Short reports

Sleep apnoea in infants with congenital stridor

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SUMMARY Indices of central apnoea, respiration rate, heart rate, and body movements were comparable in seven infants with congenital stridor (infantile larynx) and matched controls. Obstructive sleep apnoea was only observed in three of seven infants with stridor. Periodic breathing was less common in infants with stridor than in controls.

Guilleminault et al have reported death (attributed to sudden infant death syndrome) of an infant with congenital stridor in whom obstructive apnoea was shown during life. As congenital stridor is not uncommon in infancy and as its severity is influenced by coincidental respiratory infection, we set out to study sleep apnoea in seven otherwise healthy infants with this condition.

Patients and methods

Seven infants with a history of stridor from birth or the early weeks of life were studied. There were four boys and three girls, with a mean gestation of 40 weeks (range 39–40 weeks) and mean birth weight of 3100 g (range 2670–4410 g), whose mean chronological and postconceptional ages were 10 weeks (range 7–15 weeks) and 50 weeks (range 47–55 weeks), respectively, when initial studies were conducted. In each case the diagnosis of congenital laryngeal stridor ('infantile larynx') was made on the basis of history, examination, and findings at laryngoscopy. Chest x ray films yielded normal results and visualisation of the tracheal air column and barium filled oesophagus by lateral x ray films of the neck excluded important tracheal or oesophageal compression. There was no clinical suspicion of cardiac or neurological abnormality. One infant had mild to moderate micrognathia. Apart from stridor these infants were otherwise well when studies were carried out.

Seven healthy normal infants, matched for sex, gestation, and postconceptional age (to within 2 weeks in six cases) were selected as controls. Four had had no previous illnesses, but three had suffered from respiratory infections from which they had recovered some weeks earlier. All were free of symptoms and apparently healthy when studied. Consent for participation in the study was obtained from parents of all infants after a detailed explanation of the purpose and plan of the proposed investigation.

Details of monitoring procedures and analytical methods are described in detail on pages 1057–8 in our accompanying paper. Central apnoeic pauses were defined as periods during which there was absence of chest and abdominal movements and stopping of air flow at the nostrils for at least six seconds. Indices of central apnoea were assessed for apnoeic pauses >6 seconds in relation to sleep state. The mean duration of apnoeic pauses and the episode of apnoea of longest duration were noted. The frequency of occurrence and duration of periodic breathing and gross body movements were also computed.

Results

Table 1 gives the results for apnoea variables, per cent sleep, respiration rate, and heart rate in infants with stridor and matched controls. No differences were observed between corresponding variables. No infant had prolonged (>15 seconds) central apnoea. Recurrent brief (>3 and <6 seconds) episodes of obstructive apnoea was observed in three infants and prolonged (>6 seconds) episodes of obstructive apnoea in one. No control infant had either prolonged episodes of central or episodes of obstructive apnoea.

Table 2 gives the results for gross body movements and periodic breathing. Number and duration of gross body movements were comparable in infants with stridor and their matched controls.
Table 1  Apnoea variables, per cent sleep, and respiration and heart rates in relation to sleep phase in seven infants with stridor and their case controls. Values are mean (SE)

<table>
<thead>
<tr>
<th>Sleep phase</th>
<th>Apnoea index (n=7)</th>
<th>Apnoea attack rate (n=7)</th>
<th>Episode of longest duration (sec) (n=7)</th>
<th>Mean duration of apnoea (sec) (n=7)</th>
<th>Apnoa per cent (n=7)</th>
<th>Sleep per cent (n=7)</th>
<th>Respiration rate (n=7)</th>
<th>Heart rate (n=7)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Active</td>
<td>0.44 (0.36)</td>
<td>0.06 (0.05)</td>
<td>Active 3.81 (1.87)</td>
<td>Active 3.15 (1.49)</td>
<td>Active 1.70 (1.36)</td>
<td>Active 37.00 (3.42)</td>
<td>Active 128.20 (1.41)</td>
<td>Active 120.70 (3.98)</td>
</tr>
<tr>
<td>Quiet</td>
<td>0.36 (0.14)</td>
<td>0.05 (0.02)</td>
<td>Quiet 7.37 (1.57)</td>
<td>Quiet 7.52 (1.15)</td>
<td>Quiet 1.29 (0.50)</td>
<td>Quiet 31.40 (3.18)</td>
<td>Quiet 120.70 (3.98)</td>
<td>Quiet 120.70 (3.98)</td>
</tr>
</tbody>
</table>

Mann-Whitney U statistical test applied but not significant for each variable.

Table 2  Gross body movements and periodic breathing in relation to sleep phase in seven infants with stridor and their matched case controls. Values are mean (SE)

<table>
<thead>
<tr>
<th>Sleep phase</th>
<th>Body movements</th>
<th>Periodic breathing</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Total duration (min/100 min sleep)</td>
<td>No/100 min sleep</td>
</tr>
<tr>
<td>Active</td>
<td>3.55 (0.70)</td>
<td>23 (3.94)</td>
</tr>
<tr>
<td>Quiet</td>
<td>1.71 (1.00)</td>
<td>18 (9.47)</td>
</tr>
</tbody>
</table>

Mann-Whitney U statistical test applied but not significant for each variable.

There was less periodic breathing in infants with stridor than in controls, but this difference did not reach significance.

Discussion

The results indicate that apnoea variables, per cent sleep, respiration rate, heart rate, and gross body movements were similar in infants with mild stridor and normal healthy controls in both active and quiet stages of sleep.

Brief or prolonged episodes of obstructive apnoea was occasionally observed in infants with stridor but not in matched controls. In their review of congenital stridor in infancy Kahn et al reported an infant in whom tracheomalacia had been diagnosed and who died suddenly and unexpectedly at 4–5 months of age. In that condition stridor is predominantly expiratory and not inspiratory as in the present cases. Guilleminault et al reported the sudden death of a 5 month old infant who presented as a case of ‘near miss’ sudden infant death syndrome at 3 months of age. Twenty four hour polygraphic sleep monitoring was carried out within 30 hours of her death. The data obtained were compared with those from healthy control infants and other infants with ‘near miss’ sudden infant death syndrome who were of similar age. The number of mixed and obstructive respiratory events was abnormally high in the case described. The finding of obstructive episodes of apnoea in three of the seven infants in the present series suggests that superimposed respiratory infection might increase still further the hazard of episodes of obstructive apnoea in such cases, with its attendant adverse effects on heart rate and oxygenation.

Periodic breathing, defined as two episodes of apnoea of 3 seconds or more within 20 seconds of each other, was uncommon in infants with stridor, occurring in only two index cases and in each case control. This difference is obscure in the case control comparison (Table 2), which failed to show a significant reduction in periodic breathing in infants with stridor. It is conceivable that upper airways obstruction modifies feedback loops so as to influence central respiratory control mechanisms, thereby reducing or abolishing periodic breathing. It is of interest that the case described above had less periodic breathing than the controls selected.

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References


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