

## PERSONAL VIEW

## Ethnicity and the aetiology of sudden infant death syndrome

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In spite of current reports of an encouraging decline in the incidence of sudden infant death syndrome (SIDS), the sudden and unexplained death of an apparently healthy baby, in this and other western countries,<sup>1</sup> it is still the single most important cause of death between 1 and 12 months. Especially highlighted have been its associations with social disadvantage, poor home environment, inadequate antenatal care, smoking, low birth weight, higher winter incidence, and greater male vulnerability.<sup>2,3</sup> An anthropological perspective suggests that we investigate the incidence of SIDS from a broader historical and geographical perspective, recognising its continuing rarity in many parts of the world and its association with western patterns of child rearing.

### Ethnicity and the incidence of SIDS

One intriguing, though relatively poorly explored, aspect of the epidemiology of SIDS is its variable incidence in relation to geographical and ethnic factors. For example, in Chicago from 1975–80 there was a much higher incidence in the black population (5.1 per 1000 live births) than among white (1.2) and hispanic (1.3) people.<sup>4</sup> Until recently the South Island of New Zealand had one of the highest incidences of SIDS in the developed world (7.6 per 1000),<sup>5</sup> yet in Finland, a rate of only 0.41 per 1000 was described in 1969–80.<sup>6</sup> A very low incidence was reported in Hong Kong in 1987 (0.29 per 1000),<sup>7</sup> corroborating many unpublished clinical accounts of the rarity of SIDS among the Chinese and other ethnic groups in South East Asia. In Japan, published rates then for SIDS were very low, of the order 0.15 per 1000.<sup>8</sup> In Britain then an incidence of 2–4 per 1000 was widely quoted.<sup>9</sup> In the USA, the lowest incidence has been reported among oriental Americans (0.51 per 1000)<sup>4</sup> and the highest (7 per 1000 between 1984–8) in Native American infants in Washington State.<sup>10</sup> In England and Wales in the late 1980s two important reports (by Balarajan *et al*, using data from the Office of Population Censuses and Surveys of mother's country of birth to look into ethnic differences in infant deaths in England and Wales from 1982–5<sup>11</sup> and Kyle *et al* who reported on SIDS in Birmingham 1981–3<sup>12</sup>) showed that in spite of higher postneonatal mortality among Asian

people (those coming from India, Pakistan, and Bangladesh) SIDS occurred less often among their infants than in those of mothers born in Britain and the Republic of Ireland. Other more recent studies from London and Yorkshire have extended these findings that in Britain SIDS rates are significantly lower in Asian families than in those belonging to the country's ethnic majority white population (L Hilder, C Bacon; personal communications).

### A paradox

Yet many of these examples provide something of a paradox. Asian mothers in Britain are reported poorer, have more children with shorter intervals between pregnancies, and generally live in less satisfactory housing than ethnic majority mothers,<sup>12</sup> all factors that might be expected to increase the risk of SIDS. In the Chicago study Hispanics shared many adverse social and environmental factors of black families yet their incidence of SIDS was four times lower.<sup>3</sup> Overcrowding, lack of breast feeding, and a high incidence of respiratory infection in Hong Kong could also have been considered major risk factors.<sup>13</sup> Is it therefore possible that protection can be conferred in the midst of potential high risk, just as in other situations there might be increased vulnerability? Ways in which infants are looked after are an obvious consideration. Might culturally associated infant care practices provide an important clue to sudden infant death? Of special interest in this context is a study by Grether *et al* in 1990 showing that the longer oriental families lived in the USA, the greater became the risk of SIDS, implying some influences from local socioenvironmental factors.<sup>14</sup> In 1985 when commenting on the very low rate of SIDS in Hong Kong one of us speculated 'that the possible influence of lifestyle and care taking practices such as posture and babies never being left alone were being under estimated in preference to more exotic and esoteric explanations for the aetiology of SIDS'.<sup>13</sup>

We believe there is much to learn about the aetiology of SIDS by continuing to examine its presentation in various geographical and ethnic environments. Indeed it is salutary to consider that, at a time when so much emphasis is being put into genetic and

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molecular explanations for disease, perhaps a clue to the cause of SIDS rests in how human infants are cared for in cultures which have been relatively little influenced by either western patterns of nuclear family life or a 'medical model' of infant care. It is the latter which has in the last 20 years recommended the practice of front sleeping, guidance which has now been reversed. One way of investigating how these patterns vary has been to look at the experience of Asian families living in Britain.

