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CASE REPORT

Music.ALS Case Studies: Clinical Perspectives on a Home-Based Music Therapy Treatment to Improve Breathing, Speech, Swallowing and Cough of Persons with Amyotrophic Lateral Sclerosis

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ABSTRACT

Respiratory failure, malnutrition, aspiration pneumonia and dehydration contribute to mortality in amyotrophic lateral sclerosis (ALS), and loss of verbal communication impacts quality of life. There are few interventions that help with the management of these symptoms alongside pharmacological ones. Neurologic music therapy protocols, which are biomedical interventions, have been demonstrated to be effective for the treatment of human neurodegenerative disorders, but less so with ALS.

Two case studies from a larger, published ALS study were selected for this new report to provide an insight into the practical aspects of music therapy treatment. The home-based protocol was designed to sustain bulbar and respiratory functions of persons with early and mid-stage onset. It was delivered to all participants twice-weekly for six weeks as a part of a 16-week ABA mixed methods study. Feasibility data (recruitment, retention, adherence, tolerability, self-motivation, personal impressions) and 34 biomedical outcome parameters for bulbar and respiratory changes were collected. The two studies highlight the differences in therapy process between participants – one with a spinal onset, slow progression ALS and another with a bulbar onset, rapid progression.

In both cases, music therapy was tolerated well and perceived as pleasant, although moderately challenging. For both participants, developing the sense of agency played an essential role in the therapy process. Minor treatment protocol modifications were needed. Positive changes in the objective measures of respiration, cough, speech and swallowing were observed.

Suggested individual adaptations of the experimental music therapy protocol included modifications of a sitting posture, breathing technique, consonant changes in singing exercises, additional pauses and stretching, and changes to preferred song therapeutic performance. A pilot study utilising the modified protocol is called for, followed by a randomised controlled trial to assess the clinical effectiveness of the innovative MT treatment.

Introduction

According to An Assessment of Fundamental Rights of People Living with ALS / MND Worldwide conducted by International Alliance of ALS / MND Associations in 2023, people with ALS / MND have the right to the highest quality treatments available, yet only 46% agree they have access to treatments[1]. It is estimated that the number of ALS cases in the world will increase from 222,801 in 2015 to 376,674 in 2040, representing an increase of 69%[2]. The demand for supportive treatment and care will increase proportionally. Whilst ALS impacts its sufferers profoundly in many ways, it is essential that novel treatments address the aspects of the disease most crucial for the patients' quality of life, functional independence and survival.

Weakening of the respiratory function and adherence to noninvasive ventilation (NIV) are the leading causes of anxiety of persons with ALS[3,4], and respiratory failure has been cited as the leading cause of death[5,6]. 93% of persons with ALS experience speech impairments at some point during the disease progression[7]. Loss of natural communication is regarded by the patients as one of the worst aspects of the disease[8]. Dysphagia and malnutrition contribute to 25.9% of ALS mortality and increase the risk of death by 7.7 times[9-11]. Patients with dysphagia report social isolation, fear and decreased mental health[12]. Finally, aspiration pneumonia and dehydration have also been cited among the leading factors contributing to mortality in ALS[5,13].

Swallowing, vocalisation and breathing are tightly coordinated, and a close relationship exists between these processes, in terms of location and activation of the neurons[14]. The overall emotional and physical condition of a person has a great impact on these processes as well. However, support of these functions in persons with ALS has been traditionally disunited and apportioned between various medical and allied professionals, including: speech and language therapists, nutritionists, physical therapists, ear, nose and throat specialists. Though specialist expertise is essential for the deep insight into the nature of each function, this disunion may be disadvantageous from the standpoint of integrative patient care.

A home-based music therapy (MT) protocol to support bulbar and respiratory functions of persons with early and mid-stage ALS was developed for a feasibility clinical trial[15]. This is the first study to systematically look at supporting bulbar and respiratory functions in ALS.

MT is the clinical application of music and its elements to improve the psychological, emotional, cognitive, physical and social health and wellbeing of human individuals and communities[16-19]. Recently, MT protocols have been demonstrated to be effective for the treatment of human neurodegenerative disorders[20-25].

Professional music therapists have training in vocal technique as well as basic knowledge of anatomy, physiology, neurology and psychology. When supported by multidisciplinary collaboration, which is the gold standard for ALS care[26-30], a music therapist may be in the unique position to provide the much-needed crosscutting outlook for bulbar and respiratory support in ALS.

