A PILOT STUDY OF THE EFFECTIVENESS OF A TELEHEALTH GROUP FOR IMPROVING PEER RELATIONSHIPS FOR ADOLESCENTS WITH NEUROFIBROMATOSIS TYPE 1

by

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ABSTRACT

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Research that has included children and adolescents with neurofibromatosis type 1 (NF1) suggests an elevated rate of social difficulties in comparison to same-aged peers. However, interventions for social difficulties have not been investigated in the NF1 population. The Program for the Education and Enrichment of Relational Skills (PEERS®; Laugeson & Frankel, 2010) has been widely established as an intervention to improve social functioning in autism spectrum disorders (Laugeson et al., 2012) and more recently, in a pilot study for attention-deficit/hyperactivity disorder (Gardner et al., 2015). This research aimed to 1) conduct a pilot study to examine the effectiveness of a 14-week telehealth PEERS® intervention using both an open trial and a pilot randomized controlled trial, and 2) determine the acceptability and feasibility of the intervention for adolescents with NF1. Participants were 27 adolescents with NF1 (M_{age} = 13.91 years, SD = 1.51) with social skills difficulties and at least one caregiver. Caregivers and adolescents completed several measures of social outcomes at pre-test and post-test. Following the intervention, caregiver-reported social-emotional skills, social impairment, caregiver-reported number of adolescent get-togethers, and social knowledge showed improvement. Additionally, caregiver treatment ratings following the intervention indicated a high level of acceptability and feasibility for telehealth-delivered PEERS®. The pilot data from

this investigation show the promise of the specific social skills intervention, PEERS®, to support the social and friendship skills of adolescents with NF1 who have social difficulties. Identifying an intervention strategy for improving aspects of social functioning may help to mitigate negative social outcomes.

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Introduction

Given that examinations of social skills have found that children and adolescents with neurofibromatosis type 1 (NF1) are at increased risk for social difficulties (Barton & North, 2004; Huijbregts et al., 2015; Huijbregts & de Sonneville, 2011; Loitfelder et al., 2015), social difficulties have more of an impact on older individuals with NF1 in comparison to younger individuals (Wolkenstein et al., 2001), and these difficulties are likely to continue into adulthood (Pride et al., 2013), it is important to examine the effectiveness of an intervention aimed at improving social skills and peer interactions for adolescents with NF1. This research served as a pilot study to examine the effectiveness of a 14-week telehealth-delivered Program for the Education and Enrichment of Relational Skills (PEERS®) intervention for improving social skills and peer interactions in NF1, a population where social interventions have not been explored. An additional aim of this research was to explore the acceptability of the intervention in this population using caregiver report. To emphasize the innovation of this investigation, the NF1 behavioral phenotype is described. Additionally, the current literature on social functioning in children with NF1 is discussed, with mention of the phenotypic overlap with Autism Spectrum Disorder and Attention-Deficit/Hyperactivity Disorder. Lastly, the PEERS® intervention and the available empirical support for the intervention is described to highlight the feasibility and potential of this investigation.

NF1 Phenotype

Neurofibromatosis type 1 (NF1) has a prevalence rate of 1 in 3,500 births and is caused by a genetic mutation or a deletion of the NF1-gene that encodes for the tumor suppressor protein, neurofibromin (Ferner et al., 2007; Levine et al., 2006; Miller et al., 2019). The genetic mutation of NF1 can be inherited, often referred to as familial, or occur de novo, referred to as

sporadic. NF1 has a number of physical manifestations that are used to establish a diagnosis and present increasingly as an individual gets older including café-au-lait macules, skinfold freckling, cutaneous neurofibromas, plexiform neurofibromas, iris Lisch nodules, bone dysplasia and optic gliomas (Miller et al., 2019). NF1 is also associated with medical problems including skeletal and orthopedic complications such as macrocephaly, short stature, osteopenia, hypotonia, and scoliosis (Elefteriou et al., 2009; Miller et al., 2019; Tonsgard, 2006), vascular abnormalities such as congenital heart disease, vasculopathy, and hypertension (Friedman et al., 2002; Nguyen et al., 2013), and neurological conditions including migraine headaches (Pinho et al., 2014), and seizures (Ostendorf et al., 2013).

The behavioral phenotype of NF1 is broadly characterized by cognitive, executive, academic, and psychosocial difficulties (Lehtonen et al., 2013). Overall intellectual functioning (IQ) generally falls in the low average to average range for children with NF1 (Cutting et al., 2004; Ferner et al., 1996; Hyman et al., 2005; Sangster et al., 2011), with specific deficits identified in verbal, nonverbal and visuospatial abilities (Dilts et al., 1996; Hyman et al., 2005; Loitfelder et al., 2015; Sangster et al., 2011). Parent-reported executive functioning difficulties are also often evident in school age children with NF1 in the areas of inhibition, shifting, emotional control, initiation, working memory, planning, and organization (Gilboa et al., 2014; Payne et al., 2011). Executive functioning deficits have also been documented in school age children with NF1 using performance-based measures (Gilboa et al., 2014; Hyman et al., 2005; Payne et al., 2011; Roy et al., 2010, 2014). Academically, many children with NF1 have poorer skills in reading, spelling and mathematics in comparison to unaffected siblings and normative data (Barton & North, 2004; Cutting & Levine, 2010; Hyman et al., 2006; Krab et al., 2008; North et al., 1995; Watt et al., 2008) with more impairments in math and reading as they age

during childhood (Wessel et al., 2012). Speech and language impairments have also been identified for some children with NF1 (Hyman et al., 2005; Thompson et al., 2010) with skills falling in the low average range (North et al., 1994) and specific deficits in articulation, receptive language and expressive language problems (Dilts et al., 1996; Hyman et al., 2005; Thompson et al., 2010), although the literature is inconsistent with some studies suggesting no impairment (Sangster et al., 2011). Psychosocial challenges are evident including social difficulties (Allen et al., 2016; Barton & North, 2004; Lewis et al., 2016; Loitfelder et al., 2015), poorer adaptive functioning (Barton & North, 2004; Dilts et al., 1996; Eby et al., 2019; Klein-Tasman et al., 2013), and lower quality of life (Graf et al., 2006; Krab et al., 2009; Oostenbrink et al., 2007).

Social Functioning in NF1

Investigations among typically developing children have illustrated numerous negative outcomes for children with social functioning difficulties including loneliness, aggression, isolation, and academic difficulties (Buhs & Ladd, 2001; Caemmerer & Keith, 2015; Cassidy & Asher, 1992; DeRosier & Lloyd, 2010; Hymel et al., 1990; Ladd, 1999; Woodward & Fergusson, 2000) as well as later conduct problems, mental health problems, and substance abuse (Bierman & Wargo, 1995; Coie et al., 1992; Woodward & Fergusson, 1999). Adolescence, specifically, is a time of significant social development supported by studies examining friendships and brain changes in the areas related to social cognition (Blakemore, 2012; Brizio et al., 2015; Burnett et al., 2009; Steinberg & Morris, 2001). Additionally, the establishment of friendships has been found to be related to self-esteem and the ability to cope with stress and mental health symptoms in adolescence (Buhrmester, 1990; Steinberg & Morris, 2001). Given the benefits of having social relationships and the consequences of social difficulties, it is important to understand how to intervene during adolescence to promote better social outcomes.

Within NF1, the majority of research on social functioning suggests impairments are evident based on parent and teacher report. Difficulties in social functioning broadly have been reported (Allen et al., 2016; Cipolletta et al., 2018) with specific deficits also reported in social skills and social outcomes (Barton & North, 2004; Huijbregts et al., 2015; Huijbregts & de Sonneville, 2011; Loitfelder et al., 2015), including social competence (Barton & North, 2004; Johnson et al., 1999; Lewis et al., 2016) and social problems (Allen et al., 2016; Barton & North, 2004; Cipolletta et al., 2018; Dilts et al., 1996; Huijbregts et al., 2015; Johnson et al., 1999; Loitfelder et al., 2015; van der Vaart et al., 2016). Peers indicate that children with NF1 are more sensitive, more socially isolated, show less leadership behavior, are chosen as a best friend less often, have fewer reciprocated friendships, and are less liked in comparison to classroom peers (Noll et al., 2007). Research examining a virtual mind-body intervention that incorporates relaxation strategies and coping skills has illustrated improvements in social quality of life within NF1 (Funes et al., 2019; Lester et al., 2020; Vranceanu et al., 2016), but interventions strategies to improve specific social outcomes in NF1 such as social and friendship skills have not been explored, to our knowledge. The current available literature about social functioning in children with NF1 suggests social impairments are evident, highlighting the importance of examining an extensively studied social intervention, PEERS®, for this population to potentially improve social skills and peer interactions.

ASD Symptomatology in NF1

Increased vulnerability to autism spectrum disorders (ASD) among individuals with NF1 has been identified, with continued debate about the contributors to ASD symptomatology (Garg et al., 2013; Klein-Tasman, 2020; Plasschaert et al., 2015). ASD is characterized by deficits in social communication and social interaction, and restricted, repetitive behaviors or interests that

were present beginning in an early developmental period (American Psychiatric Association, 2013). Approximately 13-33% of children with NF1 meet criteria for ASD with frequent report of subthreshold symptoms (Garg et al., 2013; Plasschaert et al., 2015; Walsh et al., 2013). ASD symptomatology is evident in children with NF1 using examiner observation and assessment on the Autism Diagnostic Observation Schedule (ADOS) and the Autism Diagnostic Interview – Revised (ADI-R) (Chisholm et al., 2022; Garg et al., 2013). However, children with NF1 typically have significantly milder social deficits compared to individuals with ASD (Adviento et al., 2013; Garg et al., 2013; Plasschaert et al., 2015; Walsh et al., 2013) with social skills scores on the Social Skills Rating System and Social Skills Improvement System often falling in the below average to average rather than the impaired range (Berkovits et al., 2017; Jamison & Schuttler, 2015; Laugeson et al., 2015). Additionally, in a study comparing parent-report measures commonly used to examine social functioning for use in NF1, the majority of children with NF1 fell in the mild to moderate range of impairment on the Social Responsiveness Scale – Second Edition (SRS-2), a broad parent questionnaire of social skills and ASD symptoms, and on the SSIS (Payne et al., 2020). One study using a broad behavioral assessment (e.g., Behavior Assessment System for Children – Second Edition) did not find greater social challenges than a contrast group using parent and teacher report (Martin et al., 2012). This suggests that children with NF1 may have more subtle ASD symptomatology related to social challenges in comparison to children with ASD only.

ADHD Symptomatology in NF1

In addition to the executive functioning concerns described previously, parents and teachers of school age children with NF1 report difficulties with inattention, hyperactivity, and impulsivity (Barton & North, 2004; Dilts et al., 1996; Gilboa et al., 2011; Huijbregts et al., 2015;

Johnson et al., 1999; Payne et al., 2011) with 30-50% meeting DSM criteria for Attention-Deficit/Hyperactivity Disorder (ADHD; Kayl et al., 2000; Koth et al., 2000; Mautner et al., 2002; Pride et al., 2012). ADHD is characterized by persistent inattention and/or hyperactivity and impulsivity that has been present before the age of 12 and impairs social and academic functioning (American Psychiatric Association, 2013). Performance based measures have also shown poorer skills in selective, divided, sustained, and switching attention in comparison to unaffected siblings (Gilboa et al., 2011; Hyman et al., 2005; Isenberg et al., 2013; Payne et al., 2011). There is some evidence to suggest inattention is more common in children with NF1 compared to hyperactivity/impulsivity (Hyman et al., 2005; Pride et al., 2012). However, despite evidence of attention difficulties, it is unclear whether ADHD as a diagnosis is highly comorbid with NF1 or a characteristic part of the NF1 phenotype (Huijbregts, 2012).

Important to the rationale of the current investigation, attention difficulties have been found to be related to social skills and social problems in children with NF1 (Allen et al., 2016; Barton & North, 2004; Mautner et al., 2002; Payne et al., 2020; van der Vaart et al., 2016). Specifically, children with NF1 and ADHD have poorer social outcomes and social skills in comparison to children with NF1 only and children with NF1 and learning difficulties (Barton & North, 2004). This finding is consistent with documentation of relations between ADHD and social difficulties in children with ADHD only (Aduen et al., 2018; Bagwell et al., 2001; Dewey & Volkovinskaia, 2018; Nijmeijer et al., 2008; Van der Oord et al., 2005). Within children and adolescents with ADHD alone (i.e., without NF1), research on social skills training has evidenced improvements in social functioning (Deboo & Prins, 2007). Notably, a pilot study of 20 adolescents, ages 11-16 years, with Attention-Deficit/Hyperactivity Disorder (ADHD) utilized the intervention approach of the current study (PEERS®) and similar outcome measures.

Improvements were demonstrated in initiations of friendships, social knowledge, and frequency of get-togethers (Gardner et al., 2015).

Empirical Support for PEERS® Intervention

The Program for the Education and Enrichment of Relational Skills (PEERS®; Laugeson & Frankel, 2010) is a well-validated, caregiver-assisted social skills intervention provided in a group format for children and adolescents who have difficulty making and keeping friends. A number of studies published out of and in conjunction with the University of California-Los Angeles demonstrate the foundational evidence of the PEERS® intervention for children and adolescents with autism spectrum disorders (ASD) with improvements in social knowledge, social responsiveness, social skills and get-togethers using the 14-week in-person intervention (Table 1; Laugeson et al., 2009, 2012; Mandelberg et al., 2014). Over time, this intervention has been extensively researched and has demonstrated improvements in social knowledge, social responsiveness, social skills, and frequency of get-togethers (Dolan et al., 2016; Hill et al., 2017; Schohl et al., 2014). Additionally, Table 2 highlights the pilot studies that are available in adolescents with ADHD (Gardner et al., 2015) and in adolescents with brain injury (Gilmore et al., 2022).

Aside from research illustrating the initial efficacy of the intervention on peer relationships, studies have also provided evidence for improvements in parental functioning and parent-adolescent relationships in ASD (Corona et al., 2019; Karst et al., 2015) and ADHD samples (Gonring et al., 2017). Several modifications have been made to the intervention that show similar outcomes including adjustments for young adults (Gantman et al., 2012; Laugeson et al., 2015; Wolstencroft et al., 2021), translation into a variety of languages (Idris et al., 2020; Rabin et al., 2020; Shum et al., 2019; Yamada, 2020; Yoo et al., 2014), delivery via a classroom

setting (Laugeson et al., 2014), delivery via a peer-mediated model (Matthews et al., 2018), and accelerated versions of the intervention that meet twice weekly (Marchica & D'Amico, 2016; Matthews et al., 2020).

While the PEERS® intervention has been extensively researched and supported, there are important limitations to this intervention. First, there is a rigorous screening process that includes a number of exclusion criteria; thus, making this intervention suitable for a very specific population. Second, this intervention is group-based with caregiver involvement rather than individualized to fit each participant's specific needs. Last, this intervention is based on neurotypical assumptions and may not promote the ongoing shift towards neurodiversity (Ortega, 2009). Even though these limitations are important, this intervention is one of very few social skills curricula for teens with ASD and related conditions with a clear evidence base.

