Sequential constriction of upper airway and vocal cords in sleep apnoea of multiple system atrophy: low field magnetic resonance fluoroscopic study

M Hirayama, H Fukatsu, H Watanabe, Y Koike, A Noda, H Ito, R Kobayashi, G Sobue

Low field magnetic resonance fluoroscopy was used to clarify temporal and spatial features of airway obstruction in sleep apnoea syndrome (SAS) in multiple system atrophy (MSA), as well as in obstructive SAS (OSAS). 20 patients with OSAS with severe obesity (mean (SD) age 66 (10) years; 16 men, 4 women) and 6 patients with SAS related to probable MSA (60 (9) years; 4 men, 2 women) were studied. In the OSAS group, body mass index, apnoea index, and desaturation index were significantly higher than in the MSA group. In OSAS, simultaneous obstruction extended from the retropalatal pharynx to the retroglossal during sleep on low field magnetic resonance fluoroscopy. In MSA, obstruction of upper airway followed a similar distribution, but obstruction of vocal cords followed upper airway obstruction. In contrast to OSAS, sequentially acting neural mechanisms are suspected in SAS with MSA.

Obstructive sleep apnoea syndrome (OSAS), commonly associated with obesity, causes obstruction of the upper airway. OSAS is considered to be a risk factor for cardiovascular disease and for traffic accidents. In patients with multiple system atrophy (MSA), a sleep apnoea syndrome (SAS) occurs frequently, occasionally causing sudden death. However, duration of apnoea generally is shorter and O2 desaturation is milder in MSA than in OSAS. The pathophysiology of SAS in MSA is incompletely understood. High speed computed tomography, magnetic resonance (MR) imaging, and laryngoscopy have been used to assess the upper airway in SAS. However, high speed computed tomography does not permit a sufficiently long period of monitoring of the upper airway during sleep. Laryngoscopy also cannot be used without anaesthesia and is difficult to use during natural sleep because of the discomfort it causes.

To clarify the pathophysiology of SAS in natural sleep, we have developed a new method to monitor the upper airway using low field MR fluoroscopy, which provided useful evaluation of SAS. In the present study we compared the temporal and spatial profile of airway obstruction in sleep apnoea in MSA with that in OSAS.

**PATIENTS AND METHODS**

Twenty patients with OSAS with severe obesity (mean (SD) age 66 (10) years; 16 men, 4 women) and six patients with SAS related to probable MSA (60 (9) years; 4 men, 2 women) were studied (table 1). In these patients OSAS or MSA was diagnosed, and they were treated at Nagoya University Hospital. Patients in both groups all were evaluated by polysomnography and fulfilled diagnostic criteria for SAS. Subjects gave informed consent in advance.

Most patients underwent MR fluoroscopic assessment during the day (1:30 to 3:30 pm) or around midnight (11 pm to 2 am) in natural sleep in the supine position. Images were recorded for at least 60 minutes, and evidence of sleep related obstructive phenomena was sought in all patients during MR imaging. Since it is difficult to record an electroencephalogram during MR imaging, we identified the sleep state by inspection and presence of snoring or stridor.

An open gantry MR imager (Toshiba, Tokyo, Japan) with a 0.35 T magnet was used. A solenoid belt coil was placed...
around the patient’s neck. MR fluoroscopy was performed using a fast spin echo-type sequence with repetition time of 28 ms, echo time of 5 ms, and flip angle of 15º. Slice thickness was 10 mm, and a 23 cm field of view was imaged continuously with a 128 × 128 matrix. The midsagittal plane and the transaxial plane at the soft palate level usually were selected for monitoring. Real time images were monitored at 0.9 second intervals. The difference in the parameter of sleep apnoea between OSAS an MSA groups was statistically analysed by Student’s t test. To identify the narrowing of the vocal cord, we assessed the maximum diameter of the vocal cord image on MR imaging that adjusted on the plane of the vocal cord, and we determined the presence of narrowing of the vocal cord when the diameter was less than half that in the awake state.

RESULTS

In the OSAS group, body mass index (BMI), apnoea index, and desaturation index were significantly higher than in the MSA group (OSAS v MSA: BMI 32 (5.5) v 23 (5.6), p < 0.01;
Obstructive sleep apnoea syndrome (OSAS) and vocal cord obstruction are common conditions. In OSAS, upper airway obstruction is typically found at the level of the palate, progressing to the glossopharynx, and may involve the vocal cords. In patients with MSA, upper airway obstruction and vocal cord obstruction have been observed sequentially during natural sleep. This is the first report of simultaneous assessment of both the upper airway and the vocal cord during natural sleep in patients with MSA. Low field MR fluoroscopy is a new method that can sequentially analyse both the upper airway and the vocal cord during natural sleep. This is particularly useful in patients with MSA, where the vocal cord obstruction is often not observed.