A biomedical approach to MT is relatively new and largely unfamiliar to healthcare professionals outside of neurologic music therapy specialisation. Experimental protocols and efficacy studies have been published, but case studies illustrating biomedical MT treatment are scarce[31,32], and none have been published on MT for functional rehabilitation in ALS care.

Heterogeneity of clinical presentations and responses to treatment among the participants, even within the narrow recruitment criteria, required treatment individualisation within the protocol. The following two case studies were selected for this report to provide an insight into the practical aspects of MT treatment and to highlight the differences in therapy process between two female patients – one with spinal onset slow progression ALS and another with bulbar onset rapid progression ALS.

Experimental music therapy treatment

The experimental treatment was delivered to participants in their homes twice weekly for six weeks by a professional music therapist trained in the neurological music therapy approach. Patients with an ALSFRS-R scale bulbar subscore of ≥ 9, but ≤ 11, and Forced Vital Capacity greater than 60%, regardless of the disease onset, could participate. Bulbar subscore is the sum of ALSFRS-R questions 1–3.

Music-based breathing, relaxation and singing exercises were tailored to ALS-specific rehabilitative goals. There was built-in potential for the exercises to be adjusted to suit the unique needs and progress of each participant. Participants could choose a soundtrack for music assisted relaxation and a song for therapeutic singing from a variety of options offered by the therapist. Study participants were encouraged, but not required, to independently practise an abbreviated version of the protocol on the days with no therapy sessions. Audio and written guidelines were provided.

Please refer to the protocol article[15] for the detailed description and notation of the MT exercises and outcome measures mentioned in these case studies. Participant 1 data set correlates to Case Study 1 of the present report, and Participant 5 to Case Study 2.

Table 1 outlines the summary of demographic and clinical presentation data from the two case studies chosen for this report. Narrative description of the case studies follows.

Table 1. Summary of demographic and clinical presentation data

	Case Study 1: Helena	**Case Study 2: Tasha**
Age	62	43
Sex	Female	Female
ALSFRS-R on recruitment	37	38
ALSFRS-R total post-treatment	39	30
ALSFRS-R bulbar subscore on recruitment	9	10
ALSFRS-R bulbar subscore post-treatment	8	9
ALS onset type	Spinal	Bulbar

Case Study 1: Helena

Profile on recruitment

Helena was a 62-year-old Eastern European white female with spinal onset, slow progression ALS. Her speech was slurred, unclear, laboured and difficult to understand. Her voice had features of strong hypernasality, severe creakiness, and breathiness. She tended to expel most of the air during the first several syllables uttered, then to gasp for air. Helena coughed often, complained about tightness in her chest and reported she coughed to relieve this tightness. She walked with support. Mobility of her arms and hands was not restricted. Helena lived alone in a small city apartment and was not employed. A social worker, a sister and adult children visited her to help with errands.

Priority treatment goals

Among many losses caused by ALS diagnosis, losing the timbre and flow of her speaking voice to disease had been especially difficult for Helena. During the first session she commented that her voice used to be "beautiful", "young" and "much higher". She used to work at a call centre and often received compliments on her voice. Helena played to the therapist a recording of her voice prior to ALS onset. The voice was in a higher range and had no strain or excessive nasality.

Increasing speech clarity and improving voice timber by eliminating the strain and breathiness became prioritised individual goals for Helena's therapy process. These resulted in the prioritised individual objectives listed in Table 1.

Choice of song for therapeutic singing

Helena's choice of a song for the exercise *XIV. Preferred Song Performance*[15] presented an issue. The goal of this exercise was to practise all the elements of breathing and vocal technique addressed during each session. Helena was eager to sing "Ding, ding, ding!" - a Russian romance difficult for performance because of its wide range, dense lyrics and fast tempo. The piece could easily become a demotivator if used in sessions. However, Helena insisted because the romance had a special meaning for her. It was agreed then to have this piece as a "work in progress", whilst adding another, simpler song ("Oy tsvetot kalina") to the therapeutic singing routine as a "warm up". The second song provided appropriate phrasing, tempo and range to practise the vocal and breathing skills.

Treatment protocol modifications

Following modifications of the treatment protocol were needed In Helena's case.

The initial consonant in the *XI. Sustained Vowels Production Exercises* and in the *XII. Laryngeal Elevation through Vocalisation (Gliding Vowels) Exercise*[15] was changed from /m/ to /h/. This switching to voiceless consonant onset helped to relax vocal cords.

