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A case-control study of unexpected infant death in Hong Kong

Introduction

Sudden infant death syndrome (SIDS) is the sudden unexpected death of an infant, in which a thorough postmortem examination fails to demonstrate an adequate cause of death. SIDS rates vary widely by country from 4.0/1000 live births in 1988 in New Zealand to 0.29/1000 live births in 1987 in Hong Kong.¹ A number of childcare practices are associated with SIDS such as placing the baby to sleep on the front (prone) position, smoking mother or father, bed sharing, the baby sleeping in a room separate from the parents, and the baby not using a pacifier. Changing the prone sleeping position has been reported to reduce SIDS incidence by more than 50% in many countries.² However, prone sleeping position alone does not explain widely disparate SIDS rates; interaction with other childcare practices appears important.¹

Aims and objectives

This study aimed to document the causes of all unexpected deaths in children under 2 years of age and to identify factors associated with an increased risk of SIDS in Hong Kong.

Methods

An audit of all child deaths (under 2 years of age) that were reported to the coroner between 1 February 1999 and 31 January 2003 was conducted. Deaths were provisionally classified into four groups: A=not SIDS, B=probably not SIDS, C=possibly SIDS, and D=SIDS. The provisional B, C, and D classifications were confirmed by a further review of the death reports and related information.

A case-control study was used to compare information on those children who died (cases) with a group of children who did not die (controls). To ensure that the controls were representative, the selected children were born in Hong Kong in both government and private hospitals; 90% of all anticipated births were included. To match controls to the expected ages of the deaths, dates of interview ('nominated date') were randomly selected using computer-generated numbers for all 1460 days of the study. The age at interview and the 'nominated time' were randomly selected according to anticipated age distribution and estimated time of death distribution of the deaths. A date of birth was then calculated from these data. The randomly allocated 'nominated time' was during one of four time periods: morning routine (6 am-12 midday); afternoon routine (12 midday-6 pm); evening routine (6 pm-12 midnight); night-time routine (12 midnight-6 am). The reference sleep was the longest sleep during this nominated time period. The mortuary staff sought agreement from families for the research nurse to contact them when the families of unexpected child deaths attended the mortuary for identification purposes. Both cases and controls were interviewed at home or at another mutually agreed venue using a standardised questionnaire. The Hong Kong Observatory provided meteorological data related to the estimated time of death or sleep.

Results

Qualitative study

Of the 183 unexpected infant deaths reported to the coroner during the 4-year

Key Messages

1. The rate of sudden infant death syndrome in Hong Kong has fallen from 0.29/1000 in 1987 to $\leq 0.16/1000$ during 1999-2002.
2. Autopsies of unexpected infant deaths by a paediatric pathologist specialising in sudden infant death syndrome increases the likelihood of identifying rare and unusual metabolic diseases. Although there is a global trend toward using the terms "unascertained" or "unknown" rather than "sudden infant death syndrome", the reasons for doing this should be carefully considered in terms of the impact on the family and in terms of monitoring the rate of these deaths.
3. Hong Kong should consider introducing a child mortality review system incorporating those used in Australia and the United Kingdom. Such a system should monitor all child deaths in a systematic fashion and provide feedback to the public, health authorities, and health professionals.

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Table 1. Comparison of the causes of death determined from the coroners' report and a detailed record review

Category	Coroners' diagnosis	Review diagnosis
Congenital heart disease	48	49
Other congenital abnormality	11	11
Inborn error of metabolism	5	5
Injury (cause unspecified)	11*	-
Homicide	2	3
Infanticide/unattended delivery	-	10
Accidental injury	8	9
Possible non-accidental injury	2	7†
Pneumonia	24	12
Gastroenteritis/bowel problem	4	4
Central nervous system infection	2	2
Related to neonatal period	18	14
Other defined illness	13	11
Sudden infant death syndrome	18	33
Unknown or unascertained	12	12
Diagnosis pending	5	1

* 4/11 multiple injury

† 6/7 unexplained cerebral or subdural haematomas

study period, the coroner requested death reports for 52% (95/183), 94 (78 with death report and 16 without) were provisionally classified as groups B, C, or D and were reviewed in detail. Causes of death were classified as those recorded on the death report or other coroner records or those considered most appropriate after detailed review (Table 1). The 15 deaths that were not classified as SIDS by the coroner (pneumonia, bronchopneumonia, aspiration pneumonia or interstitial pneumonitis [n=8], asphyxia due to wedging [n=1], unknown, unascertained or pending [n=6]) were considered potential SIDS cases after review. The SIDS incidence of 0.09-0.16/1000 (range based on whether 18 coroner-classified SIDS or 33 potential SIDS after review) was less than the 0.29/1000 estimated in 1987. As seven of the 33 potential SIDS infants were non-ethnic Chinese, the rate of SIDS in the ethnic Chinese population may be even lower.

