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Music therapy for children with Autism Spectrum Disorder

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CHAPTER 6.

Music therapy for children on the autism spectrum: improved functioning observed by multiple informants

This Chapter is under revision for:

Nordic Journal of Music Therapy

Abstract

Introduction:

In a previously published multiple case study in which ten children on the autism spectrum were offered 'Papageno Music Therapy Program' (PMTP), a favorable development was found with regard to social interaction. As an extension of our previous study, we investigate if the progress can also be observed in a larger group, and whether these developments are generalized to other situations.

Method:

A non-experimental, pre-post measurement, multiple-informant design was used to monitor the development of these children in different social contexts. The observers were therapists, parents, family members and teachers. 40 children were observed over a 20-week period.

Results:

The study shows an improvement in the social interaction of children on the autism spectrum and supports the findings of our previous ten children multiple case study. The agreement between the different informants was high. The Reliable Change Index (RCI), which examined the results at an individual level, confirmed a positive improvement for more than 32 participants ($RCI > 1.96$). All the informants saw progress, so it seemed that the development was also generalized to other situations.

Conclusion:

This study confirmed earlier research into the effects of PMTP on this group: improvements in social interaction were visible during the course of the therapy. Informants observed this improvement not only within, but also outside the therapeutic setting. Overall, there were sufficient indications for music therapy having a positive effect on social behavior, making it worthwhile to investigate PMTP with a Randomized Controlled Trial for the purpose of a more rigorous efficacy test.

Introduction

Autism Spectrum Disorder (ASD) is a complex developmental condition associated with persistent challenges in social interaction, speech and nonverbal communication, and limited/repetitive behavior. (APA, 2013). The restricted capacity to communicate and the limitation to socially interact adequately are generally the most noticeable aspects. (APA, 2013, De Bildt et al., 2007; Doreleijers, et al., 2006). For persons on the autism spectrum this is a lasting burden that is demonstrated in various social situations and in different ways (Wing, 1997, 2001). Music therapy is frequently used in treatments with children and young people who are diagnosed with ASD. Several studies report progress in social interaction by music therapy treatments. Ghasemtabar et al. (2015) showed that music therapy had a strong effect (standardized mean difference = 1.06) on the social skills of 13 children on the autism spectrum compared to 14 controls. This finding is consistent with results from similar studies, for example: Vaiouli et al. (2015) found an improvement in social skills, Katagiri (2009) showed that music has a positive effect on emotional understanding, Kim et al. (2008) showed that the tool of music is used to make contact and provide the child with unique opportunities to communicate nonverbally, Kern et al. (2007) found that individually adapted songs seemed to help in social interaction, Wimpory et al. (1995) showed that when musical interaction therapy is used an improvement is seen in engagement and making eye contact, and Edgerton (1994) revealed that improvised music therapy leads to a great improvement of communicative responses. During music therapy, the therapist stimulates the child's social and communicative skills through musical interactions, especially using the musical elements as a means of communication (Arezina, 2011)). Social interaction is the reciprocal process in which children take social initiative and respond to social stimuli from their environment (Shores 1987). Because children on the autism spectrum often find it difficult to properly understand or interpret verbal social communication, (non-verbal) music may offer a more accessible way of interaction (Geretsegger et al., 2014). A musical (non-verbal) message does not have to be converted into verbal language in order to feel what someone means (Honing, 2009), and so the focus can remain on the social aspects of communication.

Generalizing learned social skills seems to be difficult for many people with ASD. They often find it complicated to apply what they have learned in one situation to another. This is because details are so prominent that the whole picture is lost. The more one tends to notice and magnify small differences between situations, the more difficult generalizing becomes; after all, generalizing requires that one notes the similarities between situations (Brown and Bepko, 2012; Rimland, 1964). In most cases, this may be because of a weak executive functioning, that is, being able to assemble separate elements into a meaningful whole. Most people on the autism spectrum have difficulty doing this and therefore perceive the world in a fragmented way, because of their strong tendency to focus on details (Ozonoff et al., 2005). For these children, improving executive functioning is one of the most important elements for a treatment to be successful (Karkhaneh et al. 2010; Vismara & Rogers 2010; Wass & Porayska-Pomsta, 2013). Moreover, it is crucial that the skills someone learns in therapy can also be used in daily life situations, i.e. outside of the therapeutic setting. In view of this, it is unfortunate that the literature neglects whether learned social skills of children with ASD during music therapy also generalize to other social contexts.

