Obstructive Sleep-Induced Apnoea due to Bilateral Recurrent Laryngeal Nerve Palsies

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SUMMARY

A patient with obstructive sleep-induced apnoea due to bilateral incomplete recurrent laryngeal nerve palsies is described. The sleep apnoea syndrome is briefly reviewed and the potentially fatal complications are emphasized.


Sleep-induced apnoea has been increasingly recognized over the past decade. It has been described with ordinary obesity, in the Pickwickian syndrome, with idiopathic alveolar hypoventilation and with anatomical obstructions to the upper airways.

We recently investigated a patient who was experiencing excessive daytime somnolence and frequent apnoic episodes during sleep. Bilateral incomplete recurrent laryngeal nerve palsies, which were found to account for his symptoms, have to date not been documented as a cause of the sleep apnoea syndrome.

CASE HISTORY

The patient was a 53-year-old butcher. He had a 6-month history of excessive daytime sleepiness and lethargy which had been of insidious onset. He would fall asleep, for example, while sitting at work, reading, watching television, or even while driving his car, and as a result could no longer carry on his business. According to his family, his breathing during sleep had become extremely noisy, and this was accompanied by moaning, talking and abnormal movements. He admitted that he had not dreamed for some months. There was no history of head injury, epilepsy or other neurological disturbances, but his voice had become slightly hoarse. He smoked 20 cigarettes a day, and had not taken any sedatives.

The patient was a lethargic, thick-set man who weighed 83 kg and was 162 cm tall. He drifted off to sleep while being examined, but was easily roused. He was not cyanosed, and his blood pressure was 140/90 mmHg. Examination of the cardiovascular and central nervous systems and of the abdomen was normal. On auscultation of the chest, coarse rhonchi were heard. There was no stridor on normal breathing.

In the ward the patient frequently fell asleep, even while eating or reading. While asleep, by day or by night, there were episodes of marked inspiratory stridor, sufficiently loud to be heard throughout the large general ward, and lasting a few minutes, and these were followed by apnoeic episodes lasting about 1 minute. Incomplete awakening would then occur, and the sequence would be repeated. Marked abdominal and thoracic respiratory efforts persisted during both the periods of stridor and apnoea.

The patient's haemoglobin concentration was 14 g/100 ml, the white cell count was 6000/μl and the sedimentation rate was 21 mm/1st h. The urea concentration was 4.4 mmol/l (26 mg/100 ml) and the creatinine 104 mmol/l (1.2 mg/100 ml). The electrocardiogram was normal and the electro-encephalogram showed no evidence of epilepsy. Both skull radiographs and computed axial tomography of the brain were normal.

At indirect laryngoscopy, the patient was found to have a narrow laryngeal glottic airway, and the vocal cords were in the paramedian position. On attempted phonation, adduction of the cords was revealed, but there was no active abduction. The appearance was that of incomplete bilateral recurrent laryngeal nerve palsies. No cause for this could be found on chest radiographs, barium swallow or at bronchoscopy.

While awake, his \( P_{aO_2} \) was 10 kPa and his \( P_{aCO_2} \), 4.1 kPa. While asleep, however, he developed respiratory failure, and the \( P_{aO_2} \) decreased to 5.2 kPa, while the \( P_{aCO_2} \) rose to 6.9 kPa (Table 1).

The flow volume loop (Fig. 1) showed the pattern typical of a variable extrathoracic obstruction with a plateau-like inspiratory part of the curve, and an inspiratory flow rate not exceeding 84 l/min. The ratio of expiratory to inspiratory flow rates at the mid-vital capacity was 3.5 (normal about 0.9). Other pulmonary function tests (Table 1) showed only some restriction of lung volumes.

Further investigations, including polygraph recordings, were refused by the patient, who also refused tracheostomy. After he had been discharged, the symptoms continued unabated. He died suddenly in bed at home 4 months after leaving hospital. Death was immediately preceded by an episode of loud stridor. An autopsy was not performed.

DISCUSSION

The hallmark of the sleep apnoea syndrome is the occurrence of frequent apnoeic periods during sleep. There are 3 types of sleep-induced apnoea: (i) central apnoea, in which apnoea is associated with cessation of both thoracic and abdominal respiratory efforts; (ii) obstructive apnoea, where airflow is prevented by obstruction to the upper airways, and respiratory efforts persist during the apnoeic period; and (iii) mixed apnoea, in which a central phase followed by an obstructive phase occurs.
TABLE I. ARTERIAL GASES AND PULMONARY FUNCTION TESTS

<table>
<thead>
<tr>
<th>Arterial gases (kPa)</th>
<th>Lung volumes</th>
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<tbody>
<tr>
<td>( P_{O_2} ) (Awake)</td>
<td>( P_{O_2} ) (Asleep)</td>
</tr>
<tr>
<td>10.0</td>
<td>4.1</td>
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TLC — total lung capacity; RV — residual volume; VC — vital capacity; FEV\(_{1} \) — forced expiratory volume in 1 s.

