Unexpected postneonatal deaths (cot deaths) due to recognizable disease

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McWeeny, P. M., and Emery, J. L. (1975). Archives of Disease in Childhood, 50, 191. Unexpected postneonatal deaths (cot deaths) due to recognizable disease. At least a quarter of the babies dying as unexpected home 'cot deaths' do so from recognizable diseases. A study of the background of such children, compared with age and admission controls, suggests that the disease symptomatology in this group of children differs in no way from children who are successfully treated. The factors leading to the death of these children appear to lie in the general lack of ability of some parents to recognize the importance of symptoms, their negative attitude and inability to avail themselves of health services, and the amount of drive and persistence required to obtain general medical practice services in some areas. There is also the failure of some practitioners to recognize severely ill children.

During the last decade our knowledge of some aspects of children's disease has advanced with great rapidity, yet the mortality rate in infants over the age of one week has shown little or no decline. The most striking feature of these postneonatal deaths is that about half of them occur as unexpected 'cot deaths' at home (Richards and McIntosh, 1972; British Medical Journal, 1973). The extent to which an adequate cause of death is found at necropsy varies considerably in different hands, and as the extent and depth of study is so variable, a comparison of statistics is a little invidious. Ignoring clinical history it is possible to divide the findings into four groups based only on necropsy findings be they gross, histological, microbiological, immunological, or chemical.

Group A—Children with gross congenital deformities with disease of long standing in whom early death was probably inevitable and could be precipitated by a minor disease. Such children may have congenital deformities of the heart, or hydrocephalus, Down's syndrome, hydronephrosis, etc.

Group B—Children in whom a definite disease state is found, a disease that is frequently diagnosed and treated vigorously at home or in hospital.

While the precise mechanism of death may not always be known, the conditions carry a well-recognized mortality rate—such diseases are meningitis, acute obstructive tracheobronchitis, and gastroenteritis with hypernatraemia.

Group C—Children who reveal evidence of a level of disease which does not usually result in a significant mortality rate, i.e. conditions such as upper respiratory tract infections, tracheitis, mild gastroenteritis, and small areas of pneumonia from which large numbers of children suffer and recover.

Group D—Children showing none of the above features of acute disease. Within this group there are two subgroups. (1) Those with evidence of growth retardation or general metabolic disease producing fatty changes in the liver or thymic or adrenal reaction, i.e. evidence of nonspecific disease. (2) Those who show no such evidence and appear to be completely fit.

The borders between these groups must be blurred, but with increased knowledge of disease processes there should be a drift in diagnosis from groups D to B.

Between the years 1954 to 1971, in a series of 644 necropsies carried out in this department, the proportion of children in group B was 25%.

In our most recent series where we introduced the
study of the chemistry of the vitreous humour (Swift, Worthy, and Emery, 1974) and have been able to diagnose degrees of hypernatraemia and uraemia, the proportion in group B reached about 50% (Emery, Swift, and Worthy, 1974), i.e. this group now constitutes about half of all unexpected home deaths and hence, a quarter of all deaths between the ages of one week and one year. If any immediate impact is to be made in the reduction of post neonatal deaths, those in groups B require our immediate attention. The mortality rate of children admitted to hospital in Sheffield with gastroenteritis is 1·7%, and with acute diseases of the chest 3·0%, and these form the predominant conditions in our group B. It seems that deaths in these categories are predominantly preventable, and the question is why these children did not obtain full medical care and attention?

That a group of possibly preventable deaths exists has been recognized for many years. In a study of 184 post neonatal deaths in Edinburgh during the years 1961 to 1963, Selwyn and Bain (1965) considered that 23 were 'preventable', and in the confidential inquiry into post neonatal deaths by the Department of Health and Social Security in 1970, an attempt was made to elucidate factors in such deaths. Among 225 deaths with amenable factors they found that social factors were active in 36%, parental inadequacy in 35%, inadequacy in general practitioners' service in 17%, in hospital service in 6% and in the local authority in 2%. Richards and McIntosh (1972), in a recent study of 226 infant deaths in Glasgow, further annotated possible avoidable factors in 199 children. Such studies do not help greatly unless we are able to evaluate these factors and to discover how some families seem able to achieve adequate medical treatment and others do not.

The questions we asked ourselves were: (1) Was the symptomatology of the explained cot deaths similar to that of the successfully treated cases? (2) What factors make one family successfully handle a sick child and another fail?

A group of age-matched living children was studied in exactly the same way as the index cases, but as these control families had not faced the situation of an acutely ill child, we took in addition as a further group of 'disease situation' controls a series of children in the same age group who had been admitted to hospital, acutely ill, but who recovered.

