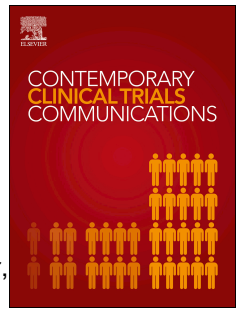

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Songwriting Intervention for Cognitively-Impaired Children with Life-Limiting Conditions: Study Protocol

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Abstract
Investigations evaluating the effectiveness of music-based interventions as a complementary approach to symptom management and health promotion for cognitively-impaired children with life-limiting conditions and their families are needed to minimize the physical and psychological burdens on individuals and their caregivers. Songwriting is one music-based strategy for a remote family-centered complementary approach to improve outcomes for cognitively-impaired children with life-limiting conditions, their parents, and families. The overall purpose of this study is to test the feasibility and preliminary efficacy of a songwriting intervention for cognitively-impaired children (ages 5-17 years) who are receiving palliative or complex care and their parents. Specific aims are to: (1) determine the feasibility of songwriting for cognitively-impaired children with life-limiting conditions and their parents, and (2) examine preliminary efficacy of songwriting for child psychological distress and physical symptoms, parent psychological distress, and family environment. The protocol for this 1-group pre-and post-intervention clinical trial (N = 25) is described.

Keywords: pediatrics, songwriting, music therapy, palliative care, palliative medicine, hospice and palliative care nursing

Background and Rationale

Nearly 500,000 children in the United States are living with life-limiting medical conditions.[1] These children and their parents represent a significant and compelling population at high risk for substantial physical discomfort, psychological distress and negative impact to the family environment.[2-13] Additional complexity occurs when life-limiting conditions are accompanied by cognitive impairment (intellectual disability), the most common developmental disorder defined by intellectual and adaptive functioning significantly below average for a child's chronological age.[14, 15] Cognitively impaired children with life-limiting medical conditions are particularly vulnerable to these risks given their limited capacity to communicate physical and psychological symptoms[16]. They may also be in home-based settings with limited access to inpatient services at the hospital.[17-19] Parents of cognitively impaired children are at higher risk for anxiety, depression, and stress than parents of typically developing ill children. [20, 21]

Music therapy and other music-based interventions have been shown to improve psychological and physical outcomes in children with life-limiting conditions.[22-30] While cognitively-impaired children with life-limiting conditions are commonly offered pediatric palliative care music therapy in clinical practice, they are often excluded from research studies because interventions do not accommodate cognitively impaired or non-verbal children. Few studies have examined parent outcomes related to music-based interventions. Research into existing music-based interventions for this population has rarely included family-centered approaches, which are vital for pediatric palliative care research and practice.

Study Aims

The overall purpose of this study is to test the feasibility and preliminary efficacy of a songwriting intervention for cognitively-impaired children (ages 5-17 years) who are receiving palliative or complex care and their parents. Specific aims are to: (1) determine the feasibility of songwriting for cognitively-impaired children with life-limiting conditions and their parents, and (2) examine preliminary efficacy of songwriting for child psychological distress and physical symptoms, parent psychological distress, and family environment. We hypothesize that the intervention will be feasible and show positive trends to improve child, parent, and family outcomes.

Research Design and Methods

Conceptual framework

Guided by Haase's model,[31] we developed a family-centered songwriting intervention that is unique for cognitively-impaired children with advanced life-limiting conditions and their parents. The scientific premise of this study is that advanced life-limiting conditions in children with cognitive impairment create an environment in which both ill children and their parents experience undesirable negative psychological and physical effects. Therapeutic intervention is needed to alleviate these negative effects. Alleviation of these effects may provide immediate benefits to both ill children and their parents, with potential long-term benefits to bereaved parents. The remarkable nature of music to influence health suggests that a family-centered songwriting intervention may be beneficial to cognitively-impaired children with progressively declining life-limiting

illnesses and their parents. As such, the conceptual framework in Figure 1 guides this proposed study.

Study Sample

The target sample for this study is 25 child-parent dyads. Children 5 to 17 years of age with cognitive impairment, receiving palliative or complex care, with progressively declining disease, and with the ability to hear are eligible to participate. Parents must be 18 years of age or older, the individuals in the role of the child's primary and secondary parent caregivers based on parent self-report, cognitively intact, and able to speak and understand English. Parents may include mothers, fathers, or other family members and surrogate parents regardless of biological relationship to the child or sexual orientation. Both parents may participate in the intervention sessions; however, the coordinator will determine the primary parent caregiver (parent who spends the most hours per week caring for the child) during the initial recruitment process to complete study assessments. Additionally, siblings of any age can participate in the intervention as determined by primary parent caregiver but will not complete any study assessments.

