Background: There is little information regarding full polysomnographic examination of infants for the evaluation of stridor or stertor.

Objective: To determine the usefulness of polysomnographic examination in the evaluation of airway disorders in infants.

Design: Case review series.

Setting: Tertiary pediatric care center and sleep disorders center.

Patients: Younger than 10 months.

Intervention: Full polysomnography and treatment with a positive-pressure assistive device or surgery if indicated.

Main Outcome Measures: Electroencephalographic findings, amount and length of apnea, percentage of desaturation and carbon dioxide retention, sleep architecture, amount of hypopnea as well as sleep arousals, episodes of gastroesophageal reflux, and clinical follow-up.

Results: Of 60 full pediatric polysomnograms performed at our institution each year, only 17 were obtained in children younger than 10 months for determination of symptoms of stridor or stertor over the past 8 years. In several cases, interpretation of 4-channel studies led to the wrong conclusions and inadequate treatment. Diagnoses made using full polysomnography included 3 children with seizure disorders, 1 with disorganized brain activity, 2 with gastroesophageal reflux, and 7 with anatomical obstructions, 3 of whom were successfully treated with a positive-pressure assistive device and 4 of whom were treated surgically.

Conclusions: Full polysomnography provides physiological data that complement anatomical data obtained via endoscopy and is a useful tool for evaluating the significance of airway disorders in infants.

PATIENTS AND METHODS

All infants younger than 10 months with signs of airway distress while awake and undergoing full PSG were included in this review. Full PSG was performed at the Minnesota Regional Sleep Disorders Center at Hennepin County Medical Center, Minneapolis. The examination consisted of respiratory plethysmography, respiratory thermistor, transcatheter carbon dioxide monitoring, electrocardiography, pulse oximetry, electroencephalography, electrooculography, chin electromyography, and esophageal pH meter. Body movements were recorded by a technician and recorded on videotape. All noises made during sleep were recorded. Prior to 6 months of age, sleep was scored as active sleep, quiet sleep, and indeterminate sleep. After 6 months of age, sleep was recorded using standard sleep staging criteria: time in bed, total sleep time, sleep efficiency, number of awakenings, percentage of wakefulness and percentage in stages 1, 2, 3/4, and rapid eye movement (REM) sleep, and latencies to stage 1, stage 2, 3-minute sleep, and REM. In addition, the results of a pH probe, oxygen saturation, carbon dioxide levels, the results of electrocardiography during wakefulness and sleep, myoclonus, arousals, snoring, and apnea were all recorded. A trial of a positive airway pressure device (continuous positive airway pressure [CPAP] or bilevel positive airway pressure [BiPAP]) was used if indicated by significant disordered breathing or hypoxemia during the study. All results were interpreted by a certified polysomnographer. Four-channel studies when used were done in the hospital or at home and recorded respiratory plethysmography, electrocardiography, and pulse oximetry.

from state-dependent hypoventilation or obstructive sleep apnea in children with neurologic as well as anatomical anomalies. The purpose of this study was to evaluate the usefulness of obtaining full PSGs in children aged 10 months or younger with airway problems other than apnea of prematurity.

RESULTS

At our institution more than 60 full pediatric PSGs are performed per year. Our review was able to retrieve only 17 performed on children younger than 10 months over the past 8 years for the evaluation of stridor or stertor. These children were initially all discharged home following birth but then seen in an outpatient setting with airway problems. Six of these children had apnea plus stertor, 6 had stridor alone, 2 had stertor and stridor, and 3 had stertor alone.

Seizure activity was discovered as the source of apnea in 4 infants (patients 1, 5, 6, and 9). Three of these infants had prior 4-channel studies because of airway problems. Obviously, the 4-channel study does not reveal nor is it looking for a seizure source. In all cases, the 4-channel study revealed apnea with bradycardia, which led to the initial mismanagement of atropine therapy until the correct diagnosis of seizure activity was made with a full PSG. It should be noted that in all 4 of these infants seizure activity was strongly suspected as the source of airway problems because of their medical history.