### Materials and methods

The 25 unexpected home deaths comprised a sequential series of explained but unexpected deaths. There were 6 children with tracheobronchitis (1 of whom also had convulsions and 2 had hypernatraemia); 3 children with meningitis—one, pneumococcal, one haemophilus, and one viral; 2 children with acute cardiorespiratory failure with mild fibroelastosis of the heart and pulmonary hypertension; 8 children with gastroenteritis (5 of whom had severe hypernatraemia and uraemia); 5 children with severe hypernatraemia and uraemia associated with minor respiratory tract infections; and 1 child with meningoccal septicemia.

The hospital admission controls who survived were also a sequential series of children in the same age group, i.e. 1 week to 1 year, who were admitted to paediatric units in the city. There were 8 with acute chest infections, 4 with severe bronchitis, 2 with bronchiolitis, and 1 with an acute upper respiratory tract infection; 9 children were admitted with gastroenteritis and 1 child was severely dehydrated with hypernatraemia.

It was not possible to match the index and admission cases exactly for time and disease as this would have entailed making the timing intervals of the cases too irregular. There was one exclusion in the admission series, a Pakistani child from a home where no English was spoken. The index cases included no recent immigrant families.

The age-matched control group consisted of the tenth birth notified before each index case.

Since the beginning of 1971 our investigation of unexpected deaths has included a standardized interview with the bereaved parents in their home, which takes place 3 weeks after the death. Parents of the disease controls were treated in exactly the same way, being interviewed 3 weeks after the child’s admission date. Social data of the family are collected, the family history, the mother’s attitude to and account of her pregnancy, labour and delivery, and details of the infant’s management, progress, and health, as seen by the parents. Details of infection in contacts with the child are also recorded. During the course of this interview an attempt is made to assess the parent’s attitude to and use of the different branches of the health service, their acceptable norm of health, and their ability to recognize the significance of signs of disturbance in the child. When taking the history, particular attention is paid to the 3 weeks before the death of the child or his admission to hospital. Note is made of the symptoms, the day and length of their occurrence, the importance attached to these by the parents, and the action taken.

A close scrutiny is also made of records relating to the case by the Children’s Hospital, the maternity hospital, midwife, health visitor, Infant Welfare Clinic, and social work agencies. The general practitioner of each case and control is contacted and his knowledge of the child and relationship with the family ascertained when possible.

### Results

#### Social class, housing, and birth rank

The index and disease controls were almost identical
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in their distribution among the social classes, each having only one family in the classes of professional and nonmanual workers, while the age-matched control group had 9 in these three classes ($X^2 - 0.001 < P < 0.01$) (Table I). The housing situation of the age-matched controls showed no unsatisfactory premises, while 8 of the disease controls and 12 of the index cases lived in inadequate premises ($P < 0.001$). The birth rank of the children in the three groups showed no significant difference. It was noted that 18 of the children in the index and disease control group and 20 in the age-matched control group were either the first or second child in the family.

**TABLE I**

Numbers of children in index, age-matched, and disease control groups related to social class, housing, and birth rank

<table>
<thead>
<tr>
<th>Social class</th>
<th>Index</th>
<th>Age-matched control</th>
<th>Disease control</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>1</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>II</td>
<td>0</td>
<td>5</td>
<td>1</td>
</tr>
<tr>
<td>III Nonmanual</td>
<td>0</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>III Manual</td>
<td>13</td>
<td>11</td>
<td>13</td>
</tr>
<tr>
<td>IV</td>
<td>4</td>
<td>4</td>
<td>6</td>
</tr>
<tr>
<td>V</td>
<td>6</td>
<td>1</td>
<td>4</td>
</tr>
<tr>
<td>VI Not known</td>
<td>1</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Housing</td>
<td></td>
<td></td>
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<tr>
<td>Satisfactory</td>
<td>13</td>
<td>25</td>
<td>17</td>
</tr>
<tr>
<td>Unsatisfactory</td>
<td>12</td>
<td>0</td>
<td>8</td>
</tr>
<tr>
<td>Birth Rank</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>8</td>
<td>12</td>
<td>7</td>
</tr>
<tr>
<td>2</td>
<td>10</td>
<td>8</td>
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<td>0</td>
<td>4</td>
</tr>
<tr>
<td>4</td>
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<td>2</td>
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</tr>
<tr>
<td>5</td>
<td>1</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>6</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Adopted</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
</tbody>
</table>

Age of parents and planned pregnancies.

The ages of the parents in all groups showed a wide range with a possible tendency for the parents of the two control groups to be older and those of the index cases to be younger than 20, but these differences are not statistically significant (Table II).

The number of children conceived outside marriage was remarkably similar in the three groups, as was the legitimacy rate. While the incidence of planned pregnancies was about twice as high in the disease control as in the index group, the difference was not statistically significant.