Recently, the PEERS® curriculum has been adapted to be offered virtually through telehealth with a 14-week protocol as well as with a 16-week protocol that covers the same skills over a slightly longer period of time. Adaptations for the telehealth version can be found in the Center for Autism and Related Disorders (CARD) manual (*PEERS Remote Manual*, n.d.) and recorded role play videos that are accessible on the UCLA website (https://www.semel.ucla.edu/peers/resources/role-play-videos). Homework assignment modifications allow for virtual opportunities (rather than in-person). While the evidence of telehealth PEERS® is still in progress, there has been some promising pilot data to suggest improvement from the intervention with samples of five adolescents with ASD. Miyake and colleagues (2018) presented at a professional conference that the telehealth delivery of PEERS® had comparable findings to in-person outcomes including decreased problem behaviors, increased social skills and improved social responsiveness in teenagers with ASD using the 14-

week protocol (Miyake et al., 2018). Similarly, Estabillo and colleagues (2021) found no significant differences between PEERS® delivered via in person and telehealth. Improvements were demonstrated using the telehealth delivery of PEERS® in social skills knowledge and number of parent-reported adolescent hosted get-togethers. Adler and researchers (2022) had a sample of 12 adolescents with ASD and found improvements in knowledge of social skills, increased parent and teen reported get-togethers, and decreased core autistic symptoms following the virtual PEERS® intervention.

Additionally, pilot studies of young adult PEERS® among young women with Turner Syndrome, a genetic neurodevelopmental disorder, similar to NF1, was conducted using a hybrid in-person and telehealth approach with a 14-week delivery as well as with an accelerated 8-week delivery with similar outcome measures to the current study (Wolstencroft et al., 2019, 2021). Improvements were found in social performance based on parent report, social knowledge, and ASD symptomatology. These initial encouraging findings within neurodevelopmental conditions highlight the potential of the intervention to be offered virtually to adolescents with NF1.

Telehealth Interventions in NF1

Research on telehealth interventions within rare populations has shown promise over the last five years. As mentioned previously, a pilot hybrid approach of young adult PEERS® among young women ages 17-20 years with Turner Syndrome showed feasibility and social improvements within this population (Wolstencroft et al., 2019, 2021). Similarly, a telehealth play-based intervention to improve language and pretend play delivered individually to children between ages 6-12 years with Prader-Willi syndrome, another genetic neurodevelopmental disorder, illustrated feasibility, acceptability and child enjoyment of the intervention (Dimitropoulos et al., 2017). In children and adolescents with William syndrome between ages

10-17 years, a telehealth response inhibition cognitive training program found the online treatment was accepted and positively received by families (Brei et al., 2020). Within NF1 specifically, as mentioned, a virtual mind-body intervention has been explored in adolescents with NF1 with a group size of five participants (Lester et al., 2020) and in adults with NF1 with a group size of 6-8 participants (Vranceanu et al., 2016). These studies have demonstrated feasibility and improvements in quality of life for different ages. These initial encouraging telehealth findings with other genetic neurodevelopmental conditions and with another intervention in NF1 highlight the potential of the present intervention to be offered virtually to adolescents with NF1.

Rationale for the Current Study

To date, there are no research studies examining the effectiveness of social skills interventions for individuals with NF1, even though social difficulties are evident for children and adolescents with NF1 (Barton & North, 2004; Huijbregts et al., 2015; Huijbregts & de Sonneville, 2011; Loitfelder et al., 2015), have more of an impact on older individuals with NF1 in comparison to younger individuals (Wolkenstein et al., 2001) and continue into adulthood (Pride et al., 2013). The lack of research on social skills interventions is likely to be due to rarity of NF1 and thus, it is challenging to do this work face-to-face for an extended period of time (e.g., for 14 weeks as in the PEERS® intervention). However, there is indication that telehealth interventions may be useful in children and adolescents with other genetic neurodevelopmental conditions (Brei et al., 2020; Dimitropoulos et al., 2017) as well as among adolescents with NF1 (Lester et al., 2020). With a telehealth approach now available for the PEERS® intervention, exploration of this intervention within NF1 is now feasible. The central aim of the proposed research was to examine the effectiveness of a 14-week telehealth-delivered PEERS®

intervention (Program for the Education and Enrichment of Relational Skills) to improve social skills and peer interactions for adolescents with NF1. PEERS® is one of only a handful of evidence-based social skills interventions with strong empirical data about efficacy and is generally provided in a group format. It has been used extensively with adolescents with ASD (Hill et al., 2017; Laugeson et al., 2012; Mandelberg et al., 2014) and also has demonstrated preliminary utility with adolescents with ADHD (Gardner et al., 2015). This intervention is conceptually ideal as adolescents with NF1 have a higher incidence of both ASD (Plasschaert et al., 2014) and ADHD (Mautner et al., 2002). Further, pilot data regarding the use of a telehealth version of this intervention are promising (Miyake et al., 2018; Estabillo et al., 2021; Wolstencroft et al., 2019, 2021; Adler et al. 2022). This research is expected to provide a valuable resource for medical providers, school personnel, researchers, and clinicians who work with individuals with NF1.

Research Aims and Hypotheses

Research Aim 1: Examine the effectiveness of a telehealth-delivered Program for the Education and Enrichment of Relational Skills (PEERS®) intervention for improving social skills and peer interactions by examining the following areas:

a) social-emotional skills (SSIS-SEL)
b) social impairment (SRS-2)
c) quality of existing friendships (FQS)
d) number of get-togethers (QSQ)
e) social knowledge (TASSK)

Aim 1a: Using an open trial approach (Phase 1): It is hypothesized that adolescents with NF1 who complete the PEERS® intervention (with a parallel caregiver group) will show improvement from pre-test to post-test (*Hypothesis 1a*).

Aim 1b: Using a pilot randomized controlled trial design (Phase 2): It is hypothesized that the treatment group and waitlist control group will have comparable functioning at pre-test (*Hypothesis 1ba*). In addition, it is hypothesized that the treatment group will have improved performance at post-test (*Hypothesis 1bb*) in comparison to the waitlist control group.

Aim 1c: Across all participants who were enrolled in the study: It is hypothesized that adolescents with NF1 who complete the PEERS® intervention (with a parallel caregiver group) will show improvement from pre-test to post-test (*Hypothesis 1c*).

Research Aim 2: Determine the acceptability and feasibility of the PEERS® intervention with adolescents with NF1 using both caregiver report and dropout analysis. It is hypothesized that caregivers who complete the PEERS® intervention will report a high degree of acceptability and feasibility of the PEERS®.

Methods

Participants

Participants were twenty-seven adolescents with NF1 (M_{age} = 13.91 years, *SD* = 1.51) with social skills difficulties and at least one caregiver from each family. Fifty-nine percent of the sample identified as male, 33% as female, 4% as transgender, and 4% indicated they were still deciding. Eighty-five percent of the sample identified as White, 7.5% as Asian and 7.5% as biracial. Eighty-five percent of the had a sporadic NF etiology, 7.5% had a familial NF etiology and 7.5% did not provide this information. Figure 1 illustrates the flow of participants in both phases of this research. Recruitment methods included (1) contacting previous NF1 participants who have agreed to be contacted about future research studies, (2) providing fliers to

surrounding area Neurofibromatosis clinics, and (3) announcements in the Children's Tumor Foundation patient NF Registry system and NF foundation newsletters (e.g., NF Midwest, NF Northeast). Inclusion and exclusion criteria are included in Table 3.

Procedure

This investigation was a pilot study to provide proof of concept of the effectiveness, feasibility, and acceptability of the PEERS® intervention with adolescents with NF1 using two phases: Phase 1 consisted of a small open trial; Phase 2 consisted of a small pilot randomized controlled trial with random assignment to a treatment or a waitlist control group. The wait list control group subsequently crossed over and received the treatment following the wait-list period. All study activities took place virtually via phone and/or online video conferencing platforms (e.g., Zoom or Microsoft Teams). Figure 2 illustrates the study procedures in a flow chart.

Interested participants contacted us via email or phone. They were informed about the study information and screened over the phone with review of inclusion and exclusion criteria. Following screening, they were consented/assented for a caregiver intake session and adolescent intake session to further determine eligibility. The adolescent intake session included brief assessments of cognitive functioning and reading ability. Assessments and questionnaire measures were administered by well-trained graduate students or other professionals. Once eligibility for the study was determined, participants were consented/assented to participate in the study which included a pre-test session with both the caregiver and the adolescent that included electronic questionnaires focused on the outcomes of interest (Figure 3). Caregivers also completed a semi-structured interview of ADHD symptomatology with a trained study staff

member. For Phase 2, families were notified of the group they were assigned to after the pre-test assessment.

The PEERS® intervention was administered according to the PEERS® manual (Laugeson & Frankel, 2010) and the CARD telehealth manual (*PEERS Remote Manual*, n.d.). The PEERS® intervention was administered by certified PEERS® providers when possible or by well-trained graduate students or other professionals. Supervision was provided by a Licensed Psychologist who is a certified PEERS® provider. The PEERS® intervention involves separate caregiver and adolescent sessions that meet for 90 minutes each week for a 14-week period. The PEERS® curriculum includes 1) structured didactic instruction with a Socratic method of teaching focused on steps of social behavior, 2) role playing with appropriate and inappropriate demonstrations and perspective taking questions, 3) behavioral rehearsal with coaching and performance feedback, and 4) caregiver assistance to help with social coaching, expanding social opportunities and maintaining skills outside of the group. The structure of the caregiver session includes reviewing homework (the majority of time in the caregiver session is spent on this), a portion spent on didactic lessons including role play demonstrations and a portion dedicated to reunification with the adolescents with discussion of homework assignments and individual checkouts. The structure of the adolescent session includes a brief review of homework, with the majority of time spent on didactic lessons including role play demonstrations, followed by opportunities to practice skills in behavioral rehearsal and a portion dedicated to reunification with caregivers with discussion of homework assignments and individual checkouts. Table 4 describes the PEERS® telehealth intervention didactic lessons by session.

Within a week of the final session of the intervention (which consisted primarily of a graduation ceremony), adolescents and caregivers again completed a set of electronic

questionnaires (post-test). Although not included in the current investigation, a 14-week follow up was also conducted where a set of electronic questionnaires was administered again (for a third time). Families received compensation for completing and submitting electronic questionnaires at follow up. This study was approved by UWM's Institutional Review Board (Protocol # 21.039).

Measures

Figure 3 depicts the timepoint when the following measures were administered.

Cognitive Functioning and Reading Ability:

These two measures were administered to address potential exclusion criteria related to cognitive and reading abilities for the study.

Kaufman Brief Intelligence Test – Second Edition (*KBIT-2*; Kaufman & Kaufman, 2004). The KBIT-2 is a brief measure of cognitive abilities including verbal reasoning and nonverbal reasoning. The KBIT-2 demonstrates good reliability and validity. Telehealth administration guidelines of the KBIT-2 were followed. Verbal reasoning is assessed based on two subtests (receptive vocabulary and word-reasoning using "riddles"). Nonverbal reasoning is assessed using a matrices task (i.e., determining which picture completes a puzzle). The IQ composite, verbal and nonverbal standard scores (M=100, SD=15) were used to characterize cognitive functions and aid in eligibility determination for the study. Higher scores represent higher cognitive abilities. Cognitive abilities in the broadly average range or above were considered meeting eligibility criteria. This measure was administered at the adolescent intake appointment.

Wechsler Individual Achievement Test – Third Edition (WIAT-III) Word Reading subtest (Breaux, 2009). The WIAT-III is a measure used to assess academic functioning throughout childhood and adolescence. The WIAT-III Word Reading subtest examines the ability to

accurately read words. This subtest standard score (*M*=100, *SD*=15) was used to characterize reading abilities and aid in eligibility determination for the study. Higher scores represent better reading abilities. Reading abilities in the broadly average range or above were considered meeting eligibility criteria. This measure was administered at the adolescent intake appointment.

Characterization of co-occurring condition symptomatology

ADHD Symptomatology

Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version – Attention Deficit/Hyperactivity Disorder module (KSADS-ADHD; Kaufman et al., 1997). The KSADS is a semi-structured caregiver interview to assess psychopathology in children and adolescents according to the DSM-5 criteria. Strong reliability and validity have been demonstrated (Kaufman et al., 1997). The KSADS-ADHD module includes assessment of inattention, hyperactivity and impulsivity symptoms and was administered to determine if participants met research criteria for an ADHD diagnosis. Symptom descriptions are rated by the interviewer on a 4-point scale including "No information," "Not present," "Occurs occasionally," and "Occurs often." This interview was administered at the pre-test appointment to a caregiver.

ASD Symptomatology

Social Communication Questionnaire (SCQ; (Rutter et al., 2003). The SCQ is a caregiver-report questionnaire that examines ASD symptomatology. The SCQ demonstrates good reliability and validity (Berument et al., 1999; Chandler et al., 2007). The SCQ has 40 items that are rated as "Yes" or "No." The SCQ yields a raw score that is compared to specific research cutoff score (e.g., cutoff of 15 indicates the possibility of ASD). Higher scores represent more ASD symptomatology. This questionnaire was administered at pre-test to a caregiver.

Outcome Measures

Social Functioning:

Social Skills Improvement System Social-Emotional Learning (*SSIS-SEL*; Gresham & Elliott, 2017). The SSIS-SEL is a caregiver and self-report questionnaire measure examining social-emotional skills in childhood and adolescence administered at each of the three timepoints (pre-test, post-test, and follow up). Adequate internal consistency, test-retest reliability, and validity have been demonstrated by the measure developers (F. Gresham et al., 2018; F. M. Gresham & Elliott, 2017). The Parent (51 items) and Student (46 items) forms were used for the caregiver and adolescent, respectively. The reporter is asked to rate each item using a 4-point scale including "Never," "Seldom," "Often" and "Almost Always." Higher scores represent better social-emotional skills. The SEL composite standard score (M=100, SD=15) was used to assess social-emotional skills and as a measure of the effectiveness of the intervention. Internal consistency for the SEL composite standard score was high for both the Parent and Student forms (Parent: Cronbach's alpha = .87; Student: Cronbach's alpha = .95). For analyses examining frequency of social difficulties on this measure, difficulty is represented by a standard score below 85.

The SSIS-SEL provides subscales in Self-Awareness (SeA), Self-Management (SM), Social Awareness (SA), Relationship Skills (RS), and Responsible Decision Making (RDM). Standard scores on subscales were used in exploratory analyses. Cronbach alphas for subscales were calculated for the current sample. On the SSIS-SEL Parent form, the Self-Awareness subscale consisted of 7 items (α = .72), the Self-Management subscale consisted of 14 items (α = .74), the Social Awareness subscale consisted of 7 items (α = .86), the Relationship Skills subscale consisted of 14 items (α = .61), and the Responsible Decision Making subscale consisted of 9 items (α = .82). On the SSIS-SEL Teen form, the Self-Awareness subscale

consisted of 9 items (α = .79), the Self-Management subscale consisted of 9 items (α = .81), the Social Awareness subscale consisted of 7 items (α = .83), the Relationship Skills subscale consisted of 15 items (α = .89), and the Responsible Decision Making subscale consisted of 6 items (α = .65).

Social Responsiveness Scale – Second Edition (*SRS-2*; Constantino & Gruber, 2012). The SRS-2 is a caregiver-report questionnaire of social impairment and ASD symptomatology in childhood and adolescence administered at each of the three timepoints (pre-test, post-test, and follow up). During development of this measure, internal consistency, test-retest reliability, and validity were reported by the developers. The measure includes 65 items that are rated on a 4-point scale including "Not true," "Sometimes true," "Often true," and "Almost always true." Higher scores represent more ASD symptomatology and more social impairment. Given the Total Score is the most well-researched score from the SRS-2, the Total Score T-score (M=50, SD=10) was examined and used as a measure of the effectiveness of the intervention. In the current sample, internal consistency for the SRS-2 Total Score T-score was high (Cronbach's alpha = .94). Difficulty will be represented by a T-score above 60. In exploratory analyses, treatment social subscales of Social Awareness (SAw), Social Cognitive (SCog), Social Communication (SCom), and Social Motivation (SMot) as well as the DSM-5 compatible scale of Social Communication and Interaction (SCI) were examined. Importantly, the measure developers suggest that the application of treatment subscales should be limited to investigations specific to alleviation of symptoms such as when evaluating treatment effects (Constantino & Gruber, 2012). Cronbach alphas for subscales were calculated for the current sample. The Social Awareness subscale consisted of 8 items (α = .68), the Social Cognitive subscale consisted of 12 items (α = .82), the Social Communication subscale consisted of 21 items (α = .52), the Social

Motivation subscale consisted of 11 items (α = .79), and the DSM-5 compatible scale of Social Communication and Interaction subscale consisted of 6 items (α = .92).

