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Case Report: Effect of Intensive Voice Therapy in a Patient With Multiple System Atrophy

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ABSTRACT

Introduction: Multiple System Atrophy (MSA) is a degenerative neurogenic disorder characterized by multiple symptoms affecting the movements and autonomic nervous system. It reduces the function of various types of nerve cells in the brain and spinal cord. MSA causes some movement disorders such as dysarthria that is one of the most common symptoms in these patients. The first neurological sign that sometimes progresses is the complete inability to produce speech. This study aimed to evaluate the effect of an intensive voice therapy based on the principles of Lee Silverman Voice Therapy (LSVT) on improving acoustic features, intelligibility, and quality of life index of the patients with MSA.

Materials and Methods: The patient was a 57-year-old woman with MSA disorder. Acoustic assessments of voice, intelligibility, and voice handicap index were performed before and after the treatment program according to LSVT principles.

Results: Acoustic values such as intensity, frequency, and harmonic to noise ratio increased, and jitter and shimmer parameters decreased. Amount of perception of intelligibility increased. A significant decrease in the scores of the quality of life index related to voice handicap was observed.

Conclusion: After treatment by intensive voice therapy, the acoustic assessment revealed improvement in all voice parameters. The perceptual assessment showed improved intelligibility and increased the patient's quality of life. Intensive voice therapy based on LSVT principles effectively improves the acoustic features, which subsequently cause intelligibility to be more comprehensible. It generally improves verbal communication, which has a positive effect on the patient's quality of life.

Keywords: Multiple system atrophy, Dysarthria, Speech acoustics, Intelligibility, Quality of life, Voice training

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1. Introduction

MSA is a degenerative neurogenic disorder characterized by multiple symptoms affecting the movements and autonomic nervous system that controls involuntary activities, such as blood pressure or digestion. MSA reduces the function and kills various nerve cells in the brain and spinal cord [1]. This movement disorder is clinically characterized by atypical Parkinsonism, cerebellar ataxia, and pyramidal signs. It is always associated with the failure of the autonomic nervous system [2]. The early signs of MSA are often difficult to distinguish from the symptoms of Parkinson disease. Some include bradykinesia, tremor, the rigidity of muscles, lack of muscle coordination, speech disorder, trembling sound, fainting and dizziness during hypotension blood pressure, bladder control problems, and others [1, 3]. This disorder is divided into two types [4]: 1) MSA-P type, which is Parkinsonism type and dominates the left hemisphere of the brain that often has Parkinson's early features, including bradykinesia, tremor, and rigidity with problems with balance, coordination, and dysfunction of the autonomic nervous system, 2) MSA-C, the cerebellar type of MSA with the domination of the brain's right hemisphere. It has early symptoms similar to ataxia, such as difficulty with balance and coordination, difficulty swallowing, speech abnormalities with a trembling voice, and abnormal eye movements like twitching. Because speech production requires precise coordination of many muscles, speech problems are often one of the first significant problems in these patients [5].

Dysarthria is one of the most common of them, and it is often the first neurological sign that sometimes progresses to complete inability to produce speech. Hypokinetic dysarthria is the predominant type whose effect is continually decreasing loudness during speaking. Decreased loudness also causes a reduction of a person's ability to have intelligible speech and, consequently, reduces the ability to function properly in society [6]. Reduced range of movements, as well as the slowness of tongue production movements in hypokinetic dysarthria, also have a significant effect on the accurate articulation of consonants and vowels [7]. In general, these changes result in deviations in the acoustic characteristics of speech production [8]. The predominant effects of dysarthria include persistent and slow changes in voice quality, hoarseness or breathiness, excessive changes in pitch, decreased speech rate, abnormal stress patterns in words and syllables, and inaccurate word production [9]. Total symptoms listed reduce the patient's speech intelligibility which can affect the quality of life and mental health. It creates depression and feelings of isolation and impairs one's ability to keep a job [10].

There are proposed methods of voice therapy on a daily basis that provide a high intensity of treatment. Such intensive approaches focus on simple tasks with phonation attempts and often practice one parameter of voice [6]. In contrast, traditional voice therapy, e.g. Boone facilitating techniques [11], involves multiple speech systems (e.g. respiration, phonation, articulation, and resonation). Their treatment intensity is low (1 to 2 weekly sessions, few tasks repetitions) and does not provide self-perception of voice quality by an individual [12]. One of the high-intensity therapies is Lee Silverman Voice Therapy (LSVT), proposed by Ramig et al. It is a focused treatment program to improve vocal cords contact, the overall voice, and speech articulation. It is based on loudness and maximum effort during sustained vowel phonation and other speech tasks [13]. Patients treated with this method should always try to produce voice loudly and control it.