**General progress and weight.** A general assessment of the infant's progress was based on the rate of weight increase, the establishment of regular feeding habits, sleep pattern, number of incidents of disturbance, e.g. vomiting, diarrhoea, respiratory tract infections, nappy rash, etc.

As indicated in Table III, the index group and the disease controls both showed that their previous history had been unsatisfactory when compared with the age-matched controls where only 2 of the 25 cases were not satisfactory $X^2 - 0.001 < P < 0.01$).

The weights of the infants were assessed on their position on normal centile curves for age
using their last recorded weight before death, interview, or admission. Here the similarity between index and disease controls is not maintained and shows a statistical difference (P <0.001). While the weights of the disease controls appear to be similar to the age-matched group, the index group had 10 children with a weight at the 10th centile or below (Table III).

There was no difference in the early feeding in the three groups or in the method of making up the artificial feed, as described by the mother.

**Duration of symptoms.** Scrutiny of the 3 weeks before the death of each index case and before the admission to hospital of each disease control case revealed that the duration of signs of disturbance in the child, as described by the parents, was similar (see Table IV).

**TABLE IV**

*Duration of symptoms preceding death in index cases and admissions in disease control cases*

<table>
<thead>
<tr>
<th>Duration of symptoms</th>
<th>Index</th>
<th>Disease control</th>
</tr>
</thead>
<tbody>
<tr>
<td>&lt;24 h</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>24-48 h</td>
<td>4</td>
<td>5</td>
</tr>
<tr>
<td>3-4 d</td>
<td>3</td>
<td>6</td>
</tr>
<tr>
<td>5-7 d</td>
<td>4</td>
<td>3</td>
</tr>
<tr>
<td>1-2 w</td>
<td>8</td>
<td>5</td>
</tr>
<tr>
<td>&gt;2 w</td>
<td>3</td>
<td>3</td>
</tr>
<tr>
<td>Not known</td>
<td>1</td>
<td>1</td>
</tr>
</tbody>
</table>

It was not possible to assess the severity of symptoms but vomiting, diarrhoea, coughing, and poor feeding appeared to be present equally in both groups.

**Attitudes to and use of health services.** Only 15 of the index group but 23 of each control group had attended an Infant Welfare Clinic at least once in the child's lifetime, but the number of consultations with doctors showed a marked difference. When the 3 weeks before death or admission were considered the disease controls showed an increasing number of visits resulting in admission. The 16 index cases who were seen during the 3 weeks before death received a total of 21 visits (including 6 within 24 hours of death) but no use had been made of the deputizing service. A total of 58 visits were made to 24 disease control babies and this included 11 made by doctors in the deputizing service (organized by the British Medical Association locally). 18 visits resulted in admission to hospital (13 referred by the family doctor and 5 by the deputizing service). Of the remaining 7 admissions, 5 were brought directly to hospital by the parents, 1 was referred by the Infant Welfare Clinic, and another by the postnatal clinic at the lying-in hospital. 9 of the index group had not been seen by their family doctor at all during the 3-week period before death, but only one of the disease control group had not been seen and this child was brought directly to the Casualty Department of the Children's Hospital by the parents. Only one consultation with the family doctor had taken place during the past week in the age-matched controls, but the same number of consultations had taken place during the previous 2 weeks in this group, as in the index cases.

An attempt was made to assess the attitude of all parents to the medical services. Parents indicated differing degrees of willingness to consult their general practitioner, resulting from a wide range of factors and previous experience. The parents of 10 index cases expressed negative feelings and attitudes to their general practitioners, while only one family in each of the control groups came into this category. Some of the parents who had a good relationship with their doctor had not seen the necessity for consultation, as they had obviously not recognized the severity of the illness of their child though they may have clearly described the symptoms.

**Discussion**

That there is an increased mortality and morbidity in large young families living in poor, overcrowded housing conditions and in the presence of parental inadequacy and incompetence is paediatric commonplace, and was one of the starting points for the inquiry carried out by the Department of Health and Social Security in 1970. In that study of 670 children it was felt that avoidable factors were present in 187 (28%) of the deaths. The predominant avoidable factors within these were social (36%), and 74 of 81 deaths were attributed to bad housing. Parental incompetence was said to be active in 29, failure to summon medical aid in 26, and failure to appreciate the severity of the medical situation in 17. The inquiry into 226 infant deaths in Glasgow (Richards and McIntosh, 1972) divided the causes of death in somewhat the same way as we have done. They divided their total number of deaths (226) into two groups. One group had no known predisposing cause, i.e. 146 infants who included deaths due to pneumonia, gastroenteritis, as well as unexplained cot deaths; and the other group of 80 children where a predisposing cause, such as a congenital deformity, was definable. Within their 103 children falling into the group of unexpected
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In our categories (C) and (D), and the others (our group B) included 28 with pneumonia and 7 with gastroenteritis. Among the characteristic features in their group of unexpected deaths was a 'below average intelligence' of the parents, and 68% of the children had never been taken to an Infant Welfare Centre.