Four children had airway problems secondary to retrognathia. These were not obvious cases of retrognathia but more subtle and missed initially at birth. Two of these patients had normal findings on sleep studies and 2 were treated successfully with CPAP or BiPAP.

Four children who underwent rigid laryngoscopy and bronchoscopy were noted to have supraglottic problems. In 1 infant with laryngomalacia, the sleep study revealed more than 35 breathing-related arousals per hour with minimal desaturations and 7 episodes of gastrointestinal reflux below pH 4.0 during the study, but none associated with apnea. The child previously underwent a 4-channel study, the results of which were normal, and was treated with gastroesophageal reflux medication. A trial of CPAP was not tolerated. The child underwent a supraglottoplasty and has since done well. The second child was noted to have a true bifid epiglottis on endoscopy with noisy breathing. An epiglottectomy was considered but the PSG findings were normal and the child was treated conservatively. A third child with severe cerebral palsy, pooling of secretions, and laryngomalacia had mixed apnea shown on both a 4-channel and full PSG. The full PSG also showed significant carbon dioxide retention. The application of BiPAP corrected the problem but was not tolerated and the child was successfully treated with a supraglottoplasty and theophylline. A fourth infant, also with significant neurologic deficit, had a pneumogram revealing significant obstructive events. The child underwent a procedure to reduce gastroesophageal reflux as well as a supraglottoplasty to treat laryngomalacia. A follow-up full PSG revealed continued hypoxia and carbon dioxide retention now corrected by BiPAP at night with good tolerance.

Two children were diagnosed before the PSG as having significant nasal obstruction without choanal atresia. Both were treated with intranasal curettage and dilation following a PSG. One child with abnormal PSG findings before surgery could not tolerate CPAP and underwent a second sleep study following surgery, the findings of which were improved but still abnormal. However, the child was now able to tolerate CPAP.

Six infants had no evidence of physiological airway obstruction during their sleep study. One of these infants had seizure activity but there was no evidence of airway obstruction. The other 5 children had studies performed because of symptoms. The normal result on sleep study did help to influence more conservative management in all these children.

In looking at the electroencephalogram, the majority of apnea or sleep disruption occurred during REM sleep. Patient 4 had 74 hypopnea episodes (49 non-REM and 25 REM). However, the more severe episodes occurred during REM. The same was true for patient 8, who had 161 respiratory arousals (96 non-REM and 65 REM). However, his central apnea events all occurred during REM. All significant decreases in pH occurred during REM sleep.
Southall et al classify airway disorders in infants into 1 of 4 different clinical scenarios: apnea of prematurity, severe cyanotic breath-holding episodes, apparent life-threatening events (ALTE), and sleep-related upper airway obstruction. Apnea of prematurity consists of cyanotic episodes secondary to hypoxic events in premature infants. Two thirds of the time this is caused by apneic pauses and cessation of nasal airflow, which can be treated with supplemental oxygen. Cyanotic breath-holding episodes are quite familiar to those otolaryngologists who deal with infants and sometimes occur during a clinical examination. A certain proportion of these infants, however, continue to have these events while asleep, and they are thought to be secondary to intrapulmonary arterial shunting. Apparent life-threatening events are defined by an episode that is frightening to the observer and consists of a combination of color change, choking, gagging, or decreased muscle tone. Sleep-related upper airway obstruction consists of infants who appear well when awake but when asleep develop stridor and/or apnea, often leading to failure to thrive. This study looked at those infants who have stertor or stridor during the day and then determined whether the use of the PSG was helpful in assessing the significance of noisy breathing and the need for treatment.

There are 2 methods for evaluating sleep disorders in infants: the pneumogram and the PSG. The 4-channel pneumogram measures respiration and heart rate and can include continuous oximetry. It can grossly distinguish between central and obstructive apnea, and when performed with continuous oximetry, the 4-channel study becomes a 5-channel study. A pH probe can be added to assess whether gastroesophageal reflux is an issue, and in most cases the pneumogram study can be done at home (pH probe placement in an infant requires a visit to the radiology department at our institution for proper placement under fluoroscopy). The cost for a pneumogram averages around $500 to $700 with the interpretation. Currently, this so-called 6-channel study is performed at our institution as an outpatient service on infants by a pediatric home respiratory service. It is favored by the insurance carriers because of its decreased cost.

