

University of Groningen

Music therapy for children with Autism Spectrum Disorder

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DOI:
[10.33612/diss.202791160](https://doi.org/10.33612/diss.202791160)

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Document Version
Publisher's PDF, also known as Version of record

Publication date:
2022

[Link to publication in University of Groningen/UMCG research database](#)

Citation for published version (APA):
Pater, M. (2022). *Music therapy for children with Autism Spectrum Disorder: development and first evaluation of the Papageno Music Therapy Program*. [Thesis fully internal (DIV), University of Groningen]. University of Groningen. <https://doi.org/10.33612/diss.202791160>

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CHAPTER 5.

The developmental progress in social behavior of children with Autism Spectrum Disorder getting music therapy: a multiple case study

This Chapter is published in:
Pater, M., Spreen, M., & van Yperen, T. (2021). The developmental progress in social behavior of children with Autism Spectrum Disorder getting music therapy. A multiple case study. *Children and Youth Services Review*, 120, 1057-1067.
<https://doi.org/10.1016/j.childyouth.2020.105767>

Abstract

Background: Music therapy is claimed to have a positive effect on the social behavior of children with Autism Spectrum Disorder (ASD). However, in most studies the target behaviors and the interventions are insufficiently defined and described. Also, it is often doubtful whether accurate research designs were performed to study the intervention to full satisfaction.

Objective: To answer the question whether the development of social behavior of children with ASD has improved during and after finishing the intervention of the ‘Papageno Music Therapy Program’ (PMTP).

Method: A multiple case study design was used, with a baseline and intervention phase. Ten children were observed over a period of 23 weeks. The social behavior was rated prior, after 10 weeks and at the end of the therapy by the Questionnaire for the Inventory of Social Behavior of Children (VISK), as well as on a weekly basis by the Social Behavior Questionnaire (SBQ).

Results: This study shows that during 20 weeks of music therapy sessions the development of the social behavior of children with ASD has accelerated. At least five children had a moderate improvement in the areas of ‘makes eye contact’, ‘is able to focus for five minutes on a game’, ‘is able to cope with changes’, ‘behavior has adapted to the situation’, ‘communicates verbally’, ‘is able to focus on something together’, ‘is able to take another person into account’ and ‘takes initiative’ from the SBQ. The largest progress, with three children showing a strong improvement, was found in the areas of ‘is able to focus on something together’ and ‘takes initiative’. The findings on the social behavior were partly confirmed by the VISK scores. Additional statistical analyses of these aspects show a significant trend in the data that indicates growth that may be associated with the intervention phase.

Conclusion: This study confirms previous studies indicating that during music therapy the social behavior of children with ASD improves, compared to the developmental pace prior to therapy. Although this study was performed with a standardized intervention manual, a thorough description of the target behavior, and a rigor multiple case study design with a baseline and control condition, a larger study, preferably with an alternative intervention offered to a control group, is required to attribute the results more specifically to the PMTP and its elements.

Introduction

An Autism Spectrum Disorder (ASD) is a qualitative limitation in social communication skills. Furthermore, the disorder is about stereotype patterns in the behavior, interests, and activities (APA, 2013; De Bildt et al., 2007; Doreleijers et al., 2006). The restricted capacity to communicate and the limitation to socially interact adequately are generally the most noticeable aspects. For persons with ASD this is a lasting burden that is demonstrated in various social situations and in different ways (Wing, 1997, 2001)

Music therapy is presented as an appropriate approach to stimulate the development of social interaction skills (Geretsegger et al., 2014; Pater & van Yperen, 2017; Simpson & Keen 2011). By attuning the therapy to the behavior and interests of the client, new skills can be stimulated (Carpente, 2016; Geretsegger et al., 2015; Mössler et al., 2013). The American Music Therapy Association (AMTA) gives the following description of music therapy: “Music therapy is the clinical and evidence-based use of music interactions to accomplish individualized goals within a therapeutic relationship by a credentialed professional who has completed an approved music therapy program”. (source: www.musictherapy.org)

Music therapy may provide the opportunity to practice adequate social interaction in an indirect way, namely through music. A study conducted by Ghasemtabar et al. (2015) shows that music therapy has a significant effect on the social skills of children and adolescents with ASD. This is in line with results of previous studies (Edgerton, 1994; Gattino et al., 2011; Katagiri, 2009; Kim, Wigram, & Gold, 2008; Kern & Aldridge, 2006; Kern, Wolery, & Aldridge, 2007; Vaiouli, Grimmet, & Ruich, 2015). Thompson (2012) shows that music therapy has a positive effect on the communication skills of children with ASD. His study shows a small effect ($d = 0.26$) in the areas of speech and language. The studies conducted by Aldridge et al. (1995), Buday (1995), Edgerton (1994), Farmer (2003), Gattino et al. (2011) and Kim et al. (2008) all show that the use of music therapy has a positive effect on the communication skills of children with ASD.

