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Shakespeare and Autism: A Pilot Study Examining the Treatment Effects and Predictors of Treatment Response of the Hunter Heartbeat Method

Margaret H Mehling, Marc J Tassé* and Rebecca R Andridge

The Ohio State University, Columbus, USA

***Corresponding author:** Marc J Tassé, PhD, The Ohio State University Columbus, Ohio, United States, Tel: 614-685-3193; Fax: 614-366-6373; E-mail: Tasse.1@osu.edu

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Abstract

This pilot study was designed to explore the potential impact of participation in the Hunter Heartbeat Method, a drama-based social skills intervention, on the core symptoms of autism spectrum disorder (ASD). A sample of 10 children with ASD participated in this study. Children were assessed at three time-points: before the start of the intervention (T1), after 14 weeks of participation (T2) and after 42 weeks of participation (T3). Findings from this pilot study indicated that the Hunter Heartbeat Method (HHM) is an intervention that shows promise in impacting social, communication and facial emotion recognition deficits associated with ASD. We also identified the individual characteristics of the participants who were responders and non-responders to the HHM intervention. The results of these findings are discussed.

Keywords: Autism spectrum disorder; Intervention; Social skills; Drama-based; Hunter heartbeat method; Shakespeare

Introduction

Autism spectrum disorder (ASD) is a lifelong condition and the lifetime cost of caring for an individual with ASD has been estimated to be as high as $2.5 million [1]. ASD is characterized by both deficits in social communication and the presence of restricted and repetitive behaviors and interests [2]. Social skill deficits associated with ASD include poor friendship skills [3], difficulty with perspective taking [4] and inadequate social communication [5]. Social skills deficits in children with ASD are pervasive and persistent resulting in a long-term, adverse impact on community inclusion, independence and overall quality of life [6].

Many social skills interventions developed for children and adolescents with ASD explicitly teach social skills such as initiating conversation, inviting a friend to play, and recognizing facial emotions [7,8]. Research supports the efficacy of such interventions, many of which have been found to yield measurable benefits [9,10]. There are, however, several caveats to these efficacious findings. First, there is a considerable degree of variability in treatment outcomes in children with ASD even with the most supported evidence based treatments [11]. Second, many children with ASD struggle to generalize the skills learned in treatment to broader contexts such as home and school [12]. Finally, many of the gains made during treatment appear to diminish after treatment has concluded [13]. These issues of treatment response versus non-response, generalization, and maintenance are of critical importance in the social skills intervention research literature, however are under-studied as they are more difficult to operationalize and measure.

Unlike many social skills intervention, drama-based interventions rely on a more exploratory, "learn by doing" model which offers a structured approach that has the capacity to motivate hard to reach children to participate more meaningfully in social interactions [14]. Research on drama-based interventions for children with ASD is in its infancy. Promising research from Corbett et al. [15] on Social Emotional NeuroScience Endocrinology (SENSE) theatre offers support for this drama-based intervention's ability to improve social-emotional functioning and reduce stress in children with ASD. Research findings for a pilot study examining the efficacy of SENSE theatre indicate that participants showed some improvement in face identification and Theory of Mind skills [15]. Corbett et al. [16] also assessed a 2 week summer camp model of the SENSE theatre program. Results indicated significant differences in face processing, social awareness and social cognition. Furthermore, duration of interaction with familiar peers increased significantly over the course of treatment, however, engagement with novel peers outside the treatment setting remained stable [16].

Some preliminary research data on drama-based social skills interventions was published by Mehling et al. [17] examining the effects of participation in a Shakespearian theatre intervention using the Hunter Heartbeat Method. These authors examined the effects of the Hunter Heartbeat Method (HHM) on certain behavioral features associated with ASD including social engagement, expressive communication, facial emotion expression recognition, and pragmatic language. They reported encouraging finding from a sample of 14 children between the ages of 10 to 13 years who participated in the HHM intervention once a week after school for 14 weeks. Results indicated that some participants made measurable gains in social skills, facial emotion recognition, pragmatic language, and interpersonal relationship skill offering support for the potential efficacy of the HHM as a means to improve core symptoms associated with ASD [17].

This study was designed to further explore the potential impact of participation in the Hunter Heartbeat Method on core symptoms of ASD. Specifically, this study had the following aims: 1) replication of pilot study finding demonstrating ability of participation in the Hunter Heartbeat Method to impact core symptoms of ASD; 2) extension of the duration of the intervention from 14 weeks to 1.5 academic years

to gain further insight regarding optimal intervention dose; 3) exploration of differences in symptom profile and characteristics of intervention responders and non-responders to offer insight regarding children for whom this intervention may be most effective. Furthermore, this study piloted a shift in intervention delivery modality from delivery as an after-school activity to delivery embedded within the school day [17].

Methods

Hunter heartbeat method

Hunter Heartbeat Method is a drama-based intervention intended to impact social-communication deficits in children with ASD. This play-based intervention modality uses games based on Shakespeare's The Tempest that emphasize the themes of the eyes, mind and heart [18]. These games, which are learned progressively across sessions, build on one another and target foundational social communication skills including eye contact, turn-taking, facial emotion recognition, imitation, improvisation and humor using experiential, play-based learning. Children learn the games while seated in a large group circle through imitation and observation, then, children practice each game one-on-one with a facilitator after which children are encouraged to enter the middle of the circle and show their interpretation of the game to their peers. The Hunter Heartbeat Method emphasizes the low facilitator-to-child ratio so children receive individual attention, feedback, and interaction as they play the games and develop core social skills. The Hunter Heartbeat Method is described in greater detail in Mehling et al. [17] and in the intervention's published manual by Hunter [18].

