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CLINICAL STUDY

# Improvement in resiliency factors among adolescents with neurofibromatosis who participate in a virtual mind–body group program

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## Abstract

**Purpose** To examine effects of a virtual mind–body group for adolescents with neurofibromatoses (NF1 and NF2; Resilient Youth with Neurofibromatosis; RY-NF) on multiple resiliency factors against a health education attention control (Health Education for Youth with Neurofibromatosis; HE-NF) using data from a randomized controlled trial. Specifically, our research question was whether adolescents randomized to the RY-NF (versus the HE-NF) would have greater improvements in resiliency factors at post-intervention and whether these gains would be maintained at 6-month follow-up.

**Methods** Adolescents with NF ($n=51$; $M$ age 12–17) were randomly assigned to RY-NF ($n=27$) or HE-NF ($n=24$). Resiliency factors (mindfulness, coping, gratitude, optimism, and social support) were collected at baseline, post-intervention (88%), and 6-month follow-up (82%).

**Results** Participation in the RY-NF was associated with greater pre-to-post improvements in gratitude ($M_{difference}=4.38$; 95% CI—0.52–8.23; $p=.027$) and mindfulness ($M_{difference}=9.41$; 95% CI 4.40–14.42.; $p<.001$) compared to HE-NF; improvements sustained at 6 months. There were no group differences on any additional resiliency factors. However, participation in the RY-NF was associated with pre-to-post- improvements in coping ($M_{difference}=9.16$; 95% CI 2.93–15.39; $p=.005$), and social support ($M_{difference}=6.79$; 95% CI 1.96–11.63; $p=.007$); improvements sustained at 6 months.

**Conclusions** Participation in the RY-NF resulted in sustained improvement in several resiliency factors. Promoting resiliency may help adolescents successfully navigate challenges associated with NF.

**Keywords** Adolescents · Mind–body · Neurofibromatosis · Resilience · Videoconferencing

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## Introduction

The neurofibromatoses (NF; NF1, NF2) are heterogeneous genetic disorders characterized by the development of nerve sheath tumors. While most tumors are benign, risk for malignancy is high. NF is the most common genetic neurological disorder caused by a single gene, affecting over 100,000 people in the United States. Most common symptoms include cutaneous tumors and disfiguration (NF1), and hearing loss, poor gait, and facial weakness (NF2). Regardless of NF type, youth with NF have similar psychological profiles characterized by greater emotional distress (anxiety and depression) [1], more pain [2], more coping difficulties and learning disabilities [3], and lower quality of life (QoL) [4] than same age peers. Current management is primarily through surgical and palliative interventions, with no evidence-based interventions developed to address specific NF your challenges.

Resiliency—the ability to "bounce back" and maintain adaptation and effective functioning under significant adversity or challenging life conditions [5, 6]—is a multifaceted construct comprised of learned and practiced cognitive, behavioral, and social support skills [7], including mindfulness (intentional awareness without judgement; [8, 9]), coping skills (utilizing effective coping strategies) [10], social support (accessing supportive interpersonal resources) [11] gratitude (personal appreciation and thankfulness for what one has) [12], and optimism (positive expectation) [13].

Although resiliency has gained increasing interest within clinical research and practice for youth with chronic medical illness [14, 15], with programs and interventions demonstrating that resiliency factors are modifiable [16] and produce positive effects for participants [17], little research to date has focused on developing psychosocial intervention that target resiliency in NF [18], and none in adolescents with NF. To address this gap, our team has adapted the Relaxation Response Resiliency Program for adults with NF (3RP-NF) [19] which we found efficacious in improving resiliency factors in adults with NF, for the unique needs of youth (Resilient Youth with NF; RY-NF) [20]. The program teaches relaxation response methods (e.g., diaphragmatic breathing, mindfulness, meditation), appraisal and coping (stress and symptom awareness, problem solving, adaptive thinking), and growth enhancement (gratitude, acceptance, social support). In a pilot RCT we showed that RY-NF was feasible and accepted by adolescents, and was associated with sustained improvement in quality of life compared to a health education attention placebo control (Health Education for NF; HE-NF) [20]. Here, we report on secondary analyses of this data and report on changes in the resiliency factors targeted by the RY-NF.