It is important to note that not all studies point to a positive effect of music therapy for children on the autism spectrum. In a Random Controlled Trial (RCT) among 364 children with ASD aged 4 to 7 years, based on the Autism Diagnostic Observation Schedule (ADOS), Bieleninik et al. (2017) found no difference in social communication skills between the experimental group who participated in different types of music therapy and the control group receiving standard care. Broder-Fingert, Feinberg, and Silverstein (2017) argued that the lack of effect could be attributed to an overly heterogeneous experimental and control group. Another complication was the wide range of music therapies that were included. To overcome this, studying one well-described type of music therapy at a time seems a more promising approach. In addition, it is important to use a stepwise research program that begins with finding initial evidence of the effect of a particular music therapy, followed by an experimental study with a carefully matched experimental and control group (Robey, 2004).

As to the need for a well-described type of therapy, many studies on the effects of music therapy on children on the autism spectrum show a wide variety of work forms that are not well-defined. From the long history of music therapy with children and adolescents on the autism spectrum who experience severe problems in the social and community areas (Reschke-Hernandez, 2011), we know that this treatment method uses the medium of music to create experiences that stimulate people's developmental process. The therapist identifies and interprets the child's response to music, deploying the elements of musical measure, rhythm, melody, harmony and sound through the use of musical instruments, voice, musical game forms, improvisation and existing repertoire (Kern & Humpal, 2012). In practice, we see that music therapists often find it difficult to put into words what they actually do. In Chapter 2 we made an inventory of which forms of work and elements seem crucial within the music therapy treatment of children on the autism spectrum (Pater & Van Yperen 2016). In addition, video analysis with music therapists in combination with the Delphi method was used to reach consensus on which elements of the therapy were essential to improve social interaction of children on the autism spectrum. Whereas music therapists initially indicated that they work mainly intuitively, our studies revealed that there were great similarities in the working methods for these children. These findings were used to develop the well-described and manualized (Pater, 2015) (PMTP), a standardized therapy offered in the home setting (Pater, 2016).

In this study we examined the developmental progress in social interaction of children on the autism spectrum using PMTP. The Papageno Foundation is committed to support children and young people (aged between 3-30 years) on the autism spectrum in their development. This foundation, founded by Aaltje van Zweden and her husband, Dutch conductor Jaap van Zweden, offers help to children and young people on the autism spectrum using music therapy (<https://www.papageno.nl/en/>). Music therapy is one of the foundation's activities. The Foundation's ideal is to offer music therapy in a home setting. In this way, the therapy takes place close to the family, it saves stressful travelling for the child, the child remains in her or his safe environment, and there is greater involvement of the parents. In addition, other family members also directly see and/or hear what is happening during music therapy and experience the effects. Müller & Warwick (1993) looked at the influence of involvement of families of children on the autism spectrum aged 3-14 years. The results showed that turn-taking skills

increased and stereotyped behavior decreased. Another study in the family home was by Pasiali (2004). Three high functioning children on the autism spectrum aged 7-9 years participated. The music therapist worked one-on-one with the children. All problem behaviors decreased. Two of the three parents continued to use the song, which was used during therapy, to help their child to keep the improvements. Yang (2016) showed that music therapy in the home situation caused children to take more initiative verbally. In addition, parent-child synchronization also improved. The results seem to indicate that music therapy in the home situation appears to facilitate the interaction between parent and child.

The PMTP consists of five element categories that have been incorporated in the manual: Format, Communication, Musical Elements, Connect and Challenge. These elements emerged in a study of the PMTP's working elements (see Chapter 4). "Format" describes creating structure during therapy to give the child a feeling of control and predictability. The therapist ensures a recognizable structure, both during the sessions and throughout the overall therapy. "Communication" represents the therapy that includes the use of various methods of communication to make contact with the child. These are verbal communication, non-verbal communication and communication via music or a combination thereof. The main category, "musical elements", describes how music is specifically used for adaptation. The elements dynamics, rhythm, tempo and volume can be adjusted to what the child is doing or to challenge other behavior. The therapists play with timing to make it easier for the child to take the initiative. To "connect", the therapist connects to the child's interests by using topics, themes or musical instruments that he/she knows will appeal to the child. For 'challenge' the child is stimulated to exhibit different behavior when a change is made or something new is added to the music therapy. The therapist uses unpredictability, a surprise, to elicit a reaction or change. This may be a surprise in the music, such as a sudden change of tempo.

To find indications whether the program could trigger a therapeutic effect, a multiple case study was performed over a 20-week period among 10 children who were measured on a weekly basis on their social interaction before, during, and after therapy (see Chapter 5). This study showed that the children significantly improved on multiple aspects of social behavior, compared to their baseline condition (Pater, Spreen, & Van Yperen, 2020). These improvements concerned aspects that include "can deal with changes", "behavior has been adjusted", "is able to communicate verbally", "can be considerate of other people", and "takes initiative". Although this study was based on a single case experimental design with a standardized intervention manual and a description of the target behavior, the research group ($N=10$) remains small. Moreover, an important next question is whether improvements in social interaction also generalize beyond the therapeutic setting.