The Hunter Heartbeat Method uses many of the well-established components of evidence-based social skills training, such as modelling by a competent role model, practice and role-playing with feedback. Reinforcement in the Hunter Heartbeat Method is intrinsic due to the playful and humorous nature of the games. In the Hunter Heartbeat Method, children participate in social games that, while creating the opportunity for the development of core social skills, are still, ultimately, games. Children work collaboratively with actors imitating and practicing the give-and-take of social interaction; turn taking and leading and following. This intervention, to the untrained eye, appears no different than a drama class or after school activity in which a typically developing peer would participate. Thus, the Hunter Heartbeat Method allows children with ASD to learn social skills in an ecologically valid environment that mirrors both the extra-curricular activities of typically developing peers and the play-based learning environment in which social skills are acquired in typical development.

The Hunter Heartbeat Method was administered by a team of trained graduate-level theatre student-actors and a Department of Theatre faculty member, who served as the lead actor. A treatment-fidelity check was developed collaboratively by Department of Theatre faculty, Kelly Hunter (Creator of HHM), and research personnel and was completed during an observation of a Hunter Heartbeat Method session by a Department of Theatre faculty who had training in the Hunter Heartbeat Methodology but was otherwise not involved in this study. The fidelity check revealed that that treatment was being administered in keeping with key elements of the intervention associated with its efficacy as in a manner consistent with procedures outlined in the treatment manual

Participants

Participants were recruited through three local schools on the basis of having an autism spectrum disorder diagnosis, an autism educational classification, and the absence of severe behavior problems. The first ten participants (N=10) to complete the consent process were enrolled in the Treatment/Intervention group. The gender of the participants consisted of 80% boys (n=8) and 20% girls (n=2). The chronological age of our sample ranged from 9-14 years, with a mean age of 11.78 years (SD=1.85). We had a racially diverse sample, including 40% Caucasian/white (n=4), 40% (n=4) African American or Black and 20% bi-racial (n=2).

Measures

The Autism Diagnostic Observation Schedule [19,20] was used as a pre-intervention measure to validate children's ASD diagnosis. Additionally, Social Communication and Restricted and Repetitive Behavior subscale scores were used as pre-treatment participant characteristics in analyses intended to characterize differences between treatment responders and non-responders.

Outcome measures were collected at three time points (Time 1-pre intervention, Time 2-following 14 weeks of intervention and Time 3-post intervention, 1.5 academic years following Time 1 measure) to assess the impact of the Hunter Heartbeat Method on the core features of ASD. Outcome measures included the following:

Vineland adaptive behavior rating scale 2nd edition

Parent-reported measurement of change in social and communication skill level was obtained through parent-completed Vineland Adaptive Behavior Rating Scale, 2nd Edition [21,22]. Furthermore, to better understand the change in adaptive skills in children with autism spectrum disorder, researchers [23,24] have suggested examining not only the total scores of the domains and subdomains but also examining the scores of item subsets that measure specific adaptive skills. These item subsets are provided directly on the Vineland-II protocol as well as in the User's Manual [22] and were used in this study to gain a specific insight into treatment impact on social and communication skills.

Social responsiveness scale

Additionally, parents completed the Social Responsiveness Scale [25] as a direct measure of autism symptomology, specifically social communication symptoms. The SRS is a 65-item rating scale measuring the severity of autism spectrum symptoms. The SRS was normed on a sample of more than 1,600 children and demonstrates sensitivity and reliability across a wide range of symptom severity and is recommended for use as a measure of response to intervention. The SRS assesses social impairment, social awareness, social information processing, capacity for reciprocal social communication, social anxiety/avoidance, as well as repetitive behaviors [25].

Nisonger child behavior rating form

Parents competed the Nisonger Child Behavior Rating Form [26] to further characterize the sample. The parent version of the NCBRF included a total of 76 items; 10 Positive Social items and 66 Problem Behavior items. Parent respondents rate their child's behavior directly on the NCBRF rating scale using a problem rating scale of: 0=not a problem/does not occur to 3=major problem/occurs frequently.

Construct validity was reported by the authors as well as concurrent validity evidence comparing the NCBRF scores with the Aberrant Behavior Checklist [27], a well-established problem behavior scale used with this population. The authors concluded that the NCBRF's validity evidence was strong, and that the subscales of the ABC and the NCBRF that were thought to be related tap similar constructs. The factor structure of the NCBRF was established as evidence of its construct validity [28] and its psychometric properties have been independently evaluated and confirmed by several published studies [15,29,30] The NCBRF has also been widely used in published research investigating problem behavior in children with developmental disabilities [31-34] and has been translated/adapted into other languages [30,35]. The Isolate subscale of the NCBRF was of interest as an outcome measure as decreases in isolation would be an anticipated result of participation in the Hunter Heartbeat Method intervention.

Penn facial emotion recognition task

Direct assessment of changes in the core features of ASD were assessed through the administration of the Penn Facial Emotion Recognition task [36,37], an objective measure of ability to recognize facial expression of emotion. The Penn is a computer-administered measure of facial emotion recognition that was used to assess facial emotion recognition capabilities pre and post-intervention. The Penn uses colour photographs of faces expressing 4 basic emotions (happiness, sadness, anger and fear) as well as photographs of neutral or calm expressions. Participants are shown a series of faces and are asked to identify the expressed emotion from 5 possible emotion labels (happy, sad, angry, fear or calm).

Researcher-developed social validity questionnaire

Additionally, following the intervention, a brief questionnaire was included for parents, teachers, and participants to provide feedback on the intervention and possible collateral benefits (e.g. increased social interactions with peers, increased interest in social activities, etc.).

Procedure

After receiving approval from by The Ohio State University Behavioral and Social Sciences Institutional Review Board, we established a partnership with a local area school district, children from three schools within the district were recruited to participate in the Hunter Heartbeat Method intervention 1 hour per week for three 14 week sessions (42 weeks total of treatment) during their regular school day distributed across 1.5 academic years. Intervention sessions were held only during the academic year; participants did not receive in the Hunter Heartbeat Method during holiday or summer breaks. After Time 1 measures were completed participants received 14 weeks of intervention. Following the 14 weeks, Time 2 measures were completed. Time 2 measures preceded students' summer break during which no intervention sessions were held. In the autumn, when participants returned to school, participants received two 14 week sessions of intervention separated by winter break. Following the two additional 14 weeks sessions of intervention, approximately 1.5 years after Time 1 measurements, Time 3 measurements were completed.

