Section 1 The History of SIDS

Chapter 1

The History of SIDS – the Commonwealth's Contributions in its Formative Years

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The National Association of Medical Examiners (NAME) in the United States asked me recently to present the history of SIDS.[1] The context for this was an attempt to strike the term 'SIDS' from the medical lexicon, as proposed in the June 2017 theme issue of their journal *Academic Forensic Pathology* and replace this with 'undetermined'. The article[1] did not take sides on this thorny issue but was designed to provide historical context and help guide discussions within NAME. Since it was directed towards an American audience, it naturally had a somewhat American focus. My historical article caught the attention of the editors of this book, and they have asked me to write a brief historical entry highlighting important global contributions. While recognising that SIDS is not a single entity and that the use of this name, which dates back to only 1969, to classify deaths is controversial and may change in some jurisdictions, the term SIDS is used for simplicity's sake, even though the three 'SIDS investigators' I will discuss did much or all of their important work before 1969.

Prior to the 1940s and 1950s, SIDS was generally thought to be due to overlying, infanticide, thymic asthma, status thymicolymphaticus, smothering by bedclothes, and accidental suffocation[1] which will not be covered here. Fundamental work published in the 1940s suggested that SIDS is a natural entity with typical pathological and epidemiological findings. The first of these types of studies included the publications of New York City forensic pathologist Jacob Werne in 1942, Birmingham UK pathologist W. H. Davidson in 1945, and Werne, and his wife Irene Garrow from 1947 to 1953; these papers all suggested that many of these sudden, unexpected infant deaths had natural causes and that performing autopsies demonstrated explainable causes of death in some instances and provided histopathological findings demonstrating vague, mild respiratory disease processes in most of the rest.[1] These observations paved the way for the better understanding of SIDS that took place in the latter half of the twentieth century and changed the way these deaths were classified.[1]

The next wave of investigators confirmed these preliminary findings and took epidemiological analyses much further, helping establish the entity that would be named SIDS by J. Bruce Beckwith in 1969.[1] These included Melbourne forensic pathologist Keith Macrae Bowden (1908–1999), *British Medical Journal* editor Douglas Swinscow (1917–1992), and Sheffield paediatric pathologist John Lewis Emery (1915–2000). By virtue of the fact that they were working on opposite sides of the world, the convergence of their findings, combined with those of American investigators,[1] were even more compelling.

Bowden published three important papers in the *Medical Journal of Australia* from January 1950 to November 1952.[2–4] He noted that about thirty babies per year were found dead in their cots in the Melbourne area. The first paper addressed the question 'do babies accidentally suffocate in the bedclothes or face downwards on the bedding?' It provides his intriguing analyses into babies' spontaneous choices of sleep position at 2–7 months vs. 7–18 months of age and it comes very close to outlining the full 'triple-risk model'[1] currently used to explain SIDS. Bowden reported his autopsy series and concluded that if complete autopsies were performed, pathological findings are usually present. He notes that: 'although in every case the question why sudden death occurred, and although the exact mechanism of death is obscure in some cases, in practically every case natural disease was present'.[2] In one of his cases, the cause of death was determined to be an inherited metabolic disease, predicting a fertile area of research that would begin several decades later.[1] In his second paper, he added bacterial cultures and in some instances influenza virus cultures into his analyses and concluded that in twenty of forty-three cases, 'histological evidence of respiratory tract infection can be found, but in

which the aetiological agent was not isolated'.[3] He also observed that parents initially reported the infant to be 'quite well when last seen' before the death but, that when questioned later, 'a carefully taken history revealed evidence of several days or weeks of minor illness' in over two-thirds of cases.[3] Bowden's final study examined overlaying as a possible cause of death in 179 consecutive infants brought to the Melbourne City Morgue after dying suddenly and unexpectedly. In only 11 instances, was one or both parents actually sleeping with the infant and in 10 of these instances a complete autopsy established a natural cause of death. Bowden concludes that in all of Melbourne over the past four years, 'there has been one possible case of overlying'.[4] Bowden cited a forensic textbook claiming that accidental overlaying 'causes quite an appreciable annual loss of life' and that it is 'the most common form of accidental smothering', both of which he showed to be incorrect. All of Bowden's papers support his major premise: *'the more thorough the autopsy, the less the likelihood of a diagnosis of 'accidental suffocation"*.[3]