Plosive /g/ was substituted for nasal /m/ in the *VII.2. Interrupted "hah" sigh exercise* to decrease nasality during singing.

Frequent silent pauses were prompted for rest and relaxation between exercises to alleviate laryngeal muscle tension.

Initially, Helena was not able to articulate syllables /ga/ and /da/ in the following exercises: *VII.3. Voiced consonant sigh exercise*, *VIII. Consonant Range Cantillation Exercise* and *IX. Velopharyngeal Port Exercise*[15]. Holding the jaw in an open position with fingers of both hands during consonant production exercises and performing the jaw and the chin muscles self-massage often throughout the session were used as a modification, which helped to improve articulation and clarity.

When the patient was not able to articulate the lyrics in the exercise *XIV, Preferred Song Performance*[15], another modification was made: the therapist sang the verses, and Helena joined in for simpler choruses - sung on /la-la-la/ during earlier sessions, with the lyrics added later.

Developing agency

In the post-treatment interview Helena shared that she was sufficiently challenged by MT and was able to assess her own progress: "There were some problems each time, for example I could not pronounce (rolled) R sound. I am able to pronounce lower (register) vowels, there are problems with others. Some exercises worked out, some not so".

Her increasing self-awareness was notable throughout the six weeks of treatment: even before the therapist provided any feedback, Helena often commented on something that was a success ("That was good!") or on a particular detail that needed work, for example, an exhalation that had to be longer or a collapsed posture that needed changing. Helena was capable of practical observation of her bodily sensations. Once she commented: "I feel less tightness under my ribs after our sessions and less strain in my shoulders". She was instrumental in setting attainable session-to-session goals for herself and clearly formulating the ultimate purpose behind these: "I am not looking to sing that much, it's ok, but I need to save my voice".

Helena felt comfortable continuing the individual exercise routine on her own, post-therapy: "You have to practise, that's all. I have everything that I need for that, the set of exercises on a CD", "I will continue to exercise".

Generalisation of skills

By week three, it became clear that Helena's singing voice range was much wider than her speaking voice range. When she sang, the strain in her voice almost disappeared, especially in the higher register, and the consonants became distinct. These gains generalised to Helena's verbal communication outside of therapy, as perceived by herself, family and friends and medical staff. Helena also reported that she experienced less episodes of strained cough and that breathing became easier. Self-reported and objectively measured outcomes taken prior and post treatment showed positive changes in respiration, cough and speech (see Table 1).

Unique issues

During the six weeks of treatment Helena experienced several hypertension spells, when she felt weak, confused and sensitive to light. She was adamant about not letting these spells interfere with her therapy process and did not cancel any sessions. Relaxation and breathing exercises, rather than articulation and singing exercises were the focus of the sessions at times when Helena did not feel well. The light in the room was dimmed and the sessions were shortened.

Therapy closure

At the last session Helena reported that she felt MT had been useful for her speech and that she would continue exercising on her own. The emotions she demonstrated during the therapy closure were moderate and appropriate. Before parting, Helena and the therapist sang together the romance she mastered in therapy, then the therapist sang another romance for Helena, per her request. Helena recorded both songs to have as a keepsake. In a post-treatment interview Helena characterised MT as "a pleasant therapy".

Case Study 2: Tasha

Profile on recruitment

Tasha was a 43-year-old Eastern European white female with bulbar onset type ALS and an ALSFRS-R score of 38. Her speech was slurred and soft, with excessive nasality. It was very difficult to understand. She could not utter more than several syllables without running out of air: the air audibly escaped through the nose. She gasped for breath when speaking. Mobility of her limbs was limited; she could walk with support. Tasha lived in a city apartment with her husband and two young children. Her mother or her mother-in-law visited every day to help with errands. The research nurse reported that Tasha felt guilty about developing ALS. The ALS Centre staff were not successful in convincing Tasha to do anything for herself since the time of the diagnosis.

Priority treatment goals

Physical and emotional relaxation, learning abdominal breathing for speech support, increasing articulatory precision, decreasing nasal air emission and improving lip seal were identified as the priority goals for Tasha. These resulted in the prioritised individual objectives listed in Table 1.

