A Randomized Controlled Trial of Massage Therapy in Children with Sickle Cell Disease

Kathleen L. Lemanek,[1] PhD, Mark Ranalli,[1] MD, and Colleen Lukens,[2] PhD

[1]Ohio State University College of Medicine and [2]Children's Hospital of Philadelphia

Objective   This randomized controlled trial investigated the short-term effects of massage therapy on youth with SCD and their parents.   **Methods**   Thirty-four children and adolescents, and their parents were assigned to a massage therapy or an attention control group. Parents were trained in massage in their homes once a week for 4 weeks, with instructions to provide nightly massages. Families in the control group were visited weekly by a research assistant. Participants completed measures of depression and anxiety, functional status, pain intensity, medication use, and service utilization.   **Results**   Parents in the massage therapy group reported higher levels of depression and anxiety following the intervention. Youth in this group showed higher levels of functional status, and lower levels of depression, anxiety, and pain. Health service utilization rates were unchanged from pre- to post-intervention.   **Conclusions**   These results offer preliminary support for parent-delivered massage therapy as an intervention for SCD pain.

Key words   massage therapy; pain; sickle cell disease.

Introduction: Benefits of Massage Therapy in Youth with Sickle Cell Disease

Recurring or chronic pain is the hallmark clinical feature of sickle cell disease (SCD). Children and adolescents with SCD typically report pain occurring in the extremities, hip, or trunk on 7–30% of diary days, with an average duration of 2.5 days and an average pain rating of 5 on a 10-point scale (Dampier, Ely, Brodercki, & O'Neal, 2002). Events may occur spontaneously or as a consequence of environmental stress (e.g., excessive cold or heat exposure), physiological stress (e.g., infection), or psychosocial stress (e.g., school demands) (Ballas, 1998). Pain has a substantial impact on academic performance (i.e., completing school assignments, school attendance), sleep and social activities (Gil et al., 2000). The typical pain management protocol includes hydration, the use of nonsteriodal anti-inflammatory drugs (e.g., ibuprofen) and/or narcotics (e.g., Dilaudid), and cognitive-behavioral techniques (e.g., deep breathing, visual imagery), and often prayer and touch (Dampier et al., 2002; Chen, Cole, & Kato, 2004).

Touch in the form of massage has been used for centuries in such countries as China and India as a pain management strategy. Massage offers physical benefits, such as increased blood and parasympathetic circulation, and improved joint movement, along with psychological benefits, such as relaxation, daily activity promotion, and pain relief (Fritz, 2000). Massage may produce pain relief by pressure stimuli from massage competing with pain stimuli to reach the brain first, thereby, closing the gate to pain, or by restoring sleep and inhibiting the release of Substance P, which is associated with pain (Fields, 2001). A reduction in muscle rigidity and spasm also may be critical in SCD due to the role of spasms in SCD pain (Bodhise, Dejoie, Brandon, Simpkins, & Ballas, 2004). Tiffany Field and her colleagues at the Touch Institute have extensively examined the effects of massage therapy on emotional adjustment (e.g., depression, anxiety), pain responses, activities of daily living (e.g., school attendance, sleeping habits), and stress hormones levels (e.g., cortisol, epinephrine) in a range of patient populations, including infants born prematurely and individuals with painful

All correspondence concerning this article should be addressed to Kathleen L. Lemanek, PhD, Department of Psychology, Nationwide Children's Hospital, 700 Children's Drive, Columbus, OH 43205, USA. E-mail: kathleen.lemanek@nationwidechildrens.org

medical conditions (e.g., juvenile rheumatoid arthritis) (Field, 2001). In these studies, the massage technique has involved deep tissue manipulation with presumed stimulation of pressure receptors.

Massage therapy has been examined in two studies in patients with SCD, one group of adults (Myers, Robinson, Guthrie, Lamp, & Lottenberg, 1999) and a group of three adults and one child (Bodhise et al., 2004). Both studies found therapeutic changes in pain following massage therapy, along with reductions in emergency department (ED) visits and hospitalizations, and opioid use in the Bodhise study. However, both studies focused on adults within a clinic setting, and neither used a no-treatment or placebo control group.