Analyses

Exploration of intervention effects across time: Linear mixed model analyses were completed using all treatment group subject data (N=10) across the three measurement time-points (T1, T2, T3) to model trajectories over time. These models contained a random subject-specific effect to capture the within-subject correlation arising from repeated measurements on the same subjects. Due to the small sample size and limited degrees of freedom, no baseline covariates were included in the models. Testing for significant changes in scores was completed for the following planned contrasts: T1:T2, T2:T3 and T1:T3. Analyses were carried out in SAS version 9.4. Effect sizes (Cohen's d) for these changes across time were also calculated, using the estimated mean change (estimated from the mixed models) divided by the baseline standard deviation of the measure.

Exploratory analyses using Vineland Parcel Scores. Analyses of the Vineland-II Parcel scores were carried out using the same methodology described above. Effect sizes (Cohen's d) for these changes across time were also calculated. Changes across time were calculated as the estimated mean change (derived from Table 1) divided by the baseline standard deviation of the measure.

Exploration of characteristics of treatment responders and non-responders: Subjects were classified as "T2 responders" if there was a positive change from T1 to T2 for Vineland-II and Penn data and a negative change for SRS. Subjects were similarly classified as "T3 responders" if there was a positive change from T1 to T3 for Vineland-II and Penn data and a negative change from T1 to T3 for SRS data. Those subjects not classified as responders for T2 or T3 were classified as non-responders for that time point. T-tests, assuming unequal variance, were then used to compare responders and non-responders in terms of age, ADOS subscale scores, and NCBRF subscale scores. Cohen's d rather than p-values was chosen as an index of the magnitude of differences between groups as very small group size in these analyses resulted in non-significant comparisons despite large effect sizes.

Exploration of predictive relationships between t1 symptom profile and t2 and t3 outcome measures: Partial correlations between T1 predictors and outcomes at T2 and T3 adjusting for the T1 value of the outcome were used to examine predictive relationships between scores at T1 on measures of interest and subsequent outcomes at T2 and T3. For example, the predictive relationship of ADOS Social Communication Subscale at T1 and the Penn Total Score at T2 was analyzed by computing the correlation between ADOS Social Communication at T1 and Penn Total at T2 adjusting for Penn Total at T1.

Results

Exploration of intervention effects across time

Table 1 contains estimated means for Vineland-II Composite, Social Skills and Communication Skills standard scores, SRS, Penn scores, and NCBRF subscale scores at each time point (T1, T2, T3) and p-values for tests of significant change between each pair of time points as well as effect size statistics quantifying the magnitude of the change. Results for selected Vineland-II Social Skills subscales (Interpersonal Relationships, Play and Leisure, Coping Skills) and Communication Skills subscales (Receptive Language and Expressive Language) are also presented in Table 1.

Measure		T1		T2		T3		P-value and Effect Size					
		Mean	(SE)	Mean	(SE)	Mean	(SE)	T1:T2	d	T2:T3	d	T1:T3	d
Vineland	Composite	67.9	-4.7	72.6	-4.8	68.4	-5	0.08	0.4	0.18	-0.36	0.85	0.05
	Social	65.1	-5.4	71.2	-5.6	70.7	-6	0.16	0.55	0.92	-0.04	0.25	0.51
	Interpers	7.4	-1	9.1	-1	8.5	-1.1	0.04	0.74	0.54	-0.24	0.2	0.49
	Play	8	-1.1	8.3	-1.1	8.4	-1.2	0.75	0.1	0.87	0.06	0.65	0.16
	Coping	10.1	-1	11.4	-1.1	11.7	-1.1	0.12	0.6	0.738	0.14	0.09	0.74
	Commun	68.1	-3.3	70.3	-3.3	65.4	-3.4	0.05	0.2	0.002	-0.45	0.04	-0.25
	Express lang	9.2	-0.8	9.1	-0.8	9	-0.9	0.79	-0.04	0.81	-0.04	0.63	-0.08
	Recept lang	8.4	-0.8	9.7	-0.8	8.1	-0.9	0.08	0.52	0.08	-0.63	0.73	-0.11
SRS	**Total**	**80.2**	**-3.6**	**76.8**	**-3.8**	**76**	**-4.2**	**0.32**	**-0.32**	**0.83**	**-0.08**	**0.26**	**-0.4**
Penn	**Total**	**22.2**	**-1.5**	**24.8**	**-1.4**	**25.7**	**-1.5**	**0.07**	**0.61**	**0.53**	**0.21**	**0.03**	**0.82**
NCBRF	Adaptive	5.2	-0.6	4.8	-0.6	6.2	-0.7	0.62	-0.17	0.13	0.65	0.23	0.48
	Compliant	9.6	-1	8.3	-1.1	9	-1.2	0.23	-0.34	0.56	0.19	0.63	-0.15
	Conduct	10.7	-2.9	11.9	-3.1	12.1	-3.2	0.57	0.11	0.94	0.02	0.56	0.13
	Hyper	14.8	-2.4	15.1	-2.5	10.9	-2.7	0.85	0.04	0.06	-0.54	0.07	-0.5
	Insecure	8.7	-2.3	10.7	-2.4	8.5	-2.6	0.28	0.26	0.32	-0.28	0.92	-0.03
	Isolate	4.7	-1.4	7	-1.5	3.2	-1.6	0.14	0.49	0.04	-0.82	0.36	-0.33
	Self-Injury	1.3	-0.5	1.7	-0.5	1	-0.5	0.37	0.27	0.21	-0.46	0.58	-0.19
	Sensitive	5.1	-0.8	5.1	-0.9	4.2	-1	0.96	0.02	0.35	-0.45	0.35	-0.43

Table 1: Estimated means at each time point, p-values for tests of significant change between each pair of time points and effect size (Vineland-II subscales: Interpers=Interpersonal relationships; play=play and leisure; coping=coping skills; commun=communication; express lang=expressive language; recept lang=receptive language).