In this study, we examine to what extent the trend of improved social interaction we observed in our multiple case study can be replicated in a larger group of children on the autism spectrum. In addition, this study aims to determine whether improvements in social interaction during therapy can also be observed in situations outside the therapeutic setting. By having different informants from different social contexts who assess the development of the social interaction of 40 children on the autism spectrum, we aim to answer the question: does the

social interaction of children on the autism spectrum improve when PMTP is offered, and is this improvement also visible outside the treatment setting?

Method

Research design

A multiple-informant design was set up to observe and monitor the development of the more adequate social interaction skills of children in different contexts. By involving informants from the social network (e.g. parents, teachers, family) of the child, behavioral changes in daily situations can be monitored. This way the changes that occur prior, during, and after the intervention can be measured in their natural environment (Spreen, 2009). By opting for a method involving a pre-test/post-test design with multiple informants, it is possible to collect detailed information that shows to what extent the improved skills during the music therapy sessions also improve outside the treatment setting.

Intervention

The PMTP consists of 20 music therapy sessions of approximately 40 minutes at home. PMTP starts with an introduction phase of three sessions, in which the therapist will offer working methods that follow the child, especially in the beginning. The therapist follows with the pace, volume, and choices of the child, as described in the intervention guide under empathic and structuring work forms. The aim is to create a relation of mutual acceptance, that forms the basis of the treatment phase. For example, in this phase the therapist copies the child's musical expressions by adjusting his own pace and dynamics to those of the child. This way, the music therapist communicates with the child on a musical level, often by improvising with the child. This working method often creates a safe setting in a short period of time, in which the therapy can develop further.

In the next phase, the other 17 sessions, the therapist can also use a more stimulating approach and challenge the child to discover and do new things. Where the music therapist mainly connects and goes along with the child during the first three weeks, he/she will add more and more challenges over time. The therapist will do this by inducing pace changes, allowing for moments of silence, or varying widely in volume.

During a session, the therapist continuously adjusts to the child. This is done by using improvisation techniques that allow the music therapist to continue to connect with the child and thus to stimulate and challenge them to bring about change. From this connection, the therapist can challenge the children to show them a different kind of social behavior. Previous research (Pater, Van Yperen & De Graaf, 2019), Chapter 4, has shown that connecting with the child is an important precondition to change social behavior. Only if the therapist works in connection, the child will have the focus and motivation to learn new behavior. When a child shows new behavior, the therapist will compliment and encourage the child to repeat it so that they can start to adopt this behavior. Examples of new social interaction are, for example, more eye contact, more social interaction, better alignment, better cooperation and more flexibility.

Because of the use of music within the PMTP treatment, elements from the therapy can easily be integrated into daily practice. The progress within the therapy can be shared by the therapist with other people involved and these people can be given tools that work well within as well as outside the music therapy. Because the PMTP is offered at home, it is easier to apply the skills learned in daily life (Smith et al., 2015; Volkmar et al., 2014). Parents were actively involved in the music therapy, either by attending the sessions or by regular and intensive evaluation using video recordings. For each therapy series, the choice of parent participation was examined and discussed with the parents.

Participants

In this study, 40 children on the autism spectrum participated. Further details on this group are described in de Results section. The observations were made by persons from the child's personal network – a total of four persons who were divided into two groups: the inner circle and the outer circle. The inner circle was defined by a parent and the music therapist being the informants. Since PMTP is a home-based program, the inner circle experienced the music therapy sessions from up close, while the parent was also able to observe the behavior daily. The outer circle was not directly involved in the music therapy and included, for example, a schoolteacher, a family member, babysitter, or neighbor, who were able to observe the behavior of the child at least twice a week.

The PMTP was given by 10 music therapists meeting the following criteria: completion of a music therapy course; being a member of the quality register by the Dutch Register for Practicing Therapists; and having at least two years' experience with the target group. In addition, these therapists have taken a training in giving the PMTP. During the treatment phase, they kept a weekly logbook in which the working methods they have used are reported. These logs were used to check if the therapists were actually following the PMTP methods.

Ethical considerations

The parents and/or legal guardians of the children were informed about the study and were asked to give their written consent for the participation.

This study dossier was presented to the Regionale Commissie voor Patiënten Onderzoek (RTPO - Regional Committee for Patient Studies) with document number RTPO 1042. The Committee concludes that the study does not fall within the scope of the Medical Research involving Human Subjects Act (WMO). The study concerns observational research, making use of questionnaires for the parents and/or guardians and video footage made of the children.