A further review of 119 deaths not reported to the coroner did not identify any potential SIDS deaths during the period 1 February 1999 to 31 December 1999.

Case-control study

Families of 18 children who had unexpected deaths were interviewed (two of the deaths were subsequently shown to be not caused by SIDS). Of 268 planned controls, 223 were interviewed. The majority of interviews (90%) took place in the home; 73% of the controls were selected from government hospitals and 27% from private hospitals. Significant differences were noted for child's sex, parents' marital and socioeconomic status, mother's age and parents' occupation (Table 2). There was no significant difference in the mean age of infants who died (20.7 weeks at death) and controls (19.4 weeks at the 'nominated date').

Univariate and sex/socioeconomic-adjusted analyses showed that key SIDS risk factors such as sleeping position, smoking mother/father, and bed sharing were significant in this study (Table 3). Infants who shared a bed with someone other than the parent were at greater risk of death than those who shared with a parent (Table 3).

Although all infants underwent an autopsy, this information was not shared with the majority of families (14/18) and none of the families received any counselling from a paediatrician or nurse. One family had contact with a family doctor and another one with a religious leader. Eleven families had discussions or received counselling from other sources.

Discussion

Despite the small number of cases, this study confirms the key SIDS risk factors of prone sleeping position and parental smoking in Hong Kong. However, other risk factors were also significant. Smoking, particularly antenatal

Table 2. Demographic variables

Demographic variables	Infants died, n=16 No. (%)	Controls, n=223 No. (%)	Odds ratio (95% CI)
Sex			
Female	4 (25.0)	124 (55.6)	1
Male	12 (75.0)	99 (44.4)	3.8 (1.2-12.0)*
Marital status			
Married	12 (75.0)	218 (97.8)	1
Cohabitation	4 (25.0)	5 (2.2)	14.5 (3.5-61.2)†
Parents' socioeconomic status			
Average or above	8 (50.0%)	196 (87.9%)	1
Below average	8 (50.0%)	27 (12.1%)	7.3 (2.5-20.9)†
	Mean (SD)	Mean (SD)	
Baby's age at death/nominated (weeks)	20.7 (18.0)	19.4 (18.8)	0.7
Baby's birth weight (g)	2981 (702)	3219 (450)	0.2
Gestational age (weeks)	37.4 (3.3)	39.3 (4.3)	0.07
Mother's age (years)	27.6 (6.4)	31.6 (5.1)	0.02
Father's age (years)	33.8 (8.8)	35.2 (5.9)	0.2

* P<0.05

† P<0.01

Table 3. Variables showing univariate and sex/socioeconomic-adjusted relationship

	Infants who died (n=16)	Controls (n=223)	OR _U (95% CI)*	OR _A (95% CI)†
Sleeping position on last occasion (when left)				
Not front	12 (75.0%)	217 (97.7%)	1	1
Front	4 (25.0%)	5 (2.3%)	14.5 (3.4-60.9)‡	9.1 (1.8-45.1)‡
Sleeping position on last occasion (when found)				
Not front	12 (75.0%)	213 (95.5%)	1	1
Front	4 (25.0%)	10 (4.5%)	7.1 (1.9-26.0)‡	5.0 (1.2-21.0)§
Mother smokes				
Before pregnancy	3 (18.8%)	15 (6.7%)	3.2 (0.8-12.5)	1.8 (0.4-8.4)
2nd trimester	3 (18.8%)	4 (1.8%)	12.6 (2.6-62.5)‡	18.6 (3.0-117)‡
Since birth	3 (18.8%)	10 (4.5%)	4.9 (1.2-20.1)§	4.6 (0.9-22.7)
Who was looking after the baby (when found)				
Mother	6 (37.5%)	160 (71.7%)	1	1
Others	10 (62.5%)	63 (28.3%)	4.2 (1.5-12.1)‡	6.1 (1.9-20.1)‡
Bedsharing (when found)				
No	7 (43.8%)	168 (75.7%)	1	1
With at least one parent	5 (31.3%)	42 (18.9%)	2.9 (0.9-9.5)	2.6 (0.7-9.5)
With others	4 (25.0%)	12 (5.4%)	8.0 (2.1-31.2)‡	6.6 (1.5-29.3)§
Used adult pillow (when found)				
No	14 (87.5%)	220 (98.7%)	1	1
Yes	2 (12.5%)	3 (1.3%)	10.5 (1.6-67.9)§	5.8 (0.7-49.2)
Used small infant's pillow (when found)				
No	10 (66.7%)	134 (60.1%)	1	1
Yes	5 (33.3%)	89 (39.9%)	0.8 (0.2-2.3)	0.7 (0.2-2.2)
Head and body totally covered (when found)				
No	14 (87.5%)	222 (99.6%)	1	1
Yes	2 (12.5%)	1 (0.4%)	31.7 (2.7-371)‡	108 (7-1666)‡
Amount of last feed				
Not less than usual	10 (62.5%)	218 (97.8%)	1	1
Less than usual	6 (37.5%)	5 (2.2%)	26.2 (6.8-100.5)‡	63 (10.7-368)‡
The baby was ill in the past week				
No	6 (37.5%)	176 (78.9%)	1	1
Yes	10 (62.5%)	47 (21.1%)	6.2 (2.2-18.1)‡	6.1 (1.9-19.1)‡
Outside temperature				
<19.5°C	6 (42.9%)	47 (22.9%)	4.7 (1.1-19.7)§	3.9 (0.9-17.8)
19.5-27.0°C	3 (21.4%)	111 (54.1%)	1	1
>27.0°C	5 (35.7%)	47 (22.9%)	3.9 (0.9-17.1)	3.2 (0.7-15.4)