Each apnoeic episode is followed by incomplete awakening, with resumption of respiratory efforts in the central apnoeas, and relief of airways obstruction in the obstructive variety. Apnoea soon recurs, however, and the sequence is repeated between 200 and 500 times during the night, so that marked sleep deprivation occurs, inevitably leading to daytime somnolence. The incomplete awakening is triggered off by an arousal response in the brainstem reticular activating system, which can usually be seen in the electroencephalogram.\(^6\) Hypercapnia, which occurs during the apnoeic period, probably play a role in this response,\(^6\) although in the obstructive apnoeas it is conceivable that additional factors such as increased impulses from the respiratory muscles may be operative.

The most frequent type of sleep-induced apnoea found in obese patients both with and without the fully developed Pickwickian syndrome is the obstructive variety.\(^7\) Marked pharyngeal hypotonia, causing collapse of the pharyngeal muscles and retraction of the tongue, has been demonstrated during obstructive episodes,\(^7,10\) and is the mechanism primarily responsible for the interruption of airflow. The short, thick neck and jowls of such patients have been thought to contribute to the tendency towards upper airways occlusion.\(^11\)

The partial anatomical obstructions usually associated with sleep-induced apnoea are hypertrophied tonsils and adenoids (particularly in children),\(^1,6,9\) mandibular malformations\(^6\) and deviations of the nasal septum.\(^8\) In all these situations, hypopharyngeal collapse during sleep appears to play an integral role in aggravating the obstruction.\(^5,10\) It has been thought that the intrinsic laryngeal muscles are not significantly involved in the obstructive process.\(^7\) In our patient, however, each apnoeic episode was preceded by severe inspiratory stridor. We feel that the only logical explanation for this sequence of events is that additional hypotonia of the intrinsic laryngeal muscles during sleep aggravated the vocal cord palsies, thereby inducing critical obstruction.

Our patient had a number of clinical features typical of obstructive sleep apnoea. These included an underlying anatomical obstruction to the upper airways, marked respiratory efforts during apnoeic periods, and daytime somnolence and fatigue (the presenting complaints), as well as the alveolar hypoventilation and abnormal motor activity that occurred only during sleep. The absence of dreaming was probably related to the paucity of rapid eye movement sleep which occurs in the syndrome. Disturbing personality changes and irrational behaviour are further common complications, but they had not become manifest in our patient. There was no obvious cause for his recurrent laryngeal nerve palsies. In 4 - 6% of bilateral palsies no cause may be found,\(^13,14\) but it is thought that some of these 'idiopathic' varieties may be related to a viral illness.\(^15\)

Obstructive apnoea may lead to important cardiovascular complications. The hypoxaemia of the apnoeic episode results in elevated pulmonary arterial pressure,\(^5\) which may be the forerunner of cor pulmonale. Life-threatening arrhythmias have also been documented during sleep, including sinus bradycardia (in severe cases to below 30 beats/min), asystole, atrioventricular dissociation and ventricular tachycardia.\(^16\) Cor pulmonale and the arrhythmias, as well as other complications, are reversible when the obstruction is relieved, and this entails either resection of anatomical abnormalities, or a tracheostomy.

The circumstances of our patient's death suggest strongly that he died of one of the above arrhythmias, which emphasizes the grave hazards of the syndrome and the urgency for relief of the obstruction.
We should like to thank Dr S. L. Sellars, of the Department of Otolaryngology, Groote Schuur Hospital, for his advice, and Dr D. Verster-Cohen, under whose care the patient was admitted.

REFERENCES


Duchenne's Dystrophy Associated with Unusual Musculoskeletal Abnormalities

A Case Report

P. JOSHI, P. L. A. BILL

SUMMARY

A case of Duchenne's pseudohypertrophic muscular dystrophy allied with previously unreported musculoskeletal abnormalities is reported. Some of the more characteristic features of Duchenne's dystrophy are reviewed from the literature.


The Duchenne form of muscular dystrophy is a progressive degenerative disorder of muscle which usually affects boys aged 2 - 6 years, and which is inherited as an X-linked disorder. The clinical picture has been well described and is associated with a number of musculoskeletal deformities. In this report musculoskeletal abnormalities, which to our knowledge have not previously been reported in association with Duchenne's muscular dystrophy, are highlighted.

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CASE REPORT

A 13-year-old Black boy presented with a non-productive cough of 4 days' duration, severe dyspnoea which had lasted for 2 days, and vomiting which had persisted for 1 day. The patient had been confined to a wheelchair for several years, and was unable to wash or feed himself owing to weakness of the limbs. He had suffered from recurrent respiratory infections, but there was no history of tuberculosis, and no family history of muscular disease. He has a brother who is normal.

When examined, the patient was found to be obese and pyrexial (temperature 39.7°C). There was bilateral gynaecomastia, which was related to his puberty. The pulse rate was 120/min and the blood pressure was 100/60 mmHg. The heart was of normal size, with a loud first heart sound. There was evidence of respiratory distress, as manifested by tachypnoea of 56/min, and shallow breathing, flaring ala nasae and intercostal and substernal recession. There was clinical evidence of a respiratory infection.

Neurological testing revealed a fully conscious and cooperative young boy of average intelligence, whose abnormalities were confined to the motor system. There was wasting of the hand muscles, but no obvious wasting of the...