Outcome measures:

The Children's Social Behavior Questionnaire (CSBQ) and Social Responsiveness Scale (SRS) were used to operationalize the concept of social interaction and the different skills involved. For both questionnaires the Dutch version was used, i.e. VISK and SRS.

CSBQ

The CSBQ (Luteijn et al., 2007) was developed with the intention to describe the problem behavior of children with (milder) forms of pervasive development disorders. This questionnaire includes a wide range of problems in various development domains, mostly focusing on social problems. A three-point scale is used to assess the 49 statements of the assessment that can be completed by the parents/caregivers themselves where 0 indicates 'not applicable at all', 1 'a little/sometimes applicable', and 2 'clearly/often applicable'. The Dutch Committee for Testing (COTAN) assessed the reliability and the validity of the definitions of this questionnaire to be adequate. The internal consistency (Cronbach's alpha) of the CSBQ lies between 0.76 and 0.90 (Hartman et al., 2006).

The CSBQ questionnaire measures the following 6 aspects (subscales):

1. Emotion/behavior is not optimally attuned to the situation (11 items).
2. Tendency to withdraw from social situations, little need for contact (12 items).
3. Orientation problems, insufficient "automatic" orientation towards time, place, activity or person (8 items).
4. Difficulty understanding and sensing social information (7 items).
5. Stereotype movements and responses to sensorimotor information (8 items).
6. Fear of change and resistance to change (3 items).
- Total score, social problem behavior (49 items)

SRS

The SRS (Roeyers et al., 2015) maps the dimensions of interpersonal behavior, communication, and repetitive/stereotyped behavior, which are characteristic of disorders in the autistic spectrum. Based on 65 statements, parents and caregivers who have sufficient information of the child's current behavioral patterns and development history can use a four-point scale to assess the child's social interaction where 0 means never true, 1 sometimes true, 2 often true and 3 almost always true. The internal consistency of the total score of the SRS shows a value of Cronbach's alpha = .94 for children on the autism spectrum, which is 'good' (Roeyers et al., 2011). The conceptual validity was assessed as unsatisfactory because the description of the five scales was insufficiently substantiated by research. The criterion validity was assessed as good. Because there are no adequate alternatives for this instrument, we decided to use this in our research despite COTAN's assessment.

The SRS measures the following 5 aspects (subscales):

1. Social awareness: the ability to recognize social cues addressing the sensory aspects of reciprocal social interaction (8 items).
2. Social cognition: the ability to interpret social cues addressing the cognitive-interpretative aspects of reciprocal social interaction (12 items).
3. Social communication: the ability to display expressive social communication addressing the motor aspects of reciprocal social interaction (22 items).
4. Social motivation: the extent to which people are generally motivated to engage in social interpersonal contact and are hindered by fear and inhibition on a social level and a lack of empathy (11 items).

5. Autistic preoccupations: the presence of stereotypical behaviors that characterize ASD and very limited areas of interest (12 items).
 - Total score, social limitations ASD (65 items)

Procedure

The music therapy sessions were given in the home situation at a certain time during the week. In a few cases, the therapy was given at school. In that situation, the teacher was not involved in the therapy session. Parents were actively involved in the music therapy, either by attending the sessions or through a regular and intensive evaluation using video recordings.

The child's social network was used to compose the observation panel. Individuals were sought who could observe the child in different settings in order to observe the behavior both within and outside of therapy. As soon as the observation panel for each child (i.e. the parent, the therapist, the teacher, family member, neighbor, etc.) was formed and written consent was obtained, the panel received the CSBQ and the SRS. The CSBQ was completed three times: before the start of the program, after the 10th session, and after the 20th session. The SRS was completed two times: before the start of the program, and after the 20th session. We asked the panel members to complete the lists within the same week. A reminder was sent after 7 days in case of no response. The SRS only took place twice because this questionnaire looks back in time according to the user manual and longer time period. If this would be given three times as well, the period in between would be too short.

This study also included the cases studied in the earlier multiple case study of Chapter 5 (Pater, Spreen, Van Yperen, 2021). In that study, only the observations of the parents were included.

During the treatment, the therapist uses an intervention guide (Pater, 2016). This guide outlines the working methods to be used during the intervention. The techniques described in the intervention manual are consistent with Kenneth Bruscia's (1987) improvisational techniques. The following techniques are used: empathy techniques, structuring techniques, incitement techniques, techniques to take back the lead, techniques concerning intimacy, procedural techniques, referral techniques and techniques to explore their emotions, among others. In addition, structured methods are also offered that use musical parameters and the various aspects that define music (sound, rhythm, melody, dynamics and form) (Hegi1996) to facilitate change. Various working methods are described for the various sub-goals per treatment phase. Based on the child's needs, the therapist can select the most suitable working methods for each phase.