Pater & Van Yperen (2017) concluded in their literature review that many of the effect studies into music therapy and ASD have a number of shortcomings: adequate descriptions of the target behaviors are lacking, interventions are described insufficiently, and often it is doubtful whether the most accurate research designs were performed. To study interventions such as music therapy properly, it is important to have a clear description of the target behavior. What is the focal point of the treatment and what are its objectives? Effect studies into therapy on communication skills have been performed extensively without clear definitions and operationalizations of these skills. In addition, it is essential that the target group on which the intervention focuses has been clearly identified (Van Loon, Van der Meulen, & Minnaert, 2015). When these elements have been distinctly defined, it is important to choose the right instruments to report the effects.

To evaluate the effect of an intervention, Random Controlled Trials (RCTs) are regarded as the golden standard (Sibbald & Roland, 1998; Shadish, Cook & Campbell, 2002). However, studies investigating the effects of music therapy are having problems recruiting sufficient cases for a RCT (Fletcher et al., 2012; Treweek et al., 2013). This may especially be the case for children with ASD due to the enormous heterogeneity of the target group. An RCT with 364 children with

ASD between the ages of 4 and 7 years, based on the Autism Diagnostic Observation Schedule (ADOS) Bielenink et al. (2017) determined no significant difference in social communication skills between the experimental group participating in music therapy and the control group participating in standard care. This finding contrasts with several other studies which show that there is evidence of improvement in social behavior when children with ASD are participating in music therapy (Accordino, Comer & Heller, 2007; Geretsegger et. al., 2014; Pater & Van Yperen, 2016; Reschke-Hernández, 2011; Simpson & Keen, 2011; Wipple, 2004, Wigram & Gold, 2006). Broder-Fingert, Feinberg and Michael (2017) argued that the lack of effect in the study of Bielenink et al. (2017) may be attributed to a too heterogenous experimental and control group. A further complication was the wide range of music therapies included (e.g., interactive music therapy, structured receptive music therapy, relational music therapy). This demands for (1) a more focused approach by studying one well described type of music therapy at a time and (2) using a step-by-step research program that starts with finding the first indications of the effect of a particular music therapy, followed by an experimental study with a carefully matched experimental and control group.

In this study we want to find the first indications by exploring and evaluating whether children with ASD develop more social adequate behavior during a specific music therapy program, called ‘Papageno Music Therapy Program’ (PMTP). According to the model of Robey (2004), the use of multiple case study designs is very suitable for this purpose. Good examples of a similar approach are the studies of Koegel, Singh, and Koegel (2010), Mucchetti (2013), and Schweitzer, Knorth, Van Yperen and Spreen (2020). In this study, it was decided to use a multiple case study design ($N=10$) with a baseline and a control condition. This study design offers us first indications whether social behavior of children with ASD will improve during PMTP, compared to a baseline condition. Moreover, it gives us the opportunity to study individual cases in greater depth by collecting and analyzing detailed information.

Method

Participating children, parents, and therapists

Included were ten children (eight male, two female) between the ages of four and ten (mean 6.7) having an IQ ≥ 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. This foundation, founded by Aaltje van Zweden-van Buuren and her husband, Dutch conductor Jaap van Zweden, offers help to children and young people with ASD using music therapy (<https://www.papageno.nl/en/>).

If the specific need of the children was in the area of social behavior, parents were approached to participate in the study. All parents were informed about the study and asked to give their written consent for participation. None of the ten children received other social behavior improving therapies during the PMTP module. All children attended a school for special needs education where stimulating social behavior is also incorporated in the learning environment.

Because the study concerns observational research, making use of questionnaires for the parents and/or guardians and video footage made of the children, the design was submitted to the *Regionale Commissie voor Patiënten Onderzoek van het Medisch Centrum Leeuwarden* (RTPO - Regional Committee for Patient Studies from the Leeuwarden Medical Centre). The Committee concluded that the study did not fall within the scope of the Act on Medical Research with People (WMO). The study was carried out in accordance with the guidelines and regulations prescribed by the RTPO.

The six cooperating music therapists were subject to the following criteria: Completion of a music therapy course; being a member of the quality register by *Register Vaktherapie* (Register for Practicing Therapists); and having at least two years' experience with the target group. In addition, these therapists have taken a course in giving the PMTP. During the treatment phase, they keep 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.

Intervention

The PMTP consists of 20 home-based music therapy sessions of approximately 40 minutes. During treatment, the therapist uses an intervention guide. This guide outlines the work methods to be used during the module (Pater, 2016). It contains 30 work methods that focus on the improvement of the social behavior of children and adolescents with ASD through music therapy.

The intervention is completely responsive to the children and the therapist's approach adjusted to the child's capabilities. By linking the intervention to the children's capabilities, changes can be realized in the areas where the inhibitions are experienced. During the music therapy the music therapist focuses on unconditional acceptance of the music style and preferences. From this acceptance, the music therapist offers those work methods with which the child can connect. For example, the therapist is copying 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 at a musical level, often by improvising with the child. This work method often creates a safe base in a short period of time, in which the therapy can develop further. Then, the therapist can also use a more stimulating approach and challenge the child to discover new things.

