Cot death controversies

Uncertainty as to the cause or causes of sudden and unexpected infant death and difficulty in excluding the possibility of infanticide, even after the performance of a 'complete' autopsy, was graphically illustrated in the London courts recently.[1] A mother wrongly convicted of killing her two sons had her conviction quashed, and the eminent paediatrician who had given the court erroneous statistical information was struck off the medical register for serious professional misconduct.

In the developed world cot death or sudden infant death syndrome (SIDS) is the commonest cause of death among infants between 1 week and 1 year of age. Although it occurs in all countries and socioeconomic groups, the rates vary widely.

The particular poignancy of cot death and the elusiveness of its cause have engendered a vast amount of research in the Western world. Cot death associations have sprung up in many countries, and international conferences on the subject are held regularly. The list of proposed and discredited causes is a long one, including maternal overlaying, accidental mechanical suffocation, overwhelming viral or bacterial infection, a large thymus and hypersensitivity to cow's milk, among many others. But several risk factors have been identified – the 2 - 4-month age group is at highest risk, and males are more commonly affected. Deaths are more frequent in winter and over weekends. Rates are higher in poorer households and among infants with young mothers, and rates increase with parity, low birth weight, and maternal smoking and drug-taking.[2]

Because of the need for a specialised autopsy to fully exclude other causes and because of the difficulties in ruling out suffocation or wilful infanticide, expert committees have attempted various definitions of SIDS. The most recent, in July 2004, defines SIDS as: 'The sudden unexpected death of an infant under one year of age with onset of the fatal episode apparently occurring during sleep, that remains unexplained after a thorough investigation including performance of a

complete autopsy, review of the circumstances of death and the clinical history'.[3]

It seems that three converging factors operate in these tragedies: a vulnerable period, a vulnerable infant, and some additional precipitating factor; but the nature of this additional factor remains the subject of endless controversy.

What makes an infant vulnerable?

Several factors that make the infant more vulnerable to SIDS are now well accepted.

Sleeping position is considered crucial. The fall in SIDS mortality in many countries over the past two decades can be attributed almost entirely to campaigns encouraging parents to place their infants on their sides or backs rather than in the prone sleep position. Scragg and Mitchell[4] provide evidence that sleeping on the side is less safe – it doubles the risk of SIDS compared with sleeping supine (relative risk of side v. back 2.02). This is probably because infants on their sides then turn to the prone position. Infants who usually sleep supine but are placed in an unaccustomed prone position are at very high risk of SIDS.[5]

Excess bedding and clothing have been shown to increase risk of SIDS in infants sleeping prone but not in those sleeping on their side or back. Some 15 - 20% of infants who die of SIDS are found with bedclothes covering their heads. Covering of the head might cause death by overheating or by forcing the infant to re-breathe expired gases or, as suggested later, to breathe toxic gases.[5]

Infants of mothers who smoke are at almost 5-fold greater risk than infants of non-smoking mothers. This risk is probably due predominantly to the effect of tobacco smoke *in utero* rather than to inhalation of environmental tobacco smoke postnatally.

Hannah Kinney and co-workers[6] at Harvard recently found brainstem abnormalities in infants dying of SIDS. SIDS infants had a deficiency of serotonergic receptor binding in the medulla compared with controls. Kinney *et al*. have also incriminated maternal alcohol use in the periconception period and the first trimester as a possible cause of these changes.

The protective effect of breastfeeding is less clear. Some studies have shown a decreased risk of SIDS, others have found no difference, or the difference has been due to socioeconomic factors – breastfeeding in developed countries being associated with economic advantage.[5]

An unexpected finding has been that the use of pacifiers (dummies) is associated with a reduced risk of SIDS.[5] This finding – apparently confirmed – must be balanced against the possible detrimental effects of pacifiers, such as reduced breastfeeding and increased risk of otitis media.[5]

In the past there was concern that vaccination might cause SIDS, as the peak age for SIDS, viz. 2 - 4 months, coincides with the time for vaccinations. Repeated studies have shown that vaccination is not associated with sudden death.[5]

In the case of some SIDS deaths a clear cause can be identified, for example overwhelming viral or bacterial infection, errors of metabolism or cardiac arrhythmias.