Statistical analysis

A power analysis using the GPower (Bruin, 2006) program indicated that a medium effect size (0.4) with alpha level of 0.05, needed at least 18 participants. If we assume a small effect size (0.25), the minimum was set to 43. Based on the power analysis, we included 50 participants.

For each single child, the agreement between the multiple observers was analyzed by the Gower's similarity measure (Gower, 1971). The Gower coefficient takes a value between 0 (no agreement between evaluators) and 1 (perfect agreement between evaluators). A high agreement between

observers from the inner circle and the outer circle gives an indication of the extent to which the observed changes in the therapy correspond to the changes in the other contexts. The interpretation of values for the Gower coefficient is usually: 0.00 – 0.40: ‘weak agreement in the assessment’, 0.41 – 0.60: ‘moderate agreement in the assessment’, 0.61 – 0.80: ‘fair agreement in the assessment’, 0.81 – 1.00: ‘adequate agreement in the assessment’, and we follow this standard (Zegers, 1989).

Scores of the inner circle, consisting of the parent and therapist, were obtained by calculating the average. If only one respondent had given a score, this score was used. This was possible because correlation between the Gower’s of therapist-parent scores and those between family member-teacher have been found to correlate high (>.82). The same procedure was followed for the outer circle, which consisted of a family member and teacher.

For the CSBQ, the nonparametric Friedman Test was used to detect differences across the three measurement times. The paired samples t-test was applied to the SRS data to detect differences between the start and end of the program. By taking the measurement errors of the instruments into account and comparing the initial and final measurements, for each child the Reliable Change Index (RCI) was calculated to decide whether the child showed an improvement or decline in social behavior (Jacobson & Truax, 1991). The RCI makes it possible to examine, on an individual level, whether observed changes are or are not the result of errors of measurement. If not, then progress or deterioration is a reliable outcome. For interpreting the scores, we use the rule of thumb according to the Dutch Youth Care (VNG et al., 2016), which indicates that we may speak of a strong (i.e. reliable) change when the difference between initial and final measurements is greater than or equal to an RCI of + or -1.96.

Results

We selected fifty children (39 male, 11 female) between the ages of 4 and 16 (mean 6.8 years) having an IQ \geq 80. All were diagnosed with ASD according to the DSM-V criteria by a child psychiatrist or a clinical psychologist / remedial educationalist. All families had the Dutch nationality and were Dutch speaking. They were approached to participate when they applied for music therapy at the Papageno Foundation. If the specific need of the children was in the area of social behavior, their parents were requested to participate in the study at the time they applied for music therapy at the Papageno Foundation in the period 2017/2018.

Of the 50 children included in the study, two stopped music therapy prematurely and were excluded from the data processing. Data received was screened for completeness. Only when at least 3 observers had completed all measurements, cases were included in the analysis. This way there was no missing data for both the inner circle and the outer circle. Eight participants were excluded because of missing measurements from more than one observer. There appeared to be no systematic bias concerning the dropped-out participants; this is because dropouts were registered with different reasons. Some examples include illness of family, moving to another city, changing school. As a result, 40 children with ASD were eligible for analysis. There was a 100% response from both the parents and the music therapist in the inner circle. However, for the outer circle, the response was 75% for the teachers and 87.5% for the other observers. Also, all included children had attended all 20 sessions of music therapy.

Because the previously used cases from the multiple case study were also included in this study, we tested whether the parents’ scores on the CSBQ from that $N=10$ study differed from the newly collected parents ($N=30$). The mean total t-test score for the $N=10$ for the first measurement time was 47.1 (SD 10.62) and for the third measurement time was 37.9 (SD 12.63). For the parents ($N=30$), the mean total score for the first measurement moment was 48.7 (SD 13.51) and for the third measurement moment was 37.5 (SD 13.60). On the subscale level, there were no notable differences either. There was no significant difference in the averages of the two groups. This means that the positive outcomes observed in the earlier $N=10$ study were replicated in the larger group.

Table 1. GOWER CSBQ & SRS, between inner and outer circle

Time	CSBQ			SRS		
	Mean	Range	SD	Mean	Range	SD
T1	0.85	0.69 - 0.99	0.074	0.80	0.58 - 0.99	0.106
T2	0.88	0.59 - 0.99	0.091	-	-	-
T3	0.91	0.75 - 0.99	0.070	0.86	0.57 - 0.99	0.107

To investigate whether the findings within the therapy generalize in the outside settings, first the Gower coefficient was computed to examine the degree of agreement between the observations in the inner and outer circle. Table 1 shows the average Gower score of the CSBQ total and SRS total based on the various moments of measurement. The scores clearly indicate a high degree of agreement between the inner circle and the outer circle. Scores on the CSBQ show a good to very good agreement with an average value of .88. The SRS also has a good agreement with an average of 0.83. Because the observers make their observations in different environments, this high level of agreement indicates that the informants roughly observe the same degree of behaviors.