With regards to social functioning as measured by the Vineland-II, participants made improvements from Time 1 to Time 2 in their parent reported social functioning, however these improvements were not statistically significant (T1:T2, p=0.16; T1:T3, p=0.25). Gains in parent reported social functioning maintained from Time 2 to Time 3 (scores differed by less than one point; T2:T3 p=0.92), indicating that in this skill domain, as a group, intervention participants made initial skill gains in the first 14 weeks that maintained across the subsequent year of intervention. Comparable patterns of response were noted on Vineland-II Social Skills subscales. On the Interpersonal Relationships subscale, participants made significant gains from Time 1 to Time 2 (T1:T2, p=0.04) with no additional significant gain in skill from Time 2 to Time 3 (T2:T3, p=0.54) indicating that in this skill domain, as a group, intervention participants made initial skill gains in the first 14 weeks that maintained but did not increase after 42 weeks of the intervention. A similar pattern of initial gain and maintenance was noted for the Coping Skills subdomain although changes were non-significant. There was no significant increase in skill in the Play and Leisure subdomain.

SRS total scores indicated a similar pattern of participant response. Participant scores, on average, decreased across the first14 weeks of the intervention (T1:T2, p=0.32) indicating an initial reduction in ASD symptomology. This change was maintained from Time 2 to Time 3, however, no additional decrease in symptomology was measured (T1:T2, p=0.83).

With regards to participants' functional communication skills as measured by the Vineland-II Communication Skills domain, significant increases in skill were noted from Time 1 to Time 2 (T1:T2, p=0.05), however, there was a significant decrease in skill from Time 2 to Time 3 (T2:T3, p=0.002). Further examination for communication subscale scores indicated that with regards to Expressive Communication, there was no significant change in scores across the duration of the intervention. However, with regards to Expressive Language, participants scores increased notably from Time 1 to Time 2 (T1:T2, p=0.08), however these initial gains were not maintained at Time 3 with scores returning to baseline levels (T1:T3, p=0.73).

Regarding facial emotion recognition, participant data from the Penn Facial Emotion Recognition Task indicated steady skill improvement across the duration of the intervention with improvements in scores noted from Time 1 to Time 2 and from Time 2 to Time 3 that reached statistical significant across the 1.5 years in duration of the intervention (T1:T3, p=0.03).

Exploratory analyses using Vineland parcel scores

Table 2 contains estimated means for Vineland-II parcel scores for the Interpersonal Relationships, Play and Leisure, Coping, Expressive Language, and Receptive Language subscales at each time point and p-values for tests of significant change between each pair of time points as well as effect size statistics quantifying the magnitude of the change.

Vineland Parcels		T1		T2		T3				P-value and Effect Size			
Measure		Mean	(SE)	Mean	(SE)	Mean	(SE)	T1:T2	d	T2:T3	D	T1:T3	d
Coping	Apologize	2.7	-0.6	3.8	-0.7	3.4	-0.7	0.02	0.66	0.43	-0.2	0.14	0.42
	Control	6.2	-0.8	6.5	-0.9	8	-1	0.72	0.12	0.19	0.53	0.1	0.65
	Manner	9.3	-1.4	10.9	-1.4	12.4	-1.5	0.15	0.5	0.25	0.45	0.02	0.95
	Responsib	1.6	-0.5	2.1	-0.6	2.2	-0.6	0.24	0.31	0.98	0.01	0.28	0.32
	Secret	1.5	-0.5	1.1	-0.5	2	-0.5	0.34	-0.4	0.09	0.77	0.33	0.41
	SocCaution	2.4	-0.7	2.4	-0.7	3.7	-0.7	0.97	-0	0.05	0.74	0.05	0.73
	Transition	3.8	-0.5	4	-0.5	3.9	-0.5	0.6	0.13	0.81	-0.1	0.83	0.06
Expressive	BeginTalk	19.6	-1.2	19.8	-1.2	20.1	-1.3	0.79	0.07	0.78	0.08	0.6	0.15
	Express	25.1	-3.5	26.1	-3.5	25.1	-3.6	0.43	0.11	0.49	-0.1	0.98	0
	Interact	19	-2.4	19	-2.4	19.6	-2.4	0.95	0	0.35	0.08	0.36	0.08
	PreSpeech	13.5	-1.4	13	-1.5	15.5	-1.7	0.73	-0.1	0.16	0.63	0.23	0.51
	Speech	19.6	-2.3	20.1	-2.3	19.1	-2.4	0.59	0.08	0.35	-0.2	0.61	-0.1
Play	Games	7.5	-1.1	7.4	-1.2	7.7	-1.3	0.94	-0	0.8	0.08	0.84	0.06
	GoFriend	2	-0.7	2.5	-0.8	2.4	-0.8	0.46	0.3	0.9	-0.1	0.6	0.24
	Play	19.5	-2	17.6	-2.1	19	-2.2	0.16	-0.3	0.36	0.24	0.74	-0.1
	Share	5.9	-0.8	6.4	-0.9	6.6	-0.9	0.39	0.2	0.78	0.08	0.3	0.27
	SocCues	0.3	-0.3	0.7	-0.3	1.1	-0.3	0.21	0.55	0.23	0.62	0.03	1.17
Receptive	Follow Instruction	5.7	-0.6	6.4	-0.6	5.7	-0.6	0.01	0.4	0.02	-0.4	0.87	-0
	Listen Attend	7.4	-1	9.7	-1.1	10.1	-1.2	0.05	0.65	0.77	0.1	0.04	0.75
	Understanding	13.8	-0.8	13.6	-0.9	15	-1	0.74	-0.1	0.09	0.51	0.13	0.43
Relations	Dating	0.1	-0.1	0	-0.1	0	-0.1	0.33	-0.3	1	0	0.37	-0.3
	ExpEmo	12.9	-1.2	14.3	-1.2	13.5	-1.3	0.08	0.42	0.41	-0.2	0.45	0.2
	Friend	2.8	-0.8	4.3	-0.8	4.6	-0.9	0.04	0.77	0.71	0.15	0.03	0.91
	Imitate	7.1	-0.9	7.8	-0.9	7.9	-1	0.45	0.38	0.88	0.09	0.4	0.47
	Respond	10.2	-0.8	10.1	-0.8	10.8	-0.9	0.85	-0.1	0.33	0.29	0.39	0.24
	Social	7.9	-1.7	10.4	-1.8	10.6	-1.9	0.04	0.55	0.85	0.05	0.05	0.6
	Thoughtful	0.9	-0.2	1.3	-0.2	1.1	-0.3	0.12	0.7	0.48	-0.4	0.48	0.34