Friendship Qualities Scale (*FQS*; Bukowski et al., 1994). The FQS is a self-report questionnaire examining the friendship qualities of companionship, conflict, help, security and closeness at each of the three timepoints (pre-test, post-test, and follow up). During development of this questionnaire, Bukowski and colleagues reported adequate internal consistency and validity. The adolescent is asked to think of their friendship with their best friend and rate 23 items using a 5-point scale including "Not true," "A little true," "Somewhat true," "Mostly true" and "Really true. The FQS yields a total score ranging from 0 to 115 with higher total scores indicate better quality friendships. In the current sample, internal consistency for the FQS total score was high (Cronbach's alpha = .92). The FQS total score was used to gather information about the adolescent's quality of friendships.

Quality of Socialization Questionnaire (*QSQ*; Frankel & Mintz, 2008). The QSQ is a caregiver and self-report questionnaire that will be used to gather information about the adolescent's get-togethers with peers at each of the three timepoints (pre-test, post-test, and follow up). Adequate validity has been demonstrated in a sample of teenagers with ASD (Laugeson et al., 2009). Caregivers and adolescents are asked about how many get-togethers the adolescent organized, the conflict during the get-togethers and how many get-togethers the adolescent was invited to in the last month. This measure has 12 items; 10 items contribute to the conflict scale and two items are related to social initiation (i.e., organized get-togethers) and social reciprocity (i.e., invited get-togethers). Combined raw scores on the social initiation and social reciprocity items (i.e., number of total get togethers) were examined with higher raw scores representing more get-togethers.

Test of Adolescent Social Skills Knowledge (*TASSK*; Laugeson et al., 2009). The TASSK is a self-report 30-item questionnaire measure about specific social skills taught as part of the PEERS® intervention administered to adolescents at each of the three timepoints (pre-test, post-test, and follow up). Adequate internal consistency has been reported in a sample of teenagers with ASD. Higher scores represent greater social skills knowledge.

Treatment Acceptability

Treatment Acceptability Questionnaire (TAQ; (Hunsley, 1992). The TAQ is caregiver-report questionnaire of six items examining the acceptability of the treatment, how ethical the treatment is, how effective the treatment is, side effects of the treatment and the treatment providers' knowledge and trustworthiness. Internal consistency, test-retest reliability and concurrent validity have been demonstrated. Caregivers rate each item on a 7-point scale based on the question stem with 1 indicating poorer reactions to the treatment and 7 indicating better reactions to treatments. Higher raw scores indicate more acceptability of the treatment. This measure was administered to caregivers at post-test.

Treatment Evaluation Inventory Short Form (TEI-SF; (Newton, 2004). The TEI-SF is a caregiver-report questionnaire of eight items focused on reactions to the treatment including the acceptability and discomfort during treatment. The original TEI is a frequently used measure in treatment acceptability research (Carter, 2007). The revised TEI-SF was developed specifically for treatments with children with behavior difficulties and has demonstrated internal consistency (Kelley et al., 1989; Newton, 2004). Caregivers rate each item on a 5-point scale including "Strongly agree," "Disagree," "Neutral," "Agree" and "Strongly agree." Higher raw scores indicate more positive reactions to the treatment. Two items that focused on conducting the intervention without the child's consent were excluded from analyses as one requirement for

participation in this study was for the child to consent to their involvement. This measure was administered to caregivers at post-test.

Data Analytic Plan

A post-hoc power analysis was conducted using GPower 3.1 (Faul et al., 2007) to determine the power of the sample size used, with a one-tailed .05 criterion of statistical significance, and the proposed analyses. For Phase 1, our sample size (*n*=8) was based on recommended PEERS® group sizes of 7-10 (E. Laugeson & Frankel, 2010) to illustrate proof of concept. This group alone is underpowered to detect differences from pre-test to post-test (.13, .36 and .65 for small, medium and large effect sizes, respectively). Phase 2 is also underpowered to detect group differences between the treatment group and the waitlist control group for small, medium and large effect sizes with both a completer approach (TRT: *n*=6; WLC: *n*=6; Power = .09, .20 and .36) and an intent-to-treat approach (TRT: *n*=9; WLC: *n*=10; Power = .11, .27, and .51). Thus, there is not sufficient power to detect change at the specific group level. When data was combined across the groups to include all individuals who completed the intervention (*n*=19), the study continues to be underpowered to detect differences for small and medium effect sizes (.21 and .67) and adequately powered for large effect sizes (.96). When data was combined across the groups to include all participants who entered into the study (*N*=27), the study continues to be underpowered to detect differences for small effect sizes (.26) and adequately powered for medium and large effect sizes (.81 and .99). However, the goal for this pilot study is to illustrate feasibility, acceptability, and proof of concept.

To examine Research Aim 1a, paired samples t-tests were used to compare social performance on the five outcome measures (SSIS-SEL, SRS-2, FQS, QSQ, and TASSK) from pre-test to post-test. This analytic approach most parallels pilot studies published using the

PEERS® interventions (Gardner et al., 2015). Effect sizes (Hedges' g) were calculated due to the pilot nature of this study. Reliable change index scores were computed, according to Jacobson and Truax's (1991) formula, to determine whether the degree of change on the SSIS-SEL Parent Form, SSIS-SEL Student Form, and SRS-2 over time was statistically reliable.

For Research Aim 1b and 1c, analyses were run in two ways: (1) an intent-to-treat analysis with the last observation carried forward method to follow statistical best practices for intervention research (Sanders & Hunsley, 2018; Schulz et al., 2010), and (2) a completer analysis in order to compare the current findings to the PEERS® literature. For Research Aim 1b, independent samples t-tests were used in both analyses to compare social performance on the five outcome measures (SSIS-SEL, SRS-2, FQS, QSQ, and TASSK) of the treatment group and the waitlist control group at pre-test and post-test. For Research Aim 1c intent to treat analyses, all participants who enrolled in the study including the Open Trial (n=8), the Treatment Group from the Pilot RCT (n=9), and the Wait List Control Group from the RCT (n=10) were included. Wait-list control data from post-wait (i.e., pre-test) and post-test (i.e., post-intervention) were used when available (n=5). Research Aim 1c utilized paired samples t-tests to compare social performance on the five outcome measures (SSIS-SEL, SRS-2, FQS, QSQ, and TASSK) from pre-test to post-test. For both Research Aim 1b and 1c, effect sizes (Hedges g') were calculated due to the pilot nature of this study. Additionally, reliable change index scores were computed, according to Jacobson and Truax's (1991) formula, to determine whether the degree of change on the SSIS-SEL Parent Form, SSIS-SEL Student Form, and SRS-2 over time was statistically reliable at the individual participant level.

Treatment acceptability and feasibility in Research Aim 2 was assessed by examining descriptive statistics of the: 1) number of dropouts, 2) number of sessions completed prior to

treatment drop out, and 3) frequencies of caregiver ratings on the Treatment Acceptability Questionnaire (TAQ) and Treatment Evaluation Inventory (TEI-SF).

Reliable Change Calculations

To calculate reliable change, the following equations were used:

$$RCI = \frac{(x_2 - x_1)}{S_{diff}} \qquad S_{diff} = \sqrt{2(SE)^2} \qquad SE_M = SD\sqrt{1 - r_{xx}}$$

For calculating reliable change on SSIS-SEL Parent and Student forms, an improvement from pre-test to post-test was operationally defined using the standard error of measurement obtained from the measure's combined normative sample for the SEL composite score (Parent Form: SE_M = 3.00; Student Form: SE_M = 3.35; Gresham & Elliott, 2017). Importantly, the measure also provides the test–retest reliability data (r_{xx}) and the standard deviation (SD) of the sample, which can also be used to calculate SE_M. However, the calculated value for S_{diff} is different than the value provided by the measure. Ultimately, the SE_M provided by the measure to determine the appropriate reliable change index was used for analyses. For the SSIS-SEL Parent Form, the standard error of the difference between two administrations was calculated as $S_{diff} = \sqrt{2(3^2)}$, meaning that reliable change is defined as an increase of greater than 4.24. For the SSIS-SEL Student Form, the standard error of the difference between two administrations was calculated as $S_{diff} = \sqrt{2(3.35^2)}$, meaning that reliable change is defined as an increase of greater than 4.74.

For the SRS-2, the standard deviation is 10. The test–retest reliability coefficient was not provided in the measure manual and thus, a test–retest reliability coefficient (r_{xx}) of .8 was used which came from a sample that approximated the current sample and study procedures. (Constantino & Todd, 2003). Therefore, the standard error of measurement at any time point for the scale would be $SE_M = 10 \times (\sqrt{1} - .8)$, yielding 4.47. Thus, for the SRS-2, the standard error

of the differences between two administrations would be calculated as $S_{diff} = \sqrt{2(4.47^2)}$ and a reliable change is defined as an increase of greater than 6.32.

Results

The data were analyzed using IBM SPSS for Windows, version 28. A *p* value of < .05 indicated significance. Findings were interpreted with respect to both statistical significance and effect size. Interpretations of Hedges' g are as follows: negligible effect = 0 – .14; small effect = .15 – .39; medium effect = .40 – .74; large effect ≥ .75.

Treatment Effectiveness

Research Aim 1: Examine the effectiveness of a telehealth-delivered Program for the Education and Enrichment of Relational Skills (PEERS®) intervention for improving social skills and peer interactions

Aim 1a: Pilot open trial (Phase 1)

Participants. Demographic information and mean scores on outcome measures for participants in Phase 1 (Open Trial) are provided in Table 5. To characterize the sample, one-sample t-tests indicated that adolescents with NF1 had significantly lower social-emotional skills and significantly more social impairment using caregiver report compared to the normative mean with large effect sizes (SSIS-SEL Parent Form: $t(7) = -3.66$, $p = .004$, $g = -1.15$; SRS-2: $t(7) = 3.27$, $p = .007$, $g = 1.03$). Adolescent self-report of social-emotional skills was not significantly different from the normative mean (SSIS-SEL Student Form). With standard scores of <85 classified as a difficulty, difficulties were reported for three participants on the SSIS-SEL Parent Form and two participants on the SSIS-SEL Student Form at pre-test. Five participants were reported to have difficulty on the SRS-2 at pre-test with a T-score of > 60 as defined as difficulty.

Effectiveness of the Intervention. Caregiver-reported get-togethers (QSQ-Caregiver) and adolescent-reported social knowledge (TASSK) were significantly better at post-test compared to pre-test (Table 5) with a medium and large effect size, respectively. Social-emotional skills (SSIS-SEL Parent and Student Forms), quality of existing friendships (FQS), adolescent-reported get-togethers (QSQ-Adolescent) and social impairment (SRS-2) were not significantly different from pre-test to post-test based on either caregiver or adolescent report. Using a reliable change index (Jacobson & Truax, 1991), one adolescent with NF1 (12.5%) showed improvement from pre-test to post-test in caregiver-reported social-emotional skills (SSIS-SEL Parent Form) standard scores (standing relative to same-aged peers) and one participant showed worsening. Another adolescent with NF1 (12.5%) showed improvement based on self-reported social-emotional skills (SSIS-SEL Student Form). No participants showed reliable change in social impairment (SRS-2) based on caregiver-report.

Aim 1b: Pilot randomized controlled trial (Phase 2)

Intent to Treat Analyses

Participants. Demographic information and mean scores on outcome measures for participants in Phase 2 (pilot RCT) are provided in Table 6. Two participants who enrolled in the study began the pre-test appointment and were assigned to the WLC group but did not complete the SSIS-SEL Parent Form, SRS-2 and SSIS-SEL Student Form or attend the group. No group differences between the Treatment and Wait List Control groups were found in age, gender, presence of an ADHD diagnosis, ASD symptomatology or word reading skills. However, cognitive functioning of the treatment group was significantly lower than the waitlist control group ($t(17) = -2.57$, $p = .01$, $g = -1.13$).

To characterize the pilot RCT sample, one-sample t-tests indicated that adolescents with NF1 had significantly lower social-emotional skills and significantly more social impairment using caregiver report compared to the normative mean at pre-test with large effect sizes (SSIS-SEL Parent Form: $t(16) = -5.72$, $p < .001$, $g = -1.32$; SRS-2: $t(16) = 7.24$, $p < .001$, $g = 1.67$). Self-report of social-emotional skills was not significantly different from the normative mean at pre-test (SSIS-SEL Student Form). Difficulties were reported for seven participants (Treatment: $n = 3$; WLC: $n=4$) on the SSIS-SEL Parent Form and three participants (Treatment: $n = 2$; WLC: $n=1$) on the SSIS-SEL Student Form at pre-test with standard scores of <85 classified as a difficulty. Fourteen participants (Treatment: $n = 7$; WLC: $n=7$) were reported to have difficulty on the SRS-2 at pre-test with a T-score of > 60 defined as difficulty. There were no significant differences between difficulty for participants in the treatment and wait-list control group on the SSIS-SEL and SRS-2.

Comparison of Treatment and WLC group on Outcome Measures. The treatment and wait-list control groups were not significantly different at pre-test or post-test for social-emotional skills (SSIS-SEL Parent and Student Forms), quality of existing friendships (FQS), caregiver-reported get-togethers (QSQ-Caregiver), or social impairment (SRS-2). Adolescent-reported number of get-togethers (QSQ-Adolescent) and adolescent-reported social knowledge (TASSK) were not significantly different at pre-test, but wait-list control participants had significantly more get-togethers at post-test/post-wait while treatment participants reported increased social knowledge post-treatment with large effect sizes (Table 7).

Using a reliable change index for all pilot RCT participants (Jacobson & Truax, 1991), two adolescents (12%) with NF1 in the treatment group and two adolescents (12%) in the WLC group showed improvement from pre-test to post-test/post-wait in caregiver-reported social-

emotional skills (SSIS-SEL Parent Form) standard scores (standing relative to same-aged peers). Of note, two adolescents (12%) in the WLC group showed worsening during the wait period using reliable change on the SSIS-SEL Parent Form. Ratings of self-reported social-emotional skills (SSIS-SEL Student Form) evidenced one participant (6%) improved in the treatment group and one participant (6%) improved in the WLC group. Additionally, one participant (6%) in the treatment group showed improvement in social impairment (SRS-2) and one participant (6%) in the WLC group showed worsening.

Completer Analyses

Participants. Demographic information and mean scores on outcome measures for participants who completed the pilot RCT are provided in Table 6. To characterize the pilot RCT completer sample, one-sample t-tests indicated that adolescents with NF1 (n=12) had significantly lower social-emotional skills and significantly more social impairment using caregiver report compared to the normative mean with large effect sizes (SSIS-SEL Parent Form: $t(11) = -4.31$, $p < .001$, $g = -1.16$; SRS-2: $t(10) = 7.08$, $p < .001$, $g = 1.97$). Self-report of social-emotional skills was not significantly different from the normative mean (SSIS-SEL Student Form). With standard scores of <85 classified as a difficulty and ≥85 as no difficulty, difficulties were reported for four participants on the SSIS-SEL Parent Form and two participants on the SSIS-SEL Student Form at pre-test. Ten participants were reported to have difficulty on the SRS-2 at pre-test with a T-score of > 60 defined as difficulty. No group differences between the Treatment and Wait List Control groups were found in age, gender, presence of an ADHD diagnosis, ASD symptomatology or word reading skills. However, cognitive functioning of the treatment group was significantly lower than the waitlist control group ($t(10) = -3.34$, $p = .003$, $g = -1.81$).