* OR_U denotes univariate odds ratio† OR_A denotes sex and socioeconomic status-adjusted odds ratio

‡ P<0.01

§ P<0.05

|| 19.5 and 27.0°C were respectively the 25th and 75th percentiles of the temperature readings provided by the Hong Kong Observatory for the estimated time of death (cases) or nominated time (controls)

smoking by the mother, was associated with an increased risk for SIDS,³ a behaviour more difficult to change than prone sleeping practice. The association of smoking with SIDS death should provide strong motivation for mothers (and their partners) to stop (or at least reduce) smoking. Bed sharing with someone other than a parent also appeared hazardous. However, other studies have found bed sharing a risk factor associated with parents who smoke.^{4,5} While there are likely to be safe methods of bed sharing, parents need to be aware of its potential risks. Covering the baby's head with bedding was another important risk factor for SIDS death.

Review of the coroners' records showed that the terms "unknown" and "unascertained" were more likely to be used when the postmortem examination was not undertaken by the study pathologist. Although these terms may reassure the pathologist that an unnatural cause remains an option, it gives bereaved parents little comfort and probably increases their sense of guilt. These findings emphasise the importance of undertaking investigations for inborn er-

rors of metabolism that would not be identified unless specifically sought with specialised investigations. The new Hong Kong Coroners Ordinance was published in April 1997 and implemented in 1998. Review of the coroners' death reports and other information as part of this study has highlighted potential discrepancies in assignment of the cause of death (Table 1). A number of these causes of death (brain injury, suspected 'shaken baby syndrome' and 'murder-suicide') were unrelated to SIDS but have important implications, both medicolegally and in terms of preventive child health. The Hong Kong Paediatric Society has recommended a child mortality review process. Our data highlight the potential benefit that such a process could have in monitoring and investigating all child deaths, both for deaths that are reported to the coroner and those not. How such a system should be established would require discussion between the various stakeholders but the experience of systems such as the Confidential Enquiry into Stillbirths and Deaths in Infancy in the United Kingdom and the Victoria Institute of Forensic Medicine in Australia should be utilised.

Conclusion

The unexpected and unexplained death of an infant is one of the most tragic events that any parent may experience. Our data suggest that although the incidence of SIDS in Hong Kong has fallen from 0.29/1000 in 1987 to $\leq 0.16/1000$ during 1999-2002, there is still the potential to reduce these deaths further by ensuring that parents are aware of SIDS risk factors. Pregnancy and the birth of a child are intervention points to encourage parents to stop or reduce smoking. The association between smoking and SIDS may help motivate parents to quit smoking. Although bed sharing is a common practice in Hong Kong, parents should be aware that it is not always a safe practice. Parents should also be informed of the risk of other unsafe sleeping practices such as the use of adult pillows and situations that enable the baby's head to be totally covered by bedding. Support and counselling for bereaved parents is limited and providing families with more information about the autopsy findings at an earlier stage is needed. The use of the terms "unascertained" or "unknown" rather than "SIDS" should be carefully considered in terms of the impact on the family and in terms of monitoring the rates of these deaths. The current coronial system is unable to systematically involve professionals from different disciplines when determining the cause of child and other deaths and it is unable to provide systematic documentation and interpretation of factors that may help prevent some of these deaths. A recommendation is made to establish a child mortality review system to monitor all child deaths systematically in order to provide feedback to the public, health authorities, and professionals.

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