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## Sudden Infant Death Syndrome

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### Abstract

Sudden infant death has only recently begun to attract widespread publicity and medical investigation. It is still largely ignored by the standard paediatrics textbooks and the only description of it given in formal lectures to Edinburgh medical students comes in the Forensic Medicine course. This perhaps helps to serve one useful purpose in that it may be an indication as to why the sudden infant death syndrome is still almost completely unexplained. These unfortunate children are usually found dead and, if they reach hospital at all, it is only as far as the accident and emergency department on their way to the mortuary. Most go directly to a coroner's or procurator fiscal's post-mortem, and are thus relatively insulated from the research-orientated clinician's attention and interest.

This syndrome is certainly no new phenomenon. Up to the end of the last century doctors were satisfied that these deaths were due to "teething", a wide category that included many deaths for which an adequate explanation would now be found. The 1839 Registrar-General's returns showed a total of 5,016 infant deaths due to this "cause". The fashion then changed towards attributing them to "status thymolympaticus", although exactly why seems to be unknown.

Gradually, however, the position is becoming somewhat clearer. Werne and Garrow in the United States produced some of the first significant papers during and immediately after the second world war. In Britain Professor Sir Samuel Bedson and Dr. F. E. Camps (1951) submitted a memorandum to the Ministry of Health, estimating an annual toll of 200 children, and suggesting an official investigation which finally bore fruit under Professor Banks in the 1965 Report.

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# SUDDEN INFANT DEATH SYNDROME

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Sudden infant death has only recently begun to attract widespread publicity and medical investigation. It is still largely ignored by the standard paediatrics textbooks and the only description of it given in formal lectures to Edinburgh medical students comes in the Forensic Medicine course. This perhaps helps to serve one useful purpose in that it may be an indication as to why the sudden infant death syndrome is still almost completely unexplained. These unfortunate children are usually found dead and, if they reach hospital at all, it is only as far as the accident and emergency department on their way to the mortuary. Most go directly to a coroner's or procurator fiscal's post-mortem, and are thus relatively insulated from the research-orientated clinician's attention and interest.

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The 1st International Conference held in Seattle, Washington, in 1963 firmly put the sudden infant death syndrome on the medical map, and although some still persist in ascribing all these deaths to accidental suffocation, overwhelming infection or infanticide, most will now accept the existence of this syndrome as a distinct entity.

If any worthwhile comparable epidemiological studies are to be made on this topic, then a definition of the syndrome must be universally accepted. There is considerable lack of agreement even on what to call it: cot or crib death, sudden unexpected death, sudden unexplained death in infancy, undiagnosed death in infancy and many others have all been suggested and used, but the 2nd International Conference on Causes of Sudden Deaths in Infants (1969), after much discussion, finally settled for Sudden Infant Death Syndrome (SIDS) as the term causing least confusion. Pathologists have yet to come up with any consistent finding and using this as a basis Dr. J. B. Beckwith produced the following definition at this Conference: "the sudden death of any infant or young child which is unexpected by history and in which a thorough post-mortem examination fails to demonstrate an adequate cause for death".

It was also proposed that the minimal acceptable investigation into one of these deaths should include:

1. Adequate history
2. Gross examination including thorax, abdomen, brain, entire larynx ( $\pm$  spinal cord)
3. Blood culture
4. Histological examination including brain, liver, lungs, heart, kidneys and other organs as indicated by 1 and 2.

5. Ancillary studies (toxicological, chemical, special cultures, virological studies and so forth) as indicated by results of above.
6. Counselling of family.

Most of the recently-reported epidemiological investigations have used, as far as was possible, these criteria.

The first thing to determine is its actual incidence in the community. How common is it? Is this a real problem or is it just another weird and wonderful syndrome that makes good reading in the journals, but which few are ever likely to see? One major difficulty has been that something "appropriate" was usually recorded on death certificates and SIDS or its equivalent was not used, making figures impossible to obtain from this source. The Registrar-General does now allow the use of the term "cot death syndrome". The 1965 M. o. H. report estimated that 83% of deaths certified by coroners as due to "respiratory causes" were in fact "cot deaths".

Most of the papers written on SIDS have come from Western cultures and they show a remarkable uniformity (Table 1).

Very similar figures were reported by Houstek (1970) from Czechoslovakia, who thought that the incidence was about the same in Yugoslavia, Poland and the U.S.S.R., although little data has come from these nations. Doctors working in underdeveloped countries have stated that sudden death in infancy is common, but no post-mortem is

usually available and any comparison of figures is therefore impossible.