Comparison of Treatment and WLC group on Outcome Measures. Social-emotional skills (SSIS-SEL Parent and Student Forms), quality of existing friendships (FQS), number of get-togethers (QSQ), and social impairment (SRS-2) using caregiver and adolescent report were not significantly different at pre-test or post-test. Adolescent-reported social knowledge (TASSK) scores were not significantly different at pre-test but were significantly better post-treatment in the treatment group compared to the wait-list control group with a large effect size (Table 7).

Using a reliable change index with participants who completed the pilot RCT (Jacobson & Truax, 1991), two adolescents (33%) with NF1 in the treatment group and one participant with NF1 in the wait-list control group (17%) showed improvement from pre-test to post-test in caregiver-reported social-emotional skills (SSIS-SEL Parent Form) standard scores (standing relative to same-aged peers). In the wait-list control group from pre-test to post-wait, one participant (17%) showed worsening during the wait period in caregiver-reported social-emotional skills (SSIS-SEL Parent Form) standard scores. One participant (17%) from the treatment group (who also showed improvement on the SSIS-SEL Parent Form) and one WLC participant (17%) showed improvement based on self-reported social-emotional skills (SSIS-SEL Student Form). One treatment participant (17%) showed improvement in social impairment (SRS-2) while no wait-list control participants showed reliable changes in social impairment.

Aim 1c: Across all participants who were enrolled in the study.

Intent to Treat:

Participants. Characterization of the sample and mean scores on outcome measures for participants are provided in Table 8. Two participants who enrolled in the study began the pre-test appointment but did not complete the SSIS-SEL Parent Form, SRS-2 and SSIS-SEL Student Form or attend the group. There were no significant differences in age, cognitive functioning,

word reading abilities, presence of ADHD diagnosis, ASD symptomology, or on the primary outcome measures at pre-test between those who completed the intervention and those who did not. One-sample t-tests indicated that adolescents with NF1 had significantly lower overall social-emotional skills and significantly more social impairment than same-aged peers using caregiver report compared to the normative mean with large effect sizes (SSIS-SEL Parent Form: $t(24) = -7.78$, $p < .001$, $g = -1.51$; SRS-2: $t(24) = 8.21$, $p < .001$, $g = 1.59$). Self-report of overall social-emotional skills (SSIS-SEL Student Form) was not significantly different from the normative mean. With standard scores of <85 classified as a difficulty and ≥85 as no difficulty, difficulties were reported for 12 participants on the SSIS-SEL Parent Form and 6 participants on the SSIS-SEL Student Form at pre-test. Nineteen participants were reported to have difficulty on the SRS-2 at pre-test with a T-score of > 60 defined as difficulty.

Effectiveness of the Intervention. Caregiver-reported social-emotional skills (SSIS-SEL Parent Form), social impairment (SRS-2), and caregiver-reported number of adolescent get-togethers (QSQ-Caregiver) were significantly better at post-test compared to pre-test with medium effect sizes (Table 8). Social knowledge (TASSK) was also significantly better at post-test with a large effect size. Adolescent self-reported social-emotional skills (SSIS-SEL Student Form), quality of existing friendships (FQS), and adolescent-reported number of get-togethers (QSQ-Adolescent) were not significantly different from pre-test to post-test.

Seven adolescents (37% of intervention completers; 26% of sample) who received the intervention showed improvement following the intervention in caregiver-reported social-emotional skills (SSIS-SEL Parent Form) standard scores (standing relative to same-aged peers) using a reliable change index (Jacobson & Truax, 1991). Three adolescents (16% of intervention completers; 11% of sample) showed improvement in social impairment (SRS-2). Three

adolescents (16% of intervention completers; 11% of sample) showed improvement based on self-reported social-emotional skills (SSIS-SEL Student Form).

Additional exploratory analyses were conducted to examine possible effects at the subscale level on two of the outcome measures (SSIS-SEL and SRS-2) to better determine which scales may be more apt to demonstrate change (Table 9). Subscale analysis is consistent with previously published literature using PEERS® (Hill et al., 2017; Laugeson et al., 2012). One-sample t-tests were conducted with the SSIS-SEL Parent Form, SSIS-SEL Student Form, and SRS-2 subscales. Mean ratings on SSIS-SEL Parent Form subscales were significantly lower compared to the normative mean across all subscales with medium to large effect sizes (SSIS-SEL Parent Form SeA: $t(24) = -6.95$, $p < .001$, $g = -1.35$; SM: $t(24) = -4.69$, $p < .001$, $g = -.91$; SA: $t(24) = -2.26$, $p = .017$, $g = -.44$; RS: $t(24) = -6.39$, $p < .001$, $g = -1.24$; RDM: $t(24) = -2.46$, $p = .011$, $g = -.48$). On the SSIS-SEL Parent Form, ratings on the subscales of Self-Management and Relational Skills were significantly improved at post-test with medium effect sizes. Caregiver ratings on the SRS-2 subscales were significantly higher compared to normative data with large effect sizes (SAw: $t(24) = 5.33$, $p < .001$, $g = 1.03$; SCog: $t(24) = 5.66$, $p < .001$, $g = 1.09$; SCom: $t(24) = 8.10$, $p < .001$, $g = 1.57$; SMot: $t(24) = 4.41$, $p < .001$, $g = .85$; SCI: $t(24) = 7.46$, $p < .001$, $g = 1.44$). All subscales on the SRS-2 were significantly better at post-test with small to medium effect sizes. The Relational Skills subscale on the SSIS-SEL Student Form was significantly lower compared to the normative mean with a medium effect size (RS: $t(24) = -2.37$, $p = .013$, $g = -.46$). All other subscales on the SSIS-SEL Student form were not significantly different from normative data. The Self-Management subscale on the SSIS-SEL Student Form was significantly better at post-test with a small effect size while all other SSIS-SEL Student subscales were not significantly different from pre- to post-test.

Completer Analysis

Participants. Nineteen participants completed the intervention in Phase 1 and Phase 2 (See Table 8 for demographic and additional characterization of the sample). One-sample t-tests indicated that adolescents with NF1 had significantly lower overall social-emotional skills and significantly more social impairment using caregiver report compared to the normative mean with large effect sizes (SSIS-SEL Parent Form: $t(18) = -6.23$, $p < .001$, $g = -1.37$; SRS-2: $t(18) = 7.26$, $p < .001$, $g = 1.59$). Self-report of overall social-emotional skills was not significantly different from the normative mean. With standard scores of <85 classified as a difficulty and ≥85 as no difficulty, difficulties were reported for seven participants on the SSIS-SEL Parent Form and four participants on the SSIS-SEL Student Form at pre-test. Fourteen participants were reported to have difficulty on the SRS-2 at pre-test with a T-score of > 60 defined as difficulty.

Effectiveness of the Intervention. With participants who completed the intervention, caregiver-reported social-emotional skills (SSIS-SEL Parent Form) and social impairment (SRS-2) were significantly better at post-test compared to pre-test with medium effect sizes (Table 8). Caregiver-reported number of adolescent get-togethers (QSQ-Caregiver) and social knowledge (TASSK) were also significantly better at post-test compared to pre-test with large effect sizes. Self-reported social-emotional skills (SSIS-SEL Student Form), quality of existing friendships (FQS), and adolescent-reported number of get-togethers (QSQ-Adolescent) were not significantly different from pre-test to post-test.

Of those that completed the intervention (n=19), seven adolescents (37%) who received the intervention showed improvement following the intervention in caregiver-reported social-emotional skills (SSIS-SEL Parent Form) standard scores (standing relative to same-aged peers) using a reliable change index (Jacobson & Truax, 1991). Three adolescents (16%) showed

improvement in social impairment (SRS-2). Three adolescents (16%) showed improvement based on self-reported social-emotional skills (SSIS-SEL Student Form).

Mean ratings on the SSIS-SEL Parent Form subscales were significantly lower compared to the normative mean across all subscales with medium to large effect sizes (SSIS-SEL Parent Form SeA: $t(18) = -6.39$, $p < .001$, $g = -1.40$; SM: $t(18) = -4.44$, $p < .001$, $g = -.98$; SA: $t(18) = -2.31$, $p = .016$, $g = -.51$; RS: $t(18) = -5.19$, $p < .001$, $g = -1.14$; RDM: $t(18) = -2.28$, $p = .018$, $g = -.50$). On the SSIS-SEL Parent Form, ratings on the subscales of Self-Management and Relational Skills were significantly improved at post-test with medium effect sizes (Table 9). Caregiver ratings on the SRS-2 subscales were significantly higher compared to normative data with large effect sizes (SAw: $t(18) = 3.94$, $p < .001$, $g = .87$; SCog: $t(18) = 5.63$, $p < .001$, $g = 1.24$; SCom: $t(18) = 8.26$, $p < .001$, $g = 1.81$; SMot: $t(18) = 3.83$, $p < .001$, $g = .84$; SCI: $t(18) = 7.18$, $p < .001$, $g = 1.58$). All subscales on the SRS-2 were significantly better at post-test with medium effect sizes (Table 9). The Relational Skills subscale on the SSIS-SEL Student Form was significantly lower compared to the normative mean with a small effect size (RS: $t(18) = -1.76$, $p = .048$, $g = -.39$). All other subscales on the SSIS-SEL Student form were not significantly different from normative data. The Self-Management subscale on the SSIS-SEL Student Form was significantly better at post-test with a medium effect size while all other SSIS-SEL Student subscales were not significantly different from pre- to post-test (Table 9).

Treatment Acceptability

Research Aim 2: Determine the acceptability and feasibility of the PEERS® intervention with adolescents with NF1 using caregiver report.

In Phase 1, all eight adolescents who entered into the group completed the group and each measurement timepoint. In Phase 2, two adolescents who entered the Treatment group

dropped out (one after 2 sessions and one after 7 sessions). During the waiting period for our WLC group, three adolescents dropped out such that they did not complete the post-wait assessment or attend the group. One WLC participant completed the post-wait assessment but did not attend the group. Of the WLC participants who began the group, one participant began the group and dropped out after five sessions. Within the pilot RCT, there was no significant difference between drop-out rate for the treatment and WLC group. All families that dropped out were asked to participate in a post-test assessment and declined. Thus, across all participants who enrolled in the study (including those that dropped out during the wait period), 30% of the sample dropped out. When looking at just participants who began the group intervention (i.e., attending at least one session), 14% of participants dropped out.

For participants who completed the intervention and the post-test appointment (*n*=19), caregiver-reported ratings on the Treatment Acceptability Questionnaire (TAQ) and Treatment Evaluation Inventory-Short Form (TEI-SF) are presented in Figures 4 and 5. On the TAQ, all caregivers (100%) reported that the PEERS® treatment was ethical, the PEERS® treatment provider was both knowledgeable and trustworthy, and any side effects of the intervention were acceptable. Additionally, the majority of caregivers indicated on the TAQ that the PEERS® treatment was acceptable overall (89%) and effective (79%). On the TEI-SF, 89.4% of caregivers indicated they had a positive reaction to the PEERS® treatment. Specifically, the majority of caregivers agreed or strongly agreed that the treatment is an acceptable way to deal with their child's behavior (95%) and is likely to result in permanent improvement (74%). Additionally, 95% of caregivers also reported they agreed or strongly agreed that they are willing to use the treatment to change their child's behavior and liked the procedures used in the treatment. Many

caregivers disagreed or strongly disagreed that the child will experience discomfort during the treatment.

Discussion

Although social difficulties are commonly reported for adolescents with NF1, there is no current literature examining the effect of specific social skills intervention strategies on social outcomes within this population. Additionally, telehealth intervention strategies are becoming increasingly researched and have shown initial promise for individuals with NF1. The current study is the first to explore the usefulness of a telehealth social skills intervention with adolescents with NF1 and their caregivers. The primary research aim was to conduct a pilot study to examine the effectiveness of a 14-week telehealth PEERS® intervention for adolescents with NF1 for improving social skills and peer interactions. This pilot study included two phases: an open-trial and a pilot randomized controlled trial. Analyses were conducted within each phase as well as across all participants in the pilot study as a whole. Consistent with the approach of prior research, the effect of the intervention on mean group ratings for a variety of indicators of social and friendship skills were examined. In addition, the number of participants with reliable change on the norm-referenced outcome measures was explored. The secondary aim of this investigation was to determine the acceptability and feasibility of this 14-week telehealth PEERS® intervention for adolescents with NF1 using caregiver report. Group-level improvements in caregiver-reported social-emotional skills, social impairment, caregiver-reported number of adolescents get-togethers, and adolescent's social knowledge were indicated at post-test when the intervention groups were combined to allow for additional power. The intervention was also found to be feasible and acceptable based on caregiver-report and low drop-out rates.

The remainder of the Discussion is organized as follows: The central study findings and lessons learned are highlighted. Next, the findings from this pilot study are integrated into the current literature by first describing the level of social difficulties of the current sample and then, the use of self-report measures in NF1 and in the PEERS® literature. Subsequently, the importance of the analytic approach used in this investigation is discussed. Following, the novelty and significance of this telehealth investigation is highlighted. Lastly, the limitations and future directions for this research endeavor are described.

Central Study Findings

This investigation is the first to examine a specific social skills intervention, PEERS®, for use in adolescents with NF1 with social challenges. More specifically, this research simultaneously utilized an intervention that had not previously been used in NF1 and explored the use of telehealth delivery among a population with minimal telehealth focused research. When the primary outcomes measures were examined at a smaller group level (i.e., participants from Phase 1 and then separately, participants from Phase 2 only), there was not sufficient power to see effects from the intervention due to the small sample sizes in these groups. However, while Phases 1 and 2 were significantly underpowered, these phases of this research endeavor demonstrated this research team's ability to design, recruit, and conduct this virtual, social skills group intervention using both an open trial and a pilot randomized controlled trial approach. These phases of the research also highlighted a necessary discussion regarding administering social functioning measures that are most relevant to this population. Ratings on norm referenced measures were in the broadly average range and only a small number of the participants showed reliable changes in social-emotional skills and social impairment over time on the measures used; hence the social difficulties within this sample are likely subtle and variable (consistent

with previous research (Glad et al., 2021)), and improvement related to these difficulties may have been difficult to capture.

When the data were analyzed with sufficient power, by combining data from all participants who enrolled in the study and engaged with the intervention, group-level improvements following the intervention were indeed found; caregiver-reported social-emotional skills, social impairment, caregiver-reported number of adolescents get-togethers, and adolescent's social knowledge showed improvement following the intervention, partially supporting the stated hypotheses and consistent with the broad aim of this research. These findings provide evidence that this intervention is likely to be effective and helpful within this population, although further research with a control group would be needed to more definitively demonstrate the effectiveness. Additionally, as hypothesized, caregiver treatment ratings indicated a high degree of acceptability and feasibility for telehealth-delivered PEERS®. The relatively low dropout rates observed within this investigation further highlights the acceptability and feasibility for use within a sample of adolescents with NF1 and their caregivers.