The therapist keeps a log to monitor the child's progress. This log describes which work methods are used in a session and what the results are. The choice of work methods for the next session are adjusted on this information. Each music therapy session has the same structure. They start with the same beginning of the song and end with the same end of the song. In between these sessions the therapist has the freedom to offer different work methods. Hereby the therapist offers known recurrent work methods interspersed with new ones.

The PMTP module starts with a first phase of three sessions, in which the child's behavior is observed. The therapist estimates whether it would be feasible to achieve the treatment goals within the set period of time and what the child's musical preferences are. This observation period is also used to build a relationship between the child and the therapist. At the end of this phase, the therapist writes an observation report in which the findings and the objectives of the therapy are described.

In the second phase, the treatment starts with activating music therapy. Activating music therapy can be defined as jointly making rhythms, melodies, or music within a therapeutic relationship with the objective to obtain developmental improvement. This can be done by applying various methods to make music. Using percussion instruments to produce rhythm is an accessible method that is often used for this reason. Other methods are singing or the use of other instruments. The therapy will regularly focus on improvisation, but it is also possible to use existing compositions. After making music, a conversation with the child is often initiated about the experiences and/or feelings that were a result of making this music. Therefore, with active music therapy, the child goes through an experience by making music. The therapist chooses the work methods that focus on the problem(s) the child is struggling with. Examples are playing, playing together, experimenting, improvising, and fantasizing. Within the active music therapy setting, a large variety of musical instruments may be used. The therapist chooses or changes the instruments that are offered, based on the child's interests and the work methods that are used within the framework of the goals of the treatment. The work methods are focused on aspects related to social interaction. For example, in the music, elements are practiced that are also needed in conversations, such as reflecting behavior. In the music therapy, this taking turns behavior is challenged by the therapist by playing some music, it invites the child listening to it to play something in return, and then to experience how the therapist reacts, and so forth. During the sessions in the treatment phase, the therapist continuously attunes the music to the child. When a connection is made, the therapist can challenge the child to show different types of behavior. A previous study has shown that finding a connection with the child is an important prerequisite to change the child's behavior (Pater, de Graaf, & Van Yperen 2019). Only when the therapy is based on this connection, will the child have the attention and motivation to learn a new behavior. When a child shows new behavior, the therapist will compliment and challenge him to repeat this, so the child can learn to adopt this new behavior.

The third phase of PMTP is the finalization. In the last two sessions the music therapy is completed. In these sessions the most important music activities are repeated and related new behaviors are practiced. This is intended to strengthen the new experiences of the child.

Study design

In this multiple case study, ten children with ASD were measured three times (at the start of the therapy, after ten sessions, and after twenty sessions) with the validated Questionnaire for the Inventory of Social Behavior of Children (VISK) (Luteijn, Minderaa & Jackson, 2007), and twenty-three weekly measurements with the self-constructed Social Behavior Questionnaire (SBQ). We decided to apply two questionnaires to measure social behavior for several reasons:

- For weekly measurements it was expected that the 48-item VISK was too time-expensive leading to nonresponse
- The SBQ is a 10-item questionnaire, and therefore more feasible for frequent measures;
- The VISK evaluates six relatively broad aspects of social behavior, whereas the SBQ items more closely match the specific observable behaviors on which the PMTP is focused to evoke change.

In all, the use of the VISK is intended to measure the general social development of the child as experienced by parents over a longer time period of time, whereas the SBQ intends to measure short term observable behaviors.

As part of repeated measurements with the SBQ, the three weekly measurements before the start of the PMTP module and in the first two weeks of the module were set as the baseline period. This was done because a longer measurement period before starting the program was unacceptable for most parents, while a five-week baseline period may give a more realistic view of the child's behavior. Therefore, the first two weeks of the first phase of PMTP were added to the baseline phase because these sessions focus on familiarization and contain hardly any therapeutic interventions that are expected to lead to change. To validate this, we also analyzed the trends in the data based on three baseline measurements. These trends were consistent with those based on the five baseline measurements. In all, the collected data consists of five weekly baseline measurements and 18 weekly measurements in the intervention phase.

The observations and filling out of both questionnaires were done by the mothers of the children. They had the most contact with the children during the week and could therefore best observe their child's behavior in the home situation. To fill out the questionnaires, the mothers received instructions. The parents did not attend the music therapy physically, but they were allowed to read the therapist's report and could watch recorded sessions.

Outcome measures

VISK

In 2003, the Dutch Committee for Testing (COTAN) rated the quality of the testing material and the manual of the VISK as good, and both the principles of the test construction and the reliability as adequate. The VISK consists of 49 items that are valued on a three-point scale, retrospectively assessing the child's condition over the past months.

