and suggest that he comes to visit them at home one evening soon. A meeting on their territory rather than his is more likely to be acceptable and fruitful. Similarly, it must go at their pace not his, and he must be prepared to stay for at least two hours. Questions must be answered with authority and with sympathy, but listening is perhaps more important. The paediatrician must be familiar with resources for counselling and befriending bereaved parents in his area, and should put the family in touch with those he thinks most suitable. Liaison with the Foundation for the Study of Infant Deaths will be particularly helpful here.

With regard to timing, experience in the Avon and the SUDI studies has shown that, contrary to traditional opinion, a visit very soon after the death is often welcomed by parents, whose need to talk with someone knowledgeable is then most acute. If a survey of SIDS is being undertaken, it is possible to combine the roles of data collector and initial counsellor during the same encounter. If the visit is made almost immediately, the paediatrician can complement the inquiries of the coroner's officer and ensure that the pathologist is fully briefed before the necropsy. A consistent service on these lines calls for a major commitment of time and energy, plus the ability to divert other work at short notice. However its value to the families is profound, and it can be one of a paediatrician's most worthwhile roles.

Future research and intervention

It is likely that the recent large case-control studies have identified most of the current major environmental risk factors, and that fur-

ther similar studies would merely find changing patterns of risk as interventions took effect. The SUDI study does however show that the tasks of communication and education are far from complete: many mothers were still laying their infants prone, and either had not heard or did not heed the advice so widely given. The National Advisory Body stresses the importance of conveying advice on the terms of the parents not the professionals. Following the recent decline in SIDS, there seems to have been a more marked shift downwards in the social pattern of incidence. Today's main associations with SIDS—such as smoking, poverty, and unemployment—are far less amenable to change than were sleeping position and thermal environment, and further reductions will not be achieved so readily. Despite criticisms of the risk related intervention promoted in Sheffield in the 1970s,⁶ one possible way of tackling the hard social core of cot deaths now remaining might be through targeted support from health visitors of families identified by newly validated risk factors.

Although we think we have identified the main environmental risks for cot death, we still have little idea of the mechanisms by which they operate. It is now for the physiologists to catch up with the environmentalists. Meanwhile, pathologists continue their assiduous search on many fronts—histology, biochemistry, microbiology, immunology—for specific disorders that can be chipped out of the unresolved block of deaths we call SIDS. The role of pathologists in the investigation of unexpected infant deaths is crucial, but so far their contribution to the elucidation of new causes has been limited, the most notable, the identification of unsuspected metabolic disorders, accounting for about 2% of deaths that were previously unexplained. After a comparatively small advance from so much effort major breakthroughs in this sector may now seem less likely.

- 1 National Advisory Body for Confidential Enquiry into Stillbirths and Deaths in Infancy. *Third annual report*. London: Department of Health, 1996.
- 2 Fleming P J, Blair P S, Bacon C J, *et al*. Environment of infants during sleep and risk of the sudden infant death syndrome: results of 1993-5 case-control study for confidential enquiry into stillbirths and deaths in infancy. *BMJ* 1996;**313**:191-5.
- 3 Blair P S, Fleming P J, Bensley D, *et al*. Smoking and the sudden infant death syndrome: results from 1993-5 case-control study for confidential enquiry into stillbirths and deaths in infancy. *BMJ* 1996;**313**:195-8.
- 4 Royal College of Pathologists. Minimum guidelines for post-mortem investigation of post-neonatal infant deaths or sudden unexpected deaths in infancy. *Guidelines for post-mortem reports*. London: Royal College of Pathologists, 1993: appendix 1, 7, 10.
- 5 Emery JL. Infanticide, filicide and cot death. *Arch Dis Child* 1985;**60**:505-7.
- 6 Carpenter R G, Gardner A, Jepson M, *et al*. Prevention of unexpected infant death: evaluation of the first seven years of the Sheffield intervention programme. *Lancet* 1983;**i**: 723-7.