Full PSG includes the above plus electromyography, electro-oculography, determination of transcutaneous carbon dioxide or end tidal carbon dioxide, body movement recordings by an observer, and, if indicated, a trial of CPAP or BiPAP. This examination usually requires an admission to a sleep center and costs around $1500 to $2000 with the interpretation. The use of full PSG has been well described in the evaluation of infants with apnea. Guillemineault et al demonstrated 20 years ago that respiratory and cardiac abnormalities were more common in a group of infants with ALTE compared with control infants using full PSG. These full PSGs are available but are more difficult to obtain than 4-channel studies because of the prior approval required by the payer. For us, this bureaucracy will consist of letters documenting the problem, which are then reviewed by nurses or physicians inexperienced in dealing with airway disorders in infants. This results in significant delays, which affect optimal patient care in many of these children.

The current belief by some is that 4- to 6-channel pneumograms are sufficient to study these infants, applying adult data to this age group. Others who have studied these infants feel that pneumographic studies are insufficient to evaluate the effects of airway obstruction alone or obstruction plus apnea. In our opinion, this also applies to those infants with stertor or stridor. Full PSG evaluations have been shown to be of benefit in assessing infants with obstructive airway syndromes, and this opinion parallels our experience in infants with stertor or stridor. The initial pneumogram in patient 2 re-
revealed apnea and bradycardia; he was initially treated with caffeine, with some improvement. Because of his continuing failure to thrive, a full PSG was obtained, which revealed both central and obstructive events. Dramatic improvement was obtained with the addition of nasal BiPAP. Patient 7 had normal pneumogram findings despite continued complaints of airway distress and failure to thrive. Full PSG revealed 35 breathing-related arousals per hour in addition to significant gastroesophageal reflux. During the full PSG it was determined that CPAP was not tolerated. This patient benefited instead from a supraglottoplasty. Patient 15 had 4-channel studies revealing mixed apnea and a trial of BiPAP was begun, with some improvements. The child continued to have problems at home until a full PSG was obtained, demonstrating significant carbon dioxide retention because of his inability to trigger the BiPAP. A rate was then dialed into the BiPAP but continued intolerance of the device led to a surgical solution. In 7 of 8 cases, the 4-channel pneumogram results were wrong or grossly inadequate in this patient population. Patient 13 was the only patient who had normal pneumographic and PSG findings.

Certainly we do not obtain nor do we recommend obtaining a full PSG for every child with noisy breathing. We consider the use of a full PSG when seizures are suspected or when there is failure to thrive. Failure to thrive in the presence of stertor or stridor, in our opinion, indicates the need for intervention. The PSG provides further documentation and will assess the value of CPAP or BiPAP. We also consider the use of a full PSG in those children who have significant airway symptoms but mild findings on physical examination. In this scenario, a marked anatomical abnormality is lacking to warrant surgical intervention. Abnormal findings of a full PSG and failure of CPAP or BiPAP to correct the physiological abnormality or intolerance of such a device would dictate a surgical approach. Children with craniofacial abnormalities who are not failing to thrive are an additional population in whom full PSG should be used as a way to diagnose subtle hypoventilation and chronic carbon dioxide retention. Performing 4- or 6-channel pneumocardiography, in our experience, is less than adequate and significant problems are often missed. Full PSG often provides the physiological data to complement endoscopic findings.

Infants with significant apnea or ALTE indicated in the medical history or on sleep study may likewise have significant findings on physical examination. These physical causes of apnea are occasionally overlooked by physicians. Ruggins et al describe 4 infants with ALTE who underwent laryngoscopy and were noted to have severe laryngomalacia during their spells of periodic breathing. Endoscopy and full PSG complement each other in the thorough evaluation of children with airway obstruction and therefore both should be considered as useful tools in the thorough evaluation of infants with airway disorders.

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REFERENCES