To observe a high agreement between the inner and outer circle does not give information about the trend over time. Therefore, a Friedman test for the CSBQ was performed that allowed comparison of the three measurements. (see table 2).

Table 2. CSBQ scores per subscale with *p*-values

Inner Circle									
Scale:	T1		T2		T3		Friedman Chi-Square	Asymp. Sig	
	Mean	SD	Mean	SD	Mean	SD			
1. Behaviors not tuned to situation	10.59	3.73	8.81	4.01	7.33	4.02	42.95	<.001	
2. Withdrawal	11.51	4.50	9.79	4.18	7.74	3.74	47.72	<.001	
3. Orientation problems	9.28	2.79	8.34	3.17	7.31	2.79	32.84	<.001	
4. Difficulties understanding social information	8.91	2.55	7.58	2.57	6.69	2.05	29.23	<.001	
5. Stereotyped behaviors	7.23	3.50	5.65	3.30	4.58	2.99	47.84	<.001	
6. Fear of and resistance to change	5.50	1.17	2.84	1.20	2.33	1.31	35.01	<.001	
Total	51.01	13.30	42.91	13.81	35.96	12.27	56.15	<.001	
Outer Circle									
Scale:	T1		T2		T3		Friedman Chi-Square	Asymp. Sig	
	Mean	SD	Mean	SD	Mean	SD			
1. Behaviors not tuned to situation	8.39	4.82	7.25	4.13	6.41	3.97	14.47	.001	
2. Withdrawal	10.39	4.74	9.09	4.49	8.23	4.49	29.06	<.001	
3. Orientation problems	7.86	3.59	6.54	3.66	6.21	3.48	14.44	.001	
4. Difficulties understanding social information	7.16	3.24	6.38	2.53	5.91	2.74	14.91	.001	
5. Stereotyped behaviors	6.14	4.09	5.21	3.45	5.1	3.8	12.76	.002	
6. Fear of and resistance to change	2.94	1.14	2.56	1.28	2.61	1.31	2.66	.264	
Total	42.88	14.54	37.02	13.11	34.48	14.09	40.4	<.001	

Within the inner circle, the total score as well as each subscale was significant ($p < 0.05$) implying that progress was observed by the informants. The same applied to the outer circle with the exception that no progress was found for scale 6 (Fear of and resistance to change).

We see in Table 2 that the results of the outer circle are significantly somewhat lower than those of the inner circle. This may be explained by the setting in which the observations are made. Parents and the therapist probably observe different degrees of behavior than the teacher.

If we take a look at the subscales of the CSBQ in more detail, we can see that the strongest progress is on the subscales “behavior is not optimally adapted to the situation” and “tendency to withdraw from social situations”.

Table 3. individual SRS scores per subscale with p -values

Inner Circle						
Scale:	T1		T3		<i>t</i>	Sig. (2-tailed)
	Mean	SD	Mean	SD		
1. Social awareness	13.59	2.78	11.55	2.36	6.388	<.001
2. Social cognition	20.18	4.72	16.09	4.11	9.937	<.001
3. Social communication	34.30	7.43	27.56	7.04	10.868	<.001
4. Social Motivation	16.94	3.44	12.73	2.76	8.621	<.001
5. Autistic preoccupations	17.68	5.68	13.28	5.72	8.265	<.001
Total	102.68	20.73	81.20	18.82	12.266	<.001

Outer Circle						
Scale:	T1		T3		<i>t</i>	Sig. (2-tailed)
	Mean	SD	Mean	SD		
1. Social awareness	11.98	2.73	10.40	2.86	4.726	<.001
2. Social cognition	17.99	4.90	15.19	4.43	5.452	<.001
3. Social communication	30.75	8.88	25.56	8.76	5.985	<.001
4. Social Motivation	15.03	4.18	13.06	4.45	3.711	.001
5. Autistic preoccupations	15.35	5.92	11.88	6.61	4.560	<.001
Total	91.09	23.08	76.09	23.82	6.929	<.001

Table 3 shows the results of the paired samples t -test for the SRS scores. This analysis also points at an improvement in social interaction when music therapy is offered. Both in the inner circle and in the outer circle we see a significant ($p < 0.05$) improvement at group level on all sub scales and the total score. The number of significant progressive results of the CSBQ and the SRS coincide substantially. Again, we see in Table 3, that the results of the outer circle are significantly lower than those of the inner circle, as in Table 2, this can be explained by the different setting in which the observations took place.