This study was, therefore, designed to examine the short-term effects of massage therapy on youth with SCD and their caregivers, as administered by caregivers within the home setting. The exact methodology used by The Touch Institute was followed in terms of measurement and massage technique. We hypothesized that: (1) caregiver ratings of depression and anxiety would be lower and ratings of functional status would be higher in the massage therapy group versus a control group; (2) self ratings of depression and anxiety, and ratings of pain would be lower in youth receiving versus those not receiving massage; and (3) health service utilization rates would be lower for youth in the massage therapy group compared to a control group.

Methods
Participants

The participants included 34 youth with SCD and their primary caregivers, and were recruited from a Sickle Cell Disease Program located at a children's hospital. Youth with documented cognitive delays, and who were on monthly transfusions or were prescribed Hydroxyurea were not recruited because of barriers to measurement completion and positive effect on pain management. This study was approved by the hospital's institutional review board (IRB). Written informed consent and assent was obtained from all participants.

In the SCD Program a potential pool of 59 children was available who met age and exclusionary criteria. Table I provides demographic characteristics of children in both groups. Chi-squared analyses revealed no group differences for gender, for education level of parents, or for income. One-way ANOVAs showed no group differences in age of participants, age of parents, or hemoglobin level at study entry. A group difference was found on disease type, with

Table I. Demographic Characteristics of the Sample

Characteristics	Massage therapy (n = 18) M (SD)	Attention control (n = 16) M (SD)
Age	9.97 (2.47)	11.55 (1.00)
Hemoglobin level	8.84 (1.21)	9.64 (1.29)
Parent age	38.99 (6.50)	37.66 (9.23)
	N (%)	N (%)
Gender		
Male	7 (38.9)	7 (43.8)
Female	11 (61.1)	9 (56.3)
Disease type		
HbSS	13 (72.2)*	6 (37.5)
HbSC/Hb Beta thal	5 (27.7)	10 (62.5)
Parent education		
High school/GED	3 (16.7)	7 (43.8)
Vocational school/some college	8 (44.4)	7 (43.8)
College/professional degree	7 (38.9)	2 (12.5)
Income		
<$30,000	8 (44.4)	10 (71.4)
$30,000–100,000	10 (55.6)	4 (28.6)

Note. Three children diagnosed with HbBeta Thal were included within the HbSC group due to relatively asymptomatic presentation. Ten income groups were collapsed into two groups due to zero cell sizes in some groups.
*$p < .05$.

more youth diagnosed with HbSS in the massage therapy group, χ^2 (1, $N = 34$) = 4.14, $p = .05$.

Measures

Center for Epidemiological Studies of Depression Scale—Depression Scale (CES-D)
The CES-D (Radloff, 1977) is a 20-item standardized self-report scale of depressive symptoms. Items are rated on a 4-point Likert scale ranging from 0 (rarely) to 3 (most of the time), with scores ranging from 0 to 60. A score of 16 or greater differentiates clinically depressed and nondepressed. A standardized α coefficient for the CES-D for this sample was .82.

State-Trait Anxiety Inventory (STAI)
The STAI (Spielberger, Gorsuch, Lushene, Vagg, & Jacobs, 1983) is a reliable and valid self-report measure of state anxiety based on how respondents feel "right now, at this moment." Twenty items are rated on a 4-point scale from 1 (not at all) to 4 (very much so), with total scores ranging from 20 to 80. A standardized α coefficient for the S-Anxiety for this sample was .89.

Functional Status-II(R) (FS-IIR)
The FS-IIR (Stein & Jessop, 1991) is a 43-item measure of health status of newborns through adolescents. A total

score is calculated based on whether the child performs the specified activity or behavior ''never or rarely'' (0), ''some of the time'' (1), or ''almost always'' (2). The standardized α coefficient for the FS-IIR for this sample was .68.

Children's Depression Inventory (CDI)

The CDI (Kovacs, 1992) is a 27-item standardized self-rated symptom scale of depression used with school-age children and adolescents. Scores range from 0 to 54 based on item ratings of 0, 1, 2, with higher scores indicating increasing severity of depression. A cut-off score of 12 or 13 is recommended to minimize the number of false positives. Standardized α for the CDI for this sample was .77.

State-Trait Anxiety Inventory for Children (STAIC)

The STAIC (Spielberger, Edwards, Lushene, Monturoi, & Platzek, 1973) is a 20-item self-report measure of state anxiety based on how children feel at a ''particular moment in time,'' Each item is rated on a 3-point scale, with total scores ranging from 20 to 60, and where higher scores indicate greater anxiety. Standardized α coefficient for this sample was .78.

