

SHORT REPORT

Vocal fold motion impairment in patients with multiple system atrophy: evaluation of its relationship with swallowing function

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Background: Vocal fold motion impairment (VFMI), especially vocal fold abductor paralysis, is frequently seen in multiple system atrophy (MSA). Since the regulation system of laryngeal function is closely related to swallowing function, swallowing function is considered to be more involved in MSA patients with VFMI than in patients that do not have VFMI. However, the relationship between dysphagia and VFMI in MSA patients has not been systematically explored.

Objective: To elucidate the relationship between VFMI and dysphagia in MSA.

Methods: We evaluated swallowing function of 36 MSA patients with and without VFMI, by videofluoroscopy, and investigated the relationship between VFMI and pharyngeal swallowing function.

Results: VFMI was found in 17 patients (47.2%). Patients with VFMI had advanced severity of the disease. Although there was a tendency for bolus stasis at the pyriform sinus and the upper oesophageal sphincter opening to be more involved in patients with VFMI, statistical analysis did not show significant differences in swallowing function of MSA patients between with and without VFMI. In contrast, patients who underwent a tracheotomy ultimately required tube feeding or a laryngectomy.

Conclusions: Appearance of VFMI is a sign of disease progression but does not necessarily mean patients should change their way of taking nutrition. However, MSA patients who need a tracheotomy might have advanced to a high-risk group for dysphagia. Appropriate evaluation and treatment for VFMI and dysphagia are required to maintain patients' quality of life in MSA.

Vocal fold motion impairment (VFMI), especially vocal fold abductor paralysis, is frequently seen in multiple system atrophy (MSA).^{1–6} VFMI may lead to respiratory failure and cause sudden death.¹ To prevent this fatal complication, a tracheotomy is often required for MSA patients.

MSA patients often start tube feeding after a tracheotomy. The burden of a tracheotomy may lead to a change of swallowing status; however, VFMI itself might be related to swallowing status in MSA. Vocal fold motion is controlled by the vagus nerve, which arises from the nucleus ambiguus (NA) in the brainstem. Since the NA is also included in the swallowing regulation complex, involvement of the NA-vagus nerve system may affect both vocal fold and pharyngeal swallowing function. However, to our knowledge, the relationship between dysphagia and VFMI in MSA patients has not been systematically explored.

The aim of this study was to assess pharyngeal swallowing function in MSA patients with and without VFMI, and to investigate the relationship between dysphagia and VFMI in MSA.

PATIENTS AND METHODS

Patient selection

Thirty-six patients, diagnosed as having MSA at the Department of Neurology in our institute, from 1996–2001, were included in this study. Thirty-three patients were probable MSA, two were possible MSA, and the other one was definite MSA, according to Gilman's criteria.⁷ They consisted of 21 males, of ages from 53–78 years (average 62.1 years), and 15 females, of ages from 46–82 years (average 62.5 years). Twenty-eight of the 36 patients had MSA-C, and the other 8 had MSA-P. Informed consent was obtained from each patient, and the local ethics committee approved this study.

Laryngeal endoscopy and videofluoroscopy for assessment of VFMI and swallowing function

VFMI was evaluated by laryngeal endoscopy for each patient. Swallowing function was investigated by videofluoroscopy (VF). The assessment parameters in pharyngeal swallowing on VF examination were as follows: constriction of the pharynx, tongue base movement, elevation of the larynx, bolus stasis at the pyriform sinus (PS), the upper oesophageal sphincter (UOS) opening, and aspiration. A 3 point scale: 0 (normal), 1 (disturbed), and 2 (severely disturbed), was used to quantify each parameter, and for statistical analysis.

Severity stages of the disease

Severity stages of the disease were used as an assessment parameter. According to Watanabe *et al*,⁸ severity stages of the disease were determined as follows:

Stage 1: Able to walk without support.

Stage 2: Aid-required walking, with use of a walking aid or a companion's arm for support, but not at all times.

Stage 3: Aid-required walking, with use, at all times, of a walking aid or a companion's arm for support.

Stage 4: Wheelchair-bound state (wheelchair use at all times).

Stage 5: Bedridden state (complete loss of ability for independent activity).