The VISK questionnaire measures the following six 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 a 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).

The Social Behavior Questionnaire

Since no suitable validated questionnaire could be found for weekly monitoring social behavior, we decided to compile a short questionnaire. The Social Behavior Questionnaire (SBQ) consists of ten observational behavior items that most closely match the goals of PMTP (Figure 1), on a 'never' to 'always' five categories Likert scale. The mothers had the option to nuance their observations ticking multiple boxes. The average of the scores in these boxes was used to process the results.

Makes eye contact												
never		infrequent			sometimes			often			always	
example:												
never		infrequent			sometimes			often			always	
							X	X	x			

Figure 1, Example score list

The Social Behavior Questionnaire has the following ten items:

1. Makes eye contact
2. Is able to focus for five minutes on a game
3. Is able to cope with changes
4. Behavior has adapted to the situation
5. Communicates verbally
6. Communicates non-verbally
7. Is able to focus together on something
8. Is able to take another person into account
9. Takes initiative
10. Has rigid behavioral patterns

Due to time and feasibility limitations, no other outcome measures could be administered.

Analysis

To test whether the social behavior of the children improved, measured three times with the VISK, the non-parametric Friedman Test was used per sub- and total scale. To test, whether the social behavior of the children improved during the PMTP module, measured weekly with the SBQ, per child the following quantitative analysis techniques were performed: the Nonoverlap of All Pairs (NAP) (Parker et al., 2009), Simulation Modelling Analysis (SMA) (Crosbie, 1993; Borckardt & Nash, 2014), and the Tau-U index (Parker & Vannest, 2011). These techniques assess whether the trends in the data during the intervention phase differ from those during the baseline phase. Because we have a weekly observation, we also look at turning points in the data. This may give us insight into whether and when an effect of the intervention becomes visible. The NAP measures the degree to which a random score of the intervention phase exceeds a random score of the baseline phase expressed by the nonoverlap between all pairs of scores. When the NAP-index is between 0 and 0.65, the improvement compared to the baseline is weak, between 0.66 -0.92 implies a moderate improvement, and between 0.93 - 1.0 a strong improvement (Walker & Snell, 2013). In the analyses we also apply a more tolerant cut off, i.e. a NAP score between 0.8 and 1.0, because for children with ASD social interaction often proves to be a skill that is difficult to train and to acquire (Schertz & Odom, 2007). SMA is applied to assess the changes in means scores between the baseline – and intervention phase, and also to assess which hypothetical trend (slope) in two phases fits the measurements best. We consider two hypothetical trend patterns that we can expect in our study (see Figure 2). In figure 2, for slope 1 we see that, after a stable baseline phase, the development of social behavior accelerates

during the intervention phase. This may indicate a therapeutic effect. Slope 2 shows that, with a significant positive correlation, the trend during the intervention phase does not differ from the baseline phase, which suggests a more stable growth. The SMA analysis shows to what extent the data correlate with the two slopes, whereby the highest correlation indicates the best ‘fit’ (Delsing & van Yperen 2017).

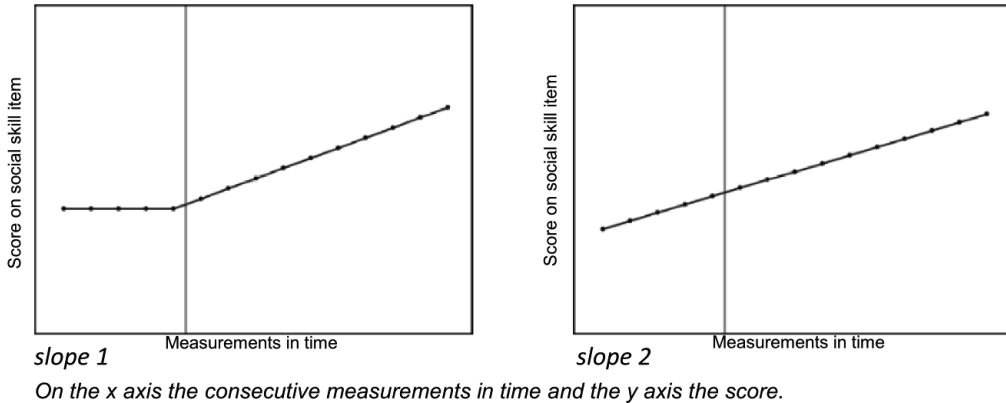


Figure 2, Example trend slope 1 and slope 2.

Tau-U is an index that is used to specifically measure changes in trends between two phases, the baseline (A) and the intervention (B; in our case the intervention and final phase) (Parker & Vannest, 2009). Tau-U is calculated based on the percentage of data that improves over time, taking both the non-overlap between the phases and the trend in phase B into account, after testing the trend in phase A. When the outcome of the Tau-U is significant, this is an indication of an acceleration of development in the intervention phase.

Results

In table 1 the individual scores on the six sub scales and the total score of the VISK are displayed.