Pediatric Pain Scale

The Pediatric Pain Scale is a reliable and valid measure of pain for children as young as 3 years of age (Baker & Wong, 1987). This depicts a series of six faces representing ascending levels of hurt from 0 (''I don't hurt at all'') to 5 (''I hurt as much as you can imagine'') (Baker & Wong, 1987).

Health Service Utilization Rates

The Hospital's Patient Care database system was reviewed for number of emergency department (ED) visits, number of hospitalizations, and number of days admitted for SCD-related complications for all children, starting 6 months before and until 6 months after the study.

Procedure/Intervention

Families were contacted by letter and then by telephone by staff of the SCD Program. Families who expressed interest were seen at the Hospital to describe the study and to obtain consent and assent. Caregivers completed the CES-D and the FS-IIR, and youth completed the CDI. These measures were completed again after each family's participation in the study. Measures of anxiety were completed by youth and caregivers on a weekly basis for 4 weeks. Youth rated their pain each morning and each evening for 30 days.

Following this meeting, youth and caregivers were randomly assigned to a massage therapy group or an attention control group. For youth in the massage therapy group, a licensed massage therapist visited the home within 1 week after signing consent/assent. During this home visit, the therapist trained one or both parents how to give a standard massage. Instruction, demonstration, and written materials were used to train parents in massage. The massage protocol was that used by Field (2001), which consisted of two phases: (1) child is in supine position and continuous stroking movements are applied to face, stomach, legs, and arms, and (2) child is in prone position and the back is massaged. The massage therapist visited families weekly for 3 more weeks (total of four home visits) to collect anxiety measures and pain ratings. The therapist also monitored caregivers' technique and provided corrective feedback in giving a massage. Caregivers were required to give a 20-minute massage using this protocol for those days between therapist visits; thus, youth received massages for a total of 30 days.

Another group of youth and caregivers served as an attention control group. Families in this group completed all forms, but caregivers were not trained to give a massage. A research assistant collected forms from families in their homes weekly. Caregivers who expressed interest in learning to give massages to their children were trained to do so at the end of the study.

Data Analysis

A series of ANCOVAs were run to examine the effects of massage therapy on parent-reported and child-reported depression and anxiety, parent-reported functional status, child-reported pain, and healthcare utilization rates within a randomized controlled clinical trial. For the CES-D, the CDI, and the FS-IIR, pre-intervention scores were used as co-variates. Parent state anxiety ratings (STAI) and child state anxiety ratings (STAIC) were averaged separately for the first weeks and the last 2 weeks to form early-intervention and late-intervention scores. Morning and evening pain ratings were added across the first 2 weeks and then the second 2 weeks of monitoring. An average early-intervention score and late-intervention score was then calculated from these two sets of pain ratings. Health service utilization rates for ED visits and hospitalizations were tabulated for 6 months before and for 6 months after the 30-day intervention. Analyses were run first with pre/early-intervention scores as co-variates and then with both pre/early-intervention scores and disease type as co-variates due to the significant difference between groups in disease type.

Table II. Group Means and Standard Deviations Pre- and Post-Intervention

	Massage therapy group ($n = 18$)				Attention control group ($n = 16$)			
	Pre/early		Post/late		Pre/early		Post/late	
	M	SD	M	SD	M	SD	M	SD
Parent measures								
CES-D	13.5	(09.8)	18.8	(13.9)***	16.5	(10.1)	15.4	(10.4)
STAI State	47.9	(12.0)	55.1	(21.0)****	45.95	(13.4)	49.2	(15.4)
FS-IIR	89.5	(09.9)	91.8	(07.8)****	86.9	(11.6)	84.4	(13.6)
Child Measures								
CDI	47.3	(07.9)	45.0	(05.4)*	50.4	(08.1)	46.7	(07.8)
STAIC State	38.2	(05.0)	40.1	(08.8)**	41.5	(08.2)	43.9	(12.8)
FS-IIR	11.3	(12.2)	11.9	(16.6)*	14.4	(14.1)	15.1	(22.5)
Utilization Rates								
Frequency of hospitalizations	0.50	(0.71)	0.94	(1.43)	0.56	(1.0)	0.63	(0.96)
Number of days hospitalized	1.8	(2.9)	3.67	(6.00)	2.8	(5.3)	3.56	(5.89)
Frequency of ED visits	0.50	(0.86)	0.50	(0.73)	0.19	(0.40)	0.50	(0.62)

Note. CES-D, FS-IIR, and CDI scores, and utilization rates were obtained before and after the 30-day intervention period. STAI-State and STAIC-State scores were collected during the first 2 weeks (early) and the last 2 weeks (late) of the intervention. CES-D: Center for Epidemiological Studies of Depression Scale; STAI: State-Trait Anxiety Inventory; FS-IIR: Functional Status II—Revised; CDI = Children's Depression Inventory; STAIC = State-Trait Anxiety Inventory for Children.
*$p < .05$; **$p < .01$; ***$p < .001$; ****$p < .0001$.