Figure 3: Low field MR imaging in the upper airway and vocal cords in a patient with obstructive sleep apnoea syndrome. The upper airway was occluded, but the glottis was not (arrowheads).

In OSAS, upper airway obstruction was found at the level of palatopharynx, with extension to the glossopharynx but without vocal cord obstruction. In MSA, however, both the upper airway and the vocal cords were obstructed sequentially. Upper airway obstruction occurred during wakefulness, but the vocal cord obstruction was slightly narrowed. In SAS episodes, snoring or stridor was notable following upper airway narrowing, when the vocal cord was additionally narrowed. Upper airway obstruction was found at the palatopharyngeal level initially and then extended to the glossopharyngeal level, progressing to complete obstruction; then the glottis was occluded completely. This sequential obstructive pattern in the upper airway extending to the vocal cords was observed in all SAS episodes in six patients with MSA, even in those with atrophy of the tongue (table 1, fig 1, fig 2). This suggested that obesity with increased BMI did not contribute to this phenomenon. Obstruction of the vocal cord alone without upper airway obstruction was not observed in any patient.

In the OSAS group, no upper airway obstruction occurred during wakefulness, but repeated episodes of upper airway obstruction were observed in natural sleep in all 20 patients. In these sleep apnoea episodes, complete obstruction occurred in a descending sequence in the upper airway, as in MSA; however, vocal cord obstruction was not observed in any patient. This obstruction pattern was similar in all 20 patients with OSAS (table 1, fig 3).

DISCUSSION

Visualising occlusive events in the upper airway is important for understanding the mechanisms of upper airway and vocal cord obstruction in patients with MSA. Vocal cord abductor paralysis (Gerhardt syndrome) has been suspected from pathological and electrophysiological viewpoints. Selective neurogenic atrophy of the posterior cricoarytenoid muscle has been documented by postmortem observations in MSA. Electromyographic evidence of a neurological abnormality also was found in the posterior cricoarytenoid muscle. Since this muscle is the sole abductor of the vocal cords, this obstruction result from paralysis of this muscle. Even during wakefulness, partial vocal cord paralysis is observed in patients with MSA, as in the present study. However, most recent electrophysiological studies have shown that the cause of stridor is hyperactivity (dystonia) rather than paralysis of the vocal cord abductors, suggesting supranuclear neurological dysfunction as the cause of glottic obstruction. These results may differ because of the stage of disease in each study. Vocal cord stenosis was seen without vocal cord abductor paralysis.

Another important question concerns why upper airway obstruction and vocal cord obstruction occur sequentially in patients with MSA. In obese patients with MSA, upper airway obstruction may partially account for sleep apnoea by the same mechanism as in OSAS patients. However, obstruction of the upper airway and at the vocal cords occurred in a patient with atrophy of the tongue and laryngopharyngeal muscles, suggesting another mechanism of upper airway obstruction distinct from OSAS. Vocal cord obstruction always occurred following upper airway obstruction in all patients with MSA. Although details of innervation of upper airway muscles from the brain stem are not entirely known in humans, motor centres in the nucleus ambiguous innervate both laryngopharyngeal and vocal cord muscles in monkey. We suspect that neural events resembling those causing vocal cord obstruction may occur in upper airway muscles. The larynx can easily be pulled inwards and downwards, narrowed by negative intrathoracic pressure causing inspiratory collapse in MSA. Once upper airway narrowing occurs, laryngeal and vocal cord stenosis can occur easily as an exacerbation. A highly synchronised neural mechanism appears to be important for sleep apnoea in MSA, which may involve excessive muscle contraction as in dystonia-like phenomena. Certain laryngeal stimuli
MR fluoroscopy study of sleep apnoea

such as contact with water or negative pressure readily induce glottic stenosis. Such stimulation induced immediate laryngeal closure and inspiratory flow limitation developed five minutes after the stimulation. Furthermore, persistent dystonia-like electromyographic activity of the cricopharyngeal muscle was seen in MSA during the hypopharyngeal dystonia-like electromyographic activity of the cricopharyngeal closure and inspiratory flow limitation developed five cardiac arrhythmia. Consequently, more prolonged apnoea induces

These results may suggest the following hypothesis that sleep apnoea in MSA occasionally causes sudden death. During sleep, hypotonia at the base of the throat causes obstruction of the upper airway, with extremely loud snoring and laboured breathing leading to apnoea in patients with OSAS. Some stimulations (hypoxaemia and hypercapnia causing apnoea) result in an arousal response, eliminating the hypotonia maintaining the upper airway. As for sleep apnoea in MSA, if the upper airway is obstructed, it stimulates more excessive upper airway and vocal cord closure. Hypoxia and hypercapnia cause an arousal response, which may cause a more excessive muscle contraction and lead to a dystonia-like reaction. Consequently, more prolonged apnoea induces cardiac arrhythmia.

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