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Does music therapy reduce distress in pediatric oncology patients?

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ABSTRACT

OBJECTIVE: The objective of this selective EBM review is to determine whether or not "Does music therapy reduce distress in pediatric oncology patients?"

STUDY DESIGN: A systematic review of three randomized controlled trials (RCTs). All studies were published in English between 2010 and 2017.

DATA SOURCES: Three RCTs were obtained from PubMed and published in peer reviewed journals. The studies were chosen based on their relevance to the clinical question and their inclusion of patient-oriented outcomes (POEMS).

OUTCOMES MEASURED: In all of the articles used for this review, the main outcome measured was distress reduction. The participants' level of distress was measured with either the Spielberger State-Trait Anxiety Inventory (STAI), the Kidcope measurement tool, or a behavioral coding form of child affect. Distress levels were reported both before and after the oncology procedure or treatment.

RESULTS: In the RCT conducted by Nguyen et al., there was a significant reduction in distress in the music group compared to the control group. The mean difference between groups was calculated to be 1.4. The p-value was < 0.001, making this study statistically significant with a large treatment effect. The RCT conducted by Barry et al. showed a p-value of 0.076 with a NNT of 2 and RBI of 1. This study approached significance with a large treatment effect. In the RCT conducted by Robb et al., the mean difference was 1.07 with a p-value of 0.040, indicating this study to be statistically significant with a large treatment effect.

CONCLUSIONS: Statistical significance was found in two of the three RCTs based on the reported p-values. The calculated p-value for one of the RCTs approached statistical significance. The music intervention group in each study showed a large treatment effect in comparison to the control groups. The results of this review suggest that music therapy can reduce distress in pediatric oncology patients. Future studies could be done to improve external validity.

KEYWORDS: pediatric oncology, music therapy

INTRODUCTION

Pediatric oncology is a medical specialty that focuses on the research and treatment of cancer in children. The types of treatment that a child will receive is largely dependent on their type of cancer and the stage of disease. Common treatments may include chemotherapy, radiation, surgery, lumbar punctures, immunotherapy, stem cell transplants, etc. Procedures associated with cancer treatment in children can often be very painful or traumatic experiences. The emotional distress associated with these treatments is a prevalent problem that can lead to prolonged changes in behavior, alterations in self-concept, fear, anxiety, and depression.[1]

Cancer in children is a common occurrence that will impact many patients and practicing physician assistants. An estimated 10,500 children in the United States under the age of 15 will have been diagnosed with cancer in the year 2021.[2] Due to recent treatment advances, 84% of children who are diagnosed with cancer now survive 5 years or more.[2] The treatment for cancer requires regular inpatient and outpatient visits. Childhood cancers are responsible for 3.5% of pediatric hospital stays and 5% of all cancer hospital stays.[3] The number of visits per year varies based on the patient specific diagnosis, prognosis, and other determinants of health. On average, a pediatric cancer hospitalization stay is 6.4 days.[3] Regardless of an individual child's treatment plan or severity of disease, the significant costs associated with care often cause further complications. In a survey conducted by The National Children's Cancer Society, 95% of families say their child's cancer caused a financial burden on their family.[4] The average medical costs associated with childhood cancer in 2018 was $833,000 for one child.[4] Even when in remission, lack of funds can shift the course of a child's future and wellbeing. With the recent increases in remission rates, more research has been allocated to studying the long-term effects of the disease. According to a study published in the Journal of Pediatric Psychology, 40% of

childhood cancer survivors have later reported traumatic stress symptoms in the moderate to severe range.[5]

In the oncology setting there is an increase in demand for adjunctive therapies to support children undergoing treatments. Current standard of care typically includes methods such as pharmacologic management, patient education, psychotherapy, or behavioral therapy. However, there is a shortcoming of empirically validated interventions that specifically address the needs of young children with cancer.[6] Music therapy may be used as an adjunctive therapeutic intervention to support pediatric oncology patients. This type of therapy involves the use of music to address the physical, emotional, cognitive, and social needs of a group or an individual.[6] Various hypotheses have been proposed to explain the mechanism by which music reduces distress, including modification of cognitive states, moods, and emotions.[7] Music has been shown to reduce s-cortisol and cause an increase in levels of s-oxytocin.[7] Additionally, relaxation from music has demonstrated to be a pleasant distraction that serves as a mild sedative.[7]

This paper evaluates three randomized controlled trials (RCTs), assessing the ability of music therapy to reduce the distress experienced by pediatric oncology patients.

