Cot deaths in Malaysia

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Cot Deaths, Crib Deaths or Sudden Infant Death Syndrome is a term which refers to the death of any infant or young child, which is unexpected by the history and in which a thorough post mortem examination fails to demonstrate an adequate cause of death. (Bergman et al 1970). In the majority of cases, the infants are found dead by their parents in cots, beds, or cribs, after having been placed there the night before. Because of the dramatic suddeness, these deaths remained until recently, within the province of forensic medicine. It was also believed that these infants had died of suffocation because of autopsy findings, suggestive of asphyxia in some cases. Even as far back as 1947, Werne and Garrow were not prepared to accept, so facile an explanation. Therehas since been a tendency, for a more critical approach, as to the cause of these deaths, the aetiology and/or mechanism of whichsis still not fully understood. The problem has assumed considerable importance especially in the Western hemisphere. In recent years cot deaths have been reported from many parts of the world, and they can be expected to assume greater prominence in those countries with a decreasing incidence of infectious diseases and nutritional disorders.

CASE REPORTS

The first case is LJH, a 2 month old, male infant of mixed Asian origin. He is reported to have been well earlier on the day of his death. Having received his last bottle feed of milk early in the afternoon, he was put to sleep in a cot. He was found dead at about 5.00 pm. According to the parents the position he was found in, did not suggest the possibility of suffocation.

LJH was an adopted child of middle-class parents, who had two other children of their own, a girl aged 2 years and a boy aged 7 months. He was seen by a general practitioner at the age of 6 weeks for a routine check up, and found to be well. He had not as yet been started on routine immunization. There was no history of any recent illness in the family. However questioning the father revealed that the maidservant, but not the infant, had been having snuffles. Information about the birth history and the early life of the infant was not available but the parents claim that he had been perfectly well since adoption at the age of 1 month.

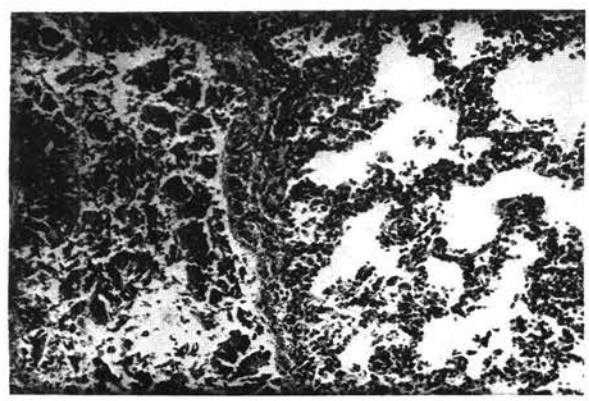

Fig. 1. Show large collections of bronchial epithelial cells in bronchus and normal aerated lung tissue.

At autopsy, the body was that of a well nourished infant with post mortem lividity over the trunk and thighs. The trachea and bronchi contained a small quantity of clear fluid but there was no aspirated material. Both lungs were expanded with a uniform pinkish appearance of the pleura on which a few petechiae were seen. The cut surfaces showed congestion. The heart was normal. The stomach was filled with a curdled mass of milk. The other abdominal viscera and brain were unremarkable. Histology revealed focal haemorrhages in the thymus. Most sections of the lungs showed aerated lung tissue with congestion, oedema, and subpleural haemorrhages. In areas groups of alveoli contained macrophages. Some of the lung sections showed that bronchi were devoid of epithelium but explosive epithelial desquamation was not seen. The other organs showed no significant histological abnormality though a detailed study of the parathyroids and cardiac conducting system was omitted.

The second case is CKF, a 2 months old male Chinese infant, who is said to have been well and cheerful on the morning of the day of his death. After being given his mid-day bottle feed on milk the mother put him to sleep, in a hammock, with a spring suspension. About 2 hours later she heard him cry and when she went to look at him he was seen closing his eyes; and looking at him again half an hour later, he was noticed to be pale and listless. He was rushed to hospital where on arrival the doctor pronounced him dead. The past history, is that a week earlier he had been having a bad cough which a private practitioner diagnosed and treated as bronchitis. However about 3 to 4 days before his death he was relatively free of cough and his bronchitis had apparently cleared up.