In reviewing the statistics in Table 1, Dr. Marie Valdés-Dapena remarked at the 1969 Conference that it would appear that the incidence of SIDS increased with urbanisation and that the lowest figures seem to occur in rural areas.

The survey showing the highest incidence was that conducted in Canada (Steel 1970), closer examination of the figures revealing that it increased the further north one went. This would suggest some relationship with the temperature or weather conditions in general.

Froggart et al (1971) have conducted a major investigation in the U.K. by studying SIDS in Northern Ireland. Their results agree with most others by showing an excess of deaths during the winter months. This peaking was greater during the first year of study (August 1965 to July 1966) and it is interesting to note that epidemics of Influenza type B and type A2 occurred in this area during January to February 1966 and February to March 1966 respectively. In the Southern Hemisphere Australian figures show a peak in June, again during the colder season. This winter increase is not due to the relatively unimportant monthly fluctuations in live births.

There is some disagreement as to whether the incidence varies with the day of the week. Some state SIDS to be commoner over the weekend, suggesting an element of neglect, but other studies do not confirm this.

TABLE 1 (Valdes-Dapena 1970)  
(re-arranged)

Author	Year	Place	No. / 1000 Live Births	Subjects
Ministry of Health (GB)	1965	England & Wales	1.4	All infants
Carpenter	1965	England & Wales	2.2	
Peterson	1966	Seattle	(2.87 (2.71 4.66)	Caucasians Non-whites (Negro and American Indians)
Steel et al.	1967	Canada	3.00	All infants
Froggart et al	1968	Northern Ireland	2.3	All infants
Valdés Dapena et al	1969	Philadelphia	2.55 (1.41 4.32)	Caucasians Non-whites (negro)
Fitzgibbons et al	1969	Olmstead Co., Minn.	1.2	All infants

Virtually all are, however, agreed that the majority of sudden infant deaths occur during the normal household sleeping hours, which is quite different from the other causes of death in infancy. Most are discovered dead when the infant is looked at for the first time in the morning.

What then is found when individual cases are looked at? The Northern Ireland study showed a slight predominance of males, but this followed a similar pattern for infant deaths generally. Results of other investigations have ranged from a male excess through equality to a female excess, so there is probably no marked difference.

The most striking epidemiological finding is undoubtedly encountered when the age of the infants is considered. It is a constant result that there is a genuinely low prevalence in the first two weeks of life, followed by a rise to a peak incidence at around three months and then a rapid decline. The very low incidence in the first two weeks of life contrasts markedly with the other causes of infant mortality. Only rare cases occur after one year of life.

Templeman (1892) was the first to record that SIDS is commoner in families of lower social class. He, however, considered that all these deaths were due to overlaying by their

mothers as a result of carelessness, drunkenness or a desire to collect small sums of insurance money (up to 45/-). He was probably right in a number of cases, but it is thought that his description tallies closely enough with the modern concept of SIDS to include it here. Most recent studies have given at least an impression of the social conditions of families in which a sudden infant death occurs (Table II).

Froggart et al (1971) put their findings on a more scientific basis, and demonstrated a clear excess of deaths in Social Class V and a deficit in Classes I and II.

Illegitimate children and those of mothers under 20 have a higher incidence. Health visitors tend to have visited the homes of victims more often than those of unaffected children and their mothers have made fewer visits to child welfare clinics. It must be remembered, however, that this reflects only a general pattern, and there are marked deviations from it: SIDS can strike at any family.

As can be seen from Table I, American workers have found a higher incidence in negroes: this holds good even when correction is applied for the higher over-all negro infant mortality. The discrepancy between Caucasians and negroes is highest amongst the poor,

TABLE II  
(Valdés-Dapena 1970)

Authors	Year	Place	Total No. of Cases	Impression
Cooke & Welch	1964	W. Hartlepool	91	Preponderance of deaths in lower social groups
Carpenter & Shaddick	1965	England & Wales	110	Lower social class of wage earner. Poorer home and mothering
Sutton & Emery	1966	Sheffield	10	The squalor of the families is outstanding
Marek et al	1966	Poland	200	Living conditions bad in a considerable percentage
Hildebrand	1967	Hamburg	216	Many poor with deficient housing, disorderly, unclean and crowded.
Steele et al	1967	Canada	66	Poor general care of the infant Mother at all levels of educational attainment
Melton et al	1968	Richmond, Va.	199	Poor economic level, poor care of infants. Disproportionate number of negroes (50% vs 20% in population)
Fitzgibbons et al	1969	Minneapolis, Minnesota	46	Lowest reported incidence among farming people and middle-class urban dwellers

leading to the conclusion that there is an "increased incidence among negroes no matter what the socioeconomic level and an increased rate among the socioeconomically disadvantaged, independent of race". (Valdés-Dapena 1970).

