Head covering – a major modifiable risk factor for sudden infant death syndrome: a systematic review

P S Blair,1 E A Mitchell,2 E M A Heckstall-Smith,1 P J Fleming1

ABSTRACT

Background: Some victims of sudden infant death syndrome (SIDS) are found with their heads covered with bedclothes, but the significance of this is uncertain. The aim of this review is to describe the prevalence of head covering, the magnitude of the risk and how far the suggested causal mechanisms agree with current epidemiological evidence.

Methods: Systematic review of population-based age-matched controlled studies.

Results: Controlled observations of head covering for the final sleep were found in 10 studies. The pooled prevalence in SIDS victims was 24.6% (95% CI 22.3% to 27.1%) compared to 3.2% (95% CI 2.7% to 3.8%) among controls. The pooled univariate odds ratio (OR) was 9.6 (95% CI 7.9 to 11.7) and the pooled adjusted OR from studies mainly conducted after the fall in SIDS rate was 16.9 (95% CI 12.6 to 22.7). The risk varied in strength but was significant across all studies. In a quarter of cases and controls head covering had occurred at least once previously (pooled adjusted OR = 1.1; 95% CI 0.9 to 1.4). The population attributable risk (27.1%; 95% CI 24.7% to 29.4%) suggests avoiding head covering might reduce SIDS deaths by more than a quarter.

Conclusions: The epidemiological evidence does not fully support postulated causal mechanisms such as hypoxia, hypercapnoea and thermal stress, but neither does it support the idea that head covering is part of some terminal struggle. Head covering is a major modifiable risk factor associated with SIDS deaths and parental advice to avoid this situation should be emphasised.

In cases of sudden infant death syndrome (SIDS) the significance of finding the head or face of the infant covered by bedclothes remains uncertain in spite of its emergence as a putative risk factor. Prior to the acceptance of the label SIDS in the 1960s, it was common to attribute these unexpected deaths to “accidental mechanical suffocation” because the sleeping infant was often found with the face down in the bedclothes, the face covered by bedclothes or next to a sleeping parent.1 2 The assumption of smothering was criticised at the time, not least for the lack of evidence when apportioning blame to parent–infant care practices.3 Post mortem investigation of suspected asphyxial deaths were then, as now, largely inconclusive and anecdotal reports of attempts to reproduce the same conditions using ordinary bedding among live infants failed to induce hypoxaemia.4 5 Neither did the emerging epidemiological profile of SIDS deaths from the United States fit with the theory of suffocation as the peak age of deaths was not during the vulnerable first few weeks after birth but at 3 months of age and although there was a winter preponderance the highest incidence occurred among the black population in the warmer southern states where infants used fewer or lighter bedclothes.6 After many years of research there is growing evidence that SIDS is more likely to be a consequence of a wide range of infant and environmental interactions than due to one particular cause. Head covering may be part of a chain of events for some of these deaths and suggested causal mechanisms include mechanical occlusion of the airways, rebreathing of expired air (both of which could lead to hypoxia or hypercapnoea) and thermal stress (leading to generalised or localised hyperthermia). This is a systematic review of the prevalence of head covering among victims of SIDS and age-matched control infants and an evaluation of how far the associated risk agrees with the epidemiological evidence.

METHODS

We have attempted to follow the MOOSE guidelines for systematic reviews.6 Customised databases at both collaborating research centres were utilised along with PubMed, which includes Medline and Old Medline citations. The primary
search used the term SIDS with words describing the risk factor (“head”, “cover” or “bedclothes”) or using the generic term “risk factor” for different study designs (“case-control”, “cohort”, “case-series”, “review”). A secondary search of all the references from the relevant papers was also conducted. Potential studies written in languages other than English were included and read for any reference to head covering. Attempts to gain access to unpublished data were made at international conferences within this field and members of the International Society for the Study and Prevention of Infant Death (ISPID), who from many different countries, were asked if there were additional studies we had not identified. Authors of included studies were contacted if specific numerators or denominators were missing from the published manuscripts.

Over 600 papers were identified along with more than 100 epidemiological investigations. Pre-existing criteria were used for inclusion in the meta-analysis: head covering had to be reported, distinct from other forms of facial occlusion and analysed across the study population, and the study had to be controlled with observations of age-matched infants and conducted between January 1950 and May 2007.

The pooled univariate estimate of the odds ratio was calculated using the Mantel-Haenszel technique. The pooled adjusted odds ratio was calculated using the statistical package Review Manager provided by the Cochrane Collaboration. The test for heterogeneity of the odds ratios was conducted using Woolf’s method.