CKF was the only son of a self-employed tradesman in the lower income group, and the mother, a housewife. He was born in a hospital at term. The delivery was normal and his birth weight was 5 lbs. 10 ozs. He was bottle fed from the beginning. He became slightly jaundiced on the fifth day but this soon cleared up.

The autopsy findings were that of a well nourished Chinese infant which showed peripheral cyanosis and lividity of the posterior chest and abdomen. The trachea and bronchi contained scanty clear fluid but no aspirated material. The pleural surfaces of both lungs were to some extent mottled pink and purple and showed a few petechial haemorrhages. The tracheo-bronchial nodes were slightly enlarged. The heart was normal except for petechiae over its mid posterior aspect on the atrioventricular groove. The stomach contained milk curds.

The liver, biliary tree, other abdominal viscera and the brain were unremarkable. Histology of the tracheo-bronchial nodes showed reactive lymphadenitis. Histology of the lungs showed oedema, congestion, sub-pleural haemorrhages and in some of the sections there were collections of macrophages within the sub-pleural alveoli. There was no evidence of a pneumonitis. Explosive desquamation of the bronchial epithelium was seen in an occasional bronchus (Fig. 1) but most of the bronchi had a normal epithelial lining. Other organs showed no significant histological abnormality. As in the previous case detailed study of the cardiac conduction system and parathyroids was omitted.

DISCUSSION

The purpose of this paper is to review briefly the current facts and theories pertaining to cot deaths and present two cot death cases which hitherto as far as we are aware have not been reported from this part of the world. We have no doubt that these deaths are far more numerous than suspected and it is hoped this paper will stimulate greater awareness. It is therefore salutary to follow this up by establishing the incidence of cot deaths here. It must be emphasized that in all suspected cases of sudden unexpected infant death, the scene, usually the home should be visited by at team of medical and social workers. The enigma of a healthy infant dying in this way must be explained to the bereaved parents amongst whom considerably recrimination and despair are the usual sequalae. The 'clearing of the air' by the team will go a long way to dispel grief and misgiving on the part of the parents. The investigation should include an interview of the family to ascertain the prior health of the dead infant, and to what extent trauma and infectious diseases in the family or in the neighbourhood may have been a contributory cause. At the onset of such an investigation it is pertinent to entertain thes question of child cruelty (battered baby syndrome) and therefore a radiographic skeletal survey followed by an autopsy should be an essential routine whenever possible. The reasons for an obligatory autopsy in cot death cases cannot be overstressed and this is discussed by Raven (1973) and others. Not only is it important to determine the exact cause of death but rule out deaths due to accident, negligence or even infanticide.

It is probable that cot deaths are less common here than in temporate countries as has been suggested by a recent study in Isreal, (Winter and Bloch 1973). In round figures the incidence in Great Britain is approximately 1,100 deaths per year and in the U.S.A. 10,000 deaths per year (Valdes-Dapena, 1963). In countries with a temperate climate there is an excess of cases in winter (Carpenter and Shaddick, 1965) and even a clustering of cases on a single day was reported in a Canadian study. In Brisbane, Australia which has sub-tropical climate, cases showed no seasonal incidence but with a tendency to occur in groups (O'Reilly and Whiley, 1967).

Most workers agree that cot deaths occur in the first six months of life, though a few occur before three weeks or after six months. There is a relative peak incidence between the ages of two and four months with a male preponderance, which varies from 53 – 62%. It will be noted that both our cases were aged about two months and were boys. Perhaps the most striking feature of this syndrome is the time of the day when the deaths occur, that is, during sleep and early in the morning. As will be seen in our cases, both had died in their sleep but in the afternoon. Some variation from the generally recognised features of the syndrome may be attributed to a tropical climate, pertinent features of which are, negligible seasonal variation and the equal duration of the day and night throughout the year.