An important cause in the latter group is the long QT syndrome. In this condition, which is often familial, a defect in the transmembrane transport of sodium and potassium results in cardiac arrhythmias, causing fainting attacks or even sudden death from ventricular fibrillation. Much is now known about the genetics of the long QT syndrome, but its frequency as a cause of sudden infant death remains uncertain. Because it can recur in subsequent children it should be mandatory to perform ECGs and if possible genetic studies on the parents of a cot death victim to exclude this syndrome. Certainly any infant who suffers a near miss or life-threatening episode must be investigated fully for this defect.[7]

In the year 2000 systemic *Helicobacter pylori* infection was incriminated as a cause of cot death and aroused heated debate (both for and against). This hypothesis finally appears to have been dismissed.[8]

Following widespread campaigns promoting the supine or side sleep position for infants and avoidance of overheating and exposure to cigarette smoke, SIDS rates have dropped in many countries, but now appear to have levelled off at a rate of about 1 in 2 000 live births. However it is clear that infants still die of SIDS in every society, and in the most optimal environments.

Bed-sharing

An important and unanswered question pertains to the role of bed-sharing, i.e. whether it is protective or potentially harmful for an infant to sleep in bed next to an adult.

In Western industrialised society, where solitary infant sleep is considered a normal and desirable arrangement, babies traditionally sleep alone and separate from the parents. Most of the recent literature referring to co-sleeping considers it a risk factor. For example, Nakamura *et al*.[9] reviewed 515 deaths among children aged under 2 who died in adult beds. Three hundred and ninety-four deaths were due to entrapment in the bed structure, and 121 were reportedly due to the parent lying on the child. These numbers were culled from all 50 states in the USA over an 8-year period, and no denominator data were provided. Nevertheless the authors warned unequivocally about potentially fatal hazards associated with children under 2 years of age sleeping in adult beds.

An Australian investigation[2] concluded that babies who shared their mother's bed were at significantly increased risk of SIDS only if the mother smoked. Additional risk factors for infant deaths while co-sleeping are alcohol, an obese parent, and an unusual sleeping arrangement, often on a sofa, with the

baby getting trapped between the back of the sofa and the parent.[10]

On the other hand, for the vast majority of non-Western people today bed-sharing is the predominant sleeping arrangement.[11] Infants are brought up in a busy environment, almost invariably sleep in the mother's bed, and for many hours of the day are in close contact with her body.

A study conducted in Cape Town[12] found that 94% of black infants slept with their mothers, compared with only 4% of white babies. Black families generally do not use or cannot afford cots or separate rooms for infants.

The SIDS Family Association in Japan conducted a survey on risk factors related to SIDS, and found that 93% of babies slept in the same room as an adult, with most of them sleeping at the adult's side. The SIDS rate in Japan at that time was 0.48 per 1 000 live births, among the lowest in the world, and the Association believes that co-sleeping should be promoted.[13]

The theory that co-sleeping may actually be protective against SIDS centres around the fact that humans are born 'neurologically immature', i.e. with a markedly undeveloped central nervous system compared with many other mammals. Centres in the brain for controlling basic functions such as breathing are still immature. Some infants may therefore slide into a state of deep sleep in which they stop breathing. Mosko *et al*.[14] showed that infants who sleep next to their mothers spend less time in the deep stages of sleep (stages 3 and 4) than those sleeping alone. Contact with the parent during sleep constantly stimulates the infant through vocalisation, body movement, radiant heat and respiratory sounds. In fact, infants who share their parents' bed exhibit synchronous arousal and co-ordination of sleep stages with the parents.

It would seem entirely reasonable biologically for the young infant to sleep in close proximity to its mother, and perhaps this is actually protective. The evidence suggests that there may be potential benefits to bed-sharing which cannot be overlooked, and this requires further study in communities where co-sleeping is common.

The 'toxic gas' theory

The hypothesis that cot deaths might be caused by inhalation of toxic gases was first suggested by a forensic chemist, J Sprott, in New Zealand in the early 1980s. He took the view that the epidemiological data pointed to environmental factors rather than a medical cause. He postulated that cot death had only one cause – gaseous poisoning. According to him the gas or gases had little or no odour, were more dense than air, and interrupted the baby's nervous system, causing cessation of breathing. Sprott proposed that the gas or gases were generated by microbiological activity on something in the baby's cot.[15]

Independently, an English industrialist, Peter Mitchell, and Barry Richardson, a consulting scientist, came to a similar conclusion, incriminating arsenical biocides used in PVC sheeting. Certain fungi thrive on these biocides and generate a toxic gas, arsine, from this substrate. Richardson engaged in a research study in which he claimed to demonstrate that the gases involved were the tri-hydrides (and/or their alkyl homologues) of the elements phosphorus, arsenic and antimony, all elements in group Vb of the periodic table of elements. This discovery had its roots in the work of an Italian chemist, Gosio, in the 1880s.