Statistical analysis

Relationships between VFMI and assessment parameters in pharyngeal swallowing on VF examination were analysed using Spearman's rank correlation. The Mann-Whitney U test was used to determine differences for age, duration of disease, and severity stages of the disease. Statistical significance was defined as $p < 0.05$.

Abbreviations: MSA, multiple system atrophy; NA, nucleus ambiguus; PD, Parkinson's disease; PS, pyriform sinus; UOS, upper oesophageal sphincter; VF, videofluoroscopy; VFMI, vocal fold motion impairment

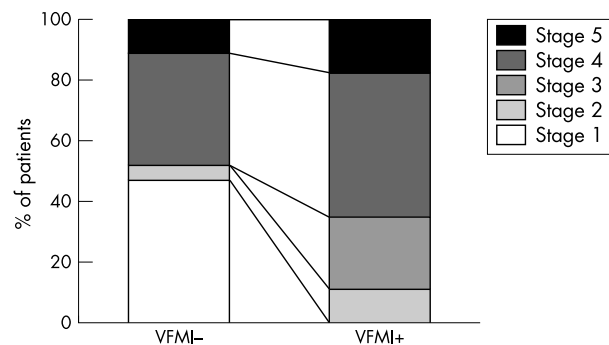


Figure 1 Graph indicates percentage of patients in each severity stage in Groups VFMI- and VFMI+. VFMI-, the group of patients that did not show vocal fold motion impairment; VFMI+, the group of patients that showed vocal fold motion impairment.

RESULTS

Laryngeal endoscopy

VFMI was found in 17 patients (47.2%). Fourteen of 17 patients had moderate or severe bilateral abductor paralysis of the vocal folds, and three of the 14 had received a tracheotomy. Two of 17 patients showed unilateral vocal fold fixation. The other patient showed bilateral vocal folds fixation at the paramedian position. Patients were divided into two groups according to having VFMI (Group VFMI+) or not (Group VFMI-). In Group VFMI-, 15 patients had MSA-C, and four had MSA-P; in Group VFMI+, 14 patients had MSA-C, and three had MSA-P. The duration of disease ranged from 1–16 years (average 5.0 years) in Group VFMI-, and from 2–10 years (average 5.4 years) in Group VFMI+.

There were no significant differences in duration of disease or age between Group VFMI- and VFMI+. Severity stages of the disease were different between Group VFMI- and VFMI+ (fig 1). The ratio of patients in advanced severity stages of the disease was significantly higher in Group VFMI+ compared with Group VFMI- (Mann-Whitney U test; $p=0.0479$).

Videofluoroscopic evaluation

Dysfunction of tongue base movement was the most common disturbance seen in 36 MSA patients, followed by disturbed elevation of the larynx (table 1). Aspiration was seen in 21.1% of Group VFMI- and 29.4% of Group VFMI+. Among various parameters, bolus stasis at the PS and UOS opening tended to be more disturbed in Group VFMI+ than in Group VFMI-; however, Spearman's rank correlation did not show significant differences in pharyngeal swallowing function between Groups VFMI- and VFMI+.

Nutrition status of tracheotomised patients

Three patients, who underwent a tracheotomy, could eat orally before the tracheotomy. The first patient could not eat orally, resulting in tube feeding right after the tracheotomy. The second patient could eat orally even after the tracheotomy in the hospital. After discharge, the patient still could eat orally to some degree; however, tube feeding to ingest sufficient nutrition was needed. The last patient maintained oral feeding for four months after the tracheotomy, but aspiration pneumonia occurred several times, and a laryngectomy ultimately had to be performed.

DISCUSSION

MSA is considered to be a parkinsonian disorder. VFMI is also seen in Parkinson's disease (PD); however, VFMI in PD is different in several aspects from that in MSA.^{1–9} Firstly, VFMI is infrequently seen in PD, compared with MSA. Secondly, VFMI in PD is usually seen during both sleep and the daytime, while VFMI in MSA is predominant during sleep and extends to the daytime as the disease progresses. Thirdly, no abnormalities of the intrinsic laryngeal muscles were found in PD, while selective neurogenic atrophy of the posterior cricoarytenoid muscle was characteristic in MSA.^{1–2}

Dysphagia, as well as VFMI, is a serious complication in both MSA and PD. As to swallowing function in PD patients with VFMI, a close relationship between VFMI and dysphagia has been reported. Isozaki *et al* reported that PD patients with VFMI had severe dysphagia requiring tube feeding.¹ A similar tendency was also found in other reports.^{9–11} However, swallowing function in PD patients with VFMI was not systematically evaluated in these reports by VF or videoendoscopy.