Level of Social Difficulties in the Current Sample

While difficulties with social functioning in young children, school-age children, and adults with NF1 have been found (Allen et al., 2016; Barton & North, 2004; Cipolletta et al., 2018; Glad et al., 2021; Huijbregts & de Sonneville, 2011; Pierpont et al., 2018), previous research has suggested that social difficulties are likely to be subtle and variable (Glad et al., 2021). The current investigation is consistent with this previous finding from the literature as social challenges were somewhat difficult to detect on norm-referenced outcome measures. Whereas 48% of adolescents in this sample had difficulty with social-emotional skills based on caregiver-report, similar to other published percentages of deficits in social functioning in NF1

with larger age ranges (Barton & North, 2004; Pierpont et al., 2018), the majority of individuals with NF1 in this sample do not have social challenges as assessed by norm-referenced measures. It is notable that to meet criteria to participate in the group, families indicate a desire to improve their adolescent's social relationships and a concern regarding their social functioning. The intake process is structured around determining what specific social challenges the adolescent has based on both caregiver and adolescent report. So, even though participants with NF1 were recruited based on perceived social difficulties and deemed eligible to participate only if they indicated social challenges, the majority of participants within the current sample have scores reported to be in the broadly average range using a gold standard recommended measurement of social functioning that has been suggested for use in NF1 (Janusz et al., 2021) and that has been frequently used in the PEERS® literature. Thus, the level of social functioning in NF1 may be more difficult to capture on norm-referenced questionnaire measures. While the researchers could have excluded participants who had broadly average social skills based on norm-referenced measures, this would not be consistent with PEERS exclusion criteria and screening processes. Furthermore, there were several participants who were excluded due to social challenges that were not substantial enough for inclusion in the group based on caregiver and/or adolescent report. This highlights again the significant subtleness and variability of social difficulties within the NF1 sample given that these families nevertheless considered participation in a social skills intervention for social challenges.

Since subtle social difficulties may be likely in NF1, it is important to note that some research among autistic individuals has suggested that individuals with milder social difficulties experienced more bullying and victimization compared to more socially impaired individuals (Rowley et al., 2012). This could be due to individuals with milder social difficulties being more

likely to engage socially and therefore, have a higher likelihood of experiencing negative social interactions whereas individuals with more significant social difficulties may avoid social settings and interactions to minimize the risk of bullying (Rowley et al., 2012; Wainscot et al., 2008). Thus, it remains possible that the subtle social challenges evident in NF1 may be associated with more negative social interactions such as bullying. However, research on bullying in NF1 has had modest exploration even though bullying is often reported among children and adolescents with ASD (Schroeder et al., 2014) and separately, with ADHD (Gardner & Gerdes, 2015; Taylor et al., 2010) and NF1 has shown overlap with each of these diagnoses. Holland and colleagues (2019) found that 62% of their sample of school-age children with NF1 reported they had been bullied at least once in the past year and 20% of the sample reported they had been bullied daily, with rates higher than the general population (Berger, 2007). In a more recent investigation that expands on the sample used in Holland and researchers (2019), risk factors for bullying victimization were explored with findings indicated that greater ADHD symptoms were associated with greater bullying victimization in NF1 (Stavinoha et al., 2021). While bullying was not an outcome explored within this investigation, results from the aforementioned research findings and the focus of bullying among the PEERS® sessions signifies that it may be an important outcome to explore within social functioning research.

Additionally, as mentioned, while improvement was evident on composite scores in several outcome measures following the intervention, mean scores on norm-referenced outcome measures often fell broadly within one standard deviation of the normative mean. This highlighted an important exploratory aim to determine whether certain subscales on the standardized outcome measures would be more apt to capture change from the intervention and further understand how to best evaluate the effectiveness of the intervention. The SSIS-SEL was

administered within this investigation to assess social-emotional skills of the adolescents and the subscales provide information about the specific social-emotional skills that are measured. An older version of the SSIS-SEL, the Social Skills Rating System (SSRS), has inconsistent findings at the subscale level for children with NF1. Loitfelder and colleagues (2015) found that the SSRS subscales of Cooperation, Assertion, Responsibility, and Self-Control were significantly worse for children with NF1 compared to healthy controls using parent-report. In contrast, Barton and North (2004) did not find significant group differences on SSRS subscales. The SSIS-SEL is the most recent update to the SSRS and SSIS with an effort to fit the measure to the Collaborative on Academic Social Emotional Learning (CASEL) framework. While the number of subscales and the subscale names on the SSIS-SEL have evolved from predecessors, the social skills items on this measure have remained consistent and the social skills composite scores are highly correlated (r = .97) (Gresham et al., 2018; Gresham & Elliott, 2017). This pilot study showed all SSIS-SEL subscales were rated by caregivers to be significantly poorer than the normative mean on the SSIS-SEL Parent Form. Similarly, all SSIS-SEL subscales showed improvement following the intervention using caregiver-report. PEERS® research with autistic adolescents has shown inconsistent findings at the subscale level using older versions of the SSIS-SEL. Hill and colleagues (2017) did not find significant differences at the subscale level, however, the sample size was relatively small with only five adolescents. On the other hand, Laugeson and colleagues (2012) found the subscales of Cooperation, Assertation, Responsibility, and Self-Control to be significantly improved at post-test and follow up.

Furthermore, subscale level explorations are available for the Social Responsiveness System (SRS), the first version of the SRS-2, in children with NF1. Within the current investigation, the SRS-2 was used to measure the level of social impairment for each adolescent

using caregiver-report. The SRS-2 focuses on aspects of social impairment that are commonly associated with ASD symptomatology (Constantino & Gruber, 2012). The current sample showed significantly worse ratings on all of the SRS-2 social subscales compared to the normative mean, which is consistent with previous literature that found the subscales of Social Awareness, Social Cognition, Reciprocal Social Communication, and Social Motivation were significantly worse for children with NF1 compared to healthy controls (Loitfelder et al., 2015). The SRS-2 Social Cognition and Social Communication subscales were significantly better following the intervention. Previous research with ASD samples has shown similar findings with improvements evident in some capacity on all SRS-2 social subscales such as at post-test in Social Cognition, Social Communication, and Social Motivation (Hill et al., 2017; Laugeson et al., 2012) as well as Social Awareness at follow-up (Laugeson et al., 2012). These exploratory findings suggest that the overall scores on the SSIS-SEL Parent Form and SRS-2 are likely to adequately capture change that may result from this investigation. Nevertheless, given the preliminary nature of this investigation and the subtleness of social difficulties in NF1, continued inclusion and exploration at the subscale level is warranted to order to ascertain if particular subscales are in fact more relevant to the PEERS® intervention within this population. This may also include examining reliable change at the subscale level to determine whether a differential pattern is evident.

Self-report of Social Functioning

Interestingly, throughout this study, improvement following the intervention was not found on self-reported outcome measures, with the exception of improvements in social knowledge. Hence, while there is evidence that adolescents did appear to gain social knowledge based on the PEERS® curriculum, they did not report significant changes in their social-

emotional skills, their quality of existing friendships, or their number of get-togethers. However, it is worth noting that while there was not a significant change on these measures, self-reported quality of existing friendships and number of get-togethers demonstrated change in the positive direction following the intervention, with negligible and small effect sizes respectively. With additional power, it is possible that these outcome measures would show an effect. Additionally, self-report ratings of social functioning on norm-referenced measures were in the solidly average range at pretest, even though a self-reported desire to improve social functioning is required to enroll in the research study. Scores in this range suggest that there may be little room to improve this score following the intervention. However, as a part of the intake process to participate in the intervention, an interview is conducted with the adolescent to gain information about the social challenges they feel they face and determine how the intervention may benefit them. For instance, adolescents are asked to report on their social relationships, what they do with other teenagers their age (if applicable), and get-togethers including how often and with whom. Challenges were often reported by adolescents in that they have no or few social relationships or get-togethers with other teenagers their age. Thus, it appears that while these challenges are not captured on self-reported standardized measures, the challenges are captured during an interview and are at a level that warrants participation in the PEERS® intervention. This points to an important direction to determine how to best capture adolescent's experience with social skills and identify more sensitive or targeted measures to highlight their social experiences.

Notably, self-report measures have shown variability in their change following the PEERS® intervention throughout the literature. Using the Quality of Socialization Questionnaire (QSQ), or an examination of get-togethers, several studies have found improvements using self-report in ASD (Hill et al., 2017; Laugeson et al., 2009, 2012, 2015; Mandelberg et al., 2014;

Schohl et al., 2014). Hosted get-togethers also significantly increased based on self-report following the intervention in ADHD (Gardner et al., 2015). However, self-reported get-togethers were not significantly improved following the intervention among a sample of individuals with TBI (Gilmore et al., 2022). Another self-report measure occasionally used in the PEERS® literature is the Friendship Qualities Scale (FQS), whose findings are highly variable. For instance, one study with an autistic sample (Laugeson et al., 2009) found a significant improvement while another study did not find change (Schohl et al., 2014). Additionally, the FQS was not significantly improved in a pilot study with adolescents with ADHD (Gardner et al., 2015). In the majority of PEERS® research with autistic samples, self-report of social skills or social-emotional skills is not utilized (Hill et al., 2017; Laugeson et al., 2009, 2012; Mandelberg et al., 2014; Schohl et al., 2014). One study that utilized self-report of social skills found the Assertion subscale to be significant immediately following the intervention (Marchica & D'Amico, 2016). Self-reported social skills were not significantly different for individuals with TBI following the PEERS® intervention (Gilmore et al., 2022). The current study findings related to the number of get-togethers (QSQ-Adolescent) are surprising given previous literature has demonstrated improvement. However, the findings related to the quality of friendships (FQS) and social-emotional skills (SSIS-SEL Student Form) are relatively consistent with previous PEERS® studies.

Correspondence between Caregiver and Adolescent Self-Report

On measures with both caregiver and self-report, findings were inconsistent across respondents, which is largely consistent with prior research that found that self-report of social functioning in children with NF1 generally indicates less difficulty than caregiver report. In this study, self-reported social-emotional skills were not significantly different from the normative

mean. This finding is consistent with previous research that has utilized self-report of social functioning (Barton & North, 2004; Noll et al., 2007). In fact, self-report has often showed that children and adolescents generally rate themselves as having good social skills with significantly better skills than the normative mean (Barton & North, 2004). Additionally, this highlights that adolescents may be less likely to report difficulties in social functioning compared to caregivers. It also remains possible adolescents with NF1 have poor awareness of their social difficulties as previous research has shown that parents indicate greater difficulty with social skills than those indicated using self-report (Barton & North, 2004; Noll et al., 2007). However, in contrast to this idea, literature on bullying victimization in NF1 evidenced child-reported rate and frequency of bullying was significantly higher than parent-report (Holland et al., 2019). Thus, while caregiver report of social skills provides valuable information, caregiver's perception of skills may be different than the adolescent's social experience themselves. Self-report of skills appears necessary to better understand how the adolescent may view their skills and capture their experience. Regardless, both caregiver and self-report may not equate to skills or building friendships in the real-world. Peer nominations may be a more useful measure of how applicable this intervention is for the adolescent's social relationships (Noll et al., 2007), although this method is challenging in rare populations.

Analysis Consideration: Intent to Treat and Completer Analysis

Within the current investigation, results are provided using both the intent to treat (ITT) and completer analytic approaches. In the PEERS® literature, completer analyses (i.e., data only from participants who completed the intervention) are often used even when drop out is present (Gardner et al., 2015; Laugeson et al., 2009, 2015). However, in intervention research more broadly, ITT approaches are often recommended as can be seen as part of the CONSORT

guidelines (Schulz et al., 2010). There are benefits and critiques for both of these approaches (Sanders & Hunsley, 2018). The completer analysis may overestimate the efficacy of an intervention if those who are not benefitting from the intervention drop out. The ITT approach has been suggested to reduces bias as the ITT conserves group randomization (i.e., when using an RCT). However, while the ITT approach is likely to underestimate treatment differences in comparison to the completer analysis, the ITT approach requires a decision for handling missing data. This is often done by statistical imputation (e.g., a complex regression) or the last observation carried forward approach (Sanders & Hunsley, 2018). Given the small sample size of the current investigation, the last observation carried forward (LOCF) approach was utilized as complex statistical analyses are unreasonable. However, it is important to note that results of the LOCF approach are likely to be biased (Lachin, 2016; Sanders & Hunsley, 2018). For instance, an assumption of the LOCF approach is that participants that drop out show no change (i.e., drop out participants did not get worse and they did not get better), which is highly unlikely to be the case in the real-world. Importantly, in the current investigation when comparing the results of the completer and ITT analyses among RCT participants and separately with all participants, statistical significance and effect sizes were generally similar for all group-level analyses.

Use of Telehealth in NF1

In the last several years, and particularly as a result of the COVID-19 pandemic, telehealth (i.e., telemedicine, telepractice, virtual videoconferencing) has become increasingly utilized in patient care settings, both medical and psychological (Hollander & Carr, 2020; Lopez et al., 2020; Lurie & Carr, 2018), and among children and adolescents (Orsolini et al., 2021). Telehealth delivered services provide several benefits such as increased access to services,

convenience and flexibility of this modality, cost-effectiveness, and higher quality care and life (Jennett et al., 2003; Pruitt et al., 2014). A systematic review of videoconferencing psychotherapy yielded generally positive results including good user satisfaction, and comparable outcomes to in-person psychotherapy (Backhaus et al., 2012). Similarly, using case studies, Pakyurek and colleagues (2010) suggested that telehealth may be preferred method of delivery for individuals with ASD and ADHD. However, critiques of telehealth from provider's perspectives have included concerns for maintaining relationships, security breaches and confidentiality, audio and video difficulties, legal issues, reimbursements and administrative burden (Gajarawala & Pelkowski, 2021; Perry et al., 2020).

Among rare populations such as NF1, telehealth is ideal to provide accessible care and resources to families. Merker and colleagues (2018) reported that NF patients have to travel a significant distance to attend a certified NF specialty care clinic and pointed to telemedicine as a means to increase outreach and accessible care to individuals with NF who live in areas with less access to certified NF clinics. Additionally, the COVID-19 pandemic has necessitated the pivot to telehealth, which has increasingly been utilized to provide NF related care with an overall positive impression from health care workers delivering telehealth care (Radtke et al., 2021). However, only 21.4% of a sample of adults with NF1 used telehealth to meet with their NF providers with half of those respondents feeling that telehealth met their needs (Wolters et al., 2022).

Telehealth research thus far within ASD suggest comparable findings with telehealth and in-person delivered PEERS® (Adler et al., 2022; Estabillo et al. 2021; Miyake et al., 2018), suggesting the telehealth delivered PEERS® is likely to be acceptable and feasible in this population but a specific exploration is not yet available. Additionally, a hybrid-delivered

PEERS® intervention in young women with Turner syndrome has shown high acceptability, high adherence to the program, and a high report of helpfulness (Wolstencroft et al., 2019). On the other hand, research on telehealth delivered interventions is still relatively novel within the NF population. As discussed, a virtual mind-body intervention has been researched over the past several years highlighting the feasibility of a virtual intervention within the NF1 population (Lester et al., 2020; Vranceanu et al., 2016) and demonstrating promising results such as improved quality of life. Continued exploration of this virtual intervention using a single-blind randomized controlled trial is underway (Reichman et al., 2020). The findings from these studies support the delivery of group therapy via telehealth in this population. Further, the current investigation demonstrates the acceptability and feasibility of utilizing a group telehealth intervention strategy among a population with minimal virtual or telehealth research. Findings in this area are two-fold. First, high acceptability and feasibility of the intervention were evident based on caregiver report and adherence to the group (i.e., low dropout rate). The dropout rate in the current study is less than or comparable to other research on in-person delivered PEERS® when reported (Laugeson et al., 2009, 2015), and in child and adolescent intervention literature broadly, which has been estimated to be on average 26-45% of study samples (de Haan et al., 2013). Second, both the effectiveness and acceptability results suggest that telehealth shows potential for usefulness within this population.