Swinscow wrote only one paper on SIDS, but it was influential.[5] 'So-called accidental mechanical suffocation of infants' was published in the *British Medical Journal* on October 1951; it called attention to the works of Davison, Werne and Garrow, and Bowden suggesting that many of these deaths had a natural cause. He then provided a detailed explanation of sex-ratio statistics and how they can be used to categorise deaths. Swinscow noted that: 'The two main factors concerned here are the sex distribution of the population exposed to risk and the differing susceptibility, characteristic of each sex, to death from a particular cause. When comparing the sex ratios of death from two or more causes in the same population, we find that the main effective reason for the difference between them is the fact that the deaths are caused by different agents'.[5] He then used England–Wales death statistics recorded by the Registrar-General for the years 1921–30, 1931–9, and 1940–9 to reveal that sex-ratio analyses showed a preponderance of male infants dying in cots and cradles when compared to infant deaths known to be accidental mechanical suffocation (eg., aspiration of food). He concluded 'many of the cot deaths are caused differently from (known accidental mechanical suffocation) deaths . . . Yet all are alleged to be due to the same cause – accidental mechanical suffocation.'[5] He further stresses 'the effect that such a diagnosis may have on the parents . . . No infant's death should be attributed to accidental mechanical suffocation unless there is clear positive evidence of it.'[5]

John Emery's contributions to the understanding of SIDS began in 1956 and he published at least another seventy-five papers on the topic after that. In fact, studying SIDS became a lifelong passion. One of his own children died in infancy, which gave him compassionate insights allowing him to deal with grieving families. According to A. H. Cameron:

> He employed two methods as the basis of his work on cot deaths. First was the meticulous morphological post-mortem study, accompanied by statistically controlled comparisons with hospital deaths. Much of his published work studied one organ at a time, for example, the lymphoid aggregates in the lung or the progress of ossification at the costochondral junction. This led inevitably to a study of normal development, which he soon discovered was based on very scanty data at that time. Paradoxically in his early investigations, he planned to use cot-death material as normal controls for other projects, but he soon realized the error of his way. His second method was an investigation of the domestic environment, again with appropriate controls. By this means he identified certain risk factors which allowed the preventive community paediatric services to concentrate their attention on particular families. He always emphasised the dependence of team-work and, in particular, the close involvement of the Health Visitor service.[6]

Emery's first SIDS paper began by citing Swinscow's mortality statistics; next, he noted that Davidson 'thought that most of these deaths were due to natural causes, and that with skilled necropsy the cause of death would be found' but concluded, 'unfortunately this is not wholly true'.[7] He noted that others have found only non-specific chronic inflammation in lungs. He then presented his own Sheffield data showing the added value of better history-taking. He notes that 'in only five of 50 infants studied was the complete correct history available at the time of necropsy' and 'in 33 the history . . . available to the pathologist was not only inadequate but misleading' (i.e., 'wrong enough to affect the diagnosis')[7] Emery also suggested that the autopsies be performed by a paediatric pathologist at the local children's hospital.

Emery published papers on the epidemiology of SIDS for forty years.[8–11] As a pathologist, he also published detailed papers on subtle pathologic findings

suggestive of chronic hypoxia (fat-laden cells in the cerebro-spinal fluid (CSF), retention of periadrenal brown fat cells, histopathological changes in the trachea) seen in SIDS starting in the mid-1970s[12–14] as well as papers showing that some pathologic changes reported by others are not specific to SIDS.[15]

Emery performed retrospective case-control studies on SIDS and control infants examining obstetric and perinatal histories in order to identify prospective 'criteria for detecting children at increased risk of dying unexpectedly'.[16] He and his team then used this to develop a scoring system to identify infants at risk.[17] He established a programme of Sheffield 'health visitors', and Emery and colleagues reported that infants who were visited and weighed frequently and received safe-sleeping advice had fewer deaths than predicted.[18] Within the identified high-risk group, they found symptoms that further predisposed to death.[19] This work resulted in an interventional study which showed excellent results after seven years,[20] as well as the Care of Next Infant (CONI) programme funded by the Foundation for the Study of Infant Deaths (now The Lullaby Trust) to provide support to families with new babies after having experienced a cot death.[21] In the 1980s, Emery published controversial estimates that about 10% of 'SIDS' cases were actually filicide;[22] while it was important to acknowledge that some cases signed out as SIDS are really filicide and that forensic science cannot always identify these cases, current data suggests that his estimate was too high.[23] However, Emery did publish data proposing a much higher incidence in families in which two or more SIDS deaths have occurred,[24] a finding supported a decade later by the high-profile multiple infanticide convictions of Wanda Hoyt in New York and Marie Noe in Philadelphia,[1] as well as chilling publications from Sir Roy Meadows and David Southall showing parents deliberately harming their infants.[1,23] Emery's contributions to SIDS were many.