Results

Table II presents the means and standard deviations for all measures. Results presented are for analyses with only pre/early-intervention scores as co-variates since the inclusion of disease type as a co-variate did not alter the results obtained.

Results of the ANCOVAs on the CES-D [(32, 2), $F = 8.21$, $p = .001$] and the STAI state anxiety scale [(22, 2), $F = 18.68$, $p = .0001$] were significant, with caregivers in the massage therapy group scoring higher than caregivers in the control group. The result for the FS-IIR was significant, where ratings of functional status for the massage therapy group were higher compared to the control group [(34, 2), $F = 14.22$, $p = .0001$].

Results of the ANCOVAs for the CDI [(34, 2), $F = 4.02$, $p = .05$], the STAIC state anxiety scale [(24, 2), $F = 6.47$, $p = .01$], and pain ratings [(21, 2), $F = 4.11$, $p = .05$] were significant, with youth in the massage therapy group rating their level of depression, anxiety, and pain lower than those youth in the control group.

The ANCOVAs for health service utilization rates were not significant for frequency of ED visits [(33, 2), $F = 2.96$, ns] or hospitalizations [(34, 2), $F = 1.68$, ns], or number of days hospitalized [(31 2), $F = 2.11$, ns]. Group differences were not found during the study period for these three variables.

Discussion

This study found decreased depression and anxiety, and pain as reported by youth with SCD who received massages during a 30-day period. Increased functioning also was rated by caregivers of these youth. This study extends previous research by providing home training to caregivers to perform the massages, by administering both self-report and parent-report measures of depression and anxiety, and by including an attention control group.

The increased reports of depression from pre to post-intervention and anxiety from early to late intervention in caregivers of youth in the massage therapy group were a surprising finding. Disease-related stresses reported by caregivers of youth with SCD have included the unpredictable nature of SCD and the demands to manage symptoms at home by recognizing symptoms and implementing treatment plans (e.g., Barakat et al., 2007). The requirement to provide a nightly massage may have added to these stresses on a short-term basis. However, if benefits are obtained over time with regular massage, caregivers may experience a sense of mastery in preventing or reducing pain, with a subsequent decrease in negative affectivity.

Several limitations are evident in this study. The primary limitation centers on treatment integrity or whether caregivers implemented the massages in a standard manner with respect to technique, frequency, and length. Caregivers, in fact, commented on the

challenges in providing nightly massages amongst other family demands. A similar issue pertains to whether the massage therapist's training was standardized across families and over time. A second limitation is the small sample size and the few types of SCD represented, which restricts the generalizability of the results. A third limitation is the measures used to assess medical and psychological variables with respect to availability of normative data, timing of data collection, and short-term versus long-term effects.

Two design issues should be considered in future investigations of massage therapy, type of control group and home- versus clinic-based treatment. A wait-list control group with ratings of caregiver competence and/or massage frequency as moderators of outcomes would be one approach. The use of sham massage (light pressure only) as a control group also would allow for examination of the relative efficacy of massage where similar treatment groups that differ on level of pressure are compared (Field, 2001; Moyer, Rounds, & Hannum, 2004). Although more costly, home-based interventions has the potential of being more culturally sensitive and enhancing the expertise of caregivers. Clinic-based interventions may be more structured but they can pose their own challenges to run, such as missing and rescheduling appointments because of illness, weather, or transportation (Myers et al., 1999).

The cost–benefit ratio will need to be examined in future studies in terms of time required by families for training and to provide the massages, employing a licensed massage therapist, and impact on social, academic, and interpersonal functioning. Those interested in using massage therapy within research or clinical practice should be mindful of two caveats (Myers et al., 1999). One caveat is that massage should not delay or replace conventional medicine in treating SCD. The second caveat is that massage therapy should not be done without medical supervision. Results of this study are preliminary but they highlight the potential benefits of massage in reducing pain and increasing functioning in youth with SCD.

Acknowledgments

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