Impact of COVID-19 Pandemic

This pilot research was conducted during the ongoing COVID-19 pandemic. While the COVID-19 pandemic presented a unique opportunity to explore a virtual social skills intervention among this population, the pandemic could have resulted in differential impact for individual participants as well as across the study as a whole. Specifically, the fluctuating total

and new cases of COVID-19 per day, varying federal and state requirements, and dissemination of a COVID-19 vaccination likely resulted in changes to how often participants engaged in social interactions (i.e., some families may have participated more in social distancing while others participated in-person activities). Additionally, it is likely that several families experienced common stressors of the COVID-19 pandemic such as changes in caregiver employment or finances as well as exposures to and contracting COVID-19 during the group. These differential experiences would be most likely to result in variable responses on the QSQ, or the measure of get-togethers, within this investigation although additional exploration is needed. However, the relatively low dropout rates experienced amidst the COVID-19 pandemic are rather remarkable and may further highlight the acceptability of this intervention for families with NF1. An additional consideration for conducting this research during the COVID-19 pandemic is related telehealth modifications. As a part of the group, families were asked to enroll their adolescent in an extracurricular activity as well as arrange get-togethers that occurred both with members of the group as well as outside of the group with family and friends. Notably, given the pandemic, these activities were allowed to occur virtually. Several families had difficulty finding virtual extracurriculars and thus, this research team deemed it appropriate to research potential online options for each participant that occurred locally or nationally. Similarly, with virtual get-togethers, families were often provided with suggestions for videoconferencing platforms as well as online games or activities that could be conducted virtually. While this level of problem-solving may not be present in the in-person PEERS protocol, it appeared crucial to the telehealth-delivered PEERS in order for participants to be able to participate in activities that most emulate the in-person version of this intervention. Lastly, this telehealth-delivered intervention provided an opportunity for families with NF1 to interact and experience this intervention together. The

rarity of NF1 makes it unlikely for families to know other families with NF1 and thus, it may be the case that this intervention provides a unique opportunity for families with NF1 to meet each other when they otherwise would not have but additional exploration of this notion is required.

Limitations and Future Directions

Several limitations are present that are central to the study design. First, while the sample size of the current pilot study is similar to other studies using PEERS® (Gardner et al., 2015; Laugeson et al., 2009, 2012, 2015), the sample size is still considerably small, resulting in underpowered analyses. This sample also lacks representation and diversity. For instance, the sample is primarily white and there are no Latinx participants which limits the generalizability of these findings to additional racial and ethnic backgrounds. There are also very few participants with a familial NF etiology which is likely not representative of the NF population as a whole. Second, for determining reliable change in this study, there do not exist test-retest reliability data for the SSIS-SEL and SRS-2 among a sample that is similar to this sample of adolescents with NF1 in age and in rate of social difficulties. Thus, it is possible that the reliable change estimates may under- or over-estimate the reliable change seen following the intervention depending on the standard samples which are usually based upon the U.S. Census (Constantino & Gruber, 2012; Gresham & Elliott, 2017). Last, given the pilot nature of this work and the desire to demonstrate feasibility of conducting this intervention, fidelity checks of the intervention were not performed at this stage in the research and serves as an important future direction in this research endeavor.

Not only is the present investigation the first to demonstrate evidence for the effectiveness, feasibility, and acceptability of a telehealth social skills intervention in adolescents with NF1 with social difficulties, this research has pointed to several future directions. On a

larger scale, broadening and replication of this study is necessary as is a sufficiently powered randomized controlled trial to be able to determine the efficacy of the intervention. A larger sample size is required to appropriately capture change from this intervention, control for drop out and missing data using sophisticated statistical techniques, as well as provide more power to detect differences in self-report measures. Similarly, while participant drop out was relatively low, it was clear over the course of this study that additional steps may be warranted to plan for intent-to-treat analyses. For instance, an interim assessment of skills such as mid-way through the intervention as well as clear procedures to encourage dropouts to complete an additional assessment may be helpful. Along the same lines, a multisite collaboration would be a beneficial next step for several reasons: to demonstrate feasibility of other research teams conducting the study procedures; to allow researcher blinding during outcome measure administration; and to serve as a comparison group to the current sample. Lastly, since the current investigation used caregiver and adolescent report, expanding this research to include rating adolescent behavior and skills using a behavior rating system during the group could provide additional information. The virtual delivery of this intervention lends itself well to this future direction as ratings could be collected during the behavioral rehearsal with adolescents and serve as an additional outcome variable to explore the immediate in-session change as well as over the course of the intervention.

More proximal future directions are also evident based on the current investigation. Exploration of follow-up data for participants is warranted for two primary reasons: to determine whether the social improvements evident following the intervention are maintained over time and to examine whether the outcome measures used may be tapping skills that are not immediately present following the intervention but emerge over time (e.g., developing quality

friendships, changes in adolescent's perceptions of their skills). Additionally, where available, investigations have evidenced that several improvements following the PEERS® intervention are likely to be maintained at a follow up assessment (e.g., 14 weeks after the intervention) (Gilmore et al., 2022; Laugeson et al., 2012, 2015). Examining follow-up data will also be beneficial given the previously described impact of COVID-19 as participants may develop more opportunities to interact over the next several months after participation. Next, while the intake process is heavily focused on the suitability of the curriculum for an adolescent using caregiver- and self-reported social difficulties and engagement, additional research into the relative utility of offering this intervention to participants at different levels of social impairment may be informative. For instance, examining the relative utility of the intervention with adolescents whose skills challenges are milder (e.g., SSIS-SEL below 100 but above 80) or who have more difficulties (e.g., SSIS-SEL score under 85) may inform whether standardized scores should be used for inclusion criteria. Similarly, other potential indicators of differential effects such as cognitive functioning or ADHD status could be examined with a larger sample.

Conclusions

This investigation is the first, to our knowledge, to explore the use of a specific social skills intervention (PEERS®; Program for the Education and Enrichment of Relational Skills) in adolescents with NF1. It is also one of very few studies to examine the utility of a telehealth delivered intervention strategy in this population. Based on caregiver-report, adolescents improved their social-emotional skills and number of get-togethers, and decreased their social impairment following the PEERS® intervention. Adolescents also exhibited increased social knowledge after completing the intervention. This intervention was reported to be highly acceptable and feasible among caregivers of adolescents with NF1 who completed the

intervention. This pilot study highlights the potential of this telehealth intervention strategy in improving social and friendship skills among adolescents with NF1 who have social difficulties. The pilot data from this investigation contribute to the limited social skills intervention research in adolescents with NF1, who are commonly reported to have social difficulties, point to the promise of utilizing telehealth within this population, and provide direction for areas of future research.