Choice of music for therapeutic singing

For the exercise *XIV. Preferred Song Performance*[15] Tasha chose a popular Soviet war song "Katyusha". It was familiar to Tasha's children and had a special meaning for her: she learned this song from her own mother as a child and they used to sing it together. At a later session another song – "Mama Bear Lullaby" – was added to the repertoire. This was the song Tasha's children, who were regularly present at MT sessions, asked for. Singing was enjoyable and not fatiguing for Tasha, so this addition was accommodated.

Treatment protocol modifications

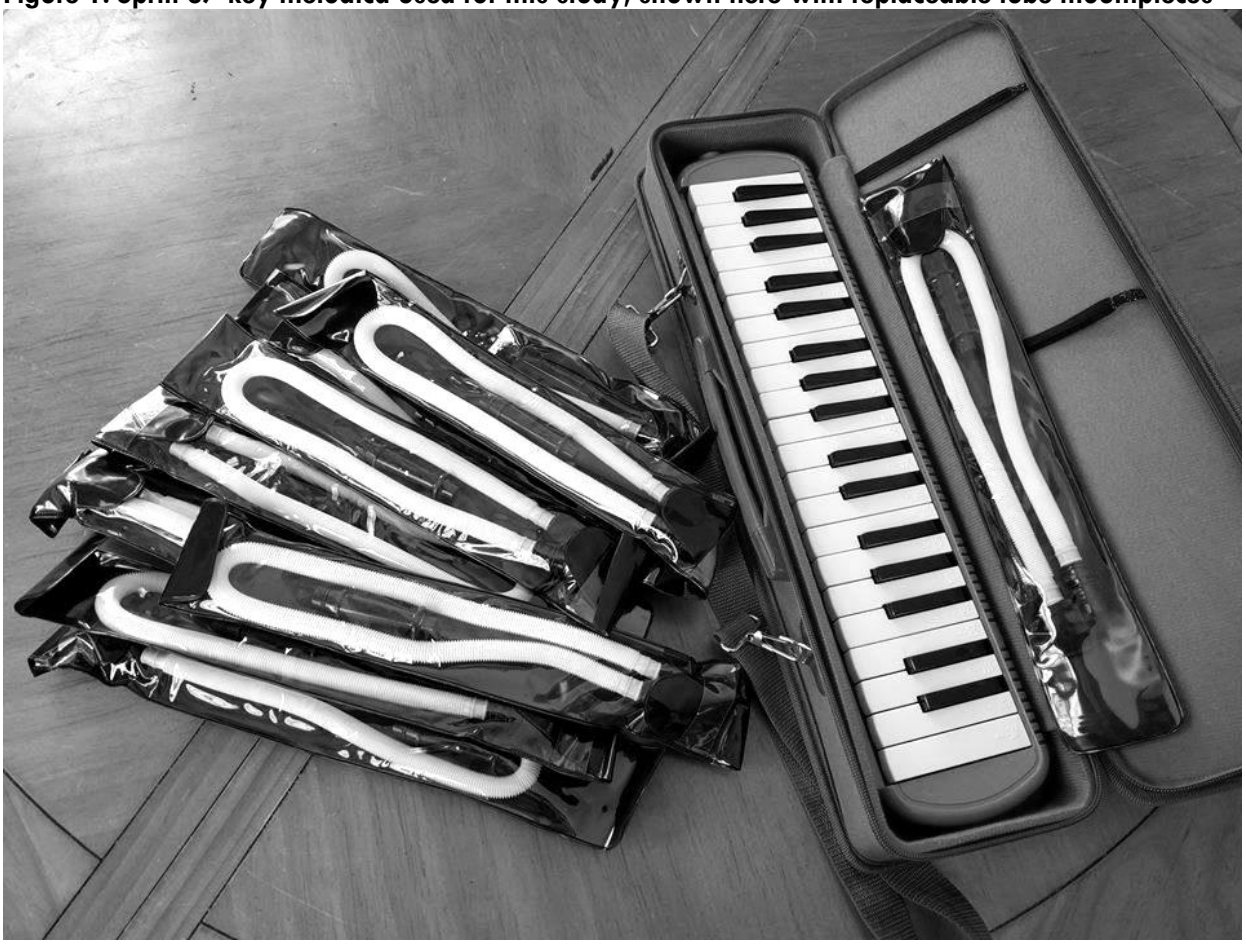
Tasha's condition progressed rapidly during the six weeks of treatment. During the first week, she could ambulate without support to open the door for the therapist. Later she used a cane, and then a walker. She mostly lost upper limb mobility in these six weeks as well.