Recruitment

Approval has been received from the Institutional Review Board (IRB). Participants will be enrolled from the pediatric palliative care and complex care departments at Vanderbilt University Medical Center. The study coordinator will work closely with the pediatric palliative care team to identify eligible participants and obtain names of potentially eligible children, including permission from the child's palliative care physician or nurse practitioner to approach the parent and child. The coordinator will either (a) mail an introductory letter to parents describing the study and providing an opt-out phone number or email address if they do not want to be contacted further, or (b) approach parents in coordination of a scheduled outpatient visit or inpatient stay for children who have scheduled appointments. Attending physicians/nurse practitioners will be encouraged to briefly introduce the study when possible.

For all eligible parents, the coordinator will obtain verbal parent consent, and confirm contact information (phone, email, mailing address) to send any study documents (e.g., study reminders, REDCap survey link). Written consent will be obtained via REDCap (a secure, web-based electronic research platform) prior to participation. Ill children are not expected to have the decision-making capacity to assent; thus, the IRB approved a waiver of child assent for the ill children. Verbal and written consent/assent will be obtained for all eligible siblings.

Study activities

We will use a 1 group pre- and post-test, repeated measures, clinical trial design. Data collection will take place at baseline (T1), within 1 week post-intervention (T2), and within 1 month of after death of a child (T3). Assessment time points were selected based on our preliminary work and similar adult and pediatric studies.[31-33]

T1 Baseline/Pre-Intervention: Parents will complete T1 measures electronically online via REDCap, a secure website for building and managing online surveys and databases.[34, 35] As soon as consent is obtained, the coordinator will send the electronic link to the REDCap survey. The coordinator will make reminder calls or send reminder emails for surveys not completed within 1 week.

T2-T3 Post-Intervention: Parents will repeat baseline questionnaires at T2 and T3. Parents will also complete a satisfaction survey to help further inform intervention outcomes (i.e., what has been helpful/non-helpful about the intervention).

Songwriting intervention

Board-certified music therapists will deliver the intervention beginning within 1 week after baseline (T1) measures are complete. Similar to previous studies,[31] child-parent dyads will participate in 4 weekly sessions over 4 weeks that will guide parents to write song lyrics about their child. The coordinator and music therapist schedule exact days and times based on family preference. A music DVD will be created by incorporating (a) the audio-recording of the song lyrics, (b) musical accompaniment, (c) photographs and video of child and family interactions, and (d) audio-recordings of physiological aspects (verbalizations, respirations) of the child. The protocol was originally developed for in-person delivery of intervention sessions, in coordination with inpatient or outpatient clinic visits. Due to COVID-19, however, the protocol has been revised to a remotely-delivered intervention using videoconferencing (Zoom). Intervention session content is summarized in Table 1.

Table 1. Songwriting Intervention: Individual Session Content

Session

Lyrics		- Sing/rehearse completed song - Discuss child physiological aspects (respirations, verbalizations) for music DVD - Discuss visual images for music DVD (artwork, photographs) – memories/importance to family
3 Sing and Record	1.5 hour	- Sing/rehearse song with music therapist providing live accompaniment - Record vocal soundtrack for video - Listen to completed soundtrack - Audio record child physiological aspects (respirations, verbalizations) that will be incorporated into song and final music DVD - Gather visual images and/or music therapist uses images/video clips recorded during intervention sessions - Complete storyboard
4 Final Music DVD	0.5 hours	- Share music video draft with family; discuss any suggested edits - Discuss thoughts about/reflections of songwriting project - Educate family on how they can use final DVD therapeutically - Finalize music DVD, create individualized DVD label for family - Mail 2 final music DVDs to family

Fidelity

Treatment fidelity will be enhanced through regular team meetings and training. We will offer all participants the same number of intervention sessions, and sessions will have the same established number of minutes. We will measure dose intensity by recording the minutes of each session and the number of sessions completed by each participant. After each intervention session, music therapists will complete a standardized fidelity checklist to monitor whether or not all study activities were followed as planned.

Measures

Instruments were selected based on outcome variables identified in previous music-based intervention studies and on our previous work.[22, 24, 25, 31, 36-40] When possible, Patient-Reported Outcomes Measurement Information System (PROMIS) measures based on sound psychometrics and developed to enhance communication among clinicians and patients in diverse research and clinical settings were selected.[41] PROMIS parent-proxy measures were developed for parents serving as proxy reporters for children ages 5-17 years; thus, these tools are ideal for measuring child outcomes for cognitively impaired children 5-17 years of age who are unable to self-report.

Table 2. Measures

Aim

2	Parent psychological distress	PROMIS Anxiety – Short Form[46, 47] NIH Toolbox Perceived Stress Survey[48, 49] PROMIS Sleep Disturbance – Short Form[50]	T1-T3 T1-T3 T1-T3
2	Family environment	Family Adaptability & Cohesions Scales (FACES IV)[31, 51]	T1-T3

*= completed by staff

Data analysis

Analysis of the feasibility and preliminary effects of songwriting on child and parent psychological distress, child physical symptoms, and family environment will be conducted using SPSS and STATA statistical software. Frequency distributions will summarize the rates of recruitment and retention to inform recruitment targets and the feasibility of protocol completion in this population for future work. If a child dies on study prior to completing the intervention, partial data (e.g., recruitment and retention data) will still be used to inform future studies. Descriptive statistical and graphical methods will be used to summarize and visualize patterns in the study outcome data distributions at each time of assessment. Given measurement error inherent in self-reported outcomes, reliable change indices (RCI) will be generated for each of the PROMIS outcomes.[52-54] Frequency distributions will summarize the distributions of reliable change (decrease, staying the same, increase) for each measure. Patterns of associations of the participant demographic and clinical characteristics, as well as PROMIS T1 values, with the RCIs will be examined graphically and via the use of effect size indices (correlations or Cohen's d as appropriate). Results from these analyses will provide invaluable information for future larger studies by informing which sets of patients with specific characteristics benefitted the most from the songwriting intervention.