Table 2: Results for selected Vineland-II parcel scores: Estimated means at each time point, p-values for tests of significant change between each pair of time points, and effect size.

Vineland-II Parcel Scores represent specific skills measured within each Vineland-II Subdomain [22]. Parcel scores were analyzed for subdomains of interest and results emphasize specific skill sets that changed across time in association with intervention participation. In the Coping subdomain, participants made gains in the apologize skill area from Time 1 to Time 2 (T1:T2, p=0.02), in the manners skill area from Time 1 to Time 3 (T1:T3, p=0.02), and in the social caution skill area from Time 2 to Time 3 (T2:T3, p=0.05) and from Time 1 to Time 3 (T1:T3, p=0.05). In the Play subdomain, participants made improvements in the Social Cues skill area from Time 1 to Time 3 (T1:T3, p=0.03). In the Receptive subdomain, participants made improvements in the Following Instructions skill area from Time 1 to

Time 2 (T1:T2, p=0.01). Participants also made improvements in the Listening and Attending skill area from Time 1 to Time 2 (T1:T2, p=0.05) and from Time 1 to Time 3 (T1:T3, p=0.04). In the Relationship subdomain, participants made significant improvement in the Friends skill area from Time 1 to time 2 (T1:T2, p=0.04) and from Time 1 to Time 3 (T1:T3, p=0.03). Participants also made significant gains in the Social skill area from Time 1 to Time 2 (T1:T2, p=0.04) and from Time 1 to Time 3 (T1:T3, p=0.05).

Characteristics of treatment responders and non-responders

Analyses were carried out to identify characteristics at baseline that differentiated treatment responders from non-responders on each outcome measure at Time 2 (Table 3) and Time 3 (Table 4). Results indicate that participants who improved in their social skills as measured by the Vineland-II Social Skills domain after 14 weeks had higher adaptive functioning as measured by NCBRF Adaptive scale at baseline and lower hyperactivity as measured by NCBRF Hyperactive scale at baseline than participants who did not demonstrate measured improvement in the Vineland-II Social Skills domain at Time 2. Participants who improved at Time 3 relative to Time 1 had lower ADOS Restricted/Repetitive Behavior (RRB) scores and higher NCBRF Adaptive scores than those participants that did not demonstrate score improvement across the full 1.5 years of the intervention.

Outcome	Significant Difference*	Direction of Difference	Effect Size
Vineland-II Social (T2)	NCBRF Adaptive (T1)	Resp>Nonresp	d=1.9
	NCBRF Hyper (T1)	Resp<Nonresp	d=-1.8
Vineland=II	Age (p=0.056)	Resp<Nonresp	d=-1.4
Communication (T2)	ADOS SBRI (T1)	Resp>Nonresp	d=1.5
	NCBRF Self Injury (T1) p=0.053)	Resp>Nonresp	d=1.5
SRS Total (T2)	NCBRF Isolate (T1) (p=0.07)	Resp>Nonresp	d=1.3
Penn Total (T2)	NCBRF Self Injury (T1) (p=0.07)	Resp>Nonresp	d=1.3

Table 3: Summary of significant differences between responders and non-responders at T2.

Outcome	Significant Difference*	Direction of Difference	Effect Size
Vineland-II Social (T3)	ADOS SBRI (T1)(p=0.07)	Resp<Nonresp	d=-1.6
	NCBRF Adaptive (T1)	Resp>Nonresp	d=3.6
	NCBRF Self Injury (T1)	Resp>Nonresp	d=2.4
Vineland-II Communication (T3)	n/a–0 responders		
SRS Total (T3)	ADOS SBRI (T1)	Resp<Nonresp	d=-5.7
Penn Total (T3)	n/a–only 1 non-responder		

Table 4: Summary of significant differences between responders and non-responders at T3.

Participants who improved from Time 1 to Time 2 in their communication skills as measured by the Vineland-II Communication Skills domain were on average younger than non-responders and were characterized by higher scores on ADOS RRB subscale and NCBRF Self-Injury subscale at baseline. In contrast, participants who showed an overall reduction in ASD symptomology as measured by the SRS at Time 2 were more self-isolated at Time 1 and those participants whose SRS scores reflected a reduction in ASD symptomology across the 1.5 years of intervention were characterized by lower ADOS RRB scores at baseline. With regards to the Penn Facial Emotion Recognition Task, participants who responded from Time 1 to Time 2 were higher on the NCBRF Self Injury subscale than non-responders. At Time 3, all participants except 1 were classified as Penn Responders so analyses were not completed to compare characteristics of responders and non-responders at this time-point.

Exploration of Predictive Relationships between T1 Symptom Profile and T2 and T3.