We hypothesized that adolescents randomized to the RY-NF (versus the HE-NF) would have greater improvements in resiliency factors (i.e., mindfulness, perceived coping, gratitude, optimism, and social support) at post-intervention and that these gains would be maintained at 6-month follow-up.

## Method

### Patient population

Participants were 51 adolescents with NF. The Massachusetts General Hospital Institutional Review Board (MGH IRB) approved all procedures related to this study, and complete methodology including recruitment was previously published [20]. Consent was obtained from all individual participants included in the study.

Briefly, we recruited adolescents through the NF registry of the Children's Tumor Foundation (CTF), who emailed our IRB-approved ad periodically to the NF community. Interested participants (adolescents or their parents) emailed study staff for study details. Study staff scheduled a brief intake session (15–20 min) with one or both parents and adolescent to provide details on the study procedures, screen for eligibility, and conduct the consent or assent process. Adolescent eligibility requirements included: (1) an NF diagnosis made by a medical professional (2) 12 to 17 years of age (3) the ability to provide consent/assent for participation, (4) third grade level English comprehension, and (5) endorsement of stress and coping difficulties related to NF. Exclusionary criteria were the presence of severe psychopathology and/or an unwillingness to participate in weekly Skype sessions (45 min).

Following consent (age 13–17) or assent (age 12), adolescent participants were emailed a secure link with the baseline questionnaires through REDCap [21] and were then randomized to their respective conditions (active or control). Randomization.com (free internet- program) randomly assigned participants to conditions (1:1 ratio within blocks of ten: 5 to RY-NF and 5 to HE-NF). After program completion (eight 45-min group sessions) and 6 months later, participants completed the questionnaires again.

### Active and control intervention conditions

Both active and control interventions were matched for time and therapist attention/support and took place over Skype. All eight sessions were 45 min long and were facilitated by the senior author who has long-standing experience delivering interventions to demographically and medically diverse patients, including NF. Groups were kept small (i.e., five or less participants) to ensure adolescents' uptake, engagement, and mastery of skills. Manuals and recordings were mailed or emailed to participants prior to the start of the study based on preference. Differences between the two conditions can be seen in the original manuscript [20].

#### Active intervention

The RY-NF intervention was developed from the adult NF program (3RP-NF) [19, 22] using qualitative information gathered from live video (Skype) focus groups with 15 adolescents (NF1 $N=11$; NF2 $N=4$). From these focus groups, specific adolescent challenges, stressors, and preferences were identified and added to the RY-NF manual. Although essential components of the original intervention remain (e.g., relaxation response skills, appraisal and coping, and positive psychology skills), the intervention for adolescents underwent major revisions for ease of use and population relevance. These included reducing session length from 90 to 45 min, lowering reading comprehension to a third-grade level, making content more relevant to adolescent stressors

(e.g., bullying, dating, academics), and using explicit behavioral strategies to positively reinforce participation and acquisition of skills. Target outcomes of the active intervention included increasing overall quality of life, reducing distress related to NF [20], as well as enhancing resiliency variables (i.e., resiliency, perceived coping, gratitude, optimism, and social support).

#### Control intervention

The HE-NF was adapted from the adult Health Enhancement Program for NF (HEP-NF) [19]. We made similar modifications in language and format. Sessions included information on lifestyle behavior changes, such as sleep, nutrition, and exercise and general psychoeducation on stress associated with NF. Although weekly goals were discussed and set by participants, no skill rehearsal or homework of any kind was administered to this condition.

### Measures

#### Mindfulness

The Child and Adolescent Mindfulness Measure (CAMM) [23] is a 10-item measure used to assess present-moment mindfulness (i.e., nonjudgmental and nonavoidant thoughts and feelings) in children and adolescents. Items on the CAMM are scored on a 5-point Likert-type scale ranging from 0 (*never true)* to 4 (*always true;* range 0–40). All items are then reverse scored and summed, with higher total scores indicating greater mindfulness.