### **Ethnicity and the care giving environment**

That Asian families living in Britain generally find themselves in a relatively poorer socio-economic environment is well known<sup>12</sup> but the care giving environment seems not to reflect this relative material poverty. The opposite in fact. Studies from Cardiff<sup>15</sup> and Birmingham<sup>16</sup> describe very different child rearing practices and immediate sensory environments for Asian babies. They are more likely to sleep in, or close to, their parents' bed at night than white babies. Throughout the day the extended family network provides an almost constant source of contact with adults and other family members. Asian households are in general busier: babies are frequently picked up, contributing to creating a home environment that is often in stark contrast with that found in the nuclear family unit of mother-father-child or mother-child which is typical in white families in this country. In these there is more emphasis on getting babies established into routines very early in life, almost as it were to hasten independent behaviour, encouraging a quiet environment. Leaving babies undisturbed for long periods both by day and night meets desire for privacy and independence of parents. Asian babies are characterised by their mothers as vulnerable and by implication in need of protection: the striving for early independence is not an issue.<sup>15</sup>

As has so vividly been described by James McKenna, an American physical anthropologist, a rich sensory environment deriving from babies being constantly close to other humans is common to most of the world's traditional cultures where customs are almost ubiquitously directed to protecting the baby from hostile external forces.<sup>17</sup> It is worth reminding ourselves of the Welsh shawl that until recently was a way mothers in Wales had of carrying their babies while continuing their daily chores, creating a very close relationship between the two. In western societies the perception of vulnerability now is different, following more a medical paradigm with protection against illness, especially infectious diseases, being more important: less intimate contact between baby and care giver could be perceived to help this. Other influential voices in infant care, such as Truby King, suggested earlier this century that too much physical contact could be harmful to infant development. The length of time

that industrial societies have occupied their niche on this planet is but a tiny fraction of the whole span of human existence. Care giving environments that encourage the close symbiotic link between immature young and care giver to persist is what has been needed for continued survival of any species: the human provides no exception. Placing babies to sleep alone for relatively long periods in a quiet environment is in evolutionary terms new. What is now exciting is to discover how close sleeping between mother and baby results in more arousals and fewer periods of deep sleep in both.<sup>18</sup>

### **Relevance of care practices to SIDS**

What relevance might these anthropological perspectives have to SIDS in this and other western countries? It is a remarkable fact that since 1988 infant mortality in Britain, and the white population of New Zealand, Australia, and other western European countries has declined significantly, due in large part to a reduction in the incidence of SIDS. This fall seems to have coincided with intense national education campaigns (spearheaded in this country by the Foundation for the Study of Infant Deaths 'Reduce the Risks' campaign<sup>19</sup> and that of the Department of Health<sup>20</sup>) to educate health care professionals and parents how to reduce the risk of SIDS, especially by discouraging prone sleeping in preference to the back or side position, avoiding overheating the baby, avoiding smoking in pregnancy or anywhere near the baby after birth, and prompt referral of a baby to primary care if there are any health worries. Avoidance of the prone sleeping position is now considered to be the single most important factor contributing to the current decline in the incidence of SIDS.<sup>21</sup> But how supine posture during sleep lessens the risk of SIDS has yet to be explained. Might it relate to less risk of overheating? Less chance of inhaling allergens from bedclothes? Less chance of suffocation? Health visitor colleagues are already commenting on the frequency with which mothers of infants sleeping on their backs report 'fractious' babies: may it be that back sleeping infants sleep for shorter periods of time and experience more arousals and fewer periods of deep sleep? Or might this change in sleeping posture even be a surrogate for something else that might be lowering risk?

### **A model for the aetiology of SIDS**

Although neither the pathophysiology nor aetiology of SIDS is known it is probable that death is somehow related to failure of respiratory regulatory mechanisms that ultimately lead to insufficient respiratory drive. The immediate sensory environment of a baby – provided by touch, warmth, sound, inhaled carbon dioxide, movement of a parent's ribcage, feel of heartbeat, etc, are likely to be important in the regulation of breathing,<sup>17</sup> acting maybe through the brain stem reticular

formation. Between 2 and 6 months there appears to be an instability in the regulation of breathing as cortical brain mechanisms begin to dominate over earlier brain stem systems. In this way, a primitive and relatively inflexible system is replaced by one in which some learning is required for its evolution. According to McKenna *et al* other unique demands are placed on this immature respiratory system in intentional crying and language.<sup>17</sup> Cardiorespiratory reflexes and chemoreceptor systems probably help to restore homeostasis after forces that upset the regulation of breathing. From this perspective SIDS is seen as more a result of subtle disordered physiology than a disease in the accepted pathological sense. The delicate control of breathing is as a tight rope walker with destabilising influences continually acting to upset a delicate balance but with counterbalancing adjustments in normal circumstances equally active. Any significant disturbance to this balance would be expected to upset the control of breathing, leading in some instances to death. We speculate also that each individual baby has a unique template for combinations of destabilising influences and inadequacies of compensatory responses that might become lethal: but in any individual baby these balances and imbalances are unpredictable. All babies are therefore theoretically at risk. Epidemiological risk factors are linked in some way with the destabilising influences such that if these can be avoided or modulated the risk of death should be reduced. The combination of the destabilising forces maybe imagined as pieces of a jigsaw puzzle: it is only when the puzzle is completed with individual pieces coming together that death will take place: or as Bergman has so elegantly put it: 'SIDS is like a nuclear explosion where a critical mass must be attained before the event is to occur'.<sup>22</sup> This critical mass is made up of biological, social, and behavioural factors.