Discussion

Songwriting may be an important strategy for a family-centered music therapy intervention to improve physical and psychological outcomes for cognitively impaired children with life-limiting conditions and their parents. We have developed a novel family-centered songwriting intervention for cognitively-impaired children with life-limiting conditions and their parents.

This study is innovative in testing an empirically-based family-centered songwriting intervention for cognitively-impaired children with life-limiting conditions and their parents. Gaps exist in the music therapy literature for evidence-based interventions that include this population. We will include children with cognitive impairments as songwriting contributors by incorporating audio-recordings of their physiological aspects (verbalizations, respirations) based on biomusic research.[55] Parents will contribute to writing song lyrics about their child, selecting the child's preferred music/song, and singing or co-singing the final audio-recording of the song to be used in the final music DVD. Other family members (e.g., siblings) can be involved throughout the process. Family interactions will be documented via photographs and video and incorporated into the final music DVD. This novel approach has strong potential to shift the paradigm of standard of care for music-based interventions such as music therapy, not only in pediatric palliative care, but also other populations who could benefit from our methods

of delivering a songwriting intervention to individuals with cognitive or verbal impairments.

This study is novel to remotely-deliver a songwriting intervention. Although previous research has utilized face-to-face music-based intervention delivery methods,[56-64] few studies have used remotely-delivered methods which are critical for home-based interventions needed for pediatric palliative care.[18, 65] Our remotely-delivered songwriting intervention will expand our potential impact and increase access for cognitively-impaired pediatric palliative care populations.

Our study is pioneering to additionally test the potential impact of family-centered songwriting on child and parent outcomes before and after a child's death. Previous work has shown that bereaved parents and siblings have found continuing bonds (maintaining connections with the deceased) as a meaningful way to promote healthy coping and adjustment.[66] Children near the end of life often desire to do or say things to be remembered, yet cognitive or verbal impairments can preclude these children from participating in services which facilitate documentation of their legacies.[66-68] Legacy-making has been explored in both adult and pediatric populations.[32, 33, 66, 68-72] Legacy-making in adults has been shown to increase patients' sense of dignity, purpose, meaning, and will to live, while decreasing suffering and depressive symptoms.[72] Our work has shown that legacy-making can improve psychological distress, physical symptoms, and family components (quality of communication) among children with cancer (ages 7-17) and their parents.[32, 33, 67, 70]

This study is led by a study team composed of pediatric palliative care nursing scientists, a music scientist, a clinical psychologist, a pediatric palliative care clinician, pediatric and parent caregiver experts, and pediatric board-certified music therapists. Our team represents a unique interdisciplinary collaboration among music, nursing, medicine, and psychology, but also a unique melding of science and alternative and complementary medicine. There is an on-going need to develop evidence-based music interventions that promote improved psychological and physical outcomes for ill children and their parents and potentially reduce the risk for long-term negative consequences. The usefulness of music-based health interventions to improve outcomes for children with cognitive impairments associated with life-limiting conditions and their parents is largely unexplored. Our study will provide rigorous evidence to fill the gap.

Limitations

This music therapy songwriting protocol was developed for cognitively-impaired children ages 5 to 17 years of age with life-limiting conditions, thus, generalizability will be limited to this population. The protocol also does not allow for extended family members, such as grandparents, to be active participants in the intervention sessions. However, parent feedback on the satisfaction survey will allow us to explore participant feedback for future modifications. Few existing measures include parent-proxy questions appropriate to assess child psychological distress for cognitively-impaired children, resulting in limited child assessments. Future work should include exploring the feasibility of additional physiological measures (e.g., salivary cortisol) to further measure songwriting effects for these children. The remotely-delivered intervention also requires participants to have internet access for Zoom sessions. We do have one study

iPad with internet access available for loan to families located nearby, however, this would likely not be feasible for a future larger multi-site study.

Conclusion

Music therapy and other music-based interventions have shown strong benefits for children with life-limiting conditions but have not been extensively tested in children with cognitive impairments and advanced illness. Our first step is to test the feasibility and preliminary efficacy of our songwriting intervention for cognitively-impaired children with advanced life-limiting conditions and their parents. We have assembled a strong multi-site interdisciplinary team with established collaboration, as well as content and methods expertise to examine songwriting in the cognitively-impaired pediatric palliative care population.

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Please see attached author declaration. We have no conflicts of interest to disclose.