Richardson's hypothesis, and his urging that only new mattresses or mattresses covered with polythene (which does not contain these elements) be used, received much publicity in the UK. Highly controversial discussion began in the *Lancet* in 1990. In the same year the Turner Committee, appointed by the British Department of Health, negated Richardson's findings, and later the Limerick Committee of Experts appointed to investigate cot death theories also could not substantiate the toxic gas theory.

Meanwhile, Dr Sprott in New Zealand promoted the use of mattresses and slip-on mattress covers known to be free of phosphorus, arsenic and antimony, and claimed that this policy has had a 100% success rate.

SIDS experts have dismissed the toxic gas theory and have labelled its authors 'overzealous proponents of a pet theory'. One might readily go along with the views of such authorities were it not for a few nagging, unanswered questions. The first is Sprott's contention that there has not been a single cot death among tens of thousands of infants sleeping on mattresses covered to his specifications, as against now more than 670 cot deaths among infants sleeping in other ways. This figure has apparently been authenticated by the New Zealand Health Ministry, yet no reference to this discrepancy is to be found in the medical literature.

Secondly, Richardson wrote a detailed refutation of the findings of the Limerick Report, but there has been no documented response from the British Department of Health.

Kapuste,[16] a strong proponent of the toxic gas theory, describes the entire controversy in a detailed article. But other than this reference, why does the topic of cot death and toxic gases make no appearance whatsoever in the 'official' medical literature?

Although only circumstantial, there are other factors that could favour the 'toxic gas' theory. One is the increasing frequency of SIDS among second and subsequent siblings, when infant mattresses might be progressively older. It has also been shown that cot deaths are more common in infants sleeping on old mattresses.[17] What better way of explaining a rare but well-known occurrence, viz. cot deaths occurring in twins at more or less the same time?

The southern African situation

What is the southern African situation with regard to SIDS? For a number of reasons the incidence in developing countries generally is difficult to establish.

Firstly, detailed autopsies by paediatric pathologists are only available at larger centres, and secondly the majority of infants reported as having died suddenly show advanced disease at autopsy, such as gastroenteritis or meningitis. These diseases may have been treated inadequately, medical treatment may not have been sought, or medical care may have been inaccessible. Accurate estimates of incidence are therefore difficult to obtain unless special population studies are carried out.

In a Cape Town study[18] of deaths below the age of 4 years, the respective incidence of SIDS was found to be 1.06 per 1 000 live births for whites, rising to 3.41 for coloured infants. At that time black infants could not be included in the study because of difficulties with home visiting.

Data from the South African Central Statistical Services[19] for the different ethnic groups for the years 1989 - 1990 showed cot death rates of 1.11 in coloured, 0.29 in white, 0.09 in Asian and 0.07 in the black population per 1 000 live births. While there were undoubtedly differences in classifying and reporting SIDS as a cause of death, it can be seen that there is a reduced relative risk associated with the black and Asian populations when compared with the coloured and white population in South Africa.

That SIDS is less common in black households is borne out further by the findings of Wolf and Ikeogu.[20] The latter conducted a well-designed prospective study on cot deaths in Bulawayo, Zimbabwe, in 1996, and found an incidence of only 0.2/1 000. The population under study was a black township with high unemployment and overcrowding, i.e. with strong risk factors for SIDS.

It is also apparent that there is a notably increased relative risk in the coloured population similar to that found in the Cape Town study. Factors that could be important are higher smoking and drinking rates among coloured women, and the high incidence of low birth weight (15 - 25%) in that group. Breastfeeding tends to be poorly sustained. In contrast, smoking and alcohol intake are unusual in black women, breastfeeding is virtually the norm, and low birth weight rates are much lower – to the order of 6 - 8%.

So what advice should we give to parents, in particular about co-sleeping and mattress coverings? Are these practices harmful, inconsequential or beneficial in the context of SIDS? There is as yet no epidemiological evidence to support a conclusion. The subject cries out for local research in South Africa.

M A Kibel
Emeritus Professor of Child Health, University of Cape Town
12 Winchester Avenue
Bishopscourt
Cape Town

C D Molteno
Professor of Mental Handicap
Department of Psychiatry
University of Cape Town

R De Decker
Division of Human Genetics
School of Child and Adolescent Health
University of Cape Town and
Red Cross Hospital
Cape Town

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