Applying this model, could social factors linked with cultural attitudes to infant care be considered important pieces of the jigsaw – stabilising or destabilising influences? Could long periods of being alone in a quiet environment be a risk factor depriving infants of sensory cues that may help regulate breathing, interfering with physiological adjustment? It is not uncommon that a baby is found dead in room away from its parents. In the animal world separating the young from the mother for long periods until weaning has been established is the exception: it is *Homo sapiens* in western society that provides this exception with babies typically placed for long periods in their own rooms quite soon after birth, a sensory poor environment with little contact with anyone. SIDS also happens to be uncommon in societies where, for social or cultural reasons, babies are not left alone. Solitary sleep has come recently in evolution. The natural care giving environment in which the human species, along with other mammalian orders, has evolved, with the young sleeping in close contact with

the mother or other care giver, provides cues optimally provided from a sensorily rich environment – an environment of physiological adaptiveness perhaps to help stabilise breathing and diminish the chances of a respiratory crisis.<sup>18</sup>

But during deep sleep respiratory drive and rhythm seem to be especially dependent on sensory input and chemical stimuli to override biological deviations in breathing control.<sup>18</sup> If, for whatever reason, such a baby has a lapse in breathing, there may be less opportunity for self correction. As Lipsitt has described SIDS babies: 'it is as though they had simply forgotten to breathe, unable to arouse themselves to take the next breath and so continue the respiratory cycle'.<sup>23</sup> Infants at greater risk, for example, through respiratory infection, premature birth, previous nervous system damage, might be expected to be particularly vulnerable to these lapses. Vulnerability might be further conferred by deficient growth before birth that compromises normal regulatory mechanisms. Prolonged periods of separation may also blunt parental awareness of minor illness that, undetected, may contribute to a baby's death. The Leicester studies remind us that in babies sleeping at home the development of mature temperature rhythms are significantly delayed in babies statistically more at risk of SIDS.<sup>24 25</sup> They have also shown that babies can demonstrate non-specific symptoms several days before fever becomes clinically obvious.

Against this background could it be that publicity advising parents to place babies to sleep on their backs has had another effect; that parents are now checking their babies more often, spending more time with them; leaving them alone much less; responding more promptly to their needs? Even detecting earlier signs of illness and providing a generally richer sensory environment that helps the baby through a difficult transition of changing breathing control? Is this a 'proxy' for back or side sleeping? It is significant that populations where SIDS is uncommon not only have their babies lying on their back, but also provide a very rich and stimulating sensory environment. Any intervention programme that has its aim to reduce the incidence of SIDS has to consider the broad range of care practices and the inter-relationships of such practices which could also reduce risk.

### Conclusion

Ethnicity in this paper has perhaps rather simplistically embraced a package of social, cultural, and educational influences modulated by biological differences between populations, and in particular its relationship to different ways of caring for young infants. Applied to SIDS the fact that in so many developing countries of the world and among certain ethnic groups in developed countries SIDS is not recognised, an event so catastrophic as to be impossible to evade description should it be occurring to any significant extent, can surely mean that

protection must be being conferred upon the very vulnerable baby. Whether we will ever be able to explain how this protection is provided is another matter. Two of the practices currently regarded as risk factors in Britain, front sleeping and cigarette smoking, are directly associated with practices of western society. In New Zealand, artificial feeding is also seen as a risk factor. The pattern of incidence of SIDS is thus associated both with western patterns of life, and a medical model of infant care. We know that Bangladeshi infants in Britain have always slept on their backs, and they are less likely to be exposed to cigarette smoke: it will be interesting to track the new epidemiological pattern of SIDS now that the majority of infants sleep on their backs.

