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## Music therapy for people with autism spectrum disorder

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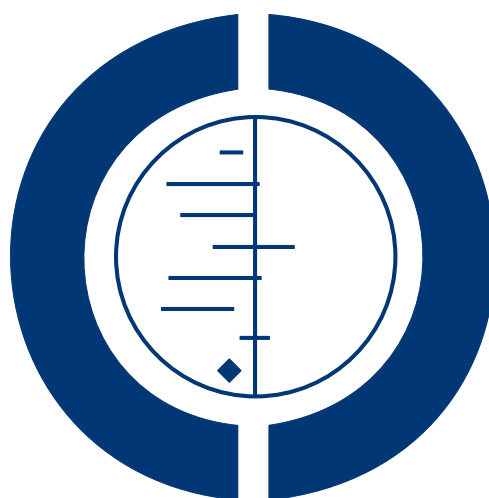
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# Music therapy for people with autism spectrum disorder (Review)

Geretsegger M, Elefant C, Mössler KA, Gold C



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# Music therapy for people with autism spectrum disorder

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## ABSTRACT

### Background

The central impairments of people with autism spectrum disorder (ASD) affect social interaction and communication. Music therapy uses musical experiences and the relationships that develop through them to enable communication and expression, thus attempting to address some of the core problems of people with ASD. The present version of this review on music therapy for ASD is an update of the original Cochrane review published in 2006.

### Objectives

To assess the effects of music therapy for individuals with ASD.

### Search methods

We searched the following databases in July 2013: CENTRAL, Ovid MEDLINE, EMBASE, LILACS, PsycINFO, CINAHL, ERIC, ASSIA, Sociological Abstracts, and Dissertation Abstracts International. We also checked the reference lists of relevant studies and contacted investigators in person.

### Selection criteria

All randomised controlled trials (RCTs) or controlled clinical trials comparing music therapy or music therapy added to standard care to 'placebo' therapy, no treatment, or standard care for individuals with ASD were considered for inclusion.

### Data collection and analysis

Two authors independently selected studies, assessed risk of bias, and extracted data from all included studies. We calculated the pooled standardised mean difference (SMD) and corresponding 95% confidence interval (CI) for continuous outcomes to allow the combination data from different scales and to facilitate the interpretation of effect sizes. Heterogeneity was assessed using the  $I^2$  statistic. In cases of statistical heterogeneity within outcome subgroups, we examined clients' age, intensity of therapy (number and frequency of therapy sessions), and treatment approach as possible sources of heterogeneity.

## Main results

We included 10 studies (165 participants) that examined the short- and medium-term effect of music therapy interventions (one week to seven months) for children with ASD. Music was superior to 'placebo' therapy or standard care with respect to the primary outcomes social interaction within the therapy context (SMD 1.06, 95% CI 0.02 to 2.10, 1 RCT, n = 10); generalised social interaction outside of the therapy context (SMD 0.71, 95% CI 0.18 to 1.25, 3 RCTs, n = 57, moderate quality evidence), non-verbal communicative skills within the therapy context (SMD 0.57, 95% CI 0.29 to 0.85, 3 RCTs, n = 30), verbal communicative skills (SMD 0.33, 95% CI 0.16 to 0.49, 6 RCTs, n = 139), initiating behaviour (SMD 0.73, 95% CI 0.36 to 1.11, 3 RCTs, n = 22, moderate quality evidence), and social-emotional reciprocity (SMD 2.28, 95% CI 0.73 to 3.83, 1 RCT, n = 10, low quality evidence). There was no statistically significant difference in non-verbal communicative skills outside of the therapy context (SMD 0.48, 95% CI -0.02 to 0.98, 3 RCTs, n = 57, low quality evidence). Music therapy was also superior to 'placebo' therapy or standard care in secondary outcome areas, including social adaptation (SMD 0.41, 95% CI 0.21 to 0.60, 4 RCTs, n = 26), joy (SMD 0.96, 95% CI 0.04 to 1.88, 1 RCT, n = 10), and quality of parent-child relationships (SMD 0.82, 95% CI 0.13 to 1.52, 2 RCTs, n = 33, moderate quality evidence). None of the included studies reported any adverse effects. The small sample sizes of the studies limit the methodological strength of these findings.

## Authors' conclusions

The findings of this updated review provide evidence that music therapy may help children with ASD to improve their skills in primary outcome areas that constitute the core of the condition including social interaction, verbal communication, initiating behaviour, and social-emotional reciprocity. Music therapy may also help to enhance non-verbal communication skills within the therapy context. Furthermore, in secondary outcome areas, music therapy may contribute to increasing social adaptation skills in children with ASD and to promoting the quality of parent-child relationships. In contrast to the studies included in an earlier version of this review published in 2006, the new studies included in this update enhanced the applicability of findings to clinical practice. More research using larger samples and generalised outcome measures is needed to corroborate these findings and to examine whether the effects of music therapy are enduring. When applying the results of this review to practice, it is important to note that the application of music therapy requires specialised academic and clinical training.

## PLAIN LANGUAGE SUMMARY

### Music therapy for people with autism spectrum disorder

#### Review Question

We reviewed the evidence about the effect of music therapy in people with autism spectrum disorder (ASD). We compared music therapy or music therapy in addition to standard care to no therapy, similar treatment without music ('placebo' therapy), or standard care.

#### Background

People with ASD have difficulties with social interaction and communication. Music therapy uses musical experiences and the relationships that develop through them to enable people to relate to others, to communicate, and to share their feelings. In this way, music therapy addresses some of the core problems of people with ASD. We wanted to discover whether music therapy helps people with ASD compared to other alternatives.

#### Study Characteristics

We included 10 studies with a total number of 165 participants. The studies examined the short- and medium-term effect of music therapy interventions (one week to seven months) for children with ASD.

#### Key Results

Music therapy was superior to 'placebo' therapy or standard care with respect to social interaction, non-verbal and verbal communicative skills, initiating behaviour, and social-emotional reciprocity. Music therapy was also superior to 'placebo' therapy or standard care in the areas of social adaptation, joy, and the quality of parent-child relationships. None of the included studies reported any side effects caused by music therapy.

#### Quality of the Evidence

The quality of the evidence was moderate for social interaction outside of the therapy context, initiating behaviour, social adaptation, and the quality of the parent-child relationship, and low for the other three main outcomes (nonverbal communicative skills outside of the therapy context, verbal communicative skills outside of the therapy context, and social-emotional reciprocity). Reasons for limited quality of the evidence were issues with study design and small number of patients who participated in the studies.

### **Authors' Conclusions**

Music therapy may help children with ASD to improve their skills in important areas such as social interaction and communication. Music therapy may also contribute to increasing social adaptation skills in children with ASD and to promoting the quality of parent-child relationships. Some of the included studies featured interventions that correspond well with treatment in clinical practice. More research with adequate design and using larger numbers of patients is needed. It is important to specifically examine how long the effects of music therapy last. The application of music therapy requires specialised academic and clinical training. This is important when applying the results of this review to practice.

## SUMMARY OF FINDINGS FOR THE MAIN COMPARISON [\[Explanation\]](#)

Music therapy compared to 'placebo' therapy or standard care for autism spectrum disorder			
<b>Patient or population:</b> Individuals with autism spectrum disorder <b>Settings:</b> Outpatient therapy centre, hospital, school, or home <b>Intervention:</b> Music therapy <b>Comparison:</b> 'Placebo' therapy or standard care			
Outcomes	Relative effect (95% CI) Music therapy versus 'placebo' therapy or standard care	Number of participants (studies)	Quality of the evidence (GRADE)
<b>Social interaction - Generalised (outside sessions, daily life)</b> CARS, PDDBI, Vineland SEEC, SRS Follow-up: 4 to 7 months	The mean social interaction - generalised (outside sessions, daily life) in the intervention groups was <b>0.71 standard deviations higher</b> (0.18 to 1.25 higher)	57 (3 studies)	⊕⊕⊕○ <b>moderate</b> <sup>1,2,4</sup>
<b>Communicative skills: non-verbal - Generalised (outside sessions, daily life)</b> CARS, ESCS, MBCDI-W&G Follow-up: 4 to 7 months	The mean communicative skills: non-verbal - generalised (outside sessions, daily life) in the intervention groups was <b>0.48 standard deviations higher</b> (0.02 lower to 0.98 higher)	57 (3 studies)	⊕⊕○○ <b>low</b> <sup>3,4</sup>
<b>Communicative skills: verbal - Generalised (outside sessions, daily life)</b> CARS, MBCDI-W&G Follow-up: 4 to 7 months	The mean communicative skills: verbal - generalised (outside sessions, daily life) in the intervention groups was <b>0.30 standard deviations higher</b> (0.28 lower to 0.89 higher)	47 (2 studies)	⊕⊕○○ <b>low</b> <sup>3,4</sup>
<b>Initiating behaviour - Non-generalised</b> Requesting (initiating joint attention), imitation of engagement frequency, requesting behaviour Follow-up: 5 weeks to 4 months	The mean initiating behaviour - non-generalised in the intervention groups was <b>0.73 standard deviations higher</b> (0.36 to 1.11 higher)	22 (3 studies)	⊕⊕⊕○ <b>moderate</b> <sup>1,2,4</sup>
<b>Social-emotional reciprocity - Non-generalised</b> Emotional and musical synchronicity, frequency, and duration	The mean social-emotional reciprocity - non-generalised in the intervention groups was <b>2.28 standard deviations higher</b>	10 (1 study)	⊕⊕⊕○ <b>low</b> <sup>2,4,5</sup>

Follow-up: 4 months	(0.73 to 3.83 higher)	
<b>Social adaptation - Non-generalised</b> Interaction (engaging in joint attention), compliant or non-compliant response frequency, no response frequency, on-task behaviour Follow-up: 5 weeks to 4 months	The mean social adaptation - 22 non-generalised in the intervention groups was <b>1.15 standard deviations higher</b> (0.69 to 1.61 higher)	⊕⊕⊕○ <b>moderate</b> <sup>1,2,4</sup>
<b>Quality of parent-child relationship</b> MPIP, PCRI Follow-up: 4 months	The mean quality of parent-child relationship in the intervention groups was <b>0.82 standard deviations higher</b> (0.13 to 1.52 higher)	⊕⊕⊕○ <b>moderate</b> <sup>2,4</sup>

**CI:** Confidence interval

GRADE Working Group grades of evidence

**High quality:** Further research is very unlikely to change our confidence in the estimate of effect.

**Moderate quality:** Further research is likely to have an important impact on our confidence in the estimate of effect and may change the estimate.

**Low quality:** Further research is very likely to have an important impact on our confidence in the estimate of effect and is likely to change the estimate.