Outcome measures

Analyses were conducted to determine if a specific baseline symptom profile was associated with treatment response in a given skill domain across time (Table 5). Results indicate that participant ADOS SBRI scores at Time 1 were significantly associated with their Time 2 Penn Score and their SRS score at Time 3. With regards to Time 2 and 3 scores on the Vineland-II Social Skills domain, participant scores on NCBRF Conduct and Hyperactivity subscales were predictive of their T2 Vineland-II Social Skills scores and scores on NCBRF Self-Injury/Stereotypic subscale were predictive of Time 3 Vineland-II Social Skills domain scores. With regards to Vineland-II Communication Skills scores at Time 2 and Time 3, NCBRF Adaptive and Self-Injury/Stereotypic scores were predictive Time 2 Vineland-II

Communication Skills scores and NCBRF Self-Injury and Sensitive subscale scores were predictive of Vineland-II Communication Skills outcomes at Time 3. Finally, participants' Vineland-II Communication Skills domain score at T1 was predictive of SRS Total Score at Time 2.

		Penn Total		SRS Total		Vineland-II Composite		Vineland-II Social		Vineland-II Communication	
		T2	T3	T2	T3	T2	T3	T2	T3	T2	T3
ADOS	Comm+Social	-0.2	0.57	0.43	0.6	0.46	-0.06	0.44	-0.23	-0.43	-0.69
	SBRI	-0.76	0.15	0.68	0.89	-0.54	-0.93	-0.57	-0.54	0.02	-0.56
NCBRF	Compliant	-0.37	0.17	0.03	0.66	0.37	-0.5	0.68	-0.08	0.08	-0.49
	Adaptive	-0.19	-0.69	-0.15	-0.62	-0.09	0.59	0.19	0.88	0.76	0.62
	Conduct	0.27	-0.17	0.03	-0.2	-0.58	-0.09	-0.81	-0.09	0.06	0.71
	Insecure	0.32	-0.25	-0.33	-0.6	-0.36	-0.17	-0.53	-0.31	-0.2	0.25
	Hyper	0.22	-0.44	0.56	0.46	-0.68	0.13	-0.83	0.43	0.39	0.61
	Self-Injury	0.32	-0.87	0.35	-0.14	-0.15	0.82	-0.32	0.92	0.94	0.92
	Isolate	0.56	0.54	-0.05	0.5	0.77	0.11	0.45	-0.21	-0.42	-0.2
	Sensitive	0.61	-0.71	-0.24	-0.3	-0.03	0.48	-0.59	0.44	0.73	0.96
Vineland	Composite	0.21	-0.58	-0.66	-0.79						
	Social	0.16	-0.53	-0.73	-0.82						
	Communication	0.09	-0.51	-0.76	-0.63						

Table 5: Pearson correlations, adjusted for T1 outcome values.

Discussion

Social skills interventions are a frontline treatment for children with ASD although evidence regarding the effectiveness of social skills interventions is mixed. Measurable treatment gains are often observed in the training environment across the duration of the intervention, however, generalization of skills to novel environments and maintenance of treatment gains across time are lacking. Furthermore, variable treatment response across individuals is common. Thus, although the effectiveness of a social skills intervention on a group level across multiple domains is compelling, it is of limited clinical utility in informing treatment recommendations for an individual child. A greater translational value may be found in the exploration of individual differences in treatment responders and non-responders over time and across treatment outcome.

This research served to further previous findings from a feasibility study that involved the participation in a 14 week Hunter Heartbeat method intervention that was associated with trends in improvement in social and communicative functioning in the home environment as reported by parents and improvements in facial emotion recognition measured directly in children outside of the training environment. Furthermore, this research extended the duration of participation in the Hunter Heartbeat Method intervention from 14 weeks to 42 weeks to examine the impact of increased dose on treatment gains by symptom class. Overall, results indicate measured skill improvement across the three measurement time points in the domains of communication, social skills, friendship skills, and facial emotion recognition.

Symptom class and treatment response

Our results indicate that various symptom domains exhibit differential response to treatments across time. Language skills, for example, improved quickly; significant change was noted in the first 14 weeks of intervention and then improvement either maintained or returned to baseline from measurement Time 2 to Time 3. Skill improvement was noted most significantly in the receptive language domain, specifically, in skill areas related to following instructions and listening and attention. Listening and attention skills made significant improvement from Time 1 to Time 2 and that improvement maintained across the next academic year of intervention. However, following instructions, which also improved from Time 1 to Time 2, returned to baseline at Time 3.

Interestingly, this pattern of response displayed by language skills differs from the pattern of response we saw in broad social skills. With the exception of apologizing, which improved most significantly during the first 14 weeks of treatment with gains maintaining until Time 3, other broad social skills including manners, social caution, and social cues, showed a pattern of more gradual improvement across the entire duration of the intervention. These skills made gradual improvement at each measurement time point reaching statistical significance in the Time 1 to Time 3 comparison.

Friendship skills distinguished themselves from these broader social skills and seemed to show a pattern of improvement more comparable to language skills, although no return to baseline occurred. Both friendship and social skills (parcels of the Vineland-II Interpersonal Relationships subdomain) showed significant improvement form Time 1 to Time 2 and also in the Time 1 to Time 3 comparison. This pattern

of results indicates that significant change occurred in the first 14 weeks that was then maintained from Time 2 to Time 3. It is interesting to consider how friendship skills display a pattern of response distinct from broader social skills that aligns more with communication skills as research indicates that level of communication impairment is closely tied to friendship quality and quantity [38].

Facial emotion recognition skills showed a different pattern of responding than friendship or communication skills. Rather than being characterized by initial skill gain followed by maintenance, facial emotion recognition abilities steadily improved across the 42 weeks of intervention. Furthermore, by Time 3, nearly all participants had made gains in this domain. Facial emotion recognition represents a critical foundational skill for effective social communication; much communicative meaning is conveyed through subtle facial expression of emotion. It must be noted that parent reported social and communicative functioning required that skill improvement generalize to environments outside of training where parents were able to observe their child's behavior and to skills that were not explicitly taught. However, facial emotion recognition was assessed directly with the child using a standardized task. It is possible that these variables contributed to the observed difference in treatment response across these skill domains.