Valdes-Dapena (1963) notes that, on autopsy with subsequent appropriate laboratory investigations, a cause of death is found in 16 percent of cot death cases. In 5 percent of the cases the basic lesion is demonstrable on gross examination. Some of these have been reported as congenital heart disease, neonatal myocarditis, endocardial fibroelastosis, bilateral purulent otitis media and pneumonia. We have on our records an infant aged ones week, apparently well, who died suddenly and unexpectedly in the post-natal ward of this hospital. Autopsy revealed a gross cardiac abnormality (cor bi-atrium triloculare). In view of its age and the severity of the cardiac lesion we considered it was not appropriate to be labelled as a cot death.

Bowden in Australia had published a list of conditions from which babies had died suddenly, including congestial heart disease and pneumonia, stating that cot death cases were all dying of natural diseases which had failed to be recognised. Camps (1972) commenting on this thinks that Bowden may have introduced a false idea, that is, to assume that *a* cause of death is necessarily *the* cause of death.

The most common naked eye findings at autopsy are petechial haemorrhages, seen in a variable proportion of cases on the lungs, heart and thymus. It was this that led to the hypothesis of suffocation as a cause of cot deaths though it cannot be denied that this may account for a negligible number. The age incidence, with a relative peak in the third to fourth month and the preponderance of cases being over six weeks of age argues against suffocation (Judge 1953). Woolley (1945) demonstrated that infants will respond to an experimentally contrived smothering by rolling over and continuing to breath well. The presence of intrathoracic petechiae does not itself lend support to suffocation as an important mechanism. Polson (1973) has discussed the significance of petechial haemorrhages. Gordon et al (1953) say that Liman's views "receive substantial support at present as pathologists have repeatedly described petechial haemorrhages in the serous membranes in may forms of deaths". More recently Guntheroth et al (1973) have repeated Handiforth's (1959) experiments in rats to determine the factors responsible for the appearance of petechiae and to establish the specifity of those lesions for laryngeal obstruction and concluded that unremitting airway obstruction is unlikely as a cause of cot deaths.

Nasopharyngeal obstruction coupled with obligatory mouth breathing (Shaw 1970) has also been claimed as a cause of death in these cases. In such a position wherein it cannot breathe through its mouth the infant would suffocate. There has been no uniform corroboration of this. Acute epiglottitis is now a well recognised paediatric entity but it has not been documented as a cause of cot deaths and likewise there is no morphologic basis for any presumptive or definitive diagnosis such as bronchiolitis, laryngospasm, laryngitis (Huntington and Jarzyn 1962).

A variety of histological findings have been reported in cot deaths. Some of these include upper respiratory tract inflammation (Valdes-Dapena 1963), intraalveolar large mono-nuclear cells with an explosive desquamation of bronchial epithelial cells (Bodian and Heslop 1956) seen in one of our cases. This however may be an artefact and has been seen in a number of non-cot deaths. Stowens (1966) has described mild diffuse alveolar over-distention and pulmonary oedema. Stowens (1966) also described a decrease in the number of eosinophils in the thymus,

a retention of its infantile and a diffuse swelling of arterioles in many organs, findings which have been uncorroborated.

In the heart, myocardial cell lesion has not been demonstrated. James (1968) described resorptive changes in the left bundle of His and the left margin of the atrioventricular nodesbut these changes were also seen in some of the controls. As current hypothesistends toward the concept of an instantaneous interruption of some basic physiologic function such as the control of cardiac action or respiratory function it would be tempting to attribute death to a transient but fatal cardiac arrhythmia based on his observations. However re-examination of the histological characteristics of the atrio-ventricular node and the bundle in infants who had died suddenly and unexpectedly and in age matched controls by Valdes-Dapena et al (1973) questions the validity of malfunction of this or any other anatomic system to features of its normal developmental histology. Cardiac electrolyte imbalances have been described and refuted in cot deaths. (Fraggot Lynas and Marshall, 1968).