**Very low quality:** We are very uncertain about the estimate.

<sup>1</sup> Limitations in the designs such as poorly reported randomisation and blinding of outcomes.

<sup>2</sup> The estimated effect was in the large or close to the large range according to [Cohen 1988](#).

<sup>3</sup> 95% confidence interval includes no effect and the upper confidence limit crosses an effect size of 0.5 ([GRADEpro 2008](#)).

<sup>4</sup> Total number of participants in this outcome is lower than 400.

<sup>5</sup> Only one study within this outcome.

## BACKGROUND

### Description of the condition

Autism spectrum disorder (ASD), as defined by the International Classification of Diseases and Related Health Problems, 10th edition (ICD-10) ([WHO 1992](#)), and the Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5) ([APA 2013](#)), is considered to be a complex neurodevelopmental disorder that is defined and diagnosed behaviourally, and usually manifests in early childhood persisting throughout life.

Individuals with ASD have difficulties in various aspects of social communication. They also have a restricted imagination and social repertoire, the latter characteristically displayed as what seems to others to be obsessional behaviour and rigidity in their own behaviour as well as in the behaviour they require from others in response to their own. In the last two decades, the key construct has been the 'triad of impairment', which affects social interaction, language and communication, and behaviour and imagination ([Wing 1997](#)), that can be identified through examination of early development and current presentation ([Wing 2002](#)). Within the ICD-10 ([WHO 1992](#)), and the DSM-IV-TR ([APA 2000](#)), the last leg of the triad was defined as restricted repetitive and stereotyped patterns of behaviour, interests, and activities. However, in



new editions of the classification systems, the recently published DSM-5 and the forthcoming ICD-11, the first two areas have been merged resulting in only two core domains of ASD: (1) social communication or social interaction and (2) restricted, repetitive behaviours and interests (Lord 2012). People with ASD also present with pervasive difficulties to 'mind-read', where a lack of perception and understanding of other people's feelings, beliefs or emotions results in a consequential inability to respond appropriately (Baron-Cohen 1995). This has particular impacts on social skills and interactions (Howlin 1998).

The clinical picture varies because individuals have different levels of ability, from profound learning disability to a spiky cognitive profile where superior skills are present in some areas of functioning. At the high-functioning end of the autism spectrum is a condition known as Asperger's syndrome, with the same fundamental core impairments as autism but also some differences in language development, motor skills, and originality of thought (Asperger 1979); with the changes in DSM-5, Asperger's syndrome was merged into the single diagnostic category of ASD (APA 2013). Recent prevalence estimates for autism spectrum conditions vary according to factors such as method of case identification, age range, or standardisation of diagnostic measures, and range from 60 to 157 children per 10,000 (Baird 2006; Baron-Cohen 2009; Fombonne 2009; Fombonne 2010), suggesting much higher prevalence rates than estimates from older studies (Chakrabarti 2001; Fombonne 1999).

## Description of the intervention

Music therapy has been defined as "a systematic process of intervention wherein the therapist helps the client to promote health, using musical experiences and the relationships that develop through them as dynamic forces of change" (Bruscia 1998, p. 20). Central music therapy techniques include free and structured improvisation, singing songs and vocalisation, and listening to both pre-recorded and live music.

Music therapy for individuals with ASD is usually provided as individual therapy, although there are also reports of group-based and peer-mediated interventions (Boso 2007; Kern 2006; Kern 2007). In recent years, family-centred approaches, where parents or other family members are included in therapy sessions, have increasingly become an important part of music therapy for children with ASD (Oldfield 2012; Pasiali 2004; Thompson 2012a; Thompson 2012b).

## How the intervention might work

The processes that occur within musical interaction may help people with ASD to develop communication skills and the capacity for social interaction. Musical interaction in music therapy, in particular musical improvisation, can be understood and described as

a non-verbal and pre-verbal language that enables verbal people to access pre-verbal experiences, enables non-verbal people to interact communicatively without words, and enables all to engage on a more emotional, relationship-oriented level than may be accessible through verbal language (Alvin 1991). Listening to music within music therapy also involves an interactive process that often includes selecting music that is meaningful for the person (e.g. relating to an issue that the person is occupied with) and, where possible, reflecting on personal issues related to the music or associations brought up by the music. For those with verbal abilities, verbal reflection on the musical processes is often an important part of music therapy (Wigram 2002).

A rationale for the use of music therapy for individuals with communication disorders is based on the findings of infancy researchers such as Stern and Trevarthen who describe sound dialogues between mothers and infants using 'musical' terms (Stern 1985; Stern 1989; Stern 2010; Trevarthen 2000). When describing tonal qualities, researchers use the terms pitch, timbre, and tonal movement, and when describing temporal qualities, they speak of pulse, tempo, rhythm, and timing (Wigram 2002). Trevarthen 1999 describes the sensitivity of very young infants to the rhythmic and melodic dimensions of maternal speech, and to its emotional tone, as demonstrating that we are born ready to engage with the 'communicative musicality' of conversation, and this premise allows music to act as an effective medium for engaging in non-verbal social exchange for children and adults with ASD. Necessary communicative behaviours, such as joint attention, eye contact, and turn-taking, are characteristic events in shared, active music making and therefore inherent components of music therapy processes. In addition to music's potential to stimulate communication, Wigram and Elefant also explain how music therapists can use music, especially improvisational music-making, to provide children with ASD with opportunities to experience foundation-giving structure combined with measured flexibility, thus helping them to find ways of coping in less predictable situations that will typically pose challenges for them (Wigram 2009).

The potential for predictability and anticipation brought about by musical structures is an element also used in behavioural approaches where music is utilised as a stimulus facilitating the perception and production of speech and language and enhancing communication skills. Another rationale for using music in this way is the increased attention and enjoyment observed in individuals when presented with musical as opposed to verbal stimuli (Buday 1995; Lim 2010; Lim 2011).

## Why it is important to do this review

This is an update of a Cochrane review first published in 2006 (Gold 2006). Before the original version of this review was published, clinical reports and pre-experimental studies had suggested that music therapy may be an effective intervention for people with ASD. For example, Edgerton 1994 examined the development of

communicative skills in 11 children with autism over the course of music therapy sessions, finding a continuous increase of communicative acts and responses in all subjects (Edgerton 1994). Schumacher described qualitatively how relationship patterns of children with autism changed and developed during long-term music therapy (Schumacher 1999a; Schumacher 1999b). Two earlier systematic reviews pertaining to the scope of this review yielded conflicting results. Whipple 2004 concluded that music therapy was effective for people with ASD. However, interventions and study designs were too heterogeneous to allow clinically meaningful and methodologically strong conclusions. Ball 2004 concluded that effects of music therapy were unclear. However, this review failed to identify many possibly relevant studies (Ball 2004). Thus, a more comprehensive systematic review of controlled studies in this area was deemed necessary.

The first version of this review concluded that music therapy may help children with ASD to improve their communicative skills, but also noted that more research was needed to investigate the effects of music therapy in typical clinical practice and within longer periods of observation (Gold 2006). A recent systematic review suggested that music therapy may be an effective treatment for young children with ASD for developing communication, interpersonal abilities, personal responsibility, and play skills (Whipple 2012). However, as in the author's previous review (Whipple 2004), the designs of the included studies lacked homogeneity and entailed various risks of bias (e.g. sample sizes of only one, lack of blinded observations).

We conducted the current update to summarise and evaluate new studies of music therapy for ASD published since the 2006 version of this review in order to provide comprehensive and up-to-date conclusions, as well as implications for practice and research that are based on recent findings.

## OBJECTIVES

To review the effects of music therapy, or music therapy added to standard care, for individuals with ASD.

## METHODS

### Criteria for considering studies for this review

#### Types of studies

All relevant randomised controlled trials (RCTs) and controlled clinical trials (CCTs) were considered for inclusion. Studies using single-case experimental designs were included if they also met the definition of RCTs or CCTs. That is if the different interventions

were provided in a different order to different participants i.e. (cross-over RCTs/CCTs). Studies in which all participants received interventions in the same order (i.e. case series) were excluded.

#### Types of participants

Individuals of any age who are diagnosed with a pervasive developmental disorder, as defined in ICD-10 or DSM-IV or DSM-IV-TR, whether identified by a psychological assessment or a psychiatric diagnosis were considered inclusion. This includes childhood autism (F84.0 in ICD-10), atypical autism (F84.1), Asperger's syndrome (F84.5), and pervasive developmental disorder not otherwise specified (F84.9). Individuals with Rett's disorder (F84.2) or childhood disintegrative disorder (F84.3) were not included as they do not conventionally fall within the autism spectrum disorders, given their significantly different clinical course.

#### Types of interventions

Interventions included music therapy (i.e. regular sessions of music therapy as defined above), delivered by a professional music therapist, compared with either 'placebo' therapy (the concept of attention placebo in psychotherapy research is discussed in Kendall 2004), no-treatment, or standard care control; or music therapy added to standard care compared with standard care (with or without 'placebo').

#### Types of outcome measures

We regarded outcome measures in all areas of social communication as primary outcomes as they refer to the core characteristics defining ASD. We regarded commonly examined outcome measures in areas not specific to defining ASD characteristics as secondary outcomes.

#### Primary outcomes

Primary outcomes included the following.

- Social interaction.
- Communicative skills (non-verbal and verbal).
- Initiating behaviour.
- Social-emotional reciprocity.
- Adverse effects.

#### Secondary outcomes

Secondary outcomes included the following.

- Social adaptation skills (including outcomes that were summarised as behavioural problems, such as stereotypic behaviour, in the 2006 version of this review).
- Quality of life in school, home, and other environments.
- Quality of family relationships.
- Cognitive ability (including attention, concentration).

- Hyperacusis (hypersensitivity to sound).

Data sources could have included non-standardised or standardised instruments (for a review of relevant standardised instruments see [Ozonoff 2005](#)), parent or teacher report, or school records. Data from rating scales were only included if the instrument was either a self report or completed by an independent rater or relative (i.e. not the therapist). We also included outcomes initially rated by the therapist and reconfirmed by an independent rater.

Changes in generalised skills that are measured outside of the immediate treatment context pose the biggest challenge for any interventions for ASD ([Warren 2011](#)). Generalised outcomes refer to changes that generalise to other behaviours and to other contexts across settings, people, or materials. Because of the importance of generalised improvements for people with ASD, we reported the results that focus solely on 'within sessions' change (hereafter referred to as 'non-generalised' outcome measures) separately from those that assess the impact of music therapy broadly in other contexts (referred to as 'generalised' outcome measures).

In the [Summary of findings for the main comparison](#), we report the results of the three generalised outcomes: social interaction, non-verbal communicative skills, verbal communicative skills; three non-generalised outcomes that relate to core areas of difficulty for children with ASD: initiating behaviour, social-emotional reciprocity, and social adaptation. Given its importance for children and their families, we also report the quality of the parent-child relationship ([Wheeler 2008](#)).