The greatest weakness in the Department of Health and Social Security and the Glasgow studies was the absence of a control group, and thus a means of assessing why one child dies in a particular social environmental circumstances and another does not. As Kincaid (1965) pointed out when discussing possibly preventable child death, there are more and less successful individuals in all classes. Our objective is to identify the potentially unsuccessful parents, ideally before the child is born.

From the evidence we have been able to obtain there appears to be no difference in the early symptomatology of the explained cot deaths and the successfully treated controls. The only significant difference in the cot death babies from the controls lay in their reduced body weight. The housing, age of parents, race, size of family, parents' age on leaving school, legitimacy of child, and family mobility do not differ in the two groups.

The evidence strongly supports the concept that had the parents of the index group used the help available to them as did the disease controls, then most of these children would have survived. The difference between these groups thus appears to be in the management of the situation.

In a study of factors concerned with children found unexpectedly dead (Prostos et al., 1973) it was found that the most significant factor distinguishing the cot death children from a control group was the failure of the mother and child to attend the postnatal follow-up clinic.

Our findings support those of Richards and McIntosh. We are particularly impressed by the nonutilization of medical services by the parents of the children who died. Richards and McIntosh attempted to assess the intelligence and efficiency of these parents. We did not do so, but are attempting to devise methods of measuring mothercraft and the standard of health accepted as normal in children (e.g. how many mothers consider it normal for an infant's ears or nose to discharge?), and a mother's ability to make up a milk feed correctly. The attitude of parents to the medical services at their disposal must also be appraised. Some parents are reluctant to seek medical advice though recognizing that it may be necessary. Some parents are reluctant to consult their doctor for a variety of reasons ranging from difficulty of making an appointment to unfavourable past experience.

In some cases it is difficult to avoid the conclusion that a factor contributing to death lies with the medical care of the child before his death. 6 children had been seen within 24 hours of death, and another 5 had been seen by their doctor once during the 10 days before death, but the initiative had been left with inadequate parents to seek further consultations. The 6 children who had been seen by the doctor within 24 hours of death had died of—pneumonia 1, heart failure with pulmonary hypertension and mild fibroelastosis of the heart 1, acute infection of the respiratory tract with uraemia and hypernatraemia 3, and gastroenteritis 1. The deaths of some of these children cannot but be attributed to the failure on the part of the doctor to diagnose an ill child, or to the giving of wrong treatment. In future studies of a larger group of child deaths it will be necessary to make a special study of these cases, and to separate them from deaths where medical attention had not been requested.

This present study illustrates the need for more detailed examination, not so much of the social and general environmental factors which have been so prominent in all statistics, but of parental attitudes and competence in all social classes. There is a need to seek ways of achieving more favourable behaviour in both the users and providers of medical services.

That 6 of the 25 infants had been seen by the general practitioner within 24 hours of death indicates considerable room for improvement in paediatric care. This is not necessarily the fault of the practitioner. In his training he has been almost entirely taught in outpatient departments or in hospital wards. The paediatricians themselves in medical schools only see the acutely ill child after illness has first been spotted by the parents and practitioner.

Conclusions

1) The duration and type of symptoms described by parents in children found unexpectedly dead from a recognizable disease differ in no way from children admitted to hospital with similar diseases.

2) The general social factors of this group of children dying unexpectedly at home were the same as those admitted to hospital, and equally different from a birth-matched control group, though birth rank was similar in all three.

3) While the general medical progress of the disease control babies was similar to the cot death
infants, a large group of the latter babies were below the 25th centile for weight.

(4) A difference in the successful and unsuccessful parent groups was in their use of the health services. Almost half the cot death babies had never been taken to a welfare clinic, and 10 of the 25 cases indicated a reluctance to take their child to see their general practitioner.

(5) The general cause of the inadequate medical care and child death in this particular group did not lie within the well recognized factors such as housing, legitimacy, and size of family usually associated with increased morbidity, but in the main in four further factors. (a) A general lack of ability in the parents to recognize the importance of symptoms. (b) Their negative attitude to and lack of energy in availing themselves of the health services. (c) The present organization of general medical practice services in some areas appears to require too great a degree of drive and persistence on the part of parents to obtain attention. (d) Failure of the medical practitioner to recognize severely ill children.

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REFERENCES


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