OBJECTIVE

The objective of this selective EBM review is to determine whether or not "Does music therapy reduce distress in pediatric oncology patients?"

METHODS

The criteria used to select the three randomized controlled trials discussed in this systematic review comprised the analysis of the use of music therapy and its ability to reduce distress in pediatric oncology patients. The population being studied in this review includes

children between the ages of 3 and 13 years old – all of whom underwent an oncologic procedure or treatment. To measure the outcome of distress reduction, the trials studied the intervention of music therapy in comparison to the use of audiobooks or to the practice of standard of care without music.

The studies chosen were found on PubMed using the key words “pediatric oncology” and “music therapy.” All three of the randomized controlled trials were published in peer-reviewed journals using the English language. The research was conducted by Kathryn Bagnardi who selected the articles based on their inclusion of patient-oriented outcomes (POEMs) and their relevance to answering the clinical question: “Does music therapy reduce distress in pediatric oncology patients?” The inclusion criteria for the selection of studies were randomized controlled trials and articles published within the last 10 years. Exclusion criteria included studies published before 2010, observational studies, and studies evaluating adults. The statistics used in the articles for measuring distress levels were reported using p-values, mean, and standard deviation. Table 1 demonstrates the demographics and characteristics of the included studies.

Table 1. Demographics and Characteristics of Included Studies

Study	Type	# Pts	Age (yrs)	Inclusion Criteria	Exclusion Criteria	W/D	Interventions
Nguyen (2010)[7]	RCT	40	7-12	Children with leukemia aged 7-12 years, undergoing lumbar puncture at the Oncology Ward at NHP, Hanoi, had undergone a lumbar puncture in conjunction with their cancer at least once before	Significant hearing or visual impairment, cognitive disorder	9	Earphones with music vs earphones without music while undergoing a lumbar puncture
Barry (2010)[8]	RCT	11	6-13	Pediatric oncology patients between the ages of 6-13 years, male and female, no prior experience of radiation therapy, outpatients, functionally and cognitively able to create a CD, attending the Peter MacCallum Cancer Centre for treatment within the ten-month recruitment phase	Clinically deaf, children sedated for their treatment, those with parents who have poor English language skills	1	Create a music CD to listen to during radiation treatment vs standard care during radiation treatment
Robb (2017)[6]	RCT	16	3-8	Children aged 3-8 years, expected hospitalization ≥ 3 days for chemotherapy, a consistent parent who could be present for all study sessions	Child and/or parent did not speak English, child had significant cognitive impairment based on physician judgment	4	Music therapy sessions vs audio books during hospital admission for chemotherapy treatment

OUTCOMES MEASURED

The major outcome measured in all the RCTs was the reduction of distress for pediatric oncology patients. The study conducted by Nguyen et al.[7] used the 6-item short form of the Speilberger State-Trait Anxiety Inventory (STAI), which is a validated scale designed to measure anxiety. The scale ranges from 6 to 24 points, with 6 points indicating no distress and 24 points indicating the highest level of distress. Study participants were evaluated using the STAI in two phases: before and after their individual lumbar punctures were performed.[7]

In the study conducted by Barry et al.[8], participants' distress levels were measured the day after their initial radiation therapy treatment using a modified Kidcope measurement tool.