The table shows that most children show improvement on the subscales ‘Not optimally tuned behavior’, ‘Difficulty with social information’ and ‘Stereotype movements’. The subscale ‘Fear of change’ shows the least progress; only four children show a small progress. For the total score, based on the Friedman test, we are seeing significant progress for the group of children ($p < 0,05$). At an individual level, children four, seven and nine do not show any or hardly any improvement in their total score.

In order to get a better view of the individual developments we have further analyzed the more detailed case study data with the weekly measurements. As described earlier, the NAP index indicates whether the values in the data during the intervention phase differ from those during the baseline phase by computing the non-overlap of the measurements between two phases.

Child	1. Not optimally tuned behaviour			2. Tendency to withdraw			3. Orientation problems			4. Difficulty with social information		
	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3
1	7	7	5	6	7	4	6	5	2	7	7	2
2	7	7	6	14	10	10	7	5	5	11	8	6
3	14	14	13	7	8	3	10	10	8	9	12	7
4	17	12	13	9	10	10	8	8	9	10	11	10
5	5	2	3	6	3	3	11	8	8	11	9	8
6	5	5	2	7	6	4	7	4	7	7	5	4
7	15	16	18	3	7	6	9	7	11	5	7	7
8	9	9	8	11	12	10	8	7	7	7	6	5
9	15	13	13	6	7	6	4	7	6	9	8	8
10	18	14	11	15	13	14	12	9	10	11	7	7
	Chi-Square 7.118 Asymp. Sig. 0.028			Chi-Square 4.389 Asymp. Sig. 0.111			Chi-Square 3.941 Asymp. Sig. 0.139			Chi-Square 8.941 Asymp. Sig. 0.012		
Child	5. Stereotype movements			6. Fear of change			Total score					
	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3			
1	13	12	9	3	2	0	42	40	22			
2	12	8	9	6	5	5	57	43	41			
3	7	6	4	2	3	0	49	53	35			
4	8	11	9	5	6	6	57	58	57			
5	7	3	4	0	1	1	40	26	27			
6	2	1	1	2	2	0	30	23	18			
7	2	4	2	6	5	6	40	46	50			
8	11	10	9	3	3	3	49	47	42			
9	3	1	1	4	4	4	41	40	38			
10	6	3	3	4	4	4	66	50	49			
	Chi-Square 7.722 Asymp. Sig. 0.021			Chi-Square 1.043 Asymp. Sig. 0.593			Chi-Square 7.538 Asymp. Sig. 0.023					

Table 2, Individual scores per scale of the NAP, SMA a Tau-U.