#### Perceived coping abilities

The Measure of Current Status—A (MOCS-A) [24] is a 13-item measure used to assess an individual's perception of their ability to use coping skills (e.g., relaxation, reappraising thoughts, work through tension and stress) effectively. Items on the MOCS-A are scored on a 5-point Likert-type scale ranging from 0 (*I cannot do this at all)* to 4 (*I can do this extremely well;* range 0–52) with higher total scores indicating greater perceived coping abilities.

#### Gratitude

The Gratitude Questionnaire (GQ-6) [25] is a 6-item measure used to assess dispositional/trait gratitude. Items on the GQ-6 are scored on a 7-point Likert-type scale ranging from 1 (*strongly disagree*) to 7 (*strongly agree;* range 7–42) with higher total scores indicating more dispositional gratitude.

#### Optimism

The Life Orientation Test – Revised (LOT-R) [26, 27] is a 10-item measure used to assess an individual's personal qualities of optimism and pessimism (bidimensional traits) [28]. Of the 10 items, 6 are scored for optimism while the other 4 are used as "filler" items (i.e., not calculated in total score). Items on the LOT-R are scored on a 5-point scale ranging from 1 (*I disagree a lot*) to 5 (*I agree a lot;* range 6–30) with higher scores indicating greater optimism.

#### Social support

The Medical Outcome Study Social Support Survey (MOS) [29] is a 19-item multidimensional measure used to assess an individual's availability/perceptions of social support within four domains: emotional, tangible, affectionate, and positive social interactions. Items on the MOS are scored on a 5-point Likert-type scale ranging from 1 (*none of the time*) to 5 (*all of the time;* range 19–95) with higher scores indicating more social support.

### Data analytic strategy

We conducted a secondary data analysis to examine the impact of RY-NF versus HE-NF on resiliency outcomes. Using a mixed-model repeated measures ANOVA, we compared the effect of the RY-NF versus HE-NF on changes from pre- to post-intervention and from post-intervention to 6-month follow-up using linear contrasts. Analyses included all randomized participants (i.e., intent-to-treat principle).

#### Power analysis

The power for this study was determined using guidelines and previous trials for pilot RCTs [19, 30–33]. For this study, 25 participants per condition (i.e., 50 participants total) was deemed adequate in allowing for stable mean estimates and for detecting a large effect size ($d = 0.80$).

## Results

### Sample characteristics and attrition

We have previously described sample demographics and attrition [20]. Briefly, of the 60 adolescents screened for study eligibility, 51 (59% male, 76% White; RY-NF $N = 27$; HE-NF $N = 24$) met study criteria and enrolled. Of these, 45 completed the intervention and provided post-intervention assessments ($\geq 6$ sessions; $n = 45$; 88%; RY-NF $N = 24$; HE-NF $N = 21$), and 39 (82%; 21 RY-NF and 18 HE-NF) completed the six-month follow-up. No

differences in demographic or main study variables were seen between completers and non-completers, or between those randomized to RY-NF versus HE–NY. This study consisted of a geographically diverse sample, with participants from Columbia ($n=1$), Canada ($n=8$), and the United States (n = 42). Sociodemographic variables are presented in Table 1, with unadjusted means for the resiliency outcomes at each time point presented in Table 2.

**Table 1** Descriptive statistics for the entire sample ($n=51$)

| Characteristic | RY-NF ($n=27$) | HE-NF ($n=24$) |
|---|---|---|
| NF Type, n (%) | | |
| NF1 | 23 (85.18) | 19 (79.16) |
| NF2 | 4 (14.81) | 5 (20.83) |
| Age, mean (SD) | 14.48 (1.34) | 14.26 (1.70) |
| Sex, n (%) | | |
| Female | 9 (33.33) | 12 (50.00) |
| Male | 18 (66.66) | 12 (50.00) |
| Race, n (%) | | |
| White/Caucasian | 21 (77.77) | 18 (75.00) |
| Other | 4 (14.81) | 3 (12.50) |
| Hispanic or Latino | 2 (7.40) | 4 (16.66) |
| Years of school completed, mean (SD) | 8.41 (1.52) | 8.39 (1.75) |
| Learning Disability, n (%) | | |
| Yes | 14 (51.85) | 13 (54.16) |
| No | 13 (48.14) | 11 (45.83) |
| Parent with NF diagnosis, n (%) | | |
| Yes | 4 (14.81) | 3 (12.50) |
| No | 23 (85.18) | 21 (87.50) |