As I have already pointed out, most of these children are found dead; few, if any, are seen to die. Some mothers claim to have found their babies alive but blue and have attempted to resuscitate them. This, however, is probably a natural reaction and the baby may well have been dead for some time. Another characteristic feature is that these children can die in the same bed as other children or in the same room as their parents and nothing is heard by them — the deaths are apparently silent. SIDS has been reported to have occurred in children in hospital without the staff detecting anything amiss (Gray 1971).

Retrospective studies have been carried out on the previous health of these infants and that of their mothers, especially during pregnancy. Prematurity may slightly increase the risk, but there is no evidence of an increase in "failure to thrive" in cases of SIDS. It appears to be commoner amongst twins but this may be a reflection of their lower birthweight, which is an important factor in infant death generally. The Northern Ireland group found no evidence of heredity playing any part, and the mothers of these children had no increase in foetal loss in their obstetric history.

Health of the children immediately prior to death is difficult to assess due to recall bias on the part of the parents and the problems of obtaining suitable control groups. Some results suggest an increase in minor illness (coryzal or digestive symptoms) in the week, and more especially the 24 hours, prior to death, but others disagree with this. There is certainly no history of major illness or anything to suggest that the child would shortly die.

This then is very briefly what epidemiology has contributed in attempting to unravel this problem. It is doubtful that it will produce much more of significance. Dr. A. B. Bergman, at the 1969 Conference, said "Epidemiology has only so much to offer — we mustn't try to squeeze out more juice than the whole orange possesses. I would make the rather dogmatic statement that except for the testing of specific hypotheses, large-scale epidemiological field investigations are apt to be repetitive and are not likely to produce new useful information."

With this in mind, combined with the pathological findings, what then can we say

about the present theories as to the cause of SIDS? No significant "at risk" factors can be isolated and this, combined with the characteristic age-range, mediates against the view that these infants have any as yet undiscovered 'disease'. Froggart et al (1971) suggests that as the infant is "passing through a developmental stage of physiological vulnerability some critical combination of extrinsic and intrinsic factors occurs which proves lethal." Just what is the "final common pathway" of these deaths causes the mystery and here epidemiology can be of little help.

I do not intend to attempt to discuss in any detail the enormous number of suggested causes, some of which are presented in Table III (an extension of one by Valdés-Dapena (1970)).

This is, of course, by no means a complete review of the literature. Evidence now exists, if not to refute, at least to throw doubt upon several of these hypotheses. That supporting others is, to put it mildly, tenuous. Some of them, although extremely ingenious, do not help at all to explain the epidemiological data. Perhaps the strongest support is given to viral infection being the major "extrinsic factor", although there is good evidence to back milk allergy. James' (1970) work on the development of the conduction pathways in the heart may well provide a clue as to the "physiological vulnerability" although a recent study has failed to confirm his findings (Valdés-Dapena 1971).

Froggart has called SIDS a "disease of theories", and until much more detailed scientific investigation is undertaken, particularly in trying to define whether the deaths are primarily pulmonary or cardiac, then little clarification can be expected.

No recipe is available for the prevention of these tragic deaths. It is thought probable by some that breast feeding is the most effective prophylaxis if continued for months, but even this by no means provides complete immunity, as 2 out of the 162 in the Northern Ireland series were entirely breast fed. The Ministry of Health report recommended that infants should sleep without pillows as their figures indicated that these were used in a significantly higher percentage of SIDS cases than in controls.

If a figure of 2.5/1,000 live births is accepted as the incidence in the United Kingdom, then this accounts for 10% of all infant mortality and 30% of post-neonatal mortality (28 days — 1 year of life) (Froggart et al 1971).