RESULTS

Included studies

Non-English manuscripts of epidemiological studies were found but did not measure head covering, while unpublished findings were sought but not discovered. Studies that had not separated observations of head covering from other forms of facial occlusion, such as infants found face down in the bedding, were excluded from the meta-analysis along with studies that had only reported observations for subgroups of SIDS infants (n = 8). A further 10 studies reported a population-based prevalence of head covering among SIDS victims but had not collected similar information from control families and thus were also excluded. Controlled observations of head covering when the SIDS infants were discovered dead or control infants woke up from a reference sleep were found in 10 studies (table 1).

Prevalence and risk

The prevalence of head covering among SIDS victims (table 1) in each study varied from 15% to 48% with a pooled mean prevalence of 24.6% (95% CI 22.3% to 27.1%). The three earliest studies conducted before the major decline in the SIDS rates reported a higher prevalence of 36.1% (95% CI 30.9% to 41.8%), although head covering remained a consistent feature in subsequent studies with a mean prevalence of 21.0% (95% CI 18.5% to 25.7%). Among the control infants the prevalence of head covering after the reference sleep was much lower, ranging from 0% to 6% with a pooled mean prevalence of 3.2% (95% CI 2.7% to 3.8%).

A test for heterogeneity of the odds ratios between studies suggests the risk associated with head covering was significantly different (p < 0.01), although this was in strength rather than direction; the univariate odds ratio varied between two-fold and almost 50-fold but denoted a significant risk in all of the studies (fig 1). The pooled univariate estimate was 9.6 (95% CI 7.9 to 11.7). In seven of the 10 studies the odds ratios for head covering were adjusted for other factors associated with SIDS (all but one of these studies were conducted after the dramatic fall in SIDS rates). The number of adjusted factors and how these were defined varied between studies (see footnote in table 1), but the multivariate risk estimates for head covering remained highly significant in each investigation (fig 2). The pooled adjusted odds ratio was 16.9 (95% CI 12.6 to 22.7). Assuming that head covering is causally related to SIDS, the data from these studies suggest the population attributable risk would be 27.1% (95% CI 24.7% to 29.4%).

Four studies also included questions to the parents regarding whether their infant had ever been found previously with bedclothes covering the face or head (table 2). Over a quarter of SIDS and control parents responded that this had occurred at least once prior to the death or reference sleep (26% SIDS vs 27% controls), although for both sets of infants head

<table>
<thead>
<tr>
<th>Study</th>
<th>SIDS, n/N (%)</th>
<th>Controls, n/N (%)</th>
<th>Unadjusted OR (95% CI)</th>
<th>Adjusted OR (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1958–1961 London and Cambridge, UK</td>
<td>28/102 (19.6)</td>
<td>1/102 (1.0)</td>
<td>38.6 (9.7 to 156.4)</td>
<td>38.6 (9.7 to 156.4)</td>
</tr>
<tr>
<td>1968–1974 Lower Saxony, Germany</td>
<td>58/130 (45.4)</td>
<td>8/130 (6.2)</td>
<td>12.5 (4.9 to 33.1)</td>
<td>12.5 (4.9 to 33.1)</td>
</tr>
<tr>
<td>1981–1983 Norway</td>
<td>10/115 (8.7)</td>
<td>11/115 (9.5)</td>
<td>1.1 (0.3 to 4.0)</td>
<td>1.1 (0.3 to 4.0)</td>
</tr>
<tr>
<td>1987–1988 Sweden, Denmark, Norway</td>
<td>65/236 (27.8)</td>
<td>4/236 (1.7)</td>
<td>37.1 (10.8 to 129.2)</td>
<td>37.1 (10.8 to 129.2)</td>
</tr>
<tr>
<td>1989–1991 Munster and Detmold, Germany</td>
<td>20/56 (39.3)</td>
<td>2/56 (3.6)</td>
<td>7.8 (2.0 to 29.8)</td>
<td>7.8 (2.0 to 29.8)</td>
</tr>
<tr>
<td>1990–1994 Munster and Detmold, Germany</td>
<td>40/100 (40.0)</td>
<td>1/100 (1.0)</td>
<td>38.0 (10.0 to 136.0)</td>
<td>38.0 (10.0 to 136.0)</td>
</tr>
<tr>
<td>1993–1994 England</td>
<td>12/30 (40.0)</td>
<td>2/30 (6.7)</td>
<td>6.7 (1.6 to 27.2)</td>
<td>6.7 (1.6 to 27.2)</td>
</tr>
<tr>
<td>1995–1996 Netherlands</td>
<td>30/150 (20.0)</td>
<td>10/150 (6.7)</td>
<td>3.8 (1.4 to 10.2)</td>
<td>3.8 (1.4 to 10.2)</td>
</tr>
<tr>
<td>1996–1997 Japan</td>
<td>10/50 (20.0)</td>
<td>1/50 (2.0)</td>
<td>7.0 (1.7 to 27.7)</td>
<td>7.0 (1.7 to 27.7)</td>
</tr>
<tr>
<td>1997–1998 Hong Kong</td>
<td>15/75 (20.0)</td>
<td>4/75 (5.3)</td>
<td>3.7 (1.2 to 11.5)</td>
<td>3.7 (1.2 to 11.5)</td>
</tr>
<tr>
<td>1998–1999 Hong Kong</td>
<td>5/30 (16.7)</td>
<td>1/30 (3.3)</td>
<td>5.0 (1.1 to 24.0)</td>
<td>5.0 (1.1 to 24.0)</td>
</tr>
<tr>
<td>1999–2000 Hong Kong</td>
<td>14/70 (20.0)</td>
<td>3/70 (4.3)</td>
<td>4.6 (1.3 to 16.0)</td>
<td>4.6 (1.3 to 16.0)</td>
</tr>
<tr>
<td>2000–2001 Hong Kong</td>
<td>20/80 (25.0)</td>
<td>4/80 (5.0)</td>
<td>4.0 (1.2 to 13.2)</td>
<td>4.0 (1.2 to 13.2)</td>
</tr>
<tr>
<td>2001–2002 Hong Kong</td>
<td>10/50 (20.0)</td>
<td>2/50 (4.0)</td>
<td>5.0 (1.3 to 18.6)</td>
<td>5.0 (1.3 to 18.6)</td>
</tr>
<tr>
<td>2002–2003 Hong Kong</td>
<td>15/75 (20.0)</td>
<td>4/75 (5.3)</td>
<td>3.7 (1.2 to 11.5)</td>
<td>3.7 (1.2 to 11.5)</td>
</tr>
<tr>
<td>Total</td>
<td>304/1234 (24.6)</td>
<td>121/1234 (9.8)</td>
<td>33.2 (9.5 to 116.7)</td>
<td>33.2 (9.5 to 116.7)</td>
</tr>
</tbody>
</table>