Modifications of the protocol included an adaptive sitting posture during the sessions (see Table 1). When excessive salivation became a problem by week four, frequent pauses in sessions were allowed for swallow control. Plosive /g/ was substituted for nasal /m/ in the *VII.2. Interrupted "hah" sigh exercise,* to decrease nasality during singing.

Most breathing and singing exercises needed to be simplified for Tasha during initial sessions. As she gained confidence, Tasha was ready to complete the standard version. The *IV. Controlled Breathing and Lip Seal Exercise*[15] adaptations described below are indicative of these changes.

Playing melodica – a small hand-held reed instrument with a keyboard, akin to a pump organ (Figure 1) - provides the immediate feedback to the player and the motivation to increase their uninterrupted exhalation time. This exercise is helpful for improving lip seal – the proper closure of lips required to swallow solid food and liquids.

Figure 1. Sprill 37-key melodica used for this study, shown here with replaceable tube mouthpieces



Initially, when playing melodica, Tasha produced no more than two brief notes on one exhale, which was discouraging for her, as she thought there was not enough air in her lungs. The therapist suggested that Tasha pinched her nose closed as she played. With this modification, she was able to produce eight notes on one exhale during the first attempt, then 13 notes during the second attempt. This demonstrated to Tasha that there was enough air in her lungs for this exercise and for breathing. With this understanding, her main goal became to control the airflow. Tasha attempted the exercise again with her nose open and could play five notes in one exhale this time. This number increased in subsequent sessions.

Other protocol modifications were needed because Tasha's children were often present during music therapy and no childcare was available. The therapist adapted the exercises *V. Music-Assisted*

Relaxation for Voice Production and *XIV. Preferred Song Performance*[15] to allow the children to participate.

Developing agency

Tasha appeared anxious about completing the exercises "just right". She often commented that it was difficult for her to relax, that she felt a constant need to be "in charge" and perceived herself as "not enough". Her body looked rigid, chin was pointed up, and facial muscles and jaw were very tense. Despite this tension, Tasha often smiled and made eye contact with the therapist. Encouragement and positive feedback were important during the sessions, as was the therapist completing all the exercises together with her. Gradually, Tasha came to regard MT as her self-care time.

During the first three weeks of treatment Tasha often reported that she couldn't find time to complete the independent practice exercises, because, in her words, she "had to care for the children" and she was "doing everything so slowly now". By the end of treatment, the number of days she performed these exercises was four out of five.

It took several sessions and a lot of encouragement for Tasha to overcome her reservations and to start singing, rather than whispering along with the song of her choice. She appeared to enjoy singing. At the end of that turning point session Tasha said she felt more air in her lungs. By session seven Tasha's singing was resonant, with clearly articulated words and organic phrasing. However, Tasha completely dismissed her own efforts and progress, saying: "I only spoil it". At session 10 Tasha sang both songs of her choice in full voice. When finished, she smiled and did not make critical comments.

The examples above demonstrate how developing agency was important for Tasha across all the therapy activities. In the post-treatment interview she said: "At first it did not turn out too well, but then I learned how to breathe abdominally. You have to get used to that". She felt that the exercises for independent practice were easy to perform: "I practised, it takes half an hour. I felt well".

Generalisation of skills

Similarly to Helena's, Tasha's comfortable singing range developed to be relatively wide at almost one octave, whilst her speaking range was limited to a fourth and was lower. Her articulation became more precise during the exercises. Although Tasha or her family did not comment on any changes in her verbal communication outside of therapy, self-reported and objectively measured outcomes taken prior and post-treatment showed positive changes in respiration, cough, speech and swallowing (see Table 1).

Issues related to home-based treatment

Tasha and her family perceived MT as an opportunity for her two primary school age children to share music activities with their mother. Since no child care was available, the therapist adapted to this expectation: when present, the children participated in some activities, such as the music assisted relaxation. They laid on the floor or sat leaning on their mother's lap during the exercise, eyes closed. The therapist adjusted the narration to suit everyone present. Sometimes the children assisted their mother in facial massage, which was pleasing for all.

During the last week of the treatment phase, Tasha moved with the family to a summer house, a two-hour drive from the city. She felt she could not affect this change and appeared distressed because of the possibility of missing the two last sessions. Upon careful consideration, the therapist made the choice to travel and see Tasha for these sessions.