Characteristics of treatment responders by skill domain

Our findings yielded three distinct classes of treatment responders based on measurement time point and skill domain: Time 2 Social Skill Responders, Time 2 Communication Responders and Time 3 Social Skills Responders. We were able to identify unique characteristics of these participants that distinguished them from non-responders at each given time point within each symptom class. Time 2 Social Skills Responders were characterized by higher adaptive functioning (response on Vineland-II social scales), less hyperactivity (response on Vineland-II social scales), more isolated behavior (response on SRS), and more rigidity (response on PENN) at baseline than non-responders (participants who did not make significant improvement on measures of social skills from Time 1 to Time 2). Time 3 Social Skills Responders showed a comparable profile in that they were characterized, like Time 2 responders, with greater adaptive functioning at baseline. However, Time 3 Social Skills Responders were less rigid at baseline. Time 2 Communication Responders, like Time 2 Social Skill Responders, were more rigid at baseline. Interestingly, Time 2 Communication Responders were also younger at baseline than non-responders in this symptom class.

Implications

Our findings suggest that the optimal treatment dose may differ by targeted symptom class and also by child characteristics. Friendship and communication skills seem to improve rapidly across the first 14 weeks whereas broader social skills, most notably facial emotion recognition, improve gradually across the 42 weeks of intervention. Furthermore, participants with higher baseline behavioral rigidity seem to make most significant gains across the first 14 weeks of intervention whereas participants with less behavioral rigidity required a longer duration of intervention to be considered "treatment responders". These findings remain preliminary and are based on a very small pilot sample but are interesting none-the-less. They serve as a starting point for the much-needed exploration of within-participant characteristics (such as greatest area of deficit or primary skill target and child characteristics such as age, rigidity, hyperactivity, etc.) that can be used to match children with optimal treatment modality, dose, and duration. Treatment for autism is costly and time consuming yet, like nearly all medical and therapeutic treatments, not all recipients make significant gains. Optimal treatment is not one-size-fits-all and consideration of treatment responder and non-responder characteristics in research has translational value in that eventually, this line of work may inform not just broad "best practice" treatment recommendations, but individualized treatment recommendations optimal for each child.

It is also interesting to consider the possibility that skill maintenance may require continued intervention. Our results indicate that although some symptom (broad Social Skills and Facial Emotion Recognition) classes made gradual improvement across the 42 weeks of intervention, others (Friendship and Communication Skills) were characterized by quick improvement during the first 14 weeks with the additional treatment serving to maintain those gains (with the exception of following instructions which returned to baseline at measurement Time 3). The Hunter Heartbeat Method, unlike many other social skills interventions, to the untrained eye, appears no different from a "drama club" or other after school activity. Rather than explicitly teaching isolated social skills, the Hunter Heartbeat Method creates a context supportive and evocative of the development of foundational social skills that is highly motivating and accessible for children with ASD. Much like a typically developing child who may make initial skill gains after beginning a new activity, maintenance of skill gains requires continued practice of the skill. It is possible that continued participation in the Hunter Heartbeat Method served to maintain initially acquired social skill.

Limitations

There are several limitations to this study that must be taken into account when interpreting the findings. This study used a pilot design and did not employ a waitlist or alternative treatment control group, which limits the interpretation of findings. Hence, without an alternative group to control for the effects of maturation, it is possible that this variable confounds the treatment effects. Furthermore, due to logistic constraints of the extended duration of this intervention, we were unable to enrol more than 10 participants. This low N impacts the generalizability of the results and the statistical power of the analyses. Due to the low number of participants and the exploratory nature of the analyses conducted, corrections for multiple testing were not used in the analyses. The lack of such a correction increases the possibility of type 1 error in the findings. However, such an exploratory approach remains appropriate as the purpose of these analyses was to identify potential characteristics of responders and non-responders to generate hypotheses to be tested in a more tightly controlled randomized controlled trial of this intervention.

Future Directions

Results of this study warrant further evaluation of the Hunter Heartbeat Method's ability to impact core deficits associated with ASD. Specifically, further research should include a randomized controlled trial with comparison to a waitlist and/or alternative treatment control group. Further exploration of optimal treatment dose and maintenance of treatment gains is also warranted. Specifically, following 14 weeks of intervention and post-test assessments, maintenance of treatment gains should be assessed following a lapse of time to determine if continued participation is required for skill maintenance.