Figure 1. Participant Flowchart

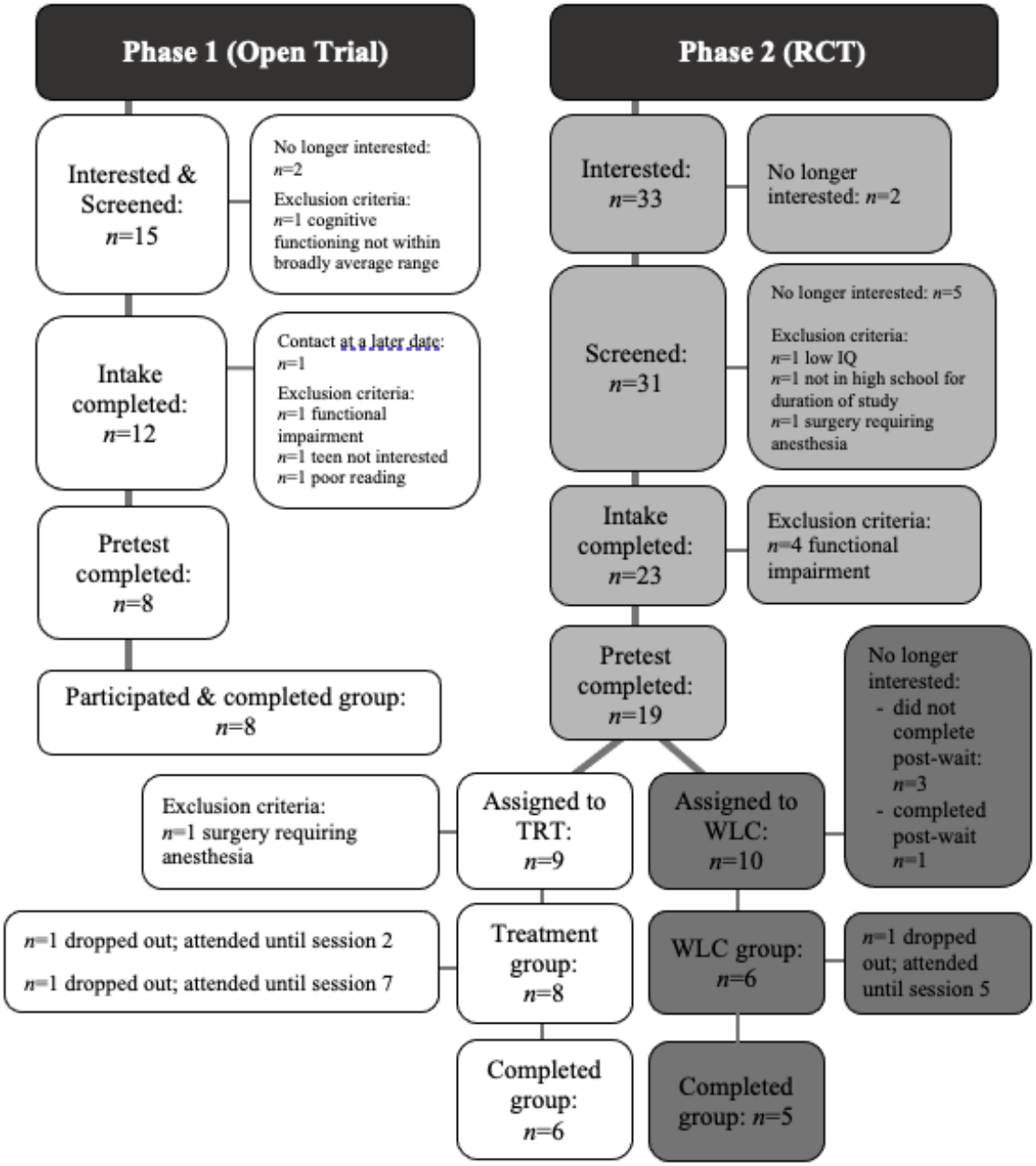


Figure 2. Study procedures flowchart for Phase 1 and Phase 2

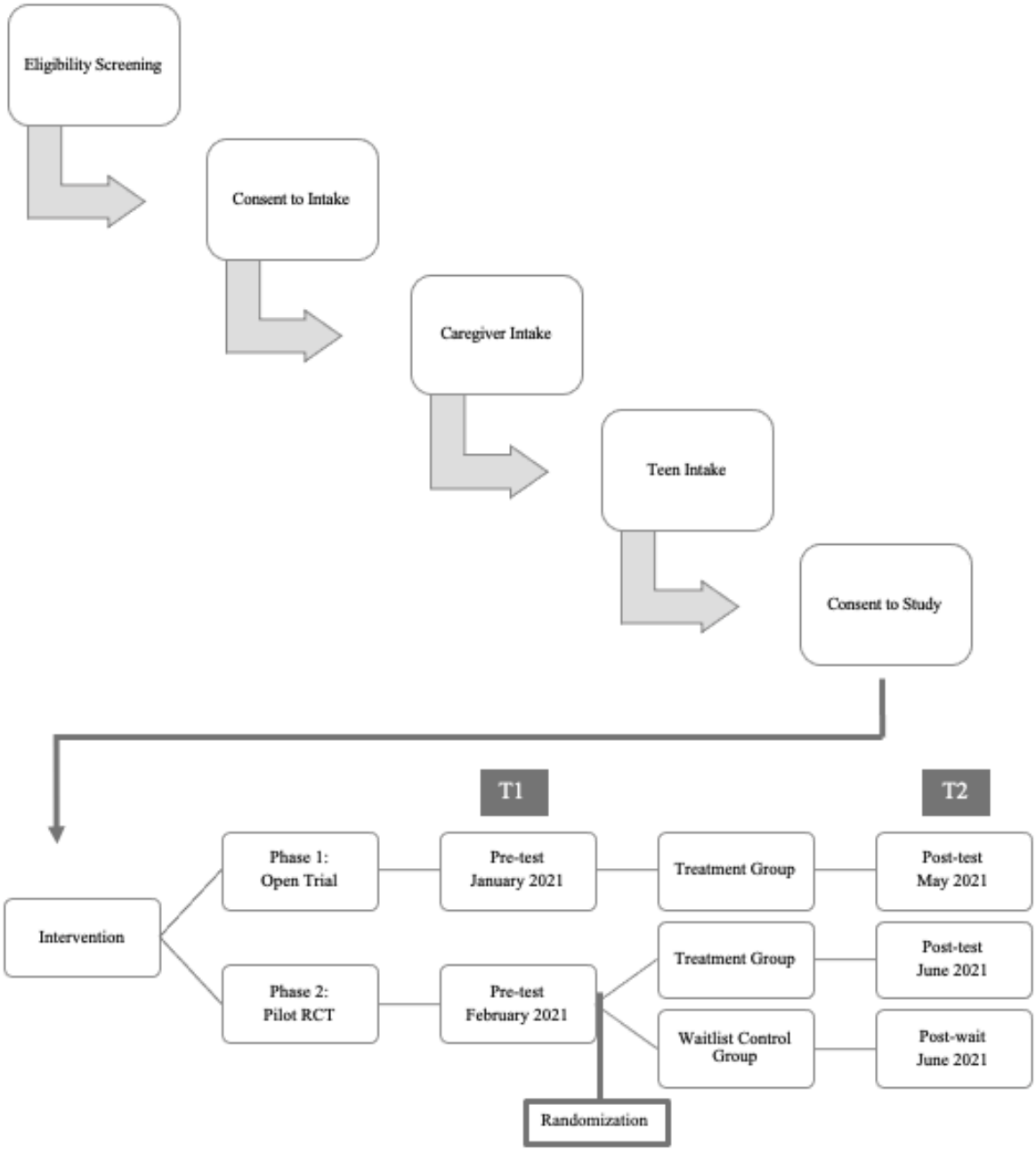


Figure 3. Illustration of measure administration at each timepoint

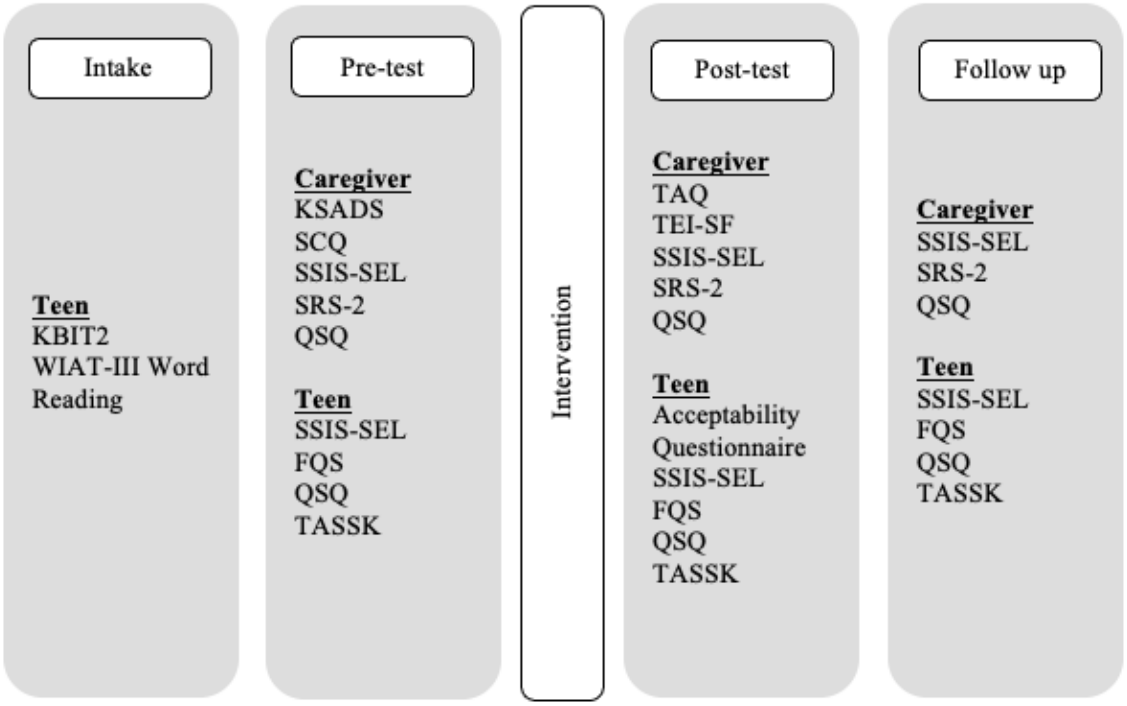
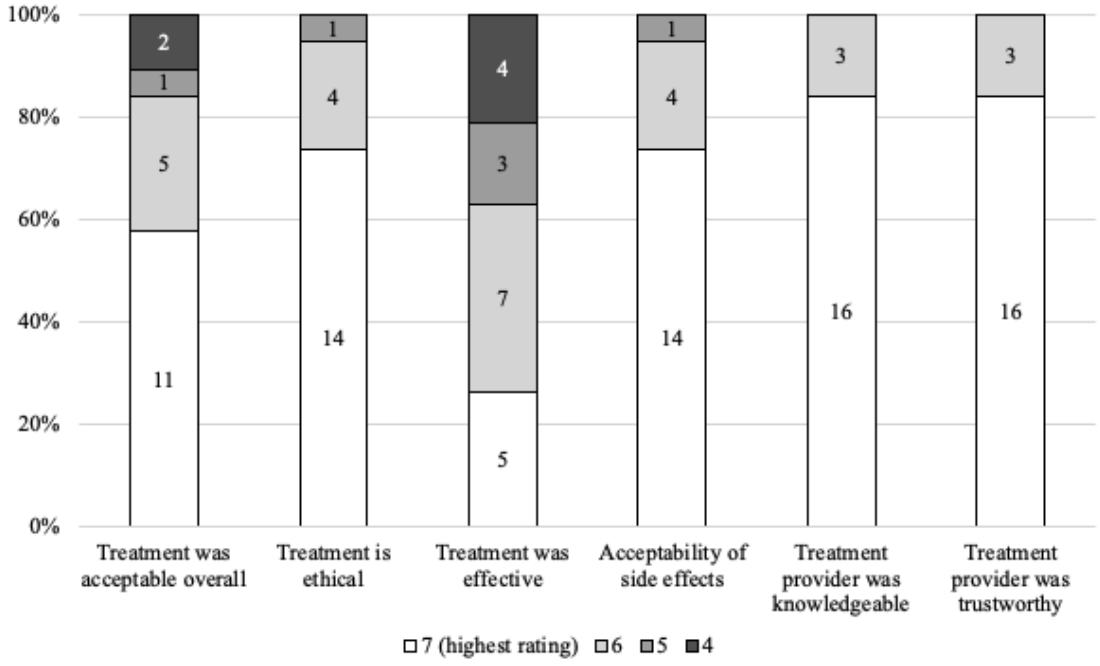
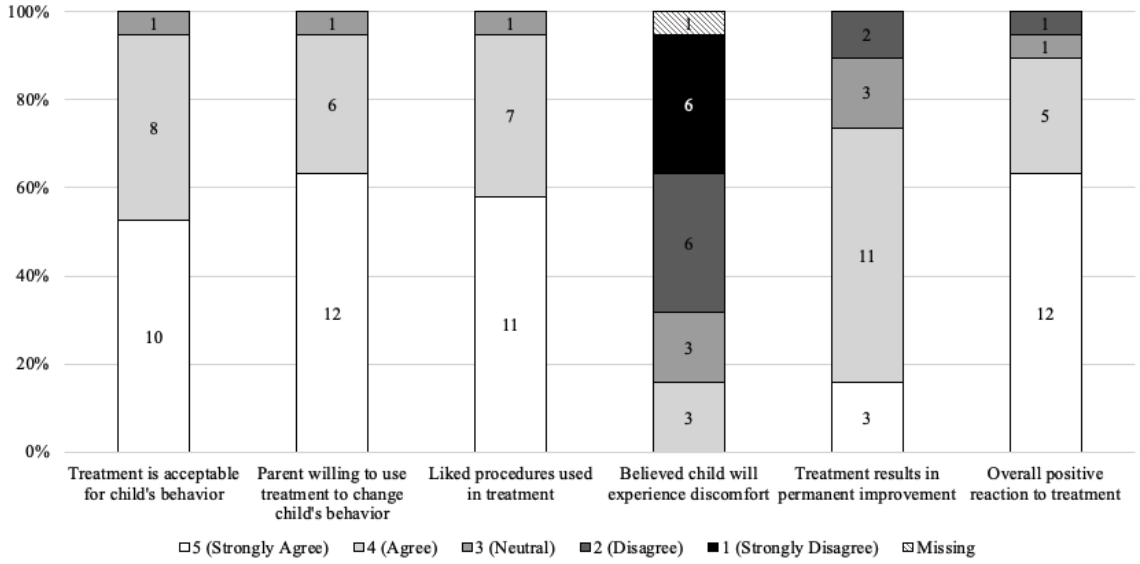


Figure 4. Caregiver Ratings on the Treatment Acceptability Questionnaire (*n*=19)



Note: Parents/caregivers reported on each item using a 7-point scale. No parental ratings of 1 (lowest rating), 2, or 3 reported for any items. The number provided in the bar indicates the number of parents/caregivers who selected the corresponding rating.

Figure 5. Caregiver Ratings on the Treatment Evaluation Inventory-Short Form (*n*=19)



Note: The number provided in the bar indicates the number of parents/caregivers who selected the corresponding rating.

Table 1. Empirical support for PEERS® intervention in adolescents with ASD

Authors	Participants & Procedure	Outcome Measures	Findings
Laugeson et al. (2009)	33 adolescents with ASD • 28 males • 5 females Range = 13-17 years Mean age = 14.6 years Treatment group (n = 17) Delayed treatment group (n = 16) 2 timepoints: • Pre-test • Post-test	*Parent:* SSRS QPQ *Teacher:* SSRS *Teen:* QPQ TASSK FQS	In comparison to the delayed treatment group, the treatment group had: • improved parent-reported socials skills • improved knowledge of social skills based on teen-report • increased hosted get-together based on parent and teen report No significant change in teacher-report of social skills (possibly due to poor return on questionnaires) The delayed treatment group showed friendship quality declined.
Laugeson et al., (2012)	28 adolescents with ASD • 23 males • 5 females Range = 12-17 years Mean age = 14.6 years Treatment group (n = 14) Delayed treatment group (n =14) 3 timepoints for treatment group: • Pre-test • Post-test • 14 week follow up (n = 12)	*Parent:* SSRS SRS QPQ *Teacher:* SSRS SRS *Teen:* QPQ TASSK	In comparison to the delayed treatment group, the treatment group had: • improved parent-reported social skills, cooperation, assertion and responsibility • decreased parent-reported ASD symptoms and autistic mannerisms as well as improved social awareness, social cognition, social communication and social motivation • increased parent-reported and teen-reported hosted get-togethers • improved knowledge of social skills Over time, the treatment group had: • improved parent-reported social skills, cooperation, assertion, responsibility and self-control • improved teacher-reported social skills and assertion • decreased parent-reported problem behaviors, externalizing behaviors, ASD symptoms and autistic mannerisms as well as improved social awareness and social communication

			• increased parent-reported and teen-reported hosted get-togethers • improved knowledge of social skills Treatment group improvements were maintained at 14 week follow up for all outcome measures except social cognition and motivation.
Mandelberg, et al., (2014)	53 adolescents with ASD • 43 males • 10 females Range = 12-18 years Mean age = 14.3 years (at time entered study) Mean age = 17.5 (at follow up) 3 timepoints for treatment group: • Pre-test • Post-test • Long-term follow up (1-5 years after treatment; average of 29 months)	*Parent:* SCQ SSRS SRS QPQ FII *Teen:* QPQ TASSK	Improvements were evident and maintained over time in: • parent-reported social skills • parent-reported social responsiveness • parent-reported problem behaviors • parent-reported frequency of get-togethers Knowledge of social skills improved from the intervention but regressed over time 1/3 of adolescents reported participating in some form of social skills training after having participated in PEERS®. 75% of adolescents reported having at least one friend with whom they were at least "pretty close" at follow-up while parents reported 87%. 79% of adolescents were involved in an extracurricular activity and 76% of parents reported these activities were "very helpful" for their adolescent socially.
Schohl et al., (2014)	58 adolescents with ASD • 47 males • 11 females Range = 11-16 years Mean age = 13.65 years Experimental group (n = 29) Waitlist control group (n = 29)	*Parent:* SSRS SRS QSQ *Teacher:* SSRS SRS *Teen:* QSQ TASSK	Compared to the waitlist control group, the experimental group had: • larger decrease in parent-reported ASD symptoms and a decrease in parent-reported and teacher-reported problem behaviors • improved knowledge of social skills • increased teen-reported hosted and invited get-togethers • decreased social anxiety

	2 timepoints: • Pre-test • Post-test	FQS SIAS	No significant differences in: • parent-reported social skills • parent-reported frequency of get-togethers • teacher-reported social skills • teacher-reported ASD symptoms • teen-report of friendship quality
Hill et al., (2017)	5 adolescents with ASD • 4 males • 1 female Range = 13-15 years Mean age = 13.4 years 2 timepoints: • Pre-test • Post-test	*Parent:* SSIS SRS-2 QPQ *Teen:* QPQ TASSK SCARED	Improvements were evident in: • parent-reported social engagement • parent-reported internalizing symptoms • parent-reported ASD symptomatology • parent reported social cognition • parent-reported social communication • parent-reported social motivation • knowledge of social skills • parent-reported and teen-reported frequency of hosted and invited get-togethers Anxiety did not change from the intervention.

Note: Autism spectrum disorder (ASD); Social Skills Rating System (SSRS) or Social Skills Improvement System (SSIS); Quality of Play Questionnaire (QPQ) or Quality of Socialization Questionnaire (QSQ); Test of Adolescent Social Skills Knowledge (TASSK); Friendship Qualities Scale (FQS). Social Responsiveness Scale (SRS) or Social Responsiveness Scale – 2nd Edition (SRS-2); Social Communication Questionnaire (SCQ); Friendships and Interventions Interview (FII); Social Interaction Anxiety Scale (SIAS); Screen for Child Anxiety Related Disorders (SCARED)

Table 2. Empirical support for PEERS® intervention in adolescents among other populations

Authors	Participants & Procedure	Outcome Measures	Findings
Gardner et al. (2015)	20 adolescents with ADHD • 14 males • 6 females Range = 11-16 years Mean age = 12.4 years 2 timepoints for treatment group: • Pre-test • Post-test	*Parent:* QSQ-R *Teen:* FQS TASSK SPPC/A QSQ-R	Improvements were evident in: • increased parent-reported and teen-reported initiation of friendships • improved teen knowledge of social skills • increased parent-reported and teen-reported hosted get-togethers No significant changes were noted in quality of existing friendships, social self-efficacy or levels of peer-conflict during get-togethers.
Gilmore et al. (2022)	36 adolescents with an acquired brain injury or cerebral palsy will be recruited • 19 males • 17 females Range = 11-17 years Mean age = 14 years Treatment group Waiting list control roup 3 timepoints for treatment group: • Pre-test • Post-test • 12-week follow up	*Parent:* SSIS-RS QSQ SRS-2 *Teen:* SSIS-RS QSQ TASSK-R	Between-group differences were evident with the treatment group having increased social knowledge at post-test and maintained at follow-up. No significant group differences were detected in get-togethers and social impairment. Pre- to post-intervention analyses found significant improvement on social competence (SSIS-RS social subscale), social knowledge, social impairment, and get-togethers.

Note: Attention-Deficit/Hyperactivity Disorder (ADHD); Quality of Socialization Questionnaire (QSQ) or QSQ-Revised (QSQ-R); Friendship Qualities Scale (FQS); Test of Adolescent Social Skills Knowledge (TASSK) or Test of Adolescent Social Skills Knowledge - Revised (TASSK-R); Self-Perception Profile for Children (SPPC) or Self-Perception Profile for Adolescent

(SPPA); Social Skills Improvement System -Rating Scale (SSIS-RS); Social Responsiveness Scale – 2nd Edition (SRS-2)

Table 3. Inclusion & Exclusion Criteria for Study Participants

Inclusion Criteria	Exclusion Criteria
Diagnosis of neurofibromatosis type 1 (NF1) by a physician	Any cognitive or developmental delays that would affect reading comprehension and/or understanding of the treatment material
Ages 12-17	First language and main language spoken in the home is NOT English (because standardized study measures and instructions are all in English)
In middle school or high school for duration of the study	Any significant behavioral concerns (e.g., violence, need of a 1-on-1 aide)
First language and main language spoken in the home is English	Any other comorbid medical conditions not commonly associated with NF1 (e.g., other illnesses; hearing or vision impaired)
Reliable access to internet	A major surgery (e.g., brain or heart surgery) within the past 6 months.
Exhibit current functional impairment in peer relationships, which will be confirmed during the parent/caregiver interview	Adolescent does not agree to participate or attend voluntarily
Willingness to attend all treatment sessions, with a maximum of two allowed absences	Prior social skills group treatment within the past 6 months
Interested in and motivated to participate in the treatment, evaluated during a structured interview at pre-assessment	

Table 4. Overview of PEERS® Telehealth Didactic Lessons

Session #	Didactic Lessons
1	Trading Information
2	Two way conversation
3	Electronic communication
4	Choosing appropriate friends
5	Appropriate use of humor
6	Starting and joining conversations
7	Exiting conversations
8	Good sportsmanship
9	Get-togethers
10	Handling disagreements
11	Handling teasing
12	Handling physical bullying & changing a reputation
13	Handling cyber bullying & minimizing rumors and gossip
14	Graduation & where to go from here

Table 5. Phase 1 Demographic Information and Mean Scores on Outcome Measures (*n*=8)

Variable (Mean (SD))						
Age (at time of entry into the study)	14.22 (1.95)					
Gender	Male – 5 (63%) Female – 3 (37%)					
Race/Ethnicity	White – 5 (62.5%) Biracial – 2 (25%) Asian – 1 (12.5%)					
NF Etiology	Familial – 1 (12.5%) Sporadic – 7 (87.5%)					
KBIT-2 SS	98.25 (8.86)					
WIAT-III Word Reading SS	94.25 (12.26)					
SCQ Lifetime Raw	5.63 (4.69)					
ADHD Diagnosis	No diagnosis – 4 (50%) Inattentive Type – 4 (50%)					
	Pretest	**Posttest**	*t*	*df*	*p*	*g*
SSIS-SEL Parent SS	*87.00* (10.04)	*86.88* (8.84)	.034	7	.49	.01
SSIS-SEL Student SS	*94.63* (11.43)	*97.75* (9.79)	-1.16	7	.14	-.39
SRS-2 T-score	62.25 (10.61)	*59.13* (7.88)	1.60	7	.08	.54
TASSK Raw	15.88 (3.4)	22.38 (4.14)	-4.40	**7**	**.002**	-1.47
QSQ-Caregiver Raw	1.63 (1.59)	2.75 (1.04)	-2.05	7	**.04**	-.69
QSQ-Adolescent Raw	2.63 (3.42)	4.38 (2.83)	-1.79	7	.58	-.59
FQS Raw	87.38 (9.93)	84.00 (14.11)	1.04	7	.17	.35

Note: SD: Standard Deviation; KBIT-2: Kaufman Brief Intelligence Test – Second Edition; SS: Standard Score; WIAT-III: Wechsler Individual Achievement Test – Third Edition; SCQ: Social Communication Questionnaire; SSIS-SEL: Social Skills Improvement System Social-Emotional Learning; SRS-2: Social Responsiveness Scale – Second Edition; TASSK: Test of Adolescent Social Skills Knowledge; QSQ: Quality of Socialization Questionnaire; FQS: Friendship Qualities Scale. Higher standard scores (Mean=100, SD=15) and higher raw scores indicate better abilities. Higher T-scores (Mean=50, SD=10) indicate more impairment.