The studies of James McKenna *et al* of mothers and babies sharing the same bed during sleep showing that there is a mutual experience of more arousals and fewer periods of deep sleep are excellent examples of the type of multidisciplinary research needed in this field.<sup>18</sup> Surely it is not wise for a young baby to be left alone for long periods during a potentially vulnerable stage of early development. These considerations inevitably lead into the current contentious issue of co-sleeping. Studies by Mitchell and his colleagues in New Zealand have highlighted possible dangers of bed sharing in the Maori population where SIDS occurs far more often than in the two other major ethnic groups in New Zealand, Pacific Island peoples and those principally of European origin.<sup>26</sup> It is important, however, to note that their more recent studies have concluded that smoking is probably more determinant of risk than bed sharing itself.<sup>27</sup> In this debate it is also paramount to define the type of social sleeping practices: that a clear distinction is made between babies sleeping always within touching distance in the same room as their parents and actual bed sharing, and also not blindly to transcribe or adopt practices from one culture to another. In the area of SIDS, epidemiology and public health are closely allied, with the emergence of apparent risk factors being translated relatively quickly into advice. It is vital that epidemiologists recognise and understand the broad cultural context of their observations, and the way in which infant care practices vary and inter-relate. Health professionals working in primary care have to present advice which is appropriate and feasible, and take account of both medical knowledge and the perceptions and experience of those to whom the advice is being given.

Is this not the time to address some fundamental attitudes in this country about how we look after young infants? Just as at one time health professionals encouraged mothers to lie babies prone so there was also the attitude that babies needed to establish independence early in life. What better way to achieve this than by encouraging the baby to endure increasingly long periods of solitude soon after birth? We have changed advice on posture; why not change it now relating to where babies should sleep in the early months

after birth? It is surely unreasonable to expect a nine month symbiotic prenatal physical bond between mother and intrauterine baby to be so abruptly served. Jelliffe and Jelliffe's concept of the 'extero-gestate fetus' the biological dependency of the baby on the mother for protection, warmth and food, is highly relevant to this debate.<sup>28</sup> Being left alone for long periods could lead to a worsening of a minor clinical illness, such as a cold, snuffles or temperature which, if detected early, could be approached and dealt with. An enhanced physical presence may make the mother more sensitive to small changes in behaviour such as might anticipate a febrile illness. Why can we not therefore now *positively encourage* mothers to have their babies in the same room at night for the first 6–9 months of life, and that the cot where they sleep by day should also be close by? There is no proof that this will protect the baby against SIDS. But at the same time there is no evidence that this care giving practice is harmful. Is it even possible that such a change in attitude in caring for young babies might also have some beneficial influence on later behavioural and emotional problems in childhood.

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### Human herpesvirus-6 and the brain

Work continues in Japan on the role of human herpesvirus-6 (HHV-6) in childhood disease (see *Archivist* 1992: 500). A case report in *Archives* in 1992<sup>1</sup> is now followed by a description of 21 children aged between 5 and 15 months who presented with convulsive seizures (Sadao Suga and colleagues, *Annals of Neurology* 1993; 33: 597-603).

All 21 children had the clinical features of exanthem subitum (roseola infantum) and HHV-6 infection was confirmed by isolating the virus from blood (six patients), by demonstrating a significant increase in antibody titre during the illness (five patients), or both (10 patients). The rash of exanthem subitum appeared two to four days after the start of fever. In two children convulsions occurred on the day before the fever. One of these had a single convulsion lasting for one hour and the other had a series of six convulsions each lasting for less than 15 minutes. Ten children had a single generalised febrile convulsion lasting less than 15 minutes and two had two such episodes during the illness. Two had prolonged left sided convulsions lasting for one and two hours followed by transient left sided hemiparesis. One child had a single generalised febrile convulsion lasting for 45 minutes. Four had focal seizures and prolonged unconsciousness and were diagnosed as having encephalitis/encephalopathy (E/E).

Nine of the 17 children without E/E had cerebrospinal fluid examined with normal findings in all of them. Of the four patients with E/E three had a modest lymphocytic pleocytosis. HHV-6 viral DNA was detected in the cerebrospinal fluid using the polymerase chain reaction in six of 11 patients tested including three of the four with E/E.

The patients with E/E all had abnormal EEGs, two of them showing periodic complexes in the temporal regions as is seen in herpes simplex virus encephalitis. These two patients both made a complete recovery. One child with E/E (the one previously described in *Archives*<sup>1</sup>) had a flat EEG by day 2 and died after 21 days. Another had 'an occasional epileptic seizure' on follow up. The remaining 19 children made a full recovery without sequelae.

Abnormalities on cerebral computed tomography were found in all four children with E/E. One showed a low density area in the left frontotemporal region on the first day of the illness, one (the one who died) had a normal scan on day 1 but low density lesions in both hemispheres and bleeding into both Sylvian fissures on day 9. The two others also had normal scans initially but later developed low density areas and atrophy.

It has long been held that exanthem subitum is an important cause of febrile convulsions. In most children it is benign but occasionally it turns violent. Why that is so will no doubt be the subject of further research.

ARCHIVIST

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