This tool is a brief assessment in which the interviewer asks a child to report on the distress felt in relation to a stressor, the child's way of coping with a stressor, and the helpfulness of coping strategies used. The specific stressor assessed in this study was undergoing their first radiation therapy treatment. The Kidcope questions were designed to assess the efficacy of 10 coping strategies by having the participants rate that the strategy they used either helped not at all (0 points), a little (1 point), or a lot (2 points). The 10 coping strategies included were: distraction, social withdrawal, cognitive restructuring, self-criticism, blaming others, problem solving, emotional regulation, wishful thinking, social support, and resignation.[8]

The third study, by Robb et al.[6], measured facial affect as an indicator of emotional distress. A behavioral coding form, was created which allowed for objective measurement of the participants' facial affect as either "positive", "negative", or "neutral." "Positive" was coded when a child smiled or laughed during the time interval. "Negative" was coded when the child cried, whined, raised their voice, or verbalized fear, unhappiness, or pain during the time interval. "Neutral" was coded when the child exhibited a flat affect during the time interval. Observers were trained in coding procedures to ensure consistency and analyzed a video recording of the patients' third session of either music therapy or attention control with audiobooks.[6]

RESULTS

Nguyen et al.[7] conducted a randomized controlled trial to evaluate the distress levels of children 7-12 years old with leukemia, all of whom were scheduled to undergo a lumbar puncture (LP) at the oncology ward of the National Hospital of Pediatrics in Hanoi, Vietnam.[7] This study was conducted between November 2007 and July 2008 and included 40 participants– 25 boys and 15 girls.[7] The children were randomly assigned to one of two groups: the music

group (n=20) or the control group (n=20).[7] Randomization was carried out using opaque envelopes, with neither the researcher nor physician knowing which group the patient was assigned to.[7] No significant differences were found between the two groups in regard to patient demographics.[7] For the music group, children chose songs that they wanted to be played through their earphones from an iPod. In the control group, children were given earphones to wear without music being played through them.[7] The children put on their earphones 10 minutes before their LP procedure began and either did or did not listen to music, dependent on their previously assigned group.[7]

Using the STAI scale, participants self-reported their anxiety levels with a range of six to 24 points – six points indicating no anxiety and 24 points indicating the highest level of anxiety.[7] These scores were measured both immediately before the LP began and directly after the procedure had finished.[7] The statistical data used to measure distress reduction was presented as p-values, mean values, and standard deviation.[7] The scores after 10 minutes of wearing earphones but before the LP, were significantly lower ($P < 0.001$) for children in the music group than for those in the control group.[7] The music group presented a mean score of 8.6 with a standard deviation of 2.78. In comparison, the control group showed a mean score of 13.25 with a standard deviation of 3.73.[7] Reductions in anxiety were also significant ($P < 0.001$) after the LP – demonstrating a mean of 8.1 with a standard deviation of 2.22 for the music group and a mean of 13.0 with a standard deviation of 4.17 for the control group.[7] The mean difference (Cohen's-*d*) between the music group and control group was calculated to be 1.4, indicating a large effect size with clinical significance.[7] Values for this study are presented in Table 2.

Table 2. Comparison of Anxiety Scores Before and After the Procedure (Nguyen et al.[7])

		Mean ± SD	P-value	Cohen's-*d*
Music Group	**Before LP**	8.6 ± 2.78	< 0.001	1.4
	After LP	8.1 ± 2.22		
Control Group	**Before LP**	13.25 ± 3.73		
	After LP	13.0 ± 4.17		

Barry et al.[8] conducted a randomized controlled trial to evaluate the usefulness of music therapy intervention for pediatric patients 6-13 years old with varying types of cancer– including brain, kidney, leukemia, and bone and soft tissue. All these patients were attending the Peter MacCallum Cancer Centre in Victoria, Australia for their initial radiation therapy treatment.[8] None of the participants had any prior experience with radiation therapy.[8] This study was conducted over a 10-month recruitment time and included 11 participants– both males and females.[8] The children were randomly assigned to one of two groups: the music group (n=5) or the control group (n=6).[8] Randomization was carried out using permuted block randomization with a block size of four and kept staff members unaware of which group the children were randomized to.[8] However, staff often became unblinded through discussions with the children and when turning on the participants' music once in the treatment room.[8]