1. Makes eye contact				2. Is able to focus for 5 minutes on a game				3. Is able to cope with changes				4. Behavior has adapted to the situation			
Child	NAP	Slope 1	Tau-U	Child	NAP	Slope 1	Tau-U	Child	NAP	Slope 1	Tau-U	Child	NAP	Slope 1	Tau-U
1	0.922 **	0.759 *	0.771 *	0.518 *	1	1.000 **	0.742 *	0.781 *	0.557 *	1	0.672	-0.003	0.672	0.061	0.012
2	0.500	0.168	0.184	0.087	2	0.317	-0.141	-0.153	-0.158	2	0.750	0.735 >	0.750	0.708 *	0.534 *
3	0.694	0.605 >	0.577 *	0.439 *	3	0.667	0.677 * >	0.652 *	0.364 *	3	0.880 **	0.854 * >	0.762 *	0.852 *	0.569 *
4	0.722	0.564 *	0.567 *	0.368 *	4	0.706	0.821 * >	0.797 *	0.565 *	4	0.700	0.612 * >	0.846 *	0.591 *	0.391 *
5	0.883 **	0.856 *	0.872 *	0.613 *	5	0.906 **	0.922 *	0.923 *	0.688 **	5	0.778	0.370	0.688 *	0.404 *	0.241
6	0.889 **	0.856 *	0.860 *	0.640 *	6	0.522	0.216	0.212	0.083	6	0.824 **	0.475 * >	0.493 *	0.862 *	0.537 *
7	0.950 **	0.732 *	0.754 *	0.542 *	7	0.900 **	0.445 *	0.447 *	0.368 *	7	0.889 **	0.777 * >	0.755 *	0.450 *	0.296 *
8	0.739	0.771 *	0.775 *	0.443 *	8	0.750	0.490 *	0.493 *	0.285 *	8	0.817 **	0.800 * >	0.799 *	0.774 *	0.569 *
9	0.750	0.634 *	0.635 *	0.340 *	9	0.694	0.155	0.182	0.103	9	0.806 **	0.979 * >	0.964 *	0.862 *	0.644 *
10	0.750	0.817 * >	0.791 *	0.514 *	10	0.833 **	0.466 *	0.489 *	0.320 *	10	0.583	0.000	0.000	0.117	0.047
5. Communicates verbally				6. Communicates non-verbally				7. Is able to focus on something together				8. Is able to take another person into account			
Child	NAP	Slope 1	Tau-U	Child	NAP	Slope 1	Tau-U	Child	NAP	Slope 1	Tau-U	Child	NAP	Slope 1	Tau-U
1	0.978 **	0.723 *	0.765 *	0.553 *	1	0.378	-0.720 *	-0.668 *	-0.498 *	1	0.993 **	0.645 *	0.671 *	0.821 *	0.850 *
2	0.850 **	0.715 *	0.715 *	0.506 *	2	0.922 **	0.818 * >	0.810 *	0.589 *	2	0.722	0.341 >	0.340	-0.074	-0.039
3	0.167	0.317	0.388 *	-0.328 *	3	0.056	-0.328 * >	-0.370 *	-0.316 *	3	0.333	0.052 >	0.018	0.896 **	0.882 *
4	0.783	0.834 * >	0.814 *	0.589 *	4	0.872	0.787 *	0.797 *	0.577 *	4	0.828 **	0.811 * >	0.812 *	0.941 * >	0.94 *
5	0.500	0.000 *	0.000 *	0.000 *	5	0.944 **	0.890 *	0.902 *	0.668 **	5	0.856 **	0.898 * >	0.881 *	0.964 * >	0.962 *
6	0.733	0.869 * >	0.853 *	0.542 *	6	0.844 **	0.895 *	0.889 *	0.640 *	6	0.889 **	0.731 * >	0.736 *	0.972 **	0.814 *
7	0.894 **	0.575 *	0.615 *	0.415 *	7	0.333	-0.046	-0.074	-0.028	7	0.794	0.666 * >	0.631 *	0.345 >	0.330
8	0.889 **	0.623 *	0.638 *	0.431 *	8	0.056	-0.328	-0.370 *	-0.316	8	0.344	0.086 >	0.048	0.715 *	0.716 *
9	0.500	0.000 *	0.000 *	0.000 *	9	0.500	0.000	0.000	0.000	9	0.750	0.788 * >	0.766 *	0.889 **	0.882 *
10	0.978 **	0.661 *	0.737 *	0.395 *	10	0.667	0.725 * >	0.694 *	0.423 *	10	0.500	0.000	0.000	0.490 *	0.531 *
9. Takes initiative				10. Has rigid behavioral patterns											
Child	NAP	Slope 1	Tau-U	Child	NAP	Slope 1	Tau-U								
1	0.961 **	0.782 *	0.785 *	0.601 *	1	0.406	-0.534 *	-0.509 *							
2	0.783	0.392 *	0.410 *	0.273 *	2	0.194	-0.364 *	-0.387 *							
3	0.961 **	0.822 *	0.841 *	0.605 *	3	0.494	0.171	0.141							
4	0.961 **	0.936 *	0.947 *	0.755 *	4	0.250	-0.538 *	-0.496 *							
5	0.694	0.782 * >	0.755 *	0.419 *	5	0.361	-0.296	-0.297							
6	0.516	0.611 * >	0.563 *	0.380 *	6	0.444	0.227	0.186							
7	0.861 **	0.745 *	0.750 *	0.526 *	7	0.561	-0.526 *	-0.449 *							
8	0.917 **	0.752 *	0.758 *	0.545 *	8	0.494	0.171	0.141							
9	0.733	0.615 *	0.635 *	0.348 *	9	0.411	-0.512 * >	-0.550 *							
10	0.722	0.738 *	0.713 *	0.455 *	10	0.117	-0.611 * >	-0.632 *							

Note:
 NAP score
 Weak improvement 0 - 0.65
 Moderate improvement 0.66 - 0.92
 Strong improvement 0.93 - 1
 ***Strong improvement 0.80 - 1
 Tau-U and SMA Slope 1 and Slope 2
 * = p<.05

In table 2 the NAP scores per item are presented. Except for the items about non-verbal communication and rigid behavioral patterns, at least seven of the ten children show a weak to moderate progress. It is noteworthy that all children show slight acceleration in the items of breaking rigid behavioral patterns. When we follow the cut-off scores of Walker & Snell (2013), we see that all children show strong progress in one or more of the investigated areas. The more tolerant cut off yields all ten children showing strong improvement in two or more areas. For each of the items ‘can cope with changes’, ‘behavior has been adjusted’, ‘is able to communicate verbally’, ‘is able to have consideration for another person’, and ‘takes initiative’, five or more children show a NAP score of 0.80 or higher. When we look more specifically at the slopes, we see that the measurements between the 12th and 15th week show a notable increase. This applies to those children who had a moderate improvement or strong improvement according to the NAP, see table 2. This means that compared to the baseline phase, these children start to show a clearly noticeable progress on the items from the weekly questionnaire between the 12th and 15th week of the PMPT module. For the children with weak improvement according to the NAP there was no notable increase in the slope.