Geertinger (1968) advanced the hypotheses that congenital incomplete development of the parathyroids is the underlying basis for such vulnerability to sudden death but other studies havesnot supported this finding. The demonstration of the fusion of parathyroids to the thymus in cot deaths has also been noted in otherwise explained infant deaths. It is apparently a structural variant and presumably of no significance. Cervical spinal epidural haemorrhage have been reported in cot death cases. These haemorrhages have also been found in controlled studies and they have been presumed to be a secondary or agonal. No underlying spinal cord pathology or changes in the cervical vertebrae have been found in such cases.

Werne and Garrow (1947), based on a study of 167 consecutive cases of infants allegedly suffocating, in the final paragraph of their paper stressed the importance of efforts to prevent the sudden death of infants by diminishing exposure to known sources of infection. Nevertheless, in general, most studies of cot deaths have not recovered a virus. Johnstone and Lowy (1966) were unable to culture a virus in 47 cases. Negative results were obtained by Parish et al (1964), who cultured fresh autopsy material in 8 cot death cases and this offsets the criticism of the deleterious effect of freezing known to kill the respiratory syncytial virus of epidemic infantile bronchilitis and possibly other viruses. Valdes-Dapena (1963) states that bacteriological studies are essentially negative and the presence of post mortem bacterial growth characterised by the absence of an inflammatory reaction has to borne in mind when a presumed pathogen is isolated from post mortem tissue. Johnstone and Lowy (1966) on the otherhand found a bacterial pathogen in 37 out of 56 cases, the offender being usually a Pneumococcus, Klebsiella pneumonia or Staph pyogenes in 37 out of 56 cases. These were pure or predominant cultures. It is not known to what extent post mortem overgrowth following terminal or agonal tracheal aspiration of gastric contents may be a factor especially as these infants are usually dead for some hours before they are found.

In 1954 Spain stated that the serum gammaglobulines in three cot deaths were lower than in two control cases. This was apparently an exceedingly attractive piece of data as it denoted an unusually low gammaglobulin for cot deaths in the period of known physiological hypogammaglobulinaemia. Recent studies have demonstrated an elevation of IgG and IgM in 15% of cot death cases (Balduzzi et al 1968). A more recent study of the cord blood from 15 out of 23 cot death cases (Clausen et al 1973) indicates that immunological mechanisms may not be of primary significance.

Hypersensitivity to cow's milk in particular became an important consideration in the aetiology and/or mechanism of cot deaths. Barret (1954) had consistently suggested the possibility of some association between these deaths and cow's milk. As a result, an immunoserological study was commenced by Parish and Coombs in 1960. They succeeded in producing a somewhat similar clinical and histological picture in milk sensitized guinea pigs by intratracheal injection of small quantities of milk under conditions simulating sleep (barbiturate sedation). The cow's milk hypersensitivity proposal is based on the contention that cot death infants are invariably bottle fed, there is a higher level of serum antibodies to milk protein, cow's ilk can be demonstrated in the lungs of cot death cases and the mechanisms of death is sudden, therefore suggestive of anaphylaxis. There is an animal model which supports the proposed aetiology and mechanism of death. The hypothesis of hypersensitivity to cow's milk though attractive has been the subject of a much critical contention and the issue remains unsettled.

SUMMARY AND CONCLUSIONS

1. The literature on Cot Deaths or the Sudden Infant Death Syndrome is reviewed with a report of the first two cot death cases from this region. A proceedure for the investigation of sudden infant deaths by a medico-social team has been recommended.

2. The main characteristics of Cot Deaths such as the peak age incidence male preponderance, clustering of deaths during the colder months in temperate countries, deaths early in the morning during sleep, all of which present a uniformly striking picture are outlined.

3. The cause and/or mechanism of Cot Deaths remains obscure for more than two decades and innumerable theories have been put forward, some of the plausible ones have been discussed briefly. At the moment the theories have not stood the test of time but it appears, either allergy to cow's milk proteins or an acute viral infection may have the greatest chance of eventually being proved correct.

4. Besides the United States and Great Britain Cot Deaths have been reported from many parts of the world, and these include Canada, Czecko-slovakia, Ireland, Sweden, Israel and Australia. With improvements in the proceedure of death certification here, these deaths can be expected to be more precisely delineated as an entity, and statistics on Cot Deaths for international Comparison and research compiled.

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