As indicated previously, to decide whether there is an improvement or decline per single child, the Reliable Change Index was calculated (RCI: Table 4).

Table 4. Distribution of RCI

Scores									
VISK									
Observer:	Declined	T1 - T2		Declined	T2 - T3		Declined	T1 - T3	
		Stable	Improved		Stable	Improved		Stable	Improved
Inner circle	12.50%	2.50%	85.00%	0.00%	0.00%	100.00%	17.50%	2.50%	80.00%
Outer circle	15.00%	2.50%	82.50%	7.50%	2.50%	90.00%	20.00%	0.00%	80.00%

SRS									
Observer:	T1 - T3			Decreased RCI < -1.96, Stable RCI between -1.96 and 1.96, Improved RCI > 1.96					
	Declined	Stable	Improved						
Inner circle	2.50%	0.00%	97.50%						
Outer circle	0.00%	0.00%	100.00%						

If we focus on the RCIs between the initial measurement (T1) and final measurement (T3) of the CSBQ, we observe that 80% (30 children) of the children clearly improved according to the inner and outer circle. By closer inspection of the data, we see that, according to the CSBQ, the music therapists have noticed significant progress in all (100%) children. The teacher observed the least progress in the children – in their view 10.34% remained stable, and 86.21% made great progress.

For the SRS, the inner circle noticed reliable progress in 97.50% children, while the outer circle noticed significant progress in all children. As with the CSBQ, we see that the scores of the different observers are close together, which indicates that the acquired skills that are observed in therapy are also observed in situations outside the therapeutic context.

Discussion

Key results

In this study, we focused on the question to what extent social interaction of children on the autism spectrum improves by and during the music therapy program ‘PMTP’. We focused specifically on the extent to which the behavior of the children in the sessions were also generalized to other contexts. 40 children on the autism spectrum were monitored during 20 sessions of PMTP. We created an observation panel with people from the children’s environment, who evaluated their social interaction at three and two different times by completing, respectively, the CSBQ and SRS questionnaires. The analysis of both instruments showed significant improvement in social interaction for the majority of the children. The social interaction during music therapy also showed significant progress for most children.

Interpretations

Therapist’s perceptions of the progress may be biased because he or she may want to see progress as an effect of his treatment. The same reasoning may apply to parents as well, because they may want to see progress among their children. But although the progress seen by participants in the outer circle is somewhat lower than those of the parents and therapists in the inner circle, it is still quite high. And since the process of generalization for children on the autism spectrum is often difficult (Brown & Bepko, 2012; Rimland 1964), it is striking that the RCI values of all observers who have reported substantial improvement in different aspects of social interaction are noticeably close to each other. For the CSBQ, the percentages lie between 88.24% and 100%, and for the SRS between 93.34% and 100%. This seems to indicate that all these observers see progress when music therapy is applied, and that this improvement can be observed in different contexts.

In the previous multiple single case study ($N=10$), of Chapter 5 (Pater, Spreen & Van Yperen, 2021) we saw that the children on the autism spectrum showed a substantial improvement in social interaction after 20 weeks with the same music therapy program PMTP. In the study presented here, we used a larger group of participants to see whether we could confirm the same effects. The results of this study support the results we found earlier. After 20 weeks of music therapy, children showed a significant improvement in social interaction compared to the initial measurement. It is also in line with previous studies that showed that music therapy has a positive effect on the social interaction of children on the autism spectrum. For example, Ghasemtabar et al. (2015) who in a Controlled Clinical Trial (CCT) saw a significant increase in social skills in the group that was offered music therapy, Vaiouli et al. (2015) observed an improvement in social skills with improvised music therapy, Gattino et al. (2011) showed that the music therapy intervention gave an improvement on non-verbal communication, and Wimporoy et al. (1995) showed that when using musical-interaction therapy there was an improvement on the involvement and the making of eye contact.

A possible explanation of the active elements that can be associated with a change in behavior is that the therapist can challenge the child to show different behavior in a non-verbal way, through the use of music. Connection seems to be an important condition for achieving this change, it ensures the children attention and motivation to learn new behavior. The versatile characteristic of music makes it a suitable medium for working with children and young people with autism, in order to provoke the change in social behavior. It offers the possibility to move and change quickly, both within and between the various musical elements. It allows the therapist to adapt to the child quickly and to vary between connecting and challenging. Music is a very flexible medium that may provoke varying social behaviors as well (Pater & Van Yperen 2017). An example of this is when the therapist is playing a fast rhythm together with the child and suddenly drops an abrupt silence in it. Often you will see some confusion at first, which then raises an alertness of the child for new play.