*Univariate odds ratio (OR) not reported but calculated from the figures given; **univariate OR not calculated because of empty cell; ***univariate OR not calculated because of empty cell; ****univariate OR not calculated because of empty cell.
covering was not described as a common event. The pooled
univariate odds ratio for the four studies was 1.0 (95% CI 0.8 to
1.3), while the pooled adjusted odds ratio from just two studies
was 1.1 (95% CI 0.9 to 1.4), suggesting no difference in previous
instances of head covering between the two groups (fig 3).

Head covering as part of an agonal event
Parental narrative accounts of the deaths and the wider
epidemiological evidence do not lend support to the idea that
head covering is simply a consequence of some terminal
struggle. Although SIDS deaths are mostly unobserved events,
it is not uncommon for the parents to be in the same room as
the infant and very rare that parents are woken or alerted to the
unfolding tragedy. A review of 300 detailed narrative accounts
from our own SIDS deaths in Avon over the last 20 years
suggests that any change in position is minimal, often described
in terms of infants rolling to the prone position or moving down
under the bed covers. Reports of observations at the time of
death by Gormally and Matthews in Ireland showed that the
bedding was undisturbed in over two thirds of SIDS cases. These
were uncontrolled observations but confirmed by the large
European study by Carpenter et al:1% of the SIDS infants
showed no movement between being put down and discovered
compared to 70% of the control infants. Prospective data of sleep
recordings among infants who eventually died of SIDS have
shown a reduced number of body movements during sleep, a
decreased propensity to arouse from sleep and a lowering of
cardiac variability. Further data of SIDS infants who actually
died while on a monitor show a fall in the heart rate before death
and decreased occurrence of partial or complete auto-resuscitation
rather than recordings suggestive of an infant struggle.

An alternative explanation might be that head covering is
common during infant sleep but the covers are often removed
during the process of waking, in which case the lack of
movement among SIDS cases may explain the association.
However, overnight video recordings in the home setting
conducted in New Zealand suggest head covering is rare among
infants sleeping in a cot: of 40 infants observed over an 8 h
period head covering occurred just once.