Other issues

When a therapeutic relationship was established, Tasha started asking questions regarding ALS aetiology, possibilities of treatment and physical abilities of other persons with ALS. She regarded the therapist as someone she could trust to safely discuss these matters. Eventually, Tasha felt safe to open up about the tension between her mother and her mother-in-law, as well as about her dissatisfaction in her marital relationship and the way it negatively affected her mental and physical health. It became apparent that Tasha experienced a great need for psychological support which the therapist was not in place to provide due to the limitations of the study protocol. After a discussion, Tasha and her family were referred to a psychologist at the local ALS centre.

Tasha missed session 11 because she had a fall with injuries to her face and needed time to recover.

Therapy closure

At the end of the last session Tasha spoke emotionally about her experience in therapy and the supportive role the therapist played for her during the treatment. Whilst speaking, she burst into tears and it was difficult for her to stop. The therapist used verbal psychotherapy techniques, including open-ended questions, active listening and mirroring, to comfort the patient. After the conversation, the therapist led an upbeat song improvisation, in which Tasha and her children

actively participated. The improvisation helped to appropriately distract Tasha from the raw emotions and to shift the mood, whilst it also provided an opportunity for the family to share an enjoyable activity. In the post-treatment interview, Tasha shared that she enjoyed MT: "It was good, comfortable, I liked it very much".

Discussion

Tolerance and sense of agency

In both cases presented above, MT was tolerated well and was perceived as pleasant, although moderately challenging. Similarly, in both cases, developing a sense of agency played an essential role in the therapy process.

Sense of agency refers to the feeling of controlling one's own actions and, through them, events in the external world[33]. Sense of agency has been often considered a constituent feature of the music therapeutic processes[34]. The MT protocol developed for this study is an instance of active music therapy: it requires active participation from the patient in order for the desired changes to take place. Mental and behavioural transition from being an object of care to being a subject of therapy, who is in charge of their own process and progress, was a necessary and discernible part of the treatment.

For both participants, developing agency started during the first treatment session when they chose their preferred music for relaxation and for the therapeutic song performance exercise. Patient preferred music has been found to be beneficial for enhancing engagement, mood and cognitive function in some patient populations, including dementia[35] and stroke[36]. It motivates the participant and provides the sense of agency - an opportunity to be in control, whilst they have no such power over disease progression and most of the medical treatment.

When working through the protocol initially, it was important that the therapist performed all the exercises together with participants, with the goal to provide guidance and to make the exercises feel safe and manageable for the participants through this shared activity. The ranges, tempos and lengths of the exercises were limited, thus requiring only a moderate effort from the participants, and then gradually expanded to increase challenge. The participants learned from the therapist about the mechanics of respiration, relaxation and sound production, and mastered self-monitoring skills. When the participant demonstrated ownership of these skills, prompts were gradually faded out and the agency was passed on to the participant.

Developing the ability to self-monitor and the sense of agency were important for participants' progress in therapy and them feeling comfortable completing the suggested exercises for independent practice.

Therapy goals and process

Albeit MT can offer a unique combination of psychological support and physical symptoms management, it is essential that the goals established on the assessment are prioritised. In Tasha's case, the psychological needs were immense and could become the primary focus of MT treatment. However, these needs could be as effectively addressed by patient's working with a psychotherapist, whilst the MT protocol allowed Tasha to maintain focus on her speech, breathing, cough and swallowing goals simultaneously - a combination not offered by any other discipline. Because of the heterogeneity and the fast progression of the ALS, it is advisable that the music therapist always collaborates closely with the multidisciplinary care team, including a speech language therapist, a social worker, a psychologist, a neurologist, a nutritionist and a physical therapist[30].

The two case studies underscore the importance of both empathetic and protocolised components of music therapy treatment. Establishing, developing and maintaining empathetic therapeutic relationships allowed to gain participants' trust, to make the therapeutic process feel safe, and to encourage and motivate them to work towards the therapy goals. At the same time, the therapist's expertise in the neurological music therapy approach and, particularly, in treatment of bulbar and respiratory symptoms of ALS, is essential for the ongoing in-session assessment and adjustments to ensure efficacy and safety. In the beginning of the treatment, individual objectives may be prioritised for each participant within the standard treatment protocol goals, as summarised in Table 2.