References

1. Buescher AS, Cidav Z, Knapp M, Mandell DS (2014) costs of autism spectrum disorders in the United Kingdom and the United States. JAMA Pediatr 168: 721-728.
2. American Psychiatric Association (2013) Diagnostic and statistical manual of mental disorders. American Psychiatric Publishing.
3. Petrina N, Carter M, Stephenson J (2014) The nature of friendship in children with autism spectrum disorders: A systematic review. Research in Autism Spectrum Disorder 8: 111-126.
4. Kana RK, Libero LE, Hu CP, Deshpande HD, Colburn JS (2014) Functional brain networks and white matter underlying theory-of-mind in autism. Soc Cogn Affect Neurosci 9: 98-105.
5. Volden J, Phillips L (2010) Measuring pragmatic language in speakers with autism spectrum disorders: Comparing the children's communication checklist - 2 and the test of pragmatic language. Am J Speech Lang Pathol 19: 204-212.
6. Carter A, Davis N, Klin A, Volkmar F (2005) Handbook of Autism and Pervasive Developmental Disorders. Wiley.
7. Elliott S, Roach A, Beddow P (2008) Best practices in school psychology. NASP Publications.
8. Volkmar FR (2014) Editorial: The importance of early intervention. J Autism Dev Disord 44: 2979-2980.
9. Kaat AJ, Lecavalier L (2014) Group-based social skills treatment: A methodological review. Research in Autism Spectrum Disorder 8: 15-24.
10. Chung KM, Reavis S, Mosconi M, Drewry J, Matthews T, et al. (2007) Peer-mediated social skills training program for young children with high-functioning autism. Res Dev Disabil 28: 423-436.
11. Weiss MJ (1999) Differential rates of skill acquisition and outcomes of early intensive behavioral intervention for autism. Behavioral Interventions 14: 3-22.
12. Barry T, Klinger L, Lee J, Palardy N, Gilmore T, et al. (2003) Examining the effectiveness of an outpatient clinic-based social skills group for high-functioning children with autism. J Autism Dev Disord 33: 685-701.
13. White SW, Keonig K, Scahill L (2007) Social skills development in children with autism spectrum disorders: A review of the intervention research. J Autism Dev Disord 37: 1858-1868.
14. Peter M (2003) Drama, narrative and early learning. Br J Special Educ 30: 21-27.
15. Corbett BA, Gunther JR, Comins D, Price J, Ryan N, et al. (2011) Brief report: Theatre as therapy for children with autism spectrum disorder. J Autism Dev Disord 41: 505-511.
16. Corbett BA, Swain DM, Coke C, Simon D, Newsom C, et al. (2014) Improvement in social deficits in autism spectrum disorders using a theatre-based, peer-mediated intervention. Autism Res 7: 4-16.
17. Mehling MH, Tassé MJ, Root R (2017) Shakespeare and autism: An exploratory evaluation of the hunter heartbeat method. Research and Practice in Intellectual and Developmental Disabilities. 4: 107-120.
18. Hunter K (2014) Shakespeare's heartbeat: Drama games for children with autism. Routledge.
19. Lord C, Rutter M, Goode S, Heemsbergen J, Jordan H, et al. (1989) Autism diagnostic observation schedule: A standardized observation of communicative and social behavior. J Autism Dev Disord 19: 185-212.
20. Gotham K, Risi S, Pickles A, Lord C (2007) The autism diagnostic observation schedule: Revised algorithms for improved diagnostic validity. J Autism Dev Disord 37: 613-627.
21. Balboni G, Tasso A, Muratori F, Cubelli R (2016) The Vineland-II in preschool children with autism spectrum disorders: An item content category analysis. J Autism Dev Disord 46: 42-52.
22. Sparrow S, Cicchetti D, Balla D (2005) Vineland adaptive behavior scales. NCS Pearson Inc.
23. Paul R, Miles S, Cicchetti D, Sparrow S, Klin A, et al. (2004) Adaptive behavior in autism and pervasive developmental disorder-not otherwise specified: Microanalysis of scores on the Vineland adaptive behavior scales. J Autism Dev Disord 34: 223-228.
24. Balboni G, Tasso A, Muratori F, Cubelli R (2016) The Vineland-II in preschool children with autism spectrum disorders: An item content category analysis. J Autism Dev Disord 46: 42-52.
25. Constantino JN, Gruber CP (2005) Social responsiveness scale. Los Angeles: Western psychological services.
26. Aman MG, Tassé MJ, Rojahn J, Hammer D (1996) The Nisonger CBRF: A child behavior rating form for children with developmental disabilities. Res Dev Disabil. 17: 41-57.
27. Aman MG and Singh NN (1986) Aberrant behavior checklist manual. East aurora, NY: Slosson educational publications.
28. Tassé MJ, Aman MG, Hammer D, Rojahn J (1996) The nisonger child behavior rating form: Age and gender effects and norms. Res Dev Disabil 17: 59-75.
29. Lecavalier L, Aman MG, Hammer D, Stoica W, Mathews GL (2004) Factor analysis of the Nisonger child behavior rating form in children with autism spectrum disorders. J Autism Dev Disord 34: 709-721.
30. Mircea CE, Rojahn J, Esbensen AJ (2010) Psychometric evaluation of Romanian translations of the behavior problems inventory-01 and the Nisonger child behavior rating form. J Ment Health Res Intellect Disabil 3: 51-65.
31. Aman MG, De Smedt G, Derivan A, Lyons B, Findling RL, et al. (2002) Double-blind, placebo-controlled study of risperidone for the treatment of disruptive behaviors in children with subaverage intelligence. Am J Psychiatry 159: 1337-1346.
32. Pandina GJ, Bilder R, Harvey PD, Keefe RSE, Aman MG, et al. (2007) Risperidone and cognitive function in children with disruptive behavior disorders. Biol Psychiatry 62: 226-234.
33. Shea S, Turgay A, Carroll A, Schulz M, Orlik H, et al. (2004) Risperidone in the treatment of disruptive behavioral symptoms in children with autistic and other pervasive developmental disorders. Pediatrics 114: 634-641.
34. Snyder R, Turgay A, Aman M, Binder C, Fisman S, et al. (2002) Effects of risperidone on conduct and disruptive behavior disorders in children with subaverage IQs. J Am Acad Child Adolesc Psychiatry 41: 1026-1036.
35. Tassé MJ, Morin IN, Girouard N (2000) Traduction et validation canadienne-française du Nisonger Child Behavior Rating Form. Can Psychol 41: 116-123.
36. Gur RC, Ragland JD, Moberg PJ,Turner TH, Bilker WB, et al. (2001) Computerized neurocognitive scanning: I. Methodology and validation in healthy people. Neuropsychopharmacology 25: 766-776.
37. Gur RC, Sara R, Hagendoorn M, Marom O, Hughett P, et al. (2002) A method for obtaining 3-dimensional facial expressions and its standardization for use in neurocognitive studies. J Neurosci Methods 115: 137-143.
38. Whitehouse AJO, Watt HJ, Line EA, Bishop DVM (2009) Adult psychosocial outcomes of children with specific language impairment, pragmatic language impairment and autism. Int J Lang Commun Disord 44: 511-528.