Keith Macrae Bowden, Douglas Swinscow, and John Lewis Emery made important pathological and/or epidemiological contributions to the establishment of SIDS as a diagnostic entity and helped advance its understanding. It is imperative that these be remembered, and their work has not been highlighted in a previous historical paper. For a more comprehensive history of SIDS and to see how the work of these three men fit into the overall picture, the readers are invited to read reference 1 below.

References

1. Wright JR Jr. A fresh look at the history of SIDS. *Acad Forensic Pathol*, 2017; **7**(2):146–62.
2. Bowden K. Sudden death or alleged accidental suffocation in babies. *Med J Austral*, 1950; **1**(3):65–72.
3. Bowden KM, French EL. Unexpected death in infants and young children: Second series. *Med J Austral*, 1951; **1**(26):925–33.
4. Bowden KM. Overlaying of infants. *Med J Austral*, 1952; **2**(18):609–11.
5. Swinscow D. So-called accidental mechanical suffocation of infants. *Br Med J*, 1951; **2**:1004–7.
6. Cameron AH. Founders of Pediatric Pathology: John Emery (1915–). In: Garvin AJ, O'Leary TJ, Bernstein J, Rosenberg HS, eds. *Perspect Pediatr Pathol*, Basel: Springer-Verlag, 1992; **16**:1–6.
7. Emery JL, Crowley EM. Clinical histories of infants reported to the coroner as cases of sudden unexpected death. *Br Med J*, 1956; **2**:1518–21.
8. Emery JL. Epidemiology of 'sudden, unexpected, or rapid' deaths in children. *Br Med J*, 1959;**2**:925–8.
9. Emery JL. 'Sudden and unexpected' death in infancy. *Proc Roy Soc Med*, 1959; **52**:890–2.
10. Sutton RNP, Emery JL. Sudden death in infancy: a microbiological and epidemiological study. *Arch Dis Child*, 1966; **41**:674–7.
11. Emery JL. 'Cot death' rates on different days of the week. *Arch Dis Child*, 1998; **79**:198–204.
12. Gadsdon DR, Emery JL. Fatty changes in the brain in perinatal and unexpected death. *Arch Dis Child*, 1976; **51**:42–8.
13. Emery JL, Dinsdale F. Structure of periadrenal brown fat in childhood in both expected and cot deaths. *Arch Dis Child*, 1978; **53**:154–8.
14. Wailoo M, Emery JL. The trachea in children with respiratory diseases including children presenting as cot deaths. *Arch Dis Child*, 1980; **55**:199–203.
15. Cullity GJ, Emery JL. Ulceration and necrosis of vocal cords in hospital and unexpected child deaths. *J Pathol*, 1975; **115**:27–31.
16. Protestos CD, Carpenter RG, McWeeny PM, Emery JL. Obstetric and perinatal histories of children who died unexpectedly (cot death). *Arch Dis Child*, 1973; **48**: 835–41.
17. Carpenter RG, Gardner A, McWeeny PM, Emery JL. Multistage scoring system for identifying infants at risk of unexpected death. *Arch Dis Child*, 1977; **52**:606–12.
18. Carpenter RG, Emery JL. Final results of study of infants at risk of sudden death. *Nature*, 1977; **268**: 724–5.

19. Carpenter RG, Gardner A, Pursall E, McWeeny PM, Emery JL. Identification of some infants at immediate risk of dying unexpectedly and justifying intensive study. *Lancet*, 1979; **2**(8138): 343–6.

20. Carpenter RG, Gardner A, Jepson M, Taylor EM, Salvin A, Sunderland R, et al. The health visitors of Sheffield. Prevention of unexpected infant death: evaluation of the first seven years of the Sheffield Intervention Programme.*Lancet*, 1983; **1** (8327):723–7.

21. Emery JL, Waite AJ. These deaths must be prevented without victimizing parents. *Br Med J*, 2000; **320**:310.

22. Taylor EM, Emery JL. Two-year study of the causes of post-perinatal infant deaths classified in term of preventability. *Arch Dis Child*, 1982; **57**:668–73.

23. Milroy CM, Kepron C. Ten percent of SIDS cases are murder – or are they? *Acad Forensic Pathol*, 2017; **7**(2): 163–70.

24. Emery JL. Families in which two or more cot deaths have occurred. *Lancet*, 1986; **1**(8476):313–15.