*NF* neurofibromatosis

Post-intervention outcomes. Participation in RY-NF resulted in greater improvements in gratitude ($M_{difference}$= 4.38; 95% CI 0.52–8.23; $p=0.027$) and mindfulness ($M_{difference}$= 9.41; 95% CI 4.40–14.42.; $p<0.001$) relative to the HE-NF. Significant group differences for baseline to post-intervention change scores were not observed for any other resiliency factors (i.e. perceived coping, optimism, and social support). However, participation in RY-NF resulted in pre- to post-intervention improvements in perceived coping abilities ($M_{difference}$= 9.16; 95% CI 2.93–15.39; $p=0.005$) and social support ($M_{difference}$= 6.79; 95% CI 1.96–11.63; $p=007$). There was also a trend toward improvement in optimism ($M_{difference}$= 1.66; 95% CI − 0.012 − 3.33; $p=0.052$). These within-group improvements were not observed for adolescents in HE-NF.

## Six-month follow-up outcomes

No differences between the RY-NF and HE-NF were seen in change scores from posttreatment to 6-month follow-up for any resiliency variables ($p>0.05$). All improvements

**Table 2** Unadjusted baseline, posttreatment and 6 months follow up scores

| Assessment | Baseline | Posttreatment | *p (b–p)* | 6 M Follow-up | *p (p–6 m)* |
|---|---|---|---|---|---|
| Mindfulness | | | | | |
| RY-NF | 19.40 (6.83) | 24.25 (6.54) | < .001* | 23.68 (7.38) | .961 |
| HE-NF | 28.20 (7.94) | 24.18 (8.85) | | 22.38 (7.24) | |
| Resiliency | | | | | |
| RY-NF | 60.63 (18.70) | 70.29 (18.06) | .150 | 76.77 (12.90) | .062 |
| HE-NF | 61.75 (16.34) | 66.50 (18.89) | | 64.24 (19.83) | |
| Perceived coping | | | | | |
| RY-NF | 21.00 (14.80) | 29.88 (12.47) | .408 | 36.00 (9.63) | .093 |
| HE-NF | 19.00 (13.65) | 24.15 (16.39) | | 22.79 (15.55) | |
| Gratitude | | | | | |
| RY-NF | 28.73 (6.19) | 31.04 (6.62) | .027* | 34.30 (5.21) | .593 |
| HE-NF | 30.00 (8.35) | 29.74 (8.89) | | 31.71 (8.77) | |
| Optimism | | | | | |
| RY-NF | 12.50 (2.90) | 14.57 (4.50) | .514 | 15.00 (3.81) | .440 |
| HE-NF | 12.58 (3.27) | 14.00 (4.31) | | 13.62 (4.21) | |
| Social support | | | | | |
| RY-NF | 74.78 (11.99) | 81.63 (10.77) | .101 | 85.28 (7.29) | .213 |
| HE-NF | 74.33 (16.60) | 76.0 (13.54) | | 83.86 (9.83) | |

*RY-NF* resilient youth-neurofibromatosis; *HE-NF* health enhancement-neurofibromatosis

Values are expressed as means and SDs; *p* p value (i.e. significance) between-groups; *b–p* baseline to post-treatment; *p–6 m* posttreatment to 6-month follow-up

*$p<.05$

observed in RY-NF maintained through the 6-months follow-up.

## Discussion

There is a clear need for accessible and effective psychosocial interventions for patients with NF. Our previous work has demonstrated that a live video mind–body intervention for adults with NF (3RP-NF) is associated with greater improvement in QoL [19] and resiliency factors [18] when compared to an attention placebo control. We have also shown that when adapted for the specific needs of adolescents with NF, the RY-NF mind–body program is also associated with similar improvements in QoL in adolescents [20].