The SMA was calculated in order to assess whether the scores that were found can also be associated with a baseline and intervention phase. In the first instance, we looked which hypothesized slope did fit best. Slope 1 and slope 2 showed no markedly visible differences. It is noteworthy that the differences in the correlation between both slopes with their hypothesized slope never exceeds 0.05. An example of this is that the values of participant one on the item ‘eye contact’ for slope 1 ($r=0.759$, $p=0.001$) and slope 2 ($r=0.771$, $p=0.001$) both show a high correlation that lies close together with a value of 0.012. This does not clearly indicate an accelerating development during music therapy, compared to the growth that was already visible in the baseline phase. Another example is participant five with regard to ‘taking another into account’. This participant shows a significantly high correlation on slope 1 ($r=0.964$, $p=0.0001$), as well as on slope 2 ($r=0.962$, $p=0.0001$), that is only 0.002 lower than the value of slope 1, meaning that SMA does not clearly indicate an accelerated development.

For a more specific analysis, we used Tau-U to test the null hypothesis that the growth pattern during the intervention does not differ from the pattern from the baseline phase, as shown in table 2.

Further inspection of the data revealed that none of the Tau-U analyses indicate a significant trend in the baseline phase, whereas all trends are significant for the intervention phase for each child with a slope 1 > slope 2 pattern. This further indicates that, when the table shows a Tau-U with a significant value ($p<.05$), the child shows a significant growth for that particular developmental aspect in the intervention phase. For the items ‘coping with change’ and ‘takes initiative’, we can see that this applies to eight children. For the item ‘is able to take others into account’, this applies to seven children, and for the item ‘behavior has changed’ it applies to six children. This closer examination of the data could implicate that an acceleration of development is present during the intervention phase.

Comparing the results of the weekly measurements to those of the VISK, a striking difference can be noticed, especially for children four, seven and nine. The VISK shows little or no progress for them, but the weekly scores do show a significant improvement on various developmental aspects. For child four the VISK only shows progress on the sub scale ‘not optimally tuned behavior’. However, the weekly measurements show a moderate improvement on six items and even a strong improvement on two items. For child seven this was the same, for the VISK only the sub scales ‘not optimally tuned behavior’ and ‘Stereotype movements’ show a small improvement. The total score even shows slower progress compared to the baseline measurement. Again, in the weekly measurements, we see a moderate improvement on seven items and for one item a strong improvement. For child nine, only on the subscales of the VISK ‘not optimally tuned behavior’ and ‘difficulty with social information’ a small improvement was visible. Also here, the weekly measurements showed moderate improvement on seven items. The other seven children did show an improvement on both scoring lists. Using the Spearman rank order correlation, the correlation between the VISK and the weekly questionnaire was calculated. For the measurement in the time period where the intervention starts it is $r = .034$ and for the time period where the intervention ends it is $r = .321$.

Discussion

In this study we attempted to answer the question whether social behavior of children with ASD improves during the ‘Papageno Music Therapy Program’ (PMTP). The PMTP has been recently developed, therefore a multiple case study among 10 children was performed. The results of the NAP, SMA and the Tau-U all point, to a certain extent, at a positive improvement in social behavior between the baseline phase and the intervention phase.

All children show weak improvement in the item of breaking rigid behavioral patterns. At least five children had a NAP score between 0.66 and 0.92 in the areas of ‘makes eye contact’, ‘is able to focus for five minutes on a game’, ‘is able to cope with changes’, ‘behavior has adapted to the situation’, ‘communicates verbally’, ‘is able to focus on something together’, ‘is able to take another person into account’ and ‘takes initiative’. The biggest progress, with three children showing a NAP score of >0.93 , was found in the areas of ‘is able to focus on something together’ and ‘takes initiative’. The findings on the social behavior were partly confirmed by the VISK scores, which were taken at three different times during the therapy process. When we put the results of the weekly measurements next to those of the VISK, we see converging trends on both instruments, except for the children four, seven, and nine, which show striking differences in scores. They showed little or no progress on the VISK on the weekly questionnaire showed moderate improvement or even strong improvement on several items. The correlation between both measurement instruments is low, indicating that the two instruments focus on different aspects of social behavior. The items of the weekly SBQ fits well with the objectives of the PMTP, whereas the VISK also measures a number of points that are not on the weekly list.

Progress in behaviors observed with the SBQ may not always translate into an improvement in terms of the VISK-scores, assuming that the SBQ-behaviors do not clearly affect every child in the more general aspects of social development measured over a longer time span. For those children for which this is the case (four, seven, and nine), they may need more other therapies

than the PMTP to achieve changes will be visible on the VISK. The combination of the SBQ and the VISK may therefore be a very useful set of tools to identify these children and to find the necessary additional activities that could eventually bring in these changes.