Table 2. Prioritised individual objectives within the standard treatment protocol goals

Case Study 1: Helena	Case Study 2: Tasha
1. Eliminate vocal cords strain. 2. Master abdominal breathing. 3. Decrease nasal air emission. 4. Increase vowel articulation precision. 5. Increase consonant articulation precision.	1. Increase physical and emotional relaxation. 2. Improve lip seal. 3. Master abdominal breathing. 4. Decrease nasal air emission. 5. Increase vowel articulation precision. 6. Increase consonant articulation precision.

The therapist needs to be sensitive to the smallest changes in the quality of the patient's voice and breath, body posture, affect, behaviour and language and to respond to these changes verbally, non-verbally and musically in order to meet the therapy goals. As expected, reasonable modifications were required within the standard treatment protocol in both cases, as outlined in Table 3. Please see the study protocol article[15] for detailed description of each exercise.

Table 3. Treatment protocol modifications

Case Study 1: Helena	Case Study 2: Tasha
1. Frequent silent pauses were prompted for rest and relaxation between exercises to alleviate laryngeal muscle tension. 2. Plosive /g/ was substituted for nasal /m/ in the *VII.2. Interrupted "hah" sigh exercise* to decrease nasality: "Hah-mah-mah-mah-mah" changed to "Hah-gah-gah-gah-gah". 3. Patient advised to hold the jaw in an open position with fingers of both hands in the *VII.3. Voiced consonant sigh exercise*, *VIII. Consonant Range Cantillation Exercise* and *IX. Velopharyngeal Port Exercise* and to perform jaw and chin self-massage often in session to improve pronunciation of consonants [d], [g]. 4. Change of onset from voiced consonant /m/ to voiceless consonant /h/ in the *XI. Sustained Vowels Production Exercises* and *XII. Laryngeal Elevation through Vocalisation (Gliding Vowels) Exercise* to decrease vocal cords strain. 5. Patient sings the melody of the *XIV. Preferred Song Performance*[15] on "la" syllable until she was ready to articulate the lyrics. 6. "Air on the G string" by J.S.Bach (Orchestral Suite No. 3 in D major, BWV 1068) used for the exercise V. Music-Assisted Relaxation for Voice Production; "Ding, ding, ding!" , "Oy tsvetot kalina" used for the exercise XIV. Preferred Song Performance	1. Sitting posture was adapted for the *II. Body Alignment Exercise* and throughout each session: patient used a small pillow for back support; her arms were crossed over the knees for stability. 2. Pauses were facilitated, as needed, during the session to allow the patient to deal with increased salivation (starting week four). 3. Most breathing and singing exercises were simplified during initial sessions to allow the patient to gain confidence. 4. Plosive /g/ was substituted for nasal /m/ in the *VII.2. Interrupted "hah" sigh exercise* to decrease nasality.: "Hah-mah-mah-mah-mah" changed to "Hah-gah-gah-gah-gah". 5. The exercises *V. Music-Assisted Relaxation for Voice Production* and *XIV. Preferred Song Performance*[13] were adapted to allow family (patient's young children) to participate. Narration was changed in the first instance to address several participants. 6. "Spiegel im Spiegel" by Arvo Pärt (version for cello and piano) used for the exercise V. Music-Assisted Relaxation for Voice Production; "Katyusha", "Mama Bear Lullaby" used for the exercise XIV. Preferred Song Performance

Home-based treatment

Home-based treatment benefits the patient in many ways, e.g. it eliminates travel time and allows people with low mobility to participate in therapy with no restrictions. However, home-based MT presents unique challenges for the therapist. It requires an increase in the therapist's travel times. A lone worker policy has to be in place in order to ensure safe working for the therapist and patient. The nature of home-based treatment, when the therapist enters patients' private space, affects the therapeutic relationship, because they are sharing that space, with its personal possessions and meanings, as well as the 'order of their home' which they may have mixed feelings about sharing, particularly at the start of therapy. Additionally, there are multiple other elements to consider, including the layout of the patient's home and furniture - whether this will present any obstacles to intervention delivery, the household members present (children, in Tasha's case) - might they disrupt the focus of the patient, and the occasional acute health needs of the patient (hypertension, in Helena's case), to name a few. Refer to Table 4 to see the summary of the issues related to individual treatment of the two study participants.