Through secondary data analyses, we explored the effects of RY-NF on resiliency factors among 51 adolescents. We found that adolescents who participated in the RY-NF experienced greater improvements from baseline to post-test in gratitude compared to their HE-NF counterparts. Although nascent in child and adolescent development [34, 35], gratitude has been depicted as a powerful resiliency variable consistently associated with substantial improvement in well-being [36–39], positive affect, pride, prosocial behavior, and decrease in physical symptoms [39]. Our pilot RCT demonstrated that an 8-week virtual mind–body intervention was associated with sustained increase in gratitude over and above education and attention from therapist.

Along with gratitude, adolescents who participated in the RY-NF also experienced greater improvements in mindfulness compared to their HE-NF counterparts. Mindfulness involves increasing awareness and skillful responding to reduce cognitive vulnerabilities [40]. Mindfulness can aid adolescents in fostering attention, compassion, and emotion regulation [41], which are associated with academic success, positive prosocial behavior, and overall well-being [41, 42]. By increasing gratitude and mindfulness among adolescents with NF, there may be potential positive downstream effects (e.g., enhanced well-being, positive affect, prosocial behavior) which support developing adolescents—especially while they are navigating the challenge of living with an uncurable, chronic medical illness like NF.

Results are consistent with our adult [18, 19], where we found that participation in 3RP-NF was also associated with sustained improvement in mindfulness over and above that experienced by those in the control condition. However, our adolescent study also showed improvements in gratitude in the active intervention over and above control, which we did not see in our study with adults. Interestingly, the magnitude of improvement in the active condition in both samples was similar (− 3 points) suggesting that between group differences may have been driven primarily by change scores in the individuals randomized to the control condition. Specifically, for adults randomized to control the mean scores on gratitude were unchanged after the intervention, while adolescents randomized to control we observed a slight decrease.

Although adolescents who participated in the RY-NF active intervention did not improve more than those in the HE-NF control on any other resiliency factors measured, they did experience significant within group improvements in perceived coping and social support, while those in the HE-NF did not. These findings are also different than what we observed in our adult study, where participants in the resiliency intervention improved significantly from baseline to post-test and over and above those in the control in perceived coping abilities and social support [18]. This suggests that while both the adult and adolescent interventions improve social support and coping, the adult intervention may have a stronger effect likely due to the sessions in RY-NF being shorter (45 versus 90 min) which allowed for less practice of the skills in session.

Strengths of the current study include the adaptation of the NF-specific adult mind–body intervention (3RP-NF) for an adolescent population (RY-NF), which may facilitate not only implementation but also practice of skills within the family. Our intervention aimed to place NF in context to the adolescent experience—identifying ways of improving resiliency which were developmentally sensitive and relevant through technologies that eliminate, or at least reduce, typical barriers to care. Other strengths included recruitment and enrollment unencumbered by geographic location, the apt use of videoconferencing technology to facilitate sessions and eliminate treatment barriers, a high participant retention through 6 months, and a rigorous single blind design. Limitations include the small sample inherent in pilot studies, and the delivery of all sessions by one clinician with substantial expertise in NF. Last, although we had geographic recruitment diversity, most adolescents participating in this study were white and presented with NF1. Future research initiatives will include efforts to diversify our sample of adolescent patients.

Despite these limitations, this study provides support for the role of RY-NF in improving resiliency factors among adolescents with NF. Resiliency is particularly important for adolescents with NF who experience both the typical medical stressors that accompany an incurable disease (i.e., ongoing medical procedures, uncertain prognosis), as well as developmentally specific stressors, such as bullying, school achievement, peer stress, and parent relationship dynamics. Using these promising findings, we are now conducting a fully powered RCT of the RY-NF versus HE-NF in order to fully determine the efficacy and sustainability of the intervention, before making it available to all adolescents with NF.

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## Compliance with ethical standards

**Conflict of interest** Authors Ethan Lester, Erik Maklin, Scott Plotkin, and Ana-Maria Vranceanu declares no conflict of interest.

**Ethical approval** All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

**Informed consent** Consent was obtained from all individual participants included in the study.

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