The development we found in this study is in line with previous studies that indicate that music therapy has a positive effect on the social behavior of children with ASD, like the ones from Edgerton (1994); Gattino et al. (2011); Ghasemtabar et al. (2015); Katagiri (2009); Kern et al. (2006); Kern et al. (2007); Kim et al. (2008); Thomson (2012); Vaiouli et al. (2015), and Wimpory et al. (1995). Even though the detected progress in several areas is significant, the steps that are taken are sometimes small and it is unclear what further progress can be expected. This makes it difficult to make firm statements about the progress and, although some steps in the development may seem small, they can still have a major impact on the child's daily life. The small steps in the child's development can possibly be explained by the fact that for the children getting PMTP social interaction is difficult.

Therapists need to be very creative in order to be able to relate to the experiences of children with ASD, because they can be introverted (Schertz & Odom, 2007). Music therapy may prevent this and produce results because the therapist continuously adapts him/herself to the child during the session. When a connection has been made, the therapist can stimulate the child to show a different type of behavior. It seems that such a connection is an important prerequisite to facilitate change; it ensures that – though slowly – the child finds the focus and motivation to learn new behavior (Pater, de Graaf, & van Yperen, 2019).

Initially, the interpretation of the NAP was made based on the standards of Walker & Snell (2013). When the values are between 0.93 and 1.0, it indicates a large or strong effect. Because children with ASD are often difficult to stimulate when it comes to developing their social behavior and because they only take small steps, this study is based on the assumption that there is significant progress when the NAP value is between 0.80 and 1.0.

Also, it became apparent that investigated children with moderate improvement or strong improvement according to the NAP started to show clearly noticeable progress between the 12th and 15th week of the intervention phase, and not earlier. This can possibly be explained by the fact that children with ASD need more time to get used to new situations.

This study does have several limitations. First of all, based on this study, we may not causally conclude that it was the music therapy that had a positive effect on the social behavior of children with ASD. To do so, it is necessary to validate our results in a study with a larger group and, if possible, to compare the results with those of another intervention or care as usual. Although our results are based on a small sample ($N=10$), our multiple case study with $n>9$ can be considered as strong evidence (Chambless, et al., 1996). In our opinion, this means that our study indicates that music therapy has a promising effect that is solid enough to defend a more rigorous and demanding research.

Second, the observations in this study are only done by the mothers of the children. It is plausible that parents evaluate behavior more positive in research situations, because they want to see their child progress. Although demanding, in future research, we advise to do a multiple informant's study to collect observations from different points of view. This excludes an influenced opinion and it gives a better observation of the child's behavior in different situations.

Third, in order to make the development trend transparent, we created a short questionnaire. The VISK assesses the social behavior as observed over the past few months. Because our questionnaire had to be completed weekly, over a period of 20 weeks, it was important that it would assess the observed behavior of the past week and that it would not take too much time so it would not overload the respondents, which could lead to non-response. The compiled questionnaire made it possible to do a weekly measurement that was also feasible for parents. The questionnaire was composed based on the purpose of the intervention. This questionnaire was not validated, although in addition to this questionnaire we also used the VISK which was validated. It is important to further develop this questionnaire.

Fourth, in this study it was decided to start music therapy as soon as possible after enrollment at the Papageno Foundation. Five weeks were used a baseline phase, of which two were during the therapy period. It would be better to have an unbiased baseline measurement over a period of, for example, ten weeks. This way, possible natural growth could be excluded with greater certainty. In our cases, however, the parents strongly objected to a long waiting period before starting therapy, because they already had to wait many weeks before their child was admitted for treatment, and they wanted to do everything possible to stimulate the development of their child. In all, we think that a five-week baseline of which three weeks prior to therapy and two weeks at the start represents an acceptable solution, also considering the fact that analyses with a baseline of the first three weeks did not result in different findings.

Finally, in this study we gained insight into the course of music therapy and its possible contribution to social behavior. Through the weekly observations and the calculation of slope 1, we were able to see the development in the therapy process. Even though we could see a change during the intervention phase compared to the baseline phase, the results would be even more complete if we also did a follow-up measurement to see if the development is sustainable.

An experiential intervention such as music therapy seems to be a complicated intervention to identify the effects. The multiple case design that was used seems to be appropriate to study an intervention such as music therapy on a child-centered level. This research suggests that PMTP is promising to positively affect the social behavior of children with ASD. For further substantiation it is especially useful to test these effects with more demanding experimental research. Robey (2004) indicates that when exploratory studies like a case study provide signs of positive effectiveness, a subsequent clinical study is highly desirable. Testing by means of a Parallel group design with an experimental and control group would be a good next step to test the PMTP. Furthermore, it is necessary to further validate the brief questionnaire as a reliable instrument to enable frequent measurement moments and at the same time ensure the response in practice.

In conclusion, our study shows that PMTP is a promising intervention to help children with ASD to further develop their social behavior. In our opinion, in this stage of the research, it would be premature to draw any conclusions on the practical or clinical implications of this finding. However, in regards to further research of this intervention, to establish whether or not this intervention is effective compared to the usual treatment or other specific forms of therapy, an experimental study with an experimental and control group seems to be the best next step.