Head covering and the prone sleeping position
In the last 20 years in Avon, England, the proportion of SIDS
victims put down to sleep in the prone position has fallen from
89% to 24%, while the SIDS rate has decreased from over 2 to
less than 0.5 per 1000 live births. The concomitant fall in the
use of the prone position may partly explain the slightly reduced
prevalence of head covering as some studies have reported that
head covering was more common among infants found in the
prone sleeping position. As Real points out, when older
infants start to “crawl” in the prone position, they often move
backwards rather than forwards for the first few weeks. L’Hoir
et al demonstrated that head covered SIDS infants tended to be
older and to have moved down under the covers compared to the
cases found uncovered, an observation confirmed by the study of Fleming et al which also showed that this downward
movement was much more common among the covered SIDS
infants compared to the covered controls. Intriguingly, a recent analysis from New Zealand suggests head covering was
associated with a decreased risk for SIDS among prone sleeping
infants found face straight down in the mattress.

A study of healthy infants in the United States put down in
the prone position and covered with soft bedding has shown
that protective behaviour such as head repositioning strategies
were not so much related to infant age as to previous experience
of sleeping in the prone position; inexperienced prone sleeping
infants tended to nuzzle into the bedding rather than lift or
turn their heads. A similar study by Skadberg and Markestad

Figure 1 Forest plot of unadjusted odds ratio (and 95% CI) for infants
found with head covered by bedclothes after last sleep.

Figure 2 Forest plot of adjusted odds ratio (and 95% CI) for infants
found with head covered by bedclothes after last sleep.

Figure 3 Forest plot of unadjusted odds ratio (and 95% CI) for infants
ever previously found with head covered by bedclothes.
Wilson or unsupervised in a room during the day-time sleep. Despite infants were discovered with the bedclothes covering the head results we report in table 2. This reduced prevalence may episodes of head covering were less common among both co-
only three (7%) were found with their head covered. Data from 
UK48 suggest that fewer co-sleeping SIDS infants were 
found prone with the head down in soft bedding, but the role it 
sometimes under the bedclothes, but results from New 
Zealand suggest infants found both face down and head 
covered are less common. 45 The early claims by Woolley 4 and 
Tappin et al that a child cannot be suffocated by “ordinary 
bedclothes” has been described as anecdotal55 56 but has yet to 
be refuted. Permeability tests for airflow through various 
bedding materials suggest little resistance. Duvets perform 
slightly better than conventional blankets and although the 
resistance increased with unwashed and wet bedding. British Standards Institution tests suggest it should not pose a threat to
ventilation and infections are all associated with SIDS deaths. A 
in depth review is given by Guntheroth and Spiers 58 who conclude that current evidence
Curie et al of a highly significant interaction between duvets and 
head covering are not supportive of the rebreathing 
hypothesis, given that duvets are more permeable than 
conventional blankets. Malcolm et al also found that the 
presence of a pacifier (dummy) promoted an excess of CO₂ in 
the inspired air, yet the epidemiological evidence suggests 
pacifiers may lower the risk of SIDS. 57

Another potential mechanism of causality involves heat stress. Excessive clothing and bedding, warmer rooms, reduced ventilation and infections are all associated with SIDS deaths. A history of profuse sweating has been reported among SIDS

### Table 2 Reported risk estimates for infants ever previously found with head covered by bedclothes

<table>
<thead>
<tr>
<th>Study</th>
<th>SIDS, n/N (%)</th>
<th>Controls, n/N (%)</th>
<th>Unadjusted OR (95% CI)</th>
<th>Adjusted OR (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Wilson 1997–1990</td>
<td>80/277 (28.8)</td>
<td>338/1103 (30.6)</td>
<td>0.9 (0.7 to 1.2)</td>
<td>0.9 (0.7 to 1.2)</td>
</tr>
<tr>
<td>Brook 1992–1995</td>
<td>35/146 (24.0)</td>
<td>35/274 (12.8)</td>
<td>2.5 (1.4 to 4.3)</td>
<td>2.2 (1.0 to 4.6)</td>
</tr>
<tr>
<td>Fleming 1993–1996</td>
<td>90/316 (28.5)</td>
<td>358/1298 (27.6)</td>
<td>1.1 (0.8 to 1.5)</td>
<td>1.1 (0.9 to 1.4)</td>
</tr>
<tr>
<td>Tappin et al 1996–2000</td>
<td>23/128 (18.0)</td>
<td>74/277 (26.7)</td>
<td>0.6 (0.3 to 1.0)</td>
<td>–</td>
</tr>
<tr>
<td>Total</td>
<td>223/867 (26.3)</td>
<td>805/2952 (27.3)</td>
<td>1.0 (0.8 to 1.3)</td>
<td>1.1 (0.9 to 1.4)</td>
</tr>
</tbody>
</table>