In a case study, a patient with ataxic dysarthria produced increased (beyond a standard deviation) amounts of acoustic values, as well as improved qualitative features, such as articulatory precision, intelligibility, and speech rate, after treating with LSVT [14]. In another study by Spielman et al., 12 participants with Parkinson disease received advanced LSVT treatment, which was held in two 1-hour sessions per week for 8 weeks. The results showed similar treatment by typical LSVT in positive changes in patients' acoustic features [15]. The result of another study showed a significant increase in terms of intelligibility 6 months after the intervention of LSVT in comparison to traditional therapy for a patient with dysarthria [16]. In a single-subject study, Dromey et al. performed intensive voice therapy to create phonatory and articulatory changes in a patient with Parkinson disease over one month. The patient could increase his voice intensity after treatment [17]. Soloman et al. presented a combination of LSVT and verbal and non-verbal breathing treatments to a patient with moderate hypokinetic dysarthria with mild spastic features and considered quality of life. The results showed an increase in loudness, improved verbal breathing and intelligibility, and reduced scores of quality of life index associated with voice disability [18]. The result of a single-subject study by Rezaei showed the positive effects of intensive voice therapy during 4 weeks on increasing acoustic features and intelligibility and subsequently scores reduction of quality of life index related to voice disability [19].

Instrumental, perceptual, and qualitative evaluation showed a marked improvement in glottis closure, sound pressure level, changes in the range of the fundamental frequency, intelligibility, and Voice Handicap Index (VHI) scores after the practicing LSVT in mentioned related studies [14-19].

Due to the numerous phonatory and articulatory problems in different diseases with dysarthria symptoms and the negative impact of limited communication on the quality of life of these patients, providing an intensive and short-term voice therapy plan will have a significant effect on communication adequacy and patient's life. Accordingly, in this study, an intensive voice therapy program based on the principles of LSVT was used to evaluate the effect of treatment on voice features improvement, including increasing voice intensity, frequency, harmonic to noise ratio, and reduction of perturbations as shimmer and jitter in a patient with MSA disorder with dysarthria symptoms. The effects of improving voice features on speech intelligibility and quality of life were the secondary study objectives.

2. Materials and Methods

Study participant

This study is a single-subject case report of treating a patient who was a 57-year-old woman with MSA. There was no family history of the disease, and its onset was sudden. The patient showed fatigue, sleep disturbance, body tilt, hypokinesia, tremors, slow and short steps, staring eyes, low speech rate, and limited drooling without dysphagia problem. The acoustic symptoms included hoarseness, tremor, low loudness, decreased pitch range, breathiness, and hypernasality. According to the aim of strategies focusing on vocal intensity, low loudness was the inclusion criterion of the study, which was the main compliance of participants.

Study assessment

Observing the physical condition of the vocal cords by endoscopic examination (specifically laryngoscopy by speech therapist) was done during the production of sustained vowel /i/ phonation. It showed weak vocal cords adduction and incomplete glottis closure. Voice samples during assessment sessions have been recorded in the setting of reticence with less than 30 dB ambient noise level [20] recording by microphone then mixing and calculating by Praat [21] in a double-walled sound-attenuating room in a medical center. Recording acoustic signals were done by Soundco UPC-1250 Headmic Microphone. The microphone diaphragm was placed at a distance of less than 10 cm to the right side of the patient's mouth [20]. Next, the patient was asked to sustain the vowel /a/ phonation. Then the voice track was saved and put up on Praat software to evaluate and analyze acoustic parameters, including the fundamental frequency and voice intensity with their maximum and minimum amount, harmonic to noise ratio, jitter, and shimmer. To remove background noise on the recorded file, it was filtered by the sound filtering section at the Praat setting [22]. Speech intelligibility was assessed perceptually by presenting the patient's voice to three unfamiliar listeners to judge simultaneously and in similar circumstances. To evaluate the patient's quality of life related to voice disability, we used a Persian adapted questionnaire of VHI. It measures the physical, emotional, and functional effects of a patient's voice problems in three sub-tests which include 30 questions and a total score of 120 [23].