Limitations

This study does have several limitations. Because we wanted to take the workload on the members of the children's network into account, weekly measurements during the course of the therapy were found to be too time-consuming. Such weekly measurements could have provided more precise insight into the similarities or differences at times when the various observers notice any changes. We also found that when a participant was registered for music therapy, parents wanted to start therapy as soon as possible. As a result, and contrary to our multiple single case study of Chapter 4 it was not possible in many cases to perform a baseline measurement that would provide insight into the level and pace of development of the child prior to therapy. In this Chapter, we therefore chose to take the start of therapy as the first measurement. At the time of T1, the observers have completed the CSBQ and SRS. Both questionnaires work retrospectively, which gives an impression of the behavior before the start of music therapy. And because four observers observed the child in different contexts and these observations correlated closely, it gives a consistent picture of the initial situation.

Although we strived to limit the burden of a large number of measurement moments, we nevertheless noticed that it was difficult to obtain a response from all observers. In this study, we chose to only include cases with a complete set of data from at least three observers (including the parents and music therapist). It was especially difficult for the teachers to provide all measurements because they often indicated that they were unable to fill in their answers due to the high workload. It would be a good idea to develop a short and validated questionnaire to measure social interaction that contains less items, so that it can be more easily completed by observers.

During the period when the PMTP was offered, the participating children were not deprived from any other assistance they received. This may have influenced the developments observed by the observers. However, none of these forms of help were started concurrently with the PMTP. This may be better monitored in a future RCT design.

No child satisfaction measurement was conducted during this study. Including these measures in future research could contribute to the further development of the treatment module. It provides insight into which elements are experienced as pleasant and which may need to be adjusted. These results can also be used to test whether they correspond to the possible active elements that have emerged. It is likely that these can be further refined and used more effectively with the help of a child satisfaction survey. A recommendation would therefore be to use the measurements to enter into dialogue with (groups of) young people and parents about which elements appeal to them and seem to be particularly effective and what improvement suggestions there are (Van Yperen, Veerman & Bijl 2017).

Generalizability

The PMTP is completely attuned to the practice as offered by the Papageno Foundation for Music Therapy. There is room for heterogeneous characteristics of the target group in terms of age and level of functioning. The different ways in which parents are involved can also be taken into account. However, it should be considered that the diversity within the target group makes the research more difficult. Perhaps it is necessary in future research to make subgroups within the research population.

Although our study design deviates from the more commonly used multiple single case study and RCT, which remains a methodological limitation, we have been able to intensively monitor 40 children at an individual level with a pre-test/post-test design by mapping the changes at different times from multiple informants. In this study, we noticed that there was a high level of agreement between the assessments. Based on these findings, we can conclude that the use of multiple informants provides a reliable picture of the behavior both inside and outside of the therapeutic setting. In other words, the change in social interaction that the children show during therapy is also observed by informants outside of therapy.

We did see that teachers reported a lower score, but this does not necessarily mean that the developmental progress can be called into question. Oosterlaan and Veerman (2014) argue that the low concordance cannot always be regarded as the unreliability of the instrument. Low correlations could also indicate situation specificity of how characteristics express themselves

(e.g. a child can behave very differently at home than at school) and the informant specificity of an assessment (teachers have a different relationship with a child than the parents; see also Kroes, Veerman & De Bruyn (2000)). As we noted before, although in our study the teachers reported a lower score, in most cases they also observed a clear improvement in the individual behaviors. In future research, it is important to investigate – for example by video analyses - if these improvements reported by teachers were assessed on the basis of the same behaviors of the children as those observed by therapists and parents. This would lead to a more in-depth understanding of how the children express their improved skills in different situations.

Conclusion

This successive research on the earlier multiple case study of Chapter 4 provides first indications that the PMTP program is promising on having a positive effect on the social interaction with children on the autism spectrum. For further substantiation, it would be particularly worthwhile to test these effects with more demanding experimental research, for example, by using a RCT, i.e. with a control group. In view of the heterogeneity of the children on the autism spectrum, it should be considered to match the cases in the experimental group with cases in the control group and to exclude cases that found no matches. This is to ensure that – even after randomization – the experimental and control group do not differ on important characteristics (Pater & Van Yperen, 2017). Furthermore, it is necessary to develop a less time-consuming instrument to increase the number of measurement moments and improve the response rate in practice. It is also important to train music therapists to engage in effective reporting. If they keep a clear log of the therapy, best practice can be collected. In addition to a substantiating RCT, for example, this would provide a great deal of qualitative insight regarding the effects in daily practice.