For the music group, children worked with a music therapist researcher to create a CD of original music that embodied their preferred musical sounds.[8] These participants were provided with the music therapy CD on their day of radiation and given the option of listening to their creation while in the treatment room.[8] All five children in the music group opted to listen to their CD for the duration of their treatment.[8] In the control group, children received standard supportive care measures.[8] This standard care could include listening to music, selected by either the patient or the staff, in order to comply with existing ethical regulations of the hospital.[8] Four of the six children in the control group opted to listen to music for the duration of their treatment.[8]

Using the Kidcope measurement tool, participants evaluated 10 different coping strategies by rating their helpfulness in reducing distress as 0 (helped not at all), 1 (helped a little), or 2 (helped a lot).[8] The statistical data was presented as number of children, median score, and range of scores. Quantitative analysis indicated that the most frequently used and effective coping strategies were distraction, cognitive restructuring, wishful thinking, and emotional regulation.[8] In contrast, the least utilized and effective coping strategies were social withdrawal, self-criticism, and blaming others.[8] In rank order, Table 3 presents the efficacy of the coping strategies used. Additionally, the Kidcope measurement tool evaluated the frequency of coping strategies used by the pediatric patients during their radiation treatment. The statistical data was presented as number of children, percentage, and P-value. Social withdrawal was used by 67% of the control group (n=4) and 0% of the music group (n=5).[8] The difference in frequency between the two groups approached significance (P = 0.076).[8] The calculated NNT was 2, which showed a large treatment effect that implies clinical significance. The ABI was calculated to be 0.67 and the RBI to be 1, as shown in Table 5. This information, efficacy and frequency of coping strategies used for distress reduction, was collected the day after each child's initial treatment in an interview guided by a music therapist researcher.[8]

Table 3. Efficacy of Coping Strategies Used (Barry et al.[8])

Coping Strategy	Number of Children	Median	Range
Distraction	**8**	**2**	**1–2**
Cognitive Restructuring	**8**	**2**	**0–2**
Emotional Regulation	**7**	**2**	**1–2**
Wishful Thinking	**8**	**1.5**	**0–2**
Resignation	**4**	**1.5**	**0–2**
Problem Solving	**6**	**1**	**0–2**
Social Support	**5**	**1**	**1**
Social Withdrawal	**4**	**1**	**0–2**
Self-criticism	**1**	**1**	**1**
Blaming Others	**0**	–	–

Table 4. Frequency of Coping Strategies Used (Barry et al.[8])

Control Group n (%)	Music Group n (%)	P-value
4 (67%)	0 (0%)	0.076

Table 5. Calculations for Treatment from Barry et al.[8]

Study	CER	EER	RBI	ABI	NNT
Barry et al.	0.67	0	1	0.67	2

Robb et al.[6] conducted a randomized control trial to evaluate emotional distress of children 3-8 years old with either leukemia or a malignant tumor. The study participants were expected to be hospitalized at Riley Children's Hospital in Indianapolis, Indiana for 3 or more days to undergo chemotherapy.[6] Participants were enrolled over nine months (May–July 2011; September 2012–January 2013) and included 16 children– 8 females and 8 males.[6] The children were randomly assigned to one of two groups: the music group (n=9) or the control group (n=7).[6] Randomization was conducted by a biostatistician using a computer algorithm, and assignments were made by the project manager using sealed, opaque envelopes.[6] No significant differences were found between the two groups in regard to patient demographics.[6] All participants received three sessions with a board-certified music therapist.[6] The sessions occurred once daily over the first three consecutive days of inpatient chemotherapy and a consistent therapist delivered all the sessions.[6] All sessions were video recorded for later analysis.[6] For the music group, patient sessions were 45 minutes long and consisted of active music engagement.[6] In the control group, patient sessions were 35 minutes long and consisted of listening to selected audio storybooks.[6]

Using a behavioral coding form, trained observers objectively measured child facial affect as an indicator of emotional distress.[6] These scores were measured at varying time intervals of the third session video recordings.[6] The statistical data used to measure distress reduction was computed based on the frequency of facial affect scores coded as "positive" and was presented as p-values, mean values, and standard deviation.[6] The scores for positive facial

affect were significantly higher (P = 0.040) for children in the music group than for those in the control group.[6] The music group measured a mean of 13.56 with a standard deviation of 10.17.[6] In comparison, the control group showed a mean of 4.93 with a standard deviation of 3.86.[6] The mean difference (Cohen's-*d*) between the music group and control group was reported to be 1.07, indicating a large effect size with clinical significance.[6] Values for this study are presented in Table 5.