Swallowing function in MSA has not been explored systematically, either. We evaluated swallowing function in MSA patients with and without VFMI by VF examination, and found a tendency for UOS opening and bolus stasis at the PS to seem more involved in Group VFMI+ than in Group VFMI-. Bolus stasis at the PS is supposed to be closely related to dysfunction of the thyropharyngeal muscle (a constrictor of the pharynx), and disturbance of UOS opening is supposed to be related to dysfunction of the cricopharyngeal muscle (a sphincter of the upper oesophagus). Since both the thyropharyngeal and cricopharyngeal muscles, as well as the intrinsic laryngeal muscles, are innervated by the vagus nerve, a close relationship between dysfunction of swallowing and vocal fold motion is suspected; however, our results did not show significant relationships between pharyngeal swallowing dysfunction and VFMI in MSA. MSA patients with VFMI do not always suffer from worse dysphagia compared with those without VFMI.

Table 1 Ratio of normal, disturbed, and severely disturbed patients in swallowing function for each parameter on videofluoroscopic evaluation

Parameter	Group	Normal (%)	Disturbed (%)	Severely disturbed (%)	Statistical analysis
Constriction of the pharynx	VFMI-	73.7	15.8	10.5	$p=0.402$
	VFMI+	58.8	29.4	11.8	
Tongue base movement	VFMI-	36.8	36.8	26.3	$p=0.919$
	VFMI+	47.1	17.6	35.3	
Elevation of the pharynx	VFMI-	63.2	21.1	15.8	$p=0.509$
	VFMI+	52.9	29.4	17.6	
Bolus status at the PS	VFMI-	84.2	5.3	10.5	$p=0.136$
	VFMI+	58.8	29.4	11.8	
UOS opening	VFMI-	94.7	0.0	5.3	$p=0.133$
	VFMI+	76.5	11.8	11.8	
Aspiration	VFMI-	78.9	15.8	5.3	$p=0.586$
	VFMI+	70.6	23.5	5.9	

PS, pyriform sinus; UOS, upper oesophageal sphincter.

A tracheotomy, especially with use of a cuffed tracheotomy tube, negatively influences swallowing. Mechanisms of swallowing disturbance caused by a cuffed tracheotomy tube are considered as follows: (1) a cuffed tracheotomy tube disturbs elevation of the larynx; (2) a cuff that presses the oesophagus disturbs a bolus passing through the esophagus; (3) subglottic pressure is lost, and a bolus is susceptible to penetrate into the glottis. In this study, three patients ultimately required tube feeding or a laryngectomy after the tracheotomy. Since they received no other intervention besides a tracheotomy, the tracheotomy seems to directly affect swallowing function in these patients; however, another factor should be also taken into consideration.

Kurisaki commented that activity of daily life of MSA patients becomes rapidly worse after a tracheotomy.¹² He discussed that this phenomenon resulted from progress of the disease. In our study, severity stages of the disease were worse in Group VFMI+ compared with Group VFMI-. These facts suggest that appearance of VFMI is a sign of disease progression, and the disease has almost already progressed to the status of requiring tube feeding, by the time MSA patients need a tracheotomy. The timing of a tracheotomy might be a kind of MSA-specific borderline between oral and tube feeding.

Isozaki *et al* found that almost half of patients could take all nutrition orally even after VFMI was identified, in MSA,⁴ and our results also showed that there were no significant differences in swallowing function of MSA patients between with and without VFMI. As such, unnecessary "prophylactic" tube feeding for MSA patients with VFMI, out of fear of aspiration pneumonia, should be avoided; however, MSA patients who undergo a tracheotomy frequently require tube feeding. Appropriate evaluation and treatment for VFMI and dysphagia during follow ups, especially to judge indications for a tracheotomy and to introduce tube feeding, are important for patients' quality of life.

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