Italics = Standardized measure with a mean score within 1 standard deviation; **Bold** = $p < .05$

Table 6. Phase 2 Demographic Information and Mean Scores on Outcome Measures (*n*=12)

Intent to Treat Analysis (*n*=19)				
Variable (Mean (SD))	Treatment (*n*=9)		Wait-list Control (*n*=10)	
Age (at time of entry into the study)	13.84 (1.29)		13.74 (1.40)	
Gender	Male – 5 (56%) Female – 3 (33%) Transgender – 1 (11%)		Male – 6 (60%) Female – 3 (30%) Still Deciding – 1 (10%)	
Race/Ethnicity	White – 8 (89%) Asian – 1 (11%)		White – 10 (100%)	
NF Etiology	Sporadic – 9 (100%)		Familial – 1 (10%) Sporadic – 7 (70%) Missing – 2 (20%)	
KBIT-2 SS	95.78 (16.38)		111.40 (9.57)	
WIAT-III Word Reading SS	94.11 (18.59)		103.10 (9.57)	
SCQ Lifetime Raw	8.00 (5.34)		6.13 (5.46)*	
ADHD Diagnosis	No diagnosis – 5 (56%) Inattentive Type – 2 (22%) Combined Type – 1 (11%) Diagnosis Missing – 1 (11%)		No diagnosis – 3 (30%) Inattentive Type – 2 (20%) Hyperactive/Impulsive Type – 1 (10%) Combined Type – 4 (40%)	
Timepoints	**Pre-test**	**Post-test**	**Pre-test**	**Post-wait**
SSIS- SEL Parent SS	84.56 (13.26)	*88.56 (14.89)*	83.88 (9.69)	81.63 (5.09)*
SSIS-SEL Student SS	*96.89 (16.29)*	*99.89 (15.94)*	*100.25 (17.99)*	*99.25 (15.94)**
SRS-2 T-score	65.44 (10.83)	64.22 (13.10)	67.50 (7.93)	68.25 (5.57)*
TASSK Raw	14.33 (4.06)	18.67 (5.22)	14.70 (2.00)	15.60 (1.43)
QSQ-Caregiver Raw	1.11 (1.54)	2.00 (2.06)	1.29 (1.49)	2.43 (2.23)^
QSQ-Adolescent Raw	1.11 (1.96)	2.00 (2.35)	4.25 (6.86)	7.25 (7.59)*
FQS Raw	81.67 (23.49)	86.44 (15.39)	89.70 (11.62)	87.50 (12.66)
Completer Analysis (*n*=12)				
Variable (Mean (SD))	Treatment (*n*=6)		Wait-list Control (*n*=7)	
Age (at time of entry into the study)	14.31 (1.32)		13.79 (1.53)	
Gender	Male – 3 (50%) Female – 3 (50%)		Male – 5 (71%) Female – 2 (29%)	
Race/Ethnicity	White – 5 (83.3%) Asian – 1 (16.7%)		White – 7 (100%)	
NF Etiology	Sporadic – 6 (100%)		Familial – 1 (17%) Sporadic – 5 (83%)	
KBIT-2 SS	90.17 (13.93)		114.14 (8.959)	
WIAT-III Word Reading SS	91.33 (15.46)		100.00 (9.29)	
SCQ Lifetime Raw	8.17 (4.71)		6.57 (5.74)	
ADHD Diagnosis	No diagnosis – 3 (50%) Inattentive Type – 2 (33%) Diagnosis Missing – 1 (17%)		No diagnosis – 2 (29%) Inattentive Type – 2 (29%) Hyperactive – 1 (14%)	

			Combined Type – 2 (29%)	
Timepoints	**Pre-test**	**Post-test**	**Pre-test**	**Post-wait**
SSIS- SEL Parent SS	82.67 (15.42)	*88.67* (18.01)	*84.86* (10.02)	82.29 (5.12)
SSIS-SEL Student SS	*95.5* (19.99)	*100.00* (19.69)	*103.29* (17.08)	*102.14* (14.77)
SRS-2 T-score	68.0 (10.22)	66.17 (14.19)	68.29 (8.22)	69.14 (5.37)
TASSK Raw	13.83 (2.40)	20.33 (3.72)	14.57 (2.23)	15.86 (1.35)
QSQ-Caregiver Raw	.833 (1.33)	2.33 (2.25)	1.29 (1.49)	2.43 (2.23)
QSQ-Adolescent Raw	2.17 (2.40)	3.83 (1.33)	3.86 (7.31)	7.29 (8.19)
FQS Raw	80.00 (27.52)	87.17 (16.17)	89.00 (12.66)	85.86 (13.77)

Note: SD: Standard Deviation; KBIT-2: Kaufman Brief Intelligence Test – Second Edition; SS: Standard Score; WIAT-III: Wechsler Individual Achievement Test – Third Edition; SCQ: Social Communication Questionnaire; SSIS-SEL: Social Skills Improvement System Social-Emotional Learning; SRS-2: Social Responsiveness Scale – Second Edition; TASSK: Test of Adolescent Social Skills Knowledge; QSQ: Quality of Socialization Questionnaire; FQS: Friendship Qualities Scale. Higher standard scores (Mean=100, SD=15) and higher raw scores indicate better abilities. Higher T-scores (Mean=50, SD=10) indicate more impairment.

* = n=8; ^ = n=7; *Italics* = Standardized measure with a mean score within 1 standard deviation.

Table 7. Phase 2 Independent samples t-tests for treatment group versus wait-list control on outcome measures

	Intent to Treat (*n*=19)							
	Pre-test				Post-test/Post-wait			
	t	*df*	*p*	*g*	*t*	*df*	*p*	*g*
SSIS-SEL Parent SS	.12	15	.45	.06	1.25	15	.12	.58
SSIS-SEL Student SS	0.40	15	.35	-.19	.08	15	.47	.04
SRS-2 T-score	-.44	15	.33	-.20	-.81	15	.22	-.37
TASSK Raw	-.25	17	.40	-.11	1.79	17	**.046**	.79
QSQ-Caregiver Raw	-.23	14	.41	-.11	-.39	14	.35	-.19
QSQ-Adolescent Raw	-1.32	15	.10	-.61	-1.98	15	**.048**	-.91
FQS Raw	-.96	17	.18	-.42	-.16	17	.44	-.07
	Completer Analysis (*n*=13)							
	Pre-test				Post-test/Post-wait			
	t	*df*	*p*	*g*	*t*	*df*	*p*	*g*
SSIS-SEL Parent SS	-.31	11	.38	-.16	.84	5.69	.22	.50
SSIS-SEL Student SS	-.76	11	.22	-.39	-.22	11	.41	-.12
SRS-2 T-score	-.06	11	.48	-.03	-.49	6.22	.32	-.27
TASSK Raw	-.58	11	.29	-.29	2.98	11	**.006**	1.54
QSQ-Caregiver Raw	-.57	11	.22	-.29	-.21	11	.42	-.11
QSQ-Adolescent Raw	-.70	11	.25	-.36	-1.32	7.06	.11	-.64
FQS Raw	-.78	11	.23	-.40	.16	11	.44	.08

Note: SS: Standard Score; SSIS-SEL: Social Skills Improvement System Social-Emotional Learning; SRS-2: Social Responsiveness Scale – Second Edition; TASSK: Test of Adolescent Social Skills Knowledge; QSQ: Quality of Socialization Questionnaire; FQS: Friendship Qualities Scale. **Bold** = $p < .05$

Table 8. Demographic Information and Mean Scores on Outcome Measures for All Participants

Variable (Mean (SD))	**Intent to Treat (*N*=27)**					
Age (at time of entry into the study)	13.92 (1.51)					
Gender	Male – 16 (59%) Female – 9 (33%) Transgender – 1 (4%) Still Deciding – 1 (4%)					
Race/Ethnicity	White – 23 (85%) Biracial – 2 (7.5%) Asian – 2 (7.5%)					
NF Etiology	Familial – 2 (7.5%) Sporadic – 23 (85%) Missing – 2 (7.5%)					
KBIT-2 SS	102.3 (13.67)					
WIAT-III Word Reading SS	97.48 (14.07)					
SCQ Lifetime Raw	6.64 (5.07)					
ADHD Diagnosis	No diagnosis – 12 (44%) Inattentive Type – 8 (30%) Hyperactive/Impulsive Type – 1 (4%) Combined Type – 5 (18%) Diagnosis Missing – 1 (4%)					
Timepoints	**Pretest**	**Posttest**	*T*	*df*	*p*	*g*
SSIS-SEL Parent SS	84.40 (10.02)	*88.16 (10.87)*	-2.19	24	**.019**	-.43
SSIS-SEL Student SS	*96.92 (14.29)*	*94.80 (22.30)*	.57	24	.28	.11
SRS-2 T-score	65.32 (9.33)	61.72 (9.97)	2.72	24	**.006**	.54
TASSK Raw	15.19 (3.09)	19.52 (4.73)	-5.74	26	**<.001**	-1.09
QSQ-Caregiver Raw	1.67 (1.78)	2.79 (1.99)	-3.58	23	**< .001**	-.72
QSQ-Adolescent Raw	3.56 (5.35)	4.40 (5.42)	-.97	24	.17	-.19
FQS Raw	85.52 (16.11)	86.96 (12.29)	-.49	26	.31	-.09
	Completer Analysis (*n*=19)					
Age (at time of entry into the study)	13.79 (1.32)					
Gender	Male – 11 (58%) Female – 6 (32%) Transgender – 1 (5%) Still Deciding – 1 (5%)					
Race/Ethnicity	White – 18 (95%) Asian – 1 (5%)					
NF Etiology	Familial – 2 (10.5%) Sporadic – 17 (89.5%)					
KBIT-2 SS	104.0 (15.14)					
WIAT-III Word Reading SS	98.84 (14.86)					
SCQ Lifetime Raw	7.12 (5.31)					

ADHD Diagnosis		No diagnosis – 3 (50%) Inattentive Type – 2 (33%) Diagnosis Missing – 1 (17%)				
Timepoints	Pretest	Posttest	*T*	*df*	*p*	*g*
SSIS-SEL Parent SS	84.47 (10.87)	*89.42 (11.61)*	-2.25	18	**.018**	-.51
SSIS-SEL Student SS	*96.58 (14.79)*	*98.11 (13.68)*	-.69	18	.25	-.15
SRS-2 T-score	65.47 (9.29)	60.74 (9.93)	2.85	18	**.005**	.64
TASSK Raw	15.21 (2.74)	21.37 (3.73)	-8.34	18	**< .001**	-1.87
QSQ-Caregiver Raw	1.74 (1.88)	3.16 (1.97)	-3.85	18	**< .001**	-.86
QSQ-Adolescent Raw	4.32 (5.77)	5.42 (5.68)	-.97	18	.17	-.22
FQS Raw	84.58 (17.78)	86.63 (12.75)	-.49	18	.32	-.12

Note: SD: Standard Deviation; KBIT-2: Kaufman Brief Intelligence Test – Second Edition; SS: Standard Score; WIAT-III: Wechsler Individual Achievement Test – Third Edition; SCQ: Social Communication Questionnaire; SSIS-SEL: Social Skills Improvement System Social-Emotional Learning; SRS-2: Social Responsiveness Scale – Second Edition; TASSK: Test of Adolescent Social Skills Knowledge; QSQ: Quality of Socialization Questionnaire; FQS: Friendship Qualities Scale. Higher standard scores (Mean=100, SD=15) and higher raw scores indicate better abilities. Higher T-scores (Mean=50, SD=10) indicate more impairment.

Italics = Standardized measure with a mean score within 1 standard deviation; **Bold** = $p < .05$

Table 9. Exploratory paired samples t-tests for primary outcome measure subscales

Intent to Treat Analysis (*N*=27)								
	Pretest		Posttest		*t*	*df*	*p*	*g*
SSIS-SEL Parent Form SS	Mean	SD	Mean	SD				
Self-Awareness	76.80	15.98	79.68	12.78	-1.07	24	.15	-.21
Self-Management	*88.36*	11.69	*92.76*	11.63	-2.61	24	**.008**	-.51
Social Awareness	*92.52*	15.44	*94.24*	13.79	-.77	24	.23	-.15
Relationship Skills	*87.24*	10.41	*91.84*	11.89	-2.81	24	**.005**	-.55
Responsible Decision Making	*90.76*	15.84	*92.32*	13.94	-.901	24	.18	-.17
SRS-2 T-scores								
Social Awareness	63.08	13.14	60.08	10.99	1.87	24	**.037**	.37
Social Cognitive	62.20	9.96	*58.68*	10.27	2.18	24	**.02**	.43
Social Communication	65.84	8.98	61.92	9.85	2.73	24	**.006**	.54
Social Motivation	60.64	11.52	*58.16*	10.39	1.88	24	**.036**	.37
Social Communication and Interaction	65.04	9.41	61.32	9.85	2.72	24	**.006**	.54
SSIS-SEL Student Form SS								
Self-Awareness	*98.72*	13.61	*99.96*	12.41	-.69	24	.25	-.14
Self-Management	*96.68*	13.89	*99.80*	14.33	-1.89	24	**.035**	-.37
Social Awareness	*99.72*	14.67	*99.52*	15.14	.081	24	.47	.016
Relationship Skills	*92.00*	17.83	*92.20*	17.43	-.11	24	.46	-.02
Responsible Decision Making	*99.60*	12.34	*100.72*	12.82	-.61	24	.27	-.12
Completer Analysis (*n*=19)								
	Pre-test		Post-test		*t*	*df*	*p*	*G*
SSIS-SEL Parent Form SS	Mean	SD	Mean	SD				
Self-Awareness	77.21	15.55	81.0	10.58	-1.071	18	.15	-.24
Self-Management	*89.53*	10.29	*95.32*	9.03	-2.711	18	**.007**	-.61
Social Awareness	*91.0*	16.95	*93.68*	14.87	-.922	18	.18	-.21
Relationship Skills	*86.89*	11.01	*92.95*	12.66	-2.943	18	**.004**	-.66
Responsible Decision Making	*91.32*	16.6	*93.37*	14.03	-.900	18	.19	-.20
SRS-2 T-scores								
Social Awareness	62.37	13.68	*58.42*	10.41	1.902	18	**.037**	.43
Social Cognitive	63.37	10.35	*58.74*	11.02	2.238	18	**.019**	.50
Social Communication	65.84	8.36	60.68	9.23	2.850	18	**.005**	.64
Social Motivation	60.73	12.23	*57.47*	10.69	1.914	18	**.036**	.43
Social Communication and Interaction	65.26	9.26	60.37	9.65	2.846	18	**.005**	.64
SSIS-SEL Student Form SS								
Self-Awareness	*99.32*	14.07	*100.95*	12.39	-.691	18	.25	-.15
Self-Management	*95.79*	13.37	*99.89*	14.11	-1.927	18	**.035**	-.43
Social Awareness	*97.63*	14.99	*97.37*	15.57	.081	18	.47	.018
Relationship Skills	*92.89*	17.61	*93.16*	17.04	-.110	18	.46	-.03
Responsible Decision Making	*99.68*	11.93	*101.16*	12.5	-.605	18	.28	-.14

Note: SSIS-SEL: Social Skills Improvement System Social-Emotional Learning; SD: Standard Deviation; SRS-2: Social Responsiveness Scale – Second Edition; Higher standard scores (Mean=100, SD=15) indicate better abilities. Higher T-scores (Mean=50, SD=10) indicate more impairment. *Italics* = Standardized measure with a mean score within 1 standard deviation; **Bold** = $p < .05$

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