*Univariate odds ratio (OR) not reported but calculated from the figures given; †pooled univariate OR using the Mantel-Haenszel estimate; ‡pooled adjusted OR using In (effect size) and SE (ln (effect size)); §adjusted for infant age, gender, race, time of death, geographic location, season, sleeping position, birthweight, gestation, admission to special care baby unit, maternal age, attendance at antenatal class, maternal smoking, breastfeeding, marital status, parity, socio-economic status and occupation; ¶adjusted for gender, gestational age, parity, birthweight, sleeping position, routine co-sleeping, use of prescription drugs or illness in the previous week, usually found sweaty on waking, history of changing position during sleep, swaddling, tog value of bed covers, age of mattress, use of cot bumper, maternal age, whether breastfed, marital status, parental smoking, maternal schooling, deprivation score, social class and previous infant death.

### Head covering and bed sharing

Home studies of healthy infants in New Zealand with overnight video and physiological recordings showed that head covering was more common during sleep among bed-sharing infants than those sleeping alone in a cot, but also that parents tended to adjust the infant covering during the night. Data from the UK60 suggest that fewer co-sleeping SIDS infants were discovered head covered (7% vs 19% among solitary sleeping SIDS infants), a finding confirmed by a more recent study in Scotland61 where of 46 SIDS infants sharing the parental bed only three (7%) were found with their head covered. Data from the Scottish study also suggested that previously reported episodes of head covering were less common among both co-sleeping SIDS infants (15%) and controls (20%) compared to the results we report in table 2. This reduced prevalence may indicate a protective effect from the parental presence; a more recent analysis of the UK data suggests twice as many SIDS infants were discovered with the bedclothes covering the head either outside the parental bedroom during the night-time sleep or unsupervised in a room during the day-time sleep.83 Despite these observations bed sharing, particularly among parents who smoke, is strongly associated with SIDS and it would be dangerous to recommend bed sharing as a strategy to reduce the prevalence of head covering.

### DISCUSSION

The prevalence of head covering among studies conducted after the fall in SIDS rates suggests that a fifth of SIDS infants are still being discovered with bedclothes covering the face or head. The initial “Back to Sleep” intervention campaign conducted in the UK in 1991 included advice for parents to avoid infant head covering, but this seems to have had far less impact than the campaign’s main message regarding infant sleeping position. The pooled adjusted estimate suggests an almost 17-fold increased risk associated with head covering, but this seems to have had far less impact than the campaign’s main message regarding infant sleeping position. The findings by L’Hoir et al of a highly significant interaction between duvets and head covering are not supportive of the rebreathing hypothesis, given that duvets are more permeable than conventional blankets. Malcolm et al also found that the presence of a pacifier (dummy) promoted an excess of CO₂ in the inspired air, yet the epidemiological evidence suggests pacifiers may lower the risk of SIDS.57

Another potential mechanism of causality involves heat stress. Excessive clothing and bedding, warmer rooms, reduced ventilation and infections are all associated with SIDS deaths. A history of profuse sweating has been reported among SIDS
Sleeping infants that covering of the infant’s face with a bed sheet, although associated with only mild increases in body temperature, induces significant changes in cardio-respiratory and autonomic parameters. In the study by Kleemann et al. head covered infants tended to show signs of profuse sweating, an association also reported by Carpenter et al., which agrees with the interaction found by L’Hoir et al. of head covered infants found under duvets which may be more permeable but have greater heat insulation than conventional covers. Thicker covers are commonly used more often during the colder months, but incongruous to all these findings is the fact that the winter peaks of SIDS deaths have substantially diminished at a time when the prevalence of head covered SIDS infants is still quite high.

Parental advice
In the UK the “Feet to Foot” campaign, advising parents to place the feet of the infant at the foot of the cot to prevent head covering, was launched by the Foundation for the Study of Infant Death in 1997 and has subsequently been endorsed by the American Academy of Pediatrics (AAP). Although this advice seems intuitively sensible there is, as yet, no published evidence that this practice reduces the risk of head covering or lowers the risk of SIDS. Findings from the study by L’Hoir et al. suggest that a lightweight cotton sleeping sack (“trappelzak”) used by the majority of Dutch parents may be protective against SIDS both in terms of preventing the infant from turning prone and avoiding head covering. Further evidence than just this one study may be needed but such a practice deserves closer scrutiny given the continued low SIDS rates in Holland.

SUMMARY
The population attributable risk suggests that more that a quarter of SIDS deaths might be avoided if the possibility of infant head covering were eliminated. Similar to the prone sleeping position, head covering is a modifiable risk factor with a potential for further reducing SIDS deaths despite a lack of a complete explanation for the causal mechanism involved.

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REFERENCES