Table III  
SUGGESTED CAUSES OF SIDS

1. Accidental suffocation	Bedson, S. (1951) see Appendix I to Reference No. 13
2. Atlanto-axial occipito-atlantoid dislocation	Englander, O. (1971) B.M.J., 4, 625
3. Cardiac arrhythmia	James, T. N. (1970) in Bergman et al pp. 118-120
4. Cortisol insufficiency	Finlayson, N. B. (1964)*
5. Electrolyte imbalance	Maresch, W. (1964): McGaffey, H. (1968)*
6. Epidural haemorrhage	Towbin, A. (1968)*
7. Gamma globulin defect	Leading article (1971) B.M.J. 4, 250
8. Hyperactive dive reflex	Wolf, S. (1964, 1965, 1966, 1968)*
9. Hypersensitivity to cow's milk	Parish, W. E. (1960), Lancet, 2, 1106
10. Immune complex disease	Urquhart, G. E. D. et al (1972), Lancet, 1, 210
11. Infanticide	Asch (1968)*
12. Infection (a) viral (b) bacterial	Johnstone, J. M. (1966)* etc.
13. Inhalation of vomited milk	M.o.H. (1965) Report on Public Health and Medical Subjects No. 113
14. Nasal obstruction	Shaw, E. B. (1968)*
15. Parathyroid insufficiency	Geertinger, P. (1967)*
16. Precapillary bypass	Jaykka, S. (1971), Lancet, 2, 1315
17. Saliva	Campbell, K. (1971), Lancet, 2, 1314
18. Stress	Bohrod, M. G. (1963)*
19. Vitamin E and Selenium	Money, D. F. L. (1971), B.M.J. 4, 559

\* See original list in Bergman et al (1970) p.12.

The 1965 Ministry of Health "Enquiry into Sudden Death in Infancy" put the problem succinctly into perspective by stating that "it would appear that the risk of unexplained sudden death before reaching the age of two is about twice that of being killed on the roads before leaving school".

This is not an inconsiderable proportion of early childhood mortality. The vast majority of the victims of SIDS have appeared perfectly healthy up to their sudden demise. It is an obvious cause of great distress and needless feelings of guilt in the parents, whose acquaintances may "show or feel cruel primitive rejection" (B.M.J. Leading Article, 1971). At least the recognition of this entity by the medical profession has led to kinder treatment of the parents by the press in their reporting of inquests.

A great deal of money and research effort is at present directed towards prolonging, in an often unsatisfactory manner, the lives of congenitally deformed children, whereas little is spent in the U.K. on trying to establish the cause of this perplexing problem.

These children, some 2,000 a year in Great Britain alone, are mysteriously deprived of their

chance to become normal, intelligent, active members of the community with apparently little concerted effort to determine why.

Valuable work in publicising SIDS and raising the money for research into it is undertaken in the United States by lay organisations, e.g. The National Foundation for Sudden Infant Death (New York). Recently similar bodies have been formed in the U.K.: The British Guild for Sudden Infant Death Study and The Foundation for the Study of Infant Deaths. The latter organisation is attempting to raise £500,000 to support research.

They also provide much-needed help and comfort to families in which one of these tragedies occurs, help that is all the more valuable as many of the members of these organisations have experienced SIDS within their own family group, and are thus uniquely qualified to be of assistance to those in need of it.

Even if the cause of SIDS is never found and it can never be prevented, there is much that can be done even now towards reducing its effects on the other members of the family. Unfortunately the necessary police investigations to exclude unnatural death, the post-

mortem and inquest, if held, all add to the parents' grief.

Someone must take the time to explain to them what is known about SIDS, in particular that no blame can be attached to anyone and that there is no indication that it is likely to recur in subsequent offspring.

Emery (1972) suggests that the best person to take on the role of counsellor would be the local paediatrician. The General Practitioner would seem to be the obvious choice but approximately one-third of affected families have been found to change their G.P. after a "cot death" has occurred. The health visitor is similarly placed in a difficult position.

There is a heavy psychiatric toll, particularly amongst the mothers of these children (Bergman et al 1969). The symptoms of grief should be discussed with the parents, medication may be temporarily required for insomnia or anxiety and if possible a close watch should be kept for marital problems and symptoms of emotional disturbance in siblings.

Future children born to the couple may well be in danger from the effects of over-protection, excessive care and surveillance leading to almost inevitable consequences on their future emotional development.

The sudden infant death syndrome therefore embraces more problems than its aetiology alone and should not, to my mind, be confined to the realms of forensic medicine.

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