Table 5. Comparison of Child Distress (Positive Facial Affect) (Robb et. al[6])

	Mean ± SD	P-value	Cohen's-*d*
Music Group	13.56 ± 10.17	0.040	1.07
Control Group	4.93 ± 3.86		

DISCUSSION

Many children in the United States are diagnosed with cancer and consequently undergo strenuous treatment regimens. The distress oftentimes associated with their medical care can have lasting, negative effects on the child. This systematic review evaluated the usefulness of incorporating music therapy to reduce the distress of pediatric oncology patients between the ages of 3 and 13 years old. Two of the three RCTs were determined statistically significant based on their P-values, and one of the RCTs approached statistical significance with a P-value of 0.076.[6-8] In each study, music therapy was found to be beneficial in reducing distress among patients in the treatment groups compared to children in the control groups. Nguyen et al.[7] demonstrated a mean difference (Cohens-*d*) of 1.4 between the music group and the control group. This study showed that music medicine is a distractor that helps relieve distress endured by pediatric oncology patients. Barry et al.[8] presented that social withdrawal was one of the least efficacious and utilized coping strategies. This study also showed that 67% of the control group used this coping mechanism to inefficiently reduce distress, while 0% of the music group used it.[8] The treatment effect was large, with NNT=2.[8] This study demonstrated that the intervention

of a music therapy CD may support the efficacious coping strategies used by children to reduce their distress during treatment. Robb et al.[6] reported a mean difference (Cohens-*d*) of 1.07 between the music group and the control group. This study demonstrated that the delivery of music engagement results in lower emotional distress for the child. The effect size for Nguyen et al. and Robb et al.[6,7] was determined to be large, based on the mean differences between control groups and music groups. The quantitative findings from the study conducted by Barry et al.[8], however, are limited in their external validity due to the small sample size and a p-value greater than 0.05.

The studies used in this review consisted of several limitations. In the study conducted by Nguyen et al.[7], the short STAI scale used was originally designed and validated for adults. Although it has previously been used to study adolescence, the scale has not been officially validated for its use on children. A limitation of this study's intervention was that the earphones were not abundantly comfortable for the children throughout their procedure and carried the increased risk of transferring infection to the children.[7] A final limitation of this study is that all the children enrolled were aware of the possibility to be given music intervention during their treatment. When participants in the control group did not receive music, this may have led to a sense of missing something and affected their overall distress levels.[7] The study by Barry et al.[8], cited more general limitations of time, budget, recruitment, and, ultimately, a small sample size. Although this study can provide quality conceptual generalizations, the small sample size limits its validity and findings should consequently be viewed as exploratory.[8] Lastly, in the study conducted by Robb et al.[6], main study limitations included bias in music delivery due to music therapists conducting both the treatment and control sessions, a small sample size, and that measurement of child distress was limited to one behavioral indicator of facial affect.

CONCLUSION

All three randomized controlled trials that were utilized to conduct this systematic review demonstrated that the use of music therapy can reduce the distress of pediatric oncology patients between the ages of 3 and 13 years old. The treatment effect in each study was determined to be large based on the NNT and the mean difference between the control groups and the music groups. Statistical significance was found in two of the three RCTs based on the reported p-values. The calculated p-value for one of the RCTs approached statistical significance.

Future studies investigating the use of music therapy to reduce distress in pediatric oncology patients should use a larger sample size to include more patients in each group and allow for more conclusive findings. Additionally, it may be beneficial to have a standardized, validated measurement tool to score pediatric patient distress levels.

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