Where outcomes were measured at multiple time points during the course of therapy, we used mean values of all data from the second therapy session onwards. We determined a small effect size (i.e. 0.2) as the minimally important threshold for appreciable change for each outcome ([Cohen 1988](#); [Gold 2004](#)). If follow-up data were included, we planned to group outcome time points as follows: immediately post-intervention, one to five months post-intervention, six to 11 months post-intervention, 12 to 23 months post-intervention, and 24 to 35 months post-intervention.

## Search methods for identification of studies

We ran the searches for this update in September 2011 and again on 29 July 2013. We revised the original search strategy by adding new search terms to increase the sensitivity of the search. Searches were limited to the period since the original review (2004 onwards). We also searched the databases for the period before 2004 using only the new search terms, to be sure we had not missed any relevant studies.

## Electronic searches

We searched the following databases:

- Cochrane Central Register of Controlled Trials (CENTRAL) 2013, Issue 6, part of The Cochrane Library;

- Ovid MEDLINE 1948 to July week 3 2013;
- EMBASE 1980 to 2013 week 30;
- LILACS 1982 to current;
- PsycINFO 1806 to July week 3 2013;
- CINAHL 1937 to current;
- ERIC 1966 to current;
- ASSIA 1987 to current;
- Sociological Abstracts 1952 to current;
- Dissertation Abstracts International.

Detailed search strategies are reported in [Appendix 1](#). Search terms from the original version of the review are reported in [Appendix 2](#)

## Searching other resources

We searched the following specific sources for music therapy literature:

- [musictherapyworld.net](#). (this website, formerly maintained by the Institute for Music Therapy at the University of Witten Herdecke, Germany, was last accessed in July 2004 but was no longer being maintained at the time of this update);
- Music Therapy Research CD ROM ([AMTA 1999](#)); and
- Music Therapy World Info-CD ROM IV ([Aldridge 2002](#)).

In addition, we searched the reference lists of the studies included in this review as well as relevant review articles ([Accordino 2007](#); [Ball 2004](#); [Reschke-Hernández 2011](#); [Simpson 2011](#); [Whipple 2004](#); [Whipple 2012](#)), and proceedings of music therapy conferences to identify additional studies.

## Data collection and analysis

### Selection of studies

Three authors (CE, CG, MG) independently inspected all titles and abstracts identified from the search. We obtained potentially relevant papers and resolved any disagreement about eligibility through discussion or consultation with the other authors. If non-English study reports had been found, we would have provided for their translation. We recorded the reasons for excluding trials.

### Data extraction and management

Two reviewers (CG, MG) independently performed data extraction using a data collection form. When necessary, we contacted the study authors to provide missing data.

### Assessment of risk of bias in included studies

Two authors (KM, MG) assessed methodological quality independently using the Cochrane risk of bias tool ([Higgins 2011a](#)). Any

disagreements were resolved by discussion, or consultation with the other reviewers, or both.

For each included study, we presented the risk of bias assessments in a table where the judgement of the review authors (low, high or unclear risk of bias) was followed by a text box providing details on the available information that led to each judgement.

We assessed the following items:

- Random sequence generation;
- Allocation concealment;
- Blinding of participants and personnel;
- Blinding of outcome assessment;
- Completeness of outcome data;
- Selective reporting; and
- Other sources of bias.

### Randomisation

We judged the risk of bias for random sequence generation as follows.

- Studies were judged to be at low risk of bias if participants were allocated to treatment interventions using randomisation such as computer-generated random numbers, a random numbers table, or coin-tossing.
- Studies were judged to be at unclear risk of bias if the randomisation method was not clearly stated or was unknown.
- Studies were judged to be at high risk of bias if the method sequence generation was non-random.

Randomised as well as quasi-randomised trials were included in the review, as noted above.

### Allocation concealment

We judged the risk of bias for allocation concealment as follows.

- Studies were judged to be at low risk of bias if allocation concealment was adequate; participants and researchers were unaware of participants' future allocation to an intervention until after decisions about eligibility were made and informed consent was obtained.
- Studies were judged to be at unclear risk of bias if the methods used for allocation concealment were not described in detail.
- Studies were judged to be at high risk of bias if allocation concealment was inadequate; allocation was not concealed from either participants before informed consent or from researchers before decisions about inclusion were made (this will always be the case for quasi-randomised studies).

As this review aimed to include randomised and quasi-randomised studies, all three categories were eligible for inclusion; we only used the rating as a descriptive measure of study quality.

### Blindness of participants and personnel

Due to the nature of the intervention it was not possible to blind those who delivered music therapy or those who received it. Consequently, neither participants nor personnel of the studies under review can be declared to be blinded. However, although children with ASD were not blinded, this was unlikely to introduce bias as they are usually not fully aware of available treatment options or study design (Cheuk 2011). The possible risk of bias introduced by therapists administering the intervention was unknown. Therefore, we judged the risk of performance bias as unclear in all studies in the review.

### Blinding of outcome assessors

We determined whether those who assessed and coded the outcome measures were blind to treatment assignment using the following categories.

- Studies were judged to be at low risk of bias if the assessor was blind to treatment assignment.
- Studies were judged to be at unclear risk of bias if blinding of assessor not reported and information not available from researchers.
- Studies were judged to be at high risk of bias if the assessor was not blind to treatment assignment.

All of the above were included in the review.

### Attrition bias

We assessed whether authors adequately dealt with missing data as follows.

- Studies were judged to be at low risk of bias if the number of participants randomised to groups was clear and it was clear that all participants completed the trials in all participant groups. Studies were also judged to be at low risk of bias if outcome data were missing in both intervention groups, but reasons for these were both reported and balanced across groups.
- Studies were judged to be at unclear risk of bias if information about which participants completed the study could not be acquired by contacting the study authors.
- Studies were judged to be at high risk of bias if there was clear evidence of attrition or exclusion from analysis in at least one participant group that was likely related to the true outcome.

### Reporting bias

We judged the risk of selective outcome reporting as follows.

- Studies were judged to be at low risk of bias if all collected data seem to be reported and all expected outcomes were reported.
- Studies were judged to be at unclear risk of bias if it was not clear whether other data were collected and not reported.

- Studies were judged to be at high risk of bias if data for one or more expected outcomes were missing.

### Other bias

Through assessment, we determined whether any other bias was present in the trial including inadequate music therapy methods or inadequate music therapy training of therapists delivering the intervention.

## Measures of treatment effect

### Binary data

We had planned to calculate the risk ratio and corresponding 95% confidence interval (95% CI) for binary outcomes. The number needed to treat for an additional beneficial outcome was to be calculated where appropriate. However, no binary data were available from the included studies.

### Continuous data

For studies where outcomes were measured on several occasions during each treatment intervention, we used the mean of all measurements from the second occasion onwards. Where raw data were available, the distributions of values were visually checked for skewness. Where skewness was found, we attempted to remove it by log-transformation. We then examined how log-transformation influenced the effect size estimate and used the more conservative estimate. We calculated the standardised mean difference (SMD) and corresponding 95% CI for all continuous outcomes. When combining different scales for the same outcome, it was necessary to standardise the effects in order to make them comparable. When combining results for the same scale, either the mean difference (MD) or SMD could have been used. We decided to use SMD in order to facilitate the interpretation of effect sizes as small (up to 0.2), medium (around 0.5) or large (0.8 and above) based on guidelines that are commonly used in the behavioural sciences (Cohen 1988; Schünemann 2011). It is noted that the choice of SMD or MD does not usually affect the significance level of the results and the authors cautiously assessed whether this was the case.

All SMDs, regardless of whether the study was a parallel or a cross-over design, were standardised by the pooled standard deviation between participants, rather than the standard deviation of the difference within participants. This is the standard procedure, which enables comparisons of different scales and facilitates interpretation of the magnitude of effects (Cohen 1988; Gold 2004). The calculation of the standard error then depended on the study design. For parallel designs, the standard error was calculated using the standard formulae for SMDs as implemented in RevMan and described in the RevMan handbook (Review Manager 2012). For

cross-over studies, we took into account the correlations within the participants as recommended and described in the literature on meta-analysis of cross-over studies (Elbourne 2002; Higgins 2011b).

### Unit of analysis issues

Where appropriate, we combined the results of cross-over trials with the results of parallel-group trials. Data from washout periods in cross-over studies were excluded from the analysis. For studies comparing more than two experimental groups, such as a music therapy intervention, a comparable non-music intervention, and an independent play condition, we compared the music therapy intervention with the non-music intervention as its 'placebo' condition.

### Dealing with missing data

We assessed loss to follow-up and drop-outs in the included studies as reported in the 'Risk of bias' tables. All but two of the included studies had complete data for all participants and therefore an intention-to-treat analysis was straightforward. We did not impute missing values. For analyses containing studies where drop-outs occurred (Kim 2008; Thompson 2012a), we examined the impact of studies with high drop-out rates using sensitivity analyses where these studies were excluded.

### Assessment of heterogeneity

Because statistical tests of heterogeneity have low power, particularly when the number of studies is low, we relied primarily on descriptive analyses of heterogeneity. We visually inspected forest plots for consistency of results and calculated the  $I^2$  statistic (Higgins 2002), which describes the proportion of variation in point estimates that is due to heterogeneity rather than sampling error. We supplemented this by calculating the  $\chi^2$  statistic to determine the strength of evidence that the heterogeneity was genuine. We investigated possible sources of heterogeneity when it was detected.

### Assessment of reporting biases

We planned to use funnel plots to investigate any relationship between effect size and study precision in cases where 10 or more studies were pooled for an outcome.

### Data synthesis

We conducted a meta-analysis utilising available or calculated SMDs. A fixed-effects model was used for all analyses. If a common effect size was not tenable due to heterogeneity, we considered a random-effects model. In addition to the fixed-effects analyses, we also examined whether random-effects analyses would have

altered the statistical significance of the results and reported any such difference.

### **Subgroup analysis and investigation of heterogeneity**

When heterogeneity was identified, we examined the impact of clients' age, intensity of therapy (i.e. number and frequency of music therapy sessions), and treatment quality in subgroup analyses.

### **Sensitivity analysis**

We conducted sensitivity analyses to determine the impact of study quality on outcome for included studies of different quality (e.g. studies with high attrition rates, see above).

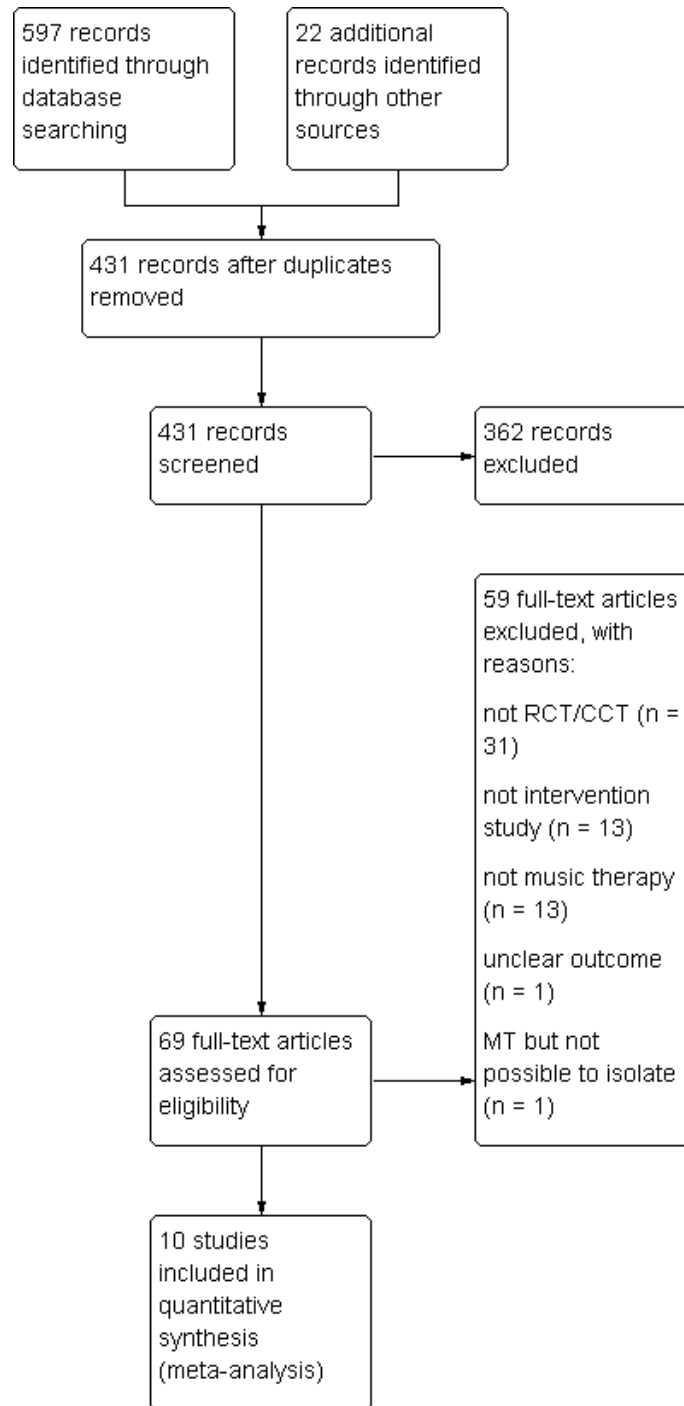
## **RESULTS**

## **Description of studies**

### **Results of the search**

Electronic searches conducted in July 2013 yielded a total of 431 records after deduplication. Sixty-nine of these were deemed potentially relevant and selected for closer inspection. Thirty-one studies were excluded because they were not RCTs or CCTs. Thirteen studies were excluded because they evaluated an assessment rather than an intervention. Thirteen studies were excluded because the intervention was not music therapy. One study was excluded because the outcome measure was unclear, and another study was excluded because it was not possible to isolate music therapy from other interventions. Ten studies met the inclusion criteria for this review. One relevant ongoing study was identified. [Figure 1](#) shows a flow diagram of search results.

**Figure 1. Study flow diagram.**





## Included studies

Ten studies met the criteria for the review (see [Characteristics of included studies](#)). Of these, three studies were included in the first version of this review ([Brownell 2002](#); [Buday 1995](#); [Farmer 2003](#)), and seven studies were added for this update ([Arezina 2011](#); [Gattino 2011](#); [Kim 2008](#); [Lim 2010](#); [Lim 2011](#); [Thomas 2003](#); [Thompson 2012a](#)). Nine were randomised trials. One study utilised a 'counterbalanced' sequence generation ([Brownell 2002](#)). Seven of the trials were short-term studies comparing music therapy to a 'placebo' type therapy, and were conducted in the USA ([Arezina 2011](#); [Brownell 2002](#); [Buday 1995](#); [Farmer 2003](#); [Lim 2010](#); [Lim 2011](#); [Thomas 2003](#)). A medium-term Korean study also compared music therapy to a 'placebo' condition of play sessions ([Kim 2008](#)). Two medium-term studies from Brazil ([Gattino 2011](#)), and Australia ([Thompson 2012a](#)), compared music therapy to standard care. Other characteristics of these studies are described below.

## Length of trials

The period under investigation in the included studies ranged from one week ([Farmer 2003](#); [Lim 2010](#)), to eight months ([Kim 2008](#)).

The duration of each treatment intervention was one week in four studies ([Brownell 2002](#); [Buday 1995](#); [Farmer 2003](#); [Lim 2010](#)), and two weeks in another study ([Lim 2011](#)). In the other studies, music therapy was applied for a period varying from five weeks ([Arezina 2011](#)), to seven months ([Gattino 2011](#)). No later follow-up assessments were included in any of the studies.

## Participants

The participants in the included studies were between two and nine years of age, with the majority being boys (range 80% to 100%). All participants had received a diagnosis of ASD. Both non-verbal and verbal children were included. In six studies symptom severity or levels of cognitive abilities, or both, were also specified ([Arezina 2011](#); [Buday 1995](#); [Gattino 2011](#); [Kim 2008](#); [Lim 2010](#); [Thompson 2012a](#)). Standardised tools for diagnosis were used in [Buday 1995](#) (i.e. participants ranging from mildly to moderately autistic according to the Childhood Autism Rating Scale, CARS), [Kim 2008](#) (i.e. participants meeting criteria for the Korean version of the CARS), [Lim 2010](#) (i.e. participants classified as being of high or low functioning level according to the CARS or the Autism Diagnostic Interview Revised), and [Thompson 2012a](#) (i.e. participants' severity of symptoms ranging from moderate to severe according to the Social Responsiveness Scale, SRS; [Constantino 2005](#)). With regard to cognitive level, [Buday 1995](#) reported participants to be ranging from mildly to severely mentally retarded

(according to DSM III-R), and [Gattino 2011](#) specified the participants' level of intelligence as ranging from intellectual disability to above average intelligence according to the Brazilian version of Raven's Coloured Progressive Matrices ([Pasquali 2002](#)).

## Setting

The participants received therapy either at home ([Thompson 2012a](#)), at school ([Brownell 2002](#); [Buday 1995](#)), in hospital ([Gattino 2011](#)), at outpatient therapy centres ([Arezina 2011](#); [Kim 2008](#)), or a combination thereof ([Farmer 2003](#); [Lim 2010](#)). For [Lim 2011](#) and [Thomas 2003](#), the therapy setting was not reported.

## Study size

Six of the studies had extremely small sample sizes, varying from four to ten participants per study ([Arezina 2011](#); [Brownell 2002](#); [Buday 1995](#); [Farmer 2003](#); [Kim 2008](#); [Thomas 2003](#)). [Farmer 2003](#) was the only study that did not use a cross-over design. Cross-over designs were used in the other studies to partly compensate for the small sample sizes. Three studies had slightly larger sample sizes of 24, 22, and 23 respectively ([Gattino 2011](#); [Lim 2011](#); [Thompson 2012a](#)). [Lim 2010](#) had a sample size of 50.

## Interventions

### Music therapy

The majority of studies included in this review examined music therapy in an individual (i.e. one-to-one) setting. [Thompson 2012a](#) applied a family-based setting where parents or other family members were also involved in therapy sessions.

In five studies music therapy was provided on a daily basis ([Brownell 2002](#); [Buday 1995](#); [Farmer 2003](#); [Lim 2010](#); [Lim 2011](#)). The duration of the music therapy intervention was only one or two weeks in all those studies. In the other studies ([Arezina 2011](#); [Gattino 2011](#); [Kim 2008](#); [Thomas 2003](#); [Thompson 2012a](#)), music therapy was provided on a weekly basis for periods ranging from five weeks ([Arezina 2011](#)) to seven months ([Gattino 2011](#)).

[Brownell 2002](#), [Buday 1995](#), [Farmer 2003](#), [Lim 2010](#), and [Lim 2011](#) utilised a highly structured approach to music therapy using mostly receptive techniques (i.e. listening to live or, in the case of [Lim 2010](#), pre-recorded music presented by the therapist). Songs sung by the music therapist were composed or chosen individually for the participants and were usually used with specific aims. For example, songs were based on a social story addressing a central problem behaviour of the particular individual in treatment



(Brownell 2002); they contained signs and words to be learned (Buday 1995; Lim 2010; Lim 2011); or they were used to build a relationship and to provide a safe and understandable structure for the participants in the study (Farmer 2003). Active music-making by the participants, which is often typical for music therapy in clinical practice (Wigram 2006), was reported in only one of those studies (Farmer 2003). Participants were allowed to play guitar and drums. Playing instruments was partly used to reinforce adjusted behaviour. The report did not specify whether, or in what ways, the therapist improvised or otherwise played music together with the client.

In the other five studies particular emphasis was put on the interactive and relational aspects of music therapy (Arezina 2011; Gattino 2011; Kim 2008; Thomas 2003; Thompson 2012a). Music therapy techniques included improvisation, songs, and structured musical games. Interventions followed a non-directive approach and focused on engaging the child in musical interaction, offering opportunities for the child to make choices and to initiate contact. Generally, the therapist's interventions were depicted as drawing on the individual child's skills, interests, preferences, and motivations as well as on their immediate expression and behaviour. By attuning to the child musically and emotionally, the therapists create moments of synchronisation that help the child to experience and recognise core elements of reciprocal communication (Kim 2008; Schumacher 1999a; Schumacher 1999b; Stephens 2008; Thompson 2012a; Wigram 2009).

Some of the studies employed specifically developed treatment guidelines in the form of a treatment contingency plan (Thompson 2012a), or a treatment manual (Kim 2008). In these protocols, principles and procedures of therapy are specified whilst allowing the therapist to adapt interventions flexibly according to the child's needs and the specific requirements of the situation.

### 'Placebo' therapy

Six of the studies included in this review compared music therapy to some kind of 'placebo' activity to control for the non-specific effects of therapeutic attention. Since in all of these studies music was considered as the specific ingredient of music therapy, the placebo conditions were constructed to closely match the music therapy condition, only that music was not used. For example, a social story was read instead of sung to the participants (Brownell 2002); rhythmic or normal speech was used instead of singing (Buday 1995; Lim 2010; Lim 2011); the same play activities were offered without using songs or music instruments (Farmer 2003); or the therapist engaged in interaction with the child by responding to the child's behaviour non-musically and using non-music toys (Arezina 2011; Kim 2008; Thomas 2003).

### Other conditions

Two of the included studies compared music therapy to standard care (Gattino 2011; Thompson 2012a). In the Thompson 2012a

study, participants received varying forms of services and support from early childhood intervention centres. Gattino 2011 reported that participants received routine clinical services, including medical examinations and consultations.

In addition to the music therapy and non-music interventions, Brownell 2002 reported outcomes during a baseline and a washout period with no intervention. These data were not used in this review. Arezina 2011 also observed behaviour in an 'independent play' group, which we considered was neither 'placebo' therapy nor 'standard care'. Therefore, data from this group were not included in this review. Lim 2010 and Lim 2011 compared music training to both a speech training and a 'no training' group. For this review, we included data from the comparison between the music and the non-music groups.

### Outcome measures

Both generalised and non-generalised outcomes were used in the included studies. Non-generalised outcomes refer to changes in the child's non-generalised behaviour in the same setting where the intervention takes place, as opposed to generalised outcomes which are observed in other settings (Warren 2011).

### Primary outcomes

#### *Social interaction*

Social interaction skills were examined in three studies (Gattino 2011; Kim 2008; Thompson 2012a). All three studies measured this outcome outside of the treatment context using published scales. Gattino 2011 utilised the 'social communication' domain of the Childhood Autism Rating Scale, Brazilian version (CARS-BR; Pereira 2008; Rapin 2008), a diagnostic behaviour observation tool administered by investigators blind to group allocation. Kim 2008 used the 'social approach' subscale of the Pervasive Developmental Disorder Behavior Inventory, Korean version (PDDBI; Cohen 1999), which was filled out by professionals (i.e. a teacher or a therapist of the child) who were blind to experimental condition. Thompson 2012a utilised social interaction measures, including the Social Responsiveness Scale (SRS; Constantino 2005), rated by parents, and the 'Interpersonal Relationships' and 'Play and Leisure Time' subscales of the Vineland Social-Emotional Early Childhood Scales (Vineland SEEC; Sparrow 1998), rated by the therapist following an interview with parents.

Kim 2008 also investigated behaviours related to social interaction in the intervention setting. These measures included frequency and duration of the child's turn-taking and frequency of imitation behaviours. The coding procedure was conducted by the lead investigator by microanalytically (second by second) observing DVD recordings, with subsequent coding supplemented by a trained research assistant who was blind to session order.

### *Communicative skills: non-verbal*

Nonverbal (i.e. gaze-related and gestural) communicative skills were examined in five studies (Buday 1995; Farmer 2003; Gattino 2011; Kim 2008; Thompson 2012a). Three studies addressed the participants' behaviour within therapy sessions (Buday 1995; Farmer 2003; Kim 2008). Independent observers counted the number of communicative gestures (e.g. imitating a sign or motion, eye contact) in the session. In the Buday 1995 study, the outcome consisted simply of the frequency count of appropriate gestures within a session. In the Farmer 2003 study, a completed gesture was given a score of two, and an attempt a score of one, and the outcome consisted of the sum of these scores for all attempted and completed gestures within a session. In the Kim 2008 study, frequency and duration of eye contact (i.e. the child looking at the therapist) was coded by microanalytic analysis of the session material. The exact criteria for what was seen as a non-verbal communicative skill were different between the three studies. The measures used for this outcome in these three studies were not published separately.

Three studies used published instruments for measuring generalised non-verbal communicative skills (Gattino 2011; Kim 2008; Thompson 2012a). Gattino 2011 applied the 'nonverbal communication' subscale of the CARS-BR as described above. Kim 2008 used the abridged version of the Early Social Communication Scales (ESCS; Mundy 2003), a structured toy play assessment yielding frequency scores of behaviours such as 'initiation of joint attention' and 'responding to joint attention'. The scoring was administered by the researcher and by two trained research assistants who were blind to group assignment. Thompson 2012a utilised the MacArthur-Bates Communicative Development Inventories - Words and Gestures (MBCDI-W&G; Fenson 2007), a parent-report measure assessing early communication skills. The subscale 'actions and gestures used' was also included in this outcome category.

### *Communicative skills: verbal*

Communicative skills in verbal communication were addressed in six studies (Buday 1995; Farmer 2003; Gattino 2011; Lim 2010; Lim 2011; Thompson 2012a). For Buday 1995, Farmer 2003, Lim 2010, and Lim 2011, independent observers rated in-session behaviour by counting the frequency of appropriate verbal responses in a manner similar to the previous outcome. The non-generalised outcome measures used in four studies were unpublished (Buday 1995; Farmer 2003; Lim 2010; Lim 2011). The other two studies used published instruments for measuring generalised verbal communicative skills. Gattino 2011 used the 'verbal communication' subscale of the CARS-BR as described above. Thompson 2012a used the subscales 'phrases understood', 'words understood' and 'words produced' of the MBCDI-W&G as described for the previous outcome.

### *Initiating behaviour*

Three studies investigated children's initiating behaviour as observed within the intervention setting using unpublished measures (Arezina 2011; Kim 2008; Thomas 2003). In Arezina 2011, the researcher coded videotaped sessions for 'requesting (initiating joint attention)' behaviours such as pointing, giving an object to the therapist, or touching the therapist while making eye contact; an independent observer additionally coded a third of the session material. In Kim 2008, the frequency of 'initiation of engagement' behaviours was coded as described above (microanalysis of DVD recordings by the researcher, supplemented by coding by a research assistant who was blind to session order). In the Thomas 2003 study, 'requesting behavior' was defined in a manner similar to the Arezina 2011 study, and coded by a trained music therapy intern using video recordings.

### *Social-emotional reciprocity*

Skills related to social-emotional reciprocity were addressed in the Kim 2008 study using behaviours within the treatment context that were coded through microanalytic analysis using unpublished measures. Child behaviours included in this outcome category were frequency and duration of both 'emotional synchronicity' and 'musical synchronicity'.

### *Secondary outcomes*

#### *Social adaptation*

Three studies investigated behaviours related to social adaptation within the interventions setting (Arezina 2011; Kim 2008; Thomas 2003). In Arezina 2011 and Thomas 2003, videotaped sessions were coded for 'interaction (engaging in joint attention)' and 'on-task behavior', respectively; this included activities such as following a direction, physically manipulating a toy in a functional manner, and imitating a movement or vocal sound. In Kim 2008, sessions were scored by frequencies of 'compliant response', 'non-compliant response', and 'no response'. Brownell 2002 addressed individually targeted repetitive behaviours. This outcome was categorised as 'Behavioural problems' in the first version of this review. Occurrence of behaviour was assessed outside therapy sessions. Independent observers (i.e. teachers) counted how often the targeted behaviour occurred in the classroom. The frequency count was used as the outcome measure. No published scale was used.

#### *Joy*

Behaviours associated with the frequency and duration of joy (i.e. smiling and laughing) on the part of the child were addressed in one study (Kim 2008). The researcher described occurrences of joy as a clinically significant motivational factor for the child to join in shared activities with the therapist. Scores were determined through microanalytic observation of videotaped sessions.

### Quality of parent-child relationship

In two studies, features of the quality of parent-child relationships were examined (Kim 2008; Thompson 2012a). Kim 2008 used the Mother Play Intervention Profile (MPIP), a measure specifically developed for her study to assess characteristics of interactions between mothers and children with ASD during a casual play situation at their home. Features such as the amount of initiation of interaction by the child and the mother and the degree of structuring activities introduced by the mother were scored on a four-point Likert scale. Scores were based on video observations conducted by the researcher, supplemented by an independent observer's coding for a third of the sessions. Thompson 2012a used the Parent-Child Relationship Inventory (PCRI; Gerard 2005), a self report questionnaire for parents to assess the parent-child relationship and parents' attitudes towards parenting.

### Excluded studies

Fifty-nine studies were excluded. Thirty-one studies were excluded because they did not have an RCT or CCT design (20 case series, i.e. studies comparing different treatments that all participants received in the same order; 11 case studies). Thirteen studies were excluded because these studies involved an assessment rather than an intervention (e.g. assessing traits of people with ASD using music therapy techniques). Thirteen studies were excluded because the intervention was not music therapy (e.g. auditory integration training). One study was excluded because the outcome measure was unclear; and one study was excluded because it was not possible to isolate music therapy from other interventions (see [Characteristics of excluded studies](#)).

### Ongoing studies

One relevant, longer-term study of improvisational music therapy was still ongoing when this review was written (ISRCTN78923965); see [Characteristics of ongoing studies](#).

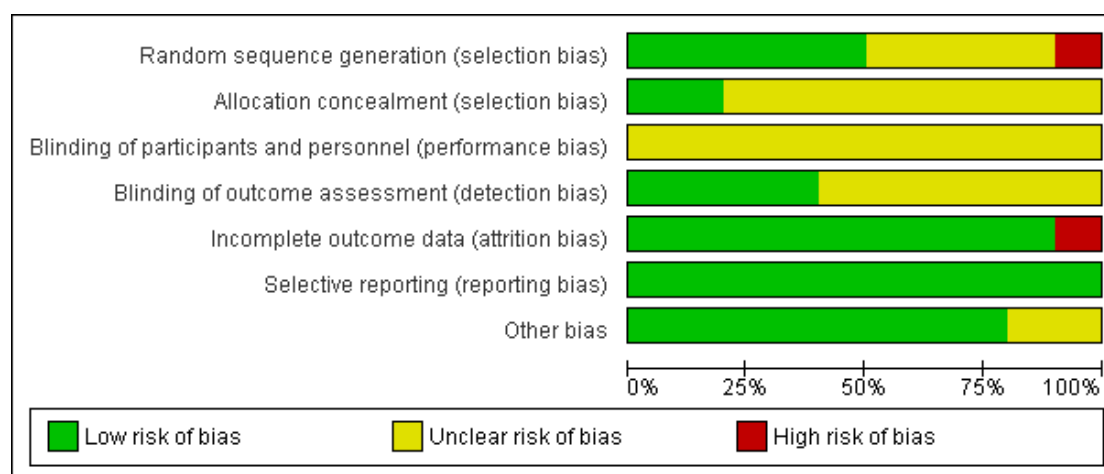
### Awaiting assessment

We were able to assess all studies for eligibility. No studies were awaiting assessment.

### Risk of bias in included studies

A visual representation of the included studies' risk of bias for each domain, as specified below, is shown in [Figure 2](#). [Figure 3](#) provides a summary of the risk of bias results for each included study.

**Figure 2. Risk of bias graph: review authors' judgements about each risk of bias item presented as percentages across all included studies.**



**Figure 3. Risk of bias summary: review authors' judgements about each risk of bias item for each included study.**

	Random sequence generation (selection bias)	Allocation concealment (selection bias)	Blinding of participants and personnel (performance bias)	Blinding of outcome assessment (detection bias)	Incomplete outcome data (attrition bias)	Selective reporting (reporting bias)	Other bias
Arezina 2011	+	?	?	?	+	+	+
Brownell 2002	-	?	?	?	+	+	+
Buday 1995	?	?	?	+	+	+	?
Farmer 2003	?	?	?	?	+	+	+
Gattino 2011	+	+	?	+	+	+	+
Kim 2008	+	?	?	?	-	+	+
Lim 2010	?	?	?	+	+	+	?
Lim 2011	+	?	?	+	+	+	+
Thomas 2003	?	?	?	?	+	+	+
Thompson 2012a	+	+	?	?	+	+	+

## Allocation

Seven of the included studies stated explicitly that randomisation was used to assign participants to treatment groups (Arezina 2011; Buday 1995; Farmer 2003; Gattino 2011; Kim 2008; Thomas 2003; Thompson 2012a). Methods of randomisation included using computer-generated random sequences for determining allocation to experimental condition (Gattino 2011; Kim 2008; Thompson 2012a), and a Latin Square for determining session order (Arezina 2011). In three studies, methods of randomisation and allocation concealment were not specified (Buday 1995; Farmer 2003; Thomas 2003). The remaining study used the term 'counterbalanced' to describe an assignment that was either random or quasi-random, but intended to be random (Brownell 2002).

## Blinding

Four of the included studies were single-blind, with blinded assessors (Buday 1995; Gattino 2011; Lim 2010; Lim 2011). In Kim 2008, some outcomes were coded by blinded assessors, while non-generalised outcome measures and two of the measures assessing generalised skills (ESCS, MPIP) were rated by the researcher and complemented by independent coders (inter-rater reliability ranging from 0.70 to 0.98). In Thompson 2012a, measures were based on parent reports; however, they contained internal safe-guards to address bias as evidenced by high correlations with non-parent rated measures and high test-retest correlations (e.g. Pearson's  $r = 0.70$ ,  $P$  value = 0.01, for the SRS's one-month test-retest reliability). No details about blinding of outcome assessment were reported in the other studies (Arezina 2011; Brownell 2002; Farmer 2003; Thomas 2003).

Five studies used more than one rater to independently assess outcomes. All of those studies reported a high inter-rater reliability for the assessment of outcomes (Arezina 2011: inter-observer agreement ranging from 85.7% to 98.9%; Brownell 2002: inter-rater reliability 0.86 to 0.94; Buday 1995: agreement rate 98%; Farmer 2003: agreement rate 91%; Kim 2008: inter-rater reliability 0.70 to 0.98, as reported above).

## Incomplete outcome data

Drop-outs were reported in two of the ten studies. In Kim 2008, five of the 15 participants initially enrolled dropped out, and data from drop-outs were excluded, yielding a high risk of bias due to attrition for this study. In Thompson 2012a, two of 23 participants dropped out, and an intention-to-treat analysis was applied, so we consider the related risk of bias to be low.

## Selective reporting

There was no evidence of selective reporting of outcomes in the included studies. In the Kim 2008 study, some outcomes were only reported in the thesis but not in the journal articles, but we included all outcomes in the meta-analysis.

## Other potential sources of bias

We considered inadequate music therapy methods and inadequate music therapy training of therapists as additional potential sources of bias. With the exception of Buday 1995, where we found music therapy methods and the training of the person delivering the intervention to be of unclear adequacy, we detected none of these sources of bias in the included studies.

## Preparation of data for meta-analysis

Buday 1995 reported means, standard deviations, and F test results for the outcomes described above. From these statistics it was possible to calculate a SMD and standard error as appropriate for cross-over studies. Similarly, we calculated SMDs from data reported in Arezina 2011, Kim 2008, Thomas 2003, and Thompson 2012a. For the other studies individual patient data were extracted from tables or graphs (Brownell 2002; Farmer 2003; Gattino 2011). We screened the data for skewness before data synthesis. Data from the Farmer 2003 study showed a skewed distribution. A log transformation would have removed the skewness, but would also have increased the effect size estimate. Therefore, we decided to use the more conservative original scale. Similarly, we found skewed distributions in 13 of the 15 non-generalised outcomes in the Kim 2008 study (all except 'compliant response frequency' and 'no response frequency'). We calculated SMDs both using log-transformed scores and raw scores and used the smaller effect size. The raw-score-based effect size was smaller than the log-transformed effect size in three of the 13 outcomes: 'frequency of eye contact', 'duration of eye contact', and 'frequency of initiation of engagement'.

## Effects of interventions

See: [Summary of findings for the main comparison Music therapy compared to 'placebo' therapy or standard care for autism spectrum disorder](#)

We used fixed-effects analysis for all outcomes, but checked whether the effect size estimate changed if a random-effects model was used. P values for each outcome indicate that results remained statistically significant using random-effects analysis. They are reported below.

## Primary outcomes

### Social interaction

Kim 2008 assessed social interaction skills within the intervention context. Post-treatment difference between the music therapy and the control group yielded an SMD effect size of 1.06 (95% CI 0.02 to 2.10), indicating a large effect (Cohen 1988; Schünemann 2011).

Three studies measured generalised social interaction skills using standardised scales (Gattino 2011; Kim 2008; Thompson 2012a). The SMD in generalised social interaction between music therapy and control groups was 0.71 (95% CI 0.18 to 1.25), indicating a moderate to large effect (Cohen 1988; Schünemann 2011). We checked whether the results changed when using a random-effects model, and found no difference (SMD 0.71, 95% CI 0.18 to 1.25,  $P$  value = 0.009). The results were homogeneous ( $\text{Chi}^2 = 1.41$ ,  $P$  value = 0.49,  $I^2 = 0\%$ ) and do not require examination of moderators (see Analysis 1.1).

We conducted a sensitivity analysis excluding data from the high-attrition study (Kim 2008), and found that the effect for generalised skills remained statistically significant ( $P$  value = 0.03). No heterogeneity was detected for this analysis ( $\text{Chi}^2 = 1.38$ ,  $P$  value = 0.24,  $I^2 = 28\%$ ).

### Communicative skills: non-verbal

Three studies used measures of non-generalised non-verbal communicative skills through continuous scales addressing observed behaviour (Buday 1995; Farmer 2003; Kim 2008). The effect size for difference in non-generalised non-verbal communicative skills between music therapy and control groups was 0.57 (95% CI 0.29 to 0.85), indicating a moderate effect. We checked whether the results changed when using a random-effects model, and found that the effect remained statistically significant (SMD 1.00, 95% CI 0.10 to 1.90,  $P$  value = 0.03). Statistically significant heterogeneity was detected for this pooled analysis ( $\text{Chi}^2 = 5.15$ ,  $P$  value = 0.08,  $I^2 = 61\%$ ). This heterogeneity may be related to the relatively high attrition rate in Kim 2008, or the unclear quality of music therapy methods and therapist's training in Buday 1995. When excluding data from either of the studies, the overall effect remained statistically significant (SMD 0.50, 95% CI 0.22 to 0.79,  $P$  value = 0.0006; and SMD 1.56, 95% CI 0.61 to 2.50,  $P$  value = 0.001, respectively), resulting in the decision to keep these studies in the pooled analysis.

Three studies assessed generalised non-verbal communicative skills using published standardised scales (Gattino 2011; Kim 2008; Thompson 2012a). The effect size for difference between music therapy and control groups was 0.48 (95% CI -0.02 to 0.98), suggesting that children receiving music therapy had similar non-verbal communicative skills after treatment as children receiving 'placebo' therapy or standard care (Analysis 1.2). Changing the

model of analysis to random-effects did not change the statistical significance of the results (SMD 0.48, 95% CI -0.02 to 0.98,  $P$  value = 0.06). No heterogeneity was detected for this comparison ( $\text{Chi}^2 = 1.33$ ,  $P$  value = 0.51,  $I^2 = 0\%$ ).

A sensitivity analysis excluding the study with a high drop-out rate (Kim 2008) did not change the statistical significance of the results for generalised non-verbal communicative skills (SMD 0.31, 95% CI -0.28 to 0.89,  $P$  value = 0.31). However, the overall effect across domains (then calculable as none of the remaining studies is represented in both domains) was significant (SMD 0.47, 95% CI 0.21 to 0.73;  $\text{Chi}^2 = 1.32$ ,  $P$  value = 0.72,  $I^2 = 0\%$ ), indicating a moderate effect.

### Communicative skills: verbal

Four studies investigated non-generalised verbal communicative skills using continuous scales addressing observed behaviour (Buday 1995; Farmer 2003; Lim 2010; Lim 2011). The effect size for difference in non-generalised verbal communicative skills was 0.33 (95% CI 0.16 to 0.50), indicating a small to moderate effect favouring music therapy over the 'placebo' intervention, suggesting that improvement in verbal communicative skills was more likely to occur with music therapy. The results did not change when using a random-effects model (SMD 0.33, 95% CI 0.16 to 0.50,  $P$  value = 0.0002). No heterogeneity was detected for this comparison ( $\text{Chi}^2 = 0.72$ ,  $P$  value = 0.87,  $I^2 = 0\%$ ).

Generalised verbal communicative skills were assessed in two studies using standardised scales (Gattino 2011; Thompson 2012a). The effect size for difference in generalised non-verbal communicative skills was 0.30 (95% CI -0.28 to 0.89), suggesting that children receiving music therapy had similar verbal communicative skills after treatment as children receiving standard care. No heterogeneity was detected for this comparison ( $\text{Chi}^2 = 0.01$ ,  $P$  value = 0.93,  $I^2 = 0\%$ ), and using a random-effects model did not change the results (SMD 0.30, 95% CI -0.28 to 0.89,  $P$  value = 0.31).

The overall effect size for difference in verbal communicative skills between music therapy and control groups was 0.33 (95% CI 0.16 to 0.49), indicating a small to moderate effect (see Analysis 1.3).

### Initiating behaviour

Three studies reported measures of non-generalised initiating behaviour using continuous scales (Arezina 2011; Kim 2008; Thomas 2003). For Arezina 2011 and Thomas 2003, we averaged participants' behaviour over all therapy sessions except the first one and calculated an SMD with a standard error. The effect size was 0.73 (95% CI 0.36 to 1.11), which indicates a close to large effect in favour of music therapy (see Analysis 1.4). Possible heterogeneity was detected for this analysis ( $\text{Chi}^2 = 3.91$ ,  $P$  value = 0.14,  $I^2 = 49\%$ ), but when the high-attrition study (Kim 2008) was excluded from analysis, the overall effect remained statistically



significant ( $P$  value = 0.009) and heterogeneity was no longer detected ( $\text{Chi}^2$  = 0.18,  $P$  value = 0.67,  $I^2$  = 0%). Using a random-effects model did not change the results (SMD 0.80, 95% CI 0.19 to 1.41,  $P$  value = 0.01).

### Social-emotional reciprocity

Kim 2008 applied measures of social-emotional reciprocity within the intervention context using continuous scores for the child displaying 'emotional synchronicity' (frequency and duration) and 'musical synchronicity' (frequency and duration). Post-treatment difference between the music therapy and the control group yielded an effect size of 2.28 (SMD 95% CI 0.73 to 3.83), indicating a large effect (see Analysis 1.5). However, this result must be interpreted with caution since data came from a study with a small sample size and a high drop-out rate.

### Adverse events

No deterioration on a primary outcome or other adverse events were reported as a result of treatment in any of the included studies.

## Secondary outcomes

### Social adaptation

Three studies used continuous scales addressing observed behaviour for examining social adaptation of children within the intervention setting (Arezina 2011; Kim 2008; Thomas 2003). This was done by observing behaviours of 'interaction (engaging in joint attention)' (Arezina 2011), 'on-task behavior' (Thomas 2003), and frequencies of 'compliant response', 'non-compliant response', and 'no response' (Kim 2008). As described above, we averaged participants' behaviour over all therapy sessions except the first one for Arezina 2011 and Thomas 2003. Data from Kim 2008 were also synthesised by calculating an SMD with a standard error. The effect size for difference in non-generalised social adaptation between music therapy and 'placebo' therapy groups was 1.15 (95% CI 0.69 to 1.61), indicating a large effect. No heterogeneity was detected for this comparison ( $\text{Chi}^2$  = 2.87,  $P$  value = 0.24,  $I^2$  = 30%). Using a random-effects model did not change the results (SMD 1.23, 95% CI 0.61 to 1.86,  $P$  value = 0.0001). The effect on non-generalised social adaptation remained statistically significant ( $P$  < 0.00001) in a sensitivity analysis excluding the high drop-out study (Kim 2008). Heterogeneity increased to 65%, but the effect remained statistically significant also when a random-effects analysis was used (SMD 1.50, 95% CI 0.24 to 2.76),  $P$  value = 0.02).

Data for generalised social adaptation were available from only one study using measures of a continuous scale for observed behaviour (Brownell 2002). We averaged participants' behaviour over all days

in therapy except the first one and calculated an SMD with a standard error. The resulting SMD effect size was 0.24 (95% CI 0.02 to 0.46), indicating a small effect, which suggests that music therapy may be slightly more beneficial than a similar verbal therapy in increasing social adaptation outside the therapy context.

The overall effect size for difference in social adaptation between music therapy and control groups was 0.41 (95% CI 0.21 to 0.60), indicating a small to moderate effect. The  $\text{Chi}^2$  and  $I^2$  statistics showed heterogeneity of studies across subcategories ( $\text{Chi}^2$  = 15.34,  $P$  value = 0.002,  $I^2$  = 80%), indicating that the Brownell 2002 study examining generalised skills was different from the more recent studies measuring non-generalised social adaptation abilities. Applying a random-effects model did not change the results (SMD 0.95, 95% CI 0.22 to 1.68,  $P$  value = 0.01).

### Quality of life in school, home and other environments

One study used an outcome that can be related to quality of life in the treatment environment by measuring frequency and duration of 'joy' displayed by the child within the therapy setting (Kim 2008). We combined data (frequency and duration) by calculating an SMD with a standard error. The resulting effect size was 0.96 (95% CI 0.04 to 1.88), indicating a large effect that suggests that an increase in displays of joy was more likely to occur in music therapy than in 'placebo' therapy. However, this result must be interpreted with considerable caution since data came from only one study with a small sample size and a high drop-out rate.

### Quality of family relationships

Two studies included measures connected to the quality of family relationships (Kim 2008; Thompson 2012a). The effect size across studies was 0.82 (95% CI 0.13 to 1.52), with no indication of heterogeneity between studies ( $\text{Chi}^2$  = 0.03,  $P$  value = 0.87,  $I^2$  = 0%). The results did not change when a random-effects model was calculated (SMD 0.82, 95% CI 0.13 to 1.52,  $P$  = 0.02). However, when excluding data from the study with high attrition (Kim 2008), the effect was no longer statistically significant ( $P$  value = 0.11), suggesting that these data must be interpreted with caution.

### Cognitive ability

None of the included studies investigated change in cognitive abilities such as concentration or intelligence.

### Hyperacusis

We did not find any reports on children's hypersensitivity to sound in any of the included studies.

## DISCUSSION

### Summary of main results

We found 10 RCTs that evaluated the effects of music therapy for children with ASD aged two to nine years using non-generalised and generalised outcomes. Non-generalised outcomes refer to changes of behaviour as observed in the treatment context, while generalised outcomes are measured outside of the therapy environment in the child's daily life. Music therapy was compared to standard care, or to a 'placebo' therapy which attempted to control for all non-specific elements of music therapy, such as the attention of a therapist or the client's motivation to participate in therapy. We calculated SMDs and conducted meta-analyses using a fixed-effect model on five primary outcomes and three secondary outcomes. The effect sizes found can be interpreted in accordance with common guidelines for interventions in the behavioural sciences (Cohen 1988; Schünemann 2011), where effect sizes of up to 0.2 are considered small, those around 0.5 are moderate, and those at 0.8 and above are large. The results show evidence of moderate to large effects of music therapy for the primary outcomes non-generalised social interaction skills, generalised social interaction skills, non-generalised non-verbal communicative skills, initiating behaviour, and social-emotional reciprocity, and for the secondary outcomes joy and quality of parent-child relationships. Small to moderate effect sizes resulted for the primary outcome verbal communicative skills and the secondary outcome social adaptation. It is interesting to note that non-verbal communicative skills, which may be more closely related to non-verbal communication within music therapy, appeared to show greater change than verbal communicative skills. However, it may also be that non-verbal communicative skills are relatively easier to address than verbal communicative skills especially in low-functioning children and through short- to medium-term interventions, and particularly regarding skills to be generalised beyond the treatment context. Results were statistically significant for all but two outcome categories under investigation, suggesting a beneficial effect of music therapy when compared to 'placebo' therapy or standard care. The only two sub-categories where the effect was not statistically significant were generalised non-verbal and generalised verbal communicative skills. Using the GRADE system (GRADEpro 2008), we rated the qual-

ity of the evidence as 'moderate' for four outcomes and 'low' for three outcomes included in the [Summary of findings for the main comparison](#). Even with Bonferroni correction, which is known to be overly conservative when outcomes are correlated, all primary outcomes that showed significant effects remained statistically significant (all P values were below Bonferroni-corrected alpha level  $0.05/5 = 0.01$ ). Therefore, alpha error accumulation can be excluded as a source of error.

### Overall completeness and applicability of evidence

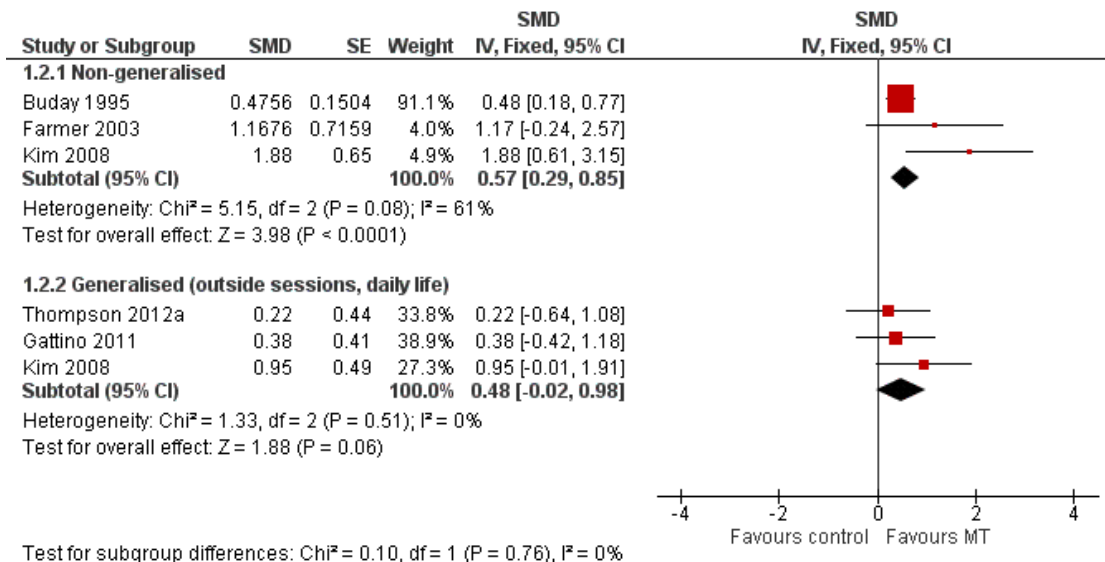
#### Music therapy conditions

Three studies that were included in the first version of this review (Gold 2006), were of limited generalisability to clinical practice (Brownell 2002; Buday 1995; Farmer 2003). These studies only used a limited subset of the music therapy techniques described in the clinical literature in the experimental treatment conditions. Receptive music therapy techniques with a high level of structuring predominated in those interventions; improvisational techniques were not utilised. However, improvisational techniques are widely used in many parts of the world (Edgerton 1994; Gattino 2011; Holck 2004; Kim 2006; Schumacher 1999a; Schumacher 1999b; Thompson 2012a; Thompson 2012b; Wigram 2006; Wigram 2009). Five of the studies added in this review update (Arezina 2011; Gattino 2011; Kim 2008; Thomas 2003; Thompson 2012a), reflect and emphasise improvisational and relational approaches to music therapy, thus considerably increasing the applicability of findings to clinical practice and hence the external validity of this review.

The findings of this review may suggest that more flexible, child-led approaches yield better outcomes, as indicated by the results for non-generalised non-verbal communicative skills, where receptive techniques as applied in Buday 1995 and Farmer 2003 yielded smaller effects than the improvisational method provided in Kim 2008 (see Figure 4). This complies with findings about musical interactions by Stephens who states that, "children with autism related reciprocally to others when they engaged in pleasurable, child-led, shared attention routines" (Stephens 2008, pp. 667-8).



**Figure 4. Forest plot of comparison: I Music therapy vs. 'placebo' therapy or standard care, outcome: I.2 Communicative skills: non-verbal.**



Generally speaking, music therapy for children with ASD should be backed by research evidence from both music therapy and related fields, aiming at cooperation with others involved in treatment and care of clients, active engagement of clients, and establishing structure, predictability, and routines. It is important to note that providing structure does not equal rigidity within interventions. Music contains rhythmic, melodic, harmonic, and dynamic structure which, when applied systematically and skilfully, can be effective in engaging children with ASD. Intervention strategies employing music improvisation are usually not pre-structured in the sense of a fixed manual. In recent years, flexible but systematic treatment protocols for music therapy have been developed in clinical practice and research investigations in ASD (Geretsegger 2012; Kim 2006; Thompson 2012a; Wigram 2006) as well as in other fields (Rolvjord 2005). As described above (see [Included studies](#)), two of the studies in this review have successfully applied such guidelines (Kim 2008; Thompson 2012a). More studies employing therapy approaches, which are close to those applied in clinical practice, will be needed in order to improve the clinical applicability of research findings.

### Control conditions

Eight of the included studies used a dismantling strategy to isolate the effect of the specific 'ingredients' of music therapy by setting up comparison conditions, which were very similar to the music therapy interventions, excluding only the music component (Arezina 2011; Brownell 2002; Buday 1995; Farmer 2003; Kim 2008; Lim 2010; Lim 2011; Thomas 2003). Any conclusion from

such comparisons will therefore address the effects of specific music therapy techniques, rather than the absolute effects of music therapy in general. This type of design is justified when exploring music therapy intervention strategies. However, such comparison conditions may introduce some artificiality into the studies through selecting out and applying a single intervention strategy. This is not typically undertaken in clinical treatment, although it does isolate specific components of music therapy. In the broader field of psychotherapy research, similar constructions of 'placebo' therapy to control for the therapist's attention and the non-specific elements have been broadly used (Kendall 2004, pp. 20-1). However, research on common factors in psychotherapy raise the question of how adequate it is conceptually, and also whether it is technically possible to separate the active from the non-active elements of therapy (Lambert 2004, pp. 150-2).

### Duration, population, and outcomes

ASD as a pervasive developmental disorder is a chronic condition, which requires sustained therapeutic intervention starting as early as possible. In clinical reports for ASD, music therapy is usually described as a longer-term intervention, and given the typical emergence of entrenched and deteriorating behaviour, therapeutic intervention relies on consolidating progress over time. With the treatment duration of included studies ranging up to seven months, we consider this review's findings as sufficiently applicable to clinical contexts.

With regards to the population addressed, the applicability of the

findings is limited to the age groups included in the studies. No direct conclusions can be drawn about music therapy in adults with ASD.

The outcomes addressed in the included studies cover areas that form the core of the condition and that we consider as highly relevant to individuals with ASD and their families.

### Quality of the evidence

As indicated by the ratings of evidence presented in the [Summary of findings for the main comparison](#) ('moderate' for four, 'low' for three of the relevant outcomes), the body of evidence identified allows for fairly robust conclusions regarding this review's objectives. Limitations to the methodological strength of the evidence are due to the small sample sizes of the 10 included studies (4 to 50 participants) and the small total number of individuals under review ( $n = 165$ ). Additionally, only some of the outcomes used in the studies were published measurement tools, which hampers replicability of findings. Moreover, some of the measures in the included studies relied on reports from parents who were aware of their children's group allocation. However, change in children's skills as assessed by parents may reflect effects of interventions that are meaningful and relevant to clients and their families.

### Potential biases in the review process

One can never be completely sure that all relevant trials have been identified. However, our searches included not only exhaustive electronic and hand searches, but relied additionally on an existing international network of leading researchers in the field. Therefore, it seems unlikely that an important trial exists that did not come to our attention. Furthermore, this field does not seem to be characterised by strongly selective publication. The trials that were unpublished or published only in the grey literature tended to have positive results and were either unpublished for reasons unrelated to study results ([Arezina 2011](#); [Thomas 2003](#)), or because they were too new ([Thompson 2012a](#)).

### Agreements and disagreements with other studies or reviews

This review's findings about music therapy's effectiveness for children with ASD fit well into the context of previous evidence in this area ([Gold 2006](#); [Wheeler 2008](#); [Whipple 2004](#); [Whipple 2012](#)), but add considerably to the external validity of previous results by including trials that employed settings and methods utilised in clinical practice. Additionally, the robustness of findings is increased by following rigorous methodology and covering a larger total sample size than previous reviews.

## AUTHORS' CONCLUSIONS

### Implications for practice

The findings of this review provide evidence that music therapy may have positive effects on social interaction and communication skills of children with ASD. Music therapy has been shown to be superior to standard care and to similar forms of therapy where music was not used, which may be indicative of a specificity of the effect of music within music therapy. In addition, the results of this review suggest that therapy approaches that focus on the relational qualities of music within interactions and on the client's own interests and motivations ([Gattino 2011](#); [Kim 2008](#); [Thompson 2012a](#)), may be effective in increasing basic skills of social communication, such as keeping eye contact or initiating interaction. However, these findings need to be corroborated by future research involving larger samples. Children and adults with ASD frequently pose considerable behavioural challenges to their parents and other family members ([Oono 2013](#)). Therefore, the increases in social adaptation skills of children and in the quality of parent-child relationships through music therapy as found in this review may be highly relevant findings for families affected by ASD. As only short- to medium-term effects have been examined, it remains unknown how enduring the effects of music therapy on social interaction, communication, and related skills are in the long term.

When applying the results of this review to practice, it is important to note that the application of music therapy requires academic and clinical training in music therapy. Trained music therapists and academic training courses are available in many countries, and information is usually accessible through professional associations. Training courses in music therapy teach not only the clinical music therapy techniques as described in the background of this review, but also aim at developing the therapist's personality and clinical sensitivity, which is necessary to apply music therapy responsibly.

### Implications for research

Future research on music therapy for people with ASD will need to pay close attention to sample size and power. Sample sizes in all included studies were small, and test power was only discussed in three studies ([Gattino 2011](#); [Kim 2008](#); [Thompson 2012a](#)). Limited sample size remains a common problem in research on interventions for ASD. As there is a lack of studies for older individuals with ASD, research is needed examining effects of music therapy for adolescents and adults with ASD. Furthermore, we recommend that future trials on music therapy in this area should be: (1) pragmatic; (2) parallel; (3) conscious of types of music therapy; (4) conscious of relevant outcome measures; and (5) include long-term follow-up assessments.

(1) Pragmatic trials of effectiveness: The earliest trials on music therapy for ASD were efficacy trials, characterised by "inflexible

experimental intervention, with strict instructions for every element”; “restricted flexibility of the comparison intervention ... [e.g.] placebo”; and a primary outcome that was “a direct and immediate consequence of the intervention ... [e.g.] a surrogate marker of another downstream outcome of interest” (Thorpe 2009, Table 1). More recent trials (Thompson 2012a; Gattino 2011) have started to use more flexible interventions, standard care comparisons, and downstream outcomes. More pragmatic trials are needed to address the question of effectiveness (i.e. whether music therapy works ‘under usual conditions’, Thorpe 2009).

(2) Parallel trials: Many of the trials to date used cross-over designs. These designs are appropriate for early trials because they have the compelling advantage of higher test power even with small sample sizes. However, this advantage is bought at the expense of additional uncertainty (Elbourne 2002). Cross-over trials are only adequate for chronic conditions (this criterion is met in ASD) and for interventions with only short-acting effects. The duration of effect is presently unknown for music therapy, where learning effects may be lasting. Parallel design trials avoid these problems but require far greater resources. The present findings appear to justify such large-scale trials in the future.

(3) Types of music therapy: As discussed in this review, various types of music therapy have been proposed. Future trials should continue to be conscious of the quality, clinical applicability and link to usual practice, and type of music therapy examined. Future trials might entail comparisons between types of music therapy, but should also continue to investigate music therapy compared to other interventions or standard care.

(4) Relevant outcome measures: There is currently no consensus about the most pertinent outcome measures to be used in ASD intervention research (Warren 2011; Wheeler 2008). However, in line with recommendation (1) above, future trials should include outcomes that address the core problems of ASD in a generalised

setting utilising standardised scales.

(5) Long-term follow-up assessments: The most notable gap in this review was a lack of trials with longer follow-up periods. Future trials should consider long-term follow-up assessments of a year or more.

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\* Indicates the major publication for the study

## CHARACTERISTICS OF STUDIES

### Characteristics of included studies [ordered by study ID]

#### Arezina 2011

Methods	Allocation: session order randomised using Latin Square Blindness: unclear; random sub-sample (33.33% of sessions) assessed by independent observer Duration: 5 weeks Design: cross-over	
Participants	Diagnosis: autism spectrum disorder N = 6 Age: range 36 to 64 months Sex: 5 males, 1 female Setting: child development program	
Interventions	1. Interactive MT (musical instrument play, songs, music books, sung and verbal responses to verbalisations), 6 ten-minute sessions, n = 6 2. Non-music interactive play (non-music toys and books, verbal responses to verbalisations), 6 ten-minute sessions, n = 6 3. Independent play, 6 ten-minute sessions, n = 6	
Outcomes	Behaviour observation of videotaped sessions: a) Interaction or engaging in joint attention (percent of 15-second intervals engaged in interaction) b) Requesting or initiating joint attention (number of requests during a given time period)	
Notes		
<i>Risk of bias</i>		
Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Order of sessions (including different therapeutic approaches) was randomised for each child using a Latin Square
Allocation concealment (selection bias)	Unclear risk	No details given
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown

**Arezina 2011** (Continued)

Blinding of outcome assessment (detection bias) All outcomes	Unclear risk	No details about blinding reported; however, a random subsample (33.33%) was assessed by an independent observer (inter-observer agreement ranged from 85.7% to 98.9%)
Incomplete outcome data (attrition bias) All outcomes	Low risk	No drop-outs No missing data reported
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Low risk	Adequate music therapy method: yes Adequate music therapy training: yes

**Brownell 2002**

Methods	Allocation: quasi-randomised, possibly randomised ('counterbalanced') Blindness: independent assessor (teacher), blinding not reported Duration: 4 weeks Design: cross-over
Participants	Diagnosis: autism N = 4 Age: range 6 to 9 years Sex: 4 males, 0 females Setting: elementary school
Interventions	1. Structured receptive MT (songs with social stories), 5 individual daily sessions, n = 4 2. Structured receptive 'story therapy' (reading of social stories), 5 individual daily sessions, n = 4 3. No intervention, 2 x 5 days, n = 4
Outcomes	Repetitive behaviours outside therapy sessions (in classroom) Inter-rater reliability 0.86 to 0.94
Notes	

***Risk of bias***

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	High risk	Assignment to a counterbalanced treatment order (either ABAC or ACAB)
Allocation concealment (selection bias)	Unclear risk	No details given

**Brownell 2002** (Continued)

Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown
Blinding of outcome assessment (detection bias) All outcomes	Unclear risk	Outcomes were assessed by a teacher or instructional associate assigned to the participant No details given on blinding of assessors
Incomplete outcome data (attrition bias) All outcomes	