Table 4. Issues related to individual treatment

	Case Study 1: Helena	Case Study 2: Tasha
Issues related to home-based treatment	None	Issue: *Patient's young children present during therapy.* Solution: *Slight adaptations to protocol to include children in some activities.* Issue: *Patient moved to a summer house outside of the service area for two weeks.* Solution: *Therapist chose to travel to see the patient.*
Other issues	Issue: *Hypertension episodes.* Solution: *Therapy sessions shortened during the episodes.*	Issue: *Psychological tension between patient and other adult family members.* Solution: *Family referred to a psychotherapist from ALS centre.*
Therapy closure	Normal	Emotional

ALS onset and progression rate

There is no evidence to assume that the difference in therapy process and progress was due to the difference in ALS onset type (spinal vs bulbar) or progression rate. More data is needed to assess the possible link. The comparison of the data on adherence to treatment and treatment outcomes for the two case studies can be found in Table 5.

Table 5. Adherence to treatment and treatment outcomes

	Case Study 1: Helena	Case Study 2: Tasha
Adherence to treatment (sessions with therapist)	100%	92%
Adherence to treatment (non-mandatory independent exercise routine between therapy sessions)	68%	33%
Generalisation of skills: functioning improved, as evidenced from these self-reported and objectively measured outcomes taken prior and post treatment (please see the study protocol[15] for detailed outcome measures, data description and analysis)	Respiration: Forced Vital Capacity, Maximal Inspiratory Pressure, Maximal Expiratory Pressure	Respiration: Forced Vital Capacity, Maximal Inspiratory Pressure, Maximal Expiratory Pressure
	Cough: Peak Expiratory Flow	Cough: Peak Expiratory Flow
	Speech: The Center for Neurologic Study Bulbar Function Scale speech subscore, Maximum Phonation Time, Jitter, Shimmer, Harmonics-to-Noise Ratio, Maximum Repetition Rate—Alternating, Fundamental frequency, Speaking rate, Speech-pause ratio, Hypernasality	Speech: The Center for Neurologic Study Bulbar Function Scale speech subscore, Maximum Phonation Time, Jitter, Shimmer, Harmonics-to-Noise Ratio, Maximum Repetition Rate—Alternating, Maximum Repetition Rate—Sequential, Vowel Space Area, Speech-pause ratio
	Swallowing: No data	Swallowing: The Center for Neurologic Study Bulbar Function Scale swallowing subscore, Time-to-Laryngeal Vestibule Closure (pudding 10 mL), Maximum Pharyngeal Constriction Area (nectar 10 mL), Maximum Pharyngeal Constriction Area (pudding 10 mL), Penetration–Aspiration Scale (worst) (nectar 10 mL), Penetration–Aspiration Scale (worst) (pudding 10 mL), Total Pharyngeal Residue C24 area (nectar 10 mL), Total Pharyngeal Residue C24 area (nectar 10 mL)

Future research

This feasibility study provided evidence that the home-based MT treatment protocol to support bulbar and respiratory functions was tolerated well by persons with early and mid-stage ALS. Minor individual modifications may be needed to address the unique needs, personalities and capabilities of every patient. The detailed treatment protocol is available as an open access publication[15]. Suggested changes to the biomedical outcome measures and parameters to measure the efficacy of the protocol are in the same publication.

The importance of rigorous clinical trials to support complementary ALS treatments has been emphasised with the dual impact of ensuring ethical medical care, as well as protecting patients from the onslaught of pseudoscientific, often harmful practices[37-39]. This research serves an immediate purpose of addressing the need for continued scientific inquiry to understand the role of exercise in supporting bulbar and respiratory functions of persons with ALS[7] and provides a foundation for higher levels of evidence[40] by delivering the necessary feasibility data for a future pilot study, after which a randomised controlled trial to assess the clinical effectiveness of this MT treatment can take place.

Conclusions

This report provides an insight into the innovative treatment to support respiratory, speech, swallowing and cough functions of persons with early and mid-stage ALS. Whilst relatively new and often perceived as exotic by mainstream healthcare, MT is being used more and more often in the care of people with neurodegenerative diseases. Protocolised treatment is one way to ensure that MT treatment results are reproducible across the field. The present case studies, however, also bring attention to the human factor at the centre of every therapeutic process and focus on the need for individualisation within an established protocol.

Conflicts of Interest Statement

The authors declare no conflict of interest. The funders had no role in the design of the study; in the collection, analyses, or interpretation of data; in the writing of the manuscript, or in the decision to publish the results.

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