Study treatment

Treatment was performed according to the principles of Lee Silverman Voice Treatment (LSVT). It is a program designed to increase vocal intensity perceived as loudness. It focuses on a simple set of tasks that are practiced intensively, 4 sessions per week during 4 weeks, resulting in maximization of phonatory and respiratory functions. LSVT emphasizes vocal effort ("think aloud, think shout") and maximizing sensory perception of loudness by therapists [13]. In this case, treatment sessions include such practices as breathing and laryngeal exercises, sustaining vowel /a/, keeping the mouth open during phonation, pitch change, expressing words, phrases, and sentences, text reading, and making conversation. Also, she was asked to focus only on the loudness while contextual noise was presented at each stage to increase practice complexity, known as Lombard effects [24]. The fifth Boone loudness level was selected as target intensity [11] at all speech practices. Each loudness practice was recorded and replayed to the patient to judge her performance. Then she modified it using auditory feedback during the sessions. The therapist provided constant positive feedback to the patient and encouraged her to self-monitor and internally calibrate her loudness. The treatment program consisted of one 1-hour individual session four times a week, and 16 sessions were held. The patient's home practice was performed once a day for 10 minutes on days she had clinical treatment sessions and twice a day for 15 minutes, on days she did not have clinical sessions and was free. Description of the protocol of the session is provided in Table 1.

Table 1. Practical LSVT protocol used for the participant

Weeks	Practical LSVT Protocol
The first	Breathing exercises: training abdominal breathing pattern, increasing breathing volume Laryngeal warm-up exercises Sustained production of vowel /a/ phonation at the fifth Boone loudness level Focusing on keeping the mouth open during sustaining vowel phonation loudly (open mouth technique) Using the Lombard effect during sustained vowel phonation Pitch changes during sustained vowel phonation Expressing words Expressing practical phrase Recording patient's voice during sustained vowel phonation which she perceived as "too loud" and then playing that for her
The second	Breathing exercises: increasing breathing volume Laryngeal warm-up exercises Sustained vowel /a/ phonation at the fifth Boone loudness level Focusing on keeping the mouth open during sustained vowel phonation and expressing words loudly (open mouth technique) Using the Lombard effect during sustained vowel phonation and expressing words Pitch changes during sustained vowel phonation Expressing sentences at the fifth Boone loudness level Expressing practical phrase Judging the loudness of recorded own voice by a patient during vowel phonation and expressing words
The third	Breathing exercises: controlling expiration air Laryngeal warm-up exercises Sustaining vowel /a/ phonation at the fifth Boone loudness level Focusing on keeping the mouth open during sustained vowel phonation, expressing words and practical phrases loudly (open mouth technique) Using the Lombard effect during sustaining vowel phonation and expressing words and practical phrases Pitch changes during sustaining vowel phonation Text reading at the fifth Boone loudness level Expressing words and practical phrases at the fifth Boone loudness level Judging the loudness of recorded own voice by a patient during vowel phonation, expressing words and practical phrases
The fourth	Breathing exercises: controlling expiration air Laryngeal warm-up exercises Sustaining vowel /a/ phonation at the fifth Boone loudness level Focusing on keeping the mouth open during sustained vowel phonation, expressing words and practical phrases loudly (open mouth technique) Using the Lombard effect during sustained vowel phonation, expressing words and practical phrases, and text reading Pitch changes during sustained vowel phonation Making conversation at the fifth Boone loudness level Judging the loudness of recorded own voice by a patient during vowel phonation, expressing words and practical phrases, and text reading Providing the therapist's opinion on the intelligibility of the heard sentences and comparing with the patient target sentences

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3. Results

Acoustic features

Loudness and pitch were measured at average, the most, and the least amount of them in patient phonation on sustained vowel /a/. Their perturbation in order as shimmer and jitter in addition to harmonic to noise ratio was measured. These features were evaluated before treatment, immediately after that, and 6 months later as a follow-up session. Although there are no acoustic features norms of old females without voice disability in Persian, the resulting participant measures compared to those reported in the study of Goy et al. [25]. Acoustic feature values are presented in Table 2.

Measurement by Praat revealed increased mean, maximum, and minimum of voice intensity almost 15 dB more than the pre-treatment phase. It raised about 75 dB as average to about 90 dB after 6 months. The frequency range has significantly increased, especially at its minimum amount, which increased to about 300 Hz after 6 months rather than 75 Hz before starting the treatment. Results showed a significant reduction in jitter and shim-

Table 2. Acoustic features before and after treatment compare to available norms

Feature	Pre-treatment	Post-treatment	Six Months Post-treatment	Mean±SD Old Females Norms
Mean of voice intensity (dB)	74.27	88.63	90.12	74.0±4.2
Max of voice intensity (dB)	76.5	88.34	92.23	Not available
Min of voice intensity (dB)	68.48	87.17	88.04	56.5±6.1
Mean of frequency (Hz)	160.68	310.89	322.21	211±42
Max of frequency (Hz)	192.36	305.71	330.42	520±120
Min of frequency (Hz)	75.70	298.79	301.02	Not available
Jitter (%)	0.85	0.08	0.07	0.47±0.34
Shimmer (%)	0.72	0.05	0.03	2.78±1.79
Harmonic to Noise (Hz)	16.05	28.18	30.71	25.3±3.8

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mer, and they reached both to below 0.08% at the follow-up session. The harmonic to noise ratio also showed a 24 Hz increase 6 months after the treatment.

Intelligibility

Speech intelligibility was assessed perceptually by presenting the patient's voice to three unfamiliar listeners to judge simultaneously and in similar circumstances immediately and 6 months after the treatment. The first and second listener perception was above 70% in the pre-treatment session, but the third one was below 50%. However, all of them reported above 80% and 90% immediately and 6 months after treatment, respectively. The results are represented in Table 3.

VHI scores

Quality of life index related to voice handicap performed for the patient immediately and 6 months after treatment. Its scores were compared to Persian adapted norms [23]. The results of that are presented in Table 4.

Emotional, physical, functional, and total parameters significantly reduced 13, 12, 9, and 34 scores, respectively, 6 months later than before treatment with LSVT.

4. Discussion

This study aimed to investigate the effects of intensive voice therapy based on the principles of LSVT in a patient with MSA disorder. Acoustic features, speech intelligibility, and quality of life index related to voice handicap were evaluated and assessed for the patient. Results showed increased parameters, such as frequency, voice intensity, harmonic to noise ratio, and reduction in jitter and shimmer amounts. Also, an endoscopic evaluation showed increased vocal cords adduction and glottis closure which helped to produce higher loudness. Due to these changes, voice quality improved. Consequently, enhanced intelligibility was obtained. Improvement in the mentioned parameters had a significant effect on the patient's functional relationship and the scores of quality of life index related to voice handicap. In general, after treatment, the patient was less negatively affected by his voice. To follow up on the treatment results, the patient

Table 3. Speech intelligibility percept by listeners before and after treatment

Listeners	%		
	Pre-treatment	Post-treatment	After 6 Months
1	83.49	90	90
2	78.54	89.34	90
3	48.44	86.2	90

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Table 4. Voice handicap index scores

Subtest	Pre-treatment	Post-treatment	Six Months Post-treatment	Mean±SD Persian Adapted Norms
Emotional score	18	10	5	10.63±9.67
Physical score	24	21	12	19.98±9.54
Functional score	18	15	9	15.24±9.81
Total score	60	46	26	46.81±27.18

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was evaluated 6 months after the last treatment session. The results showed relative stability at almost all parameters, such as frequency, intensity, harmonic to noise ratio, intelligibility, and voice handicap index scores. The study findings were similar to the results reported by other researchers, which proved an intensive voice therapy program based on principles of LSVT has a remarkable effect on improving acoustic values and consequently improves qualitative features such as intelligibility [14-17]. The findings of laryngoscopy showed increased vocal cords adduction which were similar to those studies that reported, after treatment, the patient learns to increase the adduction of the vocal cords by focusing on loudness. Thus improving the activity and cooperation of the laryngeal muscles occurs [8]. There is a direct relationship between the lengths of speech formants and loudness; in other words, a loud voice reorganizes the movement of articulators, which causes improved intelligibility [26]. The results of this study scores are similar to the result of those which represented the positive effects of LSVT on enhancing patient's quality of life and communicating performance [18, 19].

5. Conclusion

The study results showed that an intensive voice therapy program based on the principles of LSVT with a focus on voice intensity, increases the contact level of the vocal cords and improves voice quality. It improved acoustic features and thus improved speech intelligibility. Increased intelligibility also significantly improved the patient's communication quality and the quality of life index associated with voice handicap. Obviously, improved verbal communication had a positive functional effect on the patient's quality of life. The long-term effects of treatment on acoustic features, intelligibility, and quality of life were also observed after 6 months.

Study limitations

Because of the patient's physical problems, the sessions were often not completed over time, and she could not always stay rest time of practices. Because of depression arising from limited communication, she had low motivation and corporation to do exercises on the first days of treatment.

Study suggestions

It is recommended to evaluate the effects of LSVT protocol at the follow-up only to show the stability of treatment. Also, it is suggested that this program be implemented on a Group of patients with dysarthria and compare the results with a randomized control Group study of a Group with the same age and gender.

Ethical Considerations

Compliance with ethical guidelines

The research steps are based on the standards of the Ethics Committee of Isfahan University of Medical Sciences. The participant had the authority to leave the treatment and study at any time she wished. The logic of the treatment protocol was explained to the patient, and she was informed of the study objectives. The results of the quantitative evaluation were also provided to her before and after treatment.

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Authors contributions

Both authors equally contributed to preparing this article.

Conflict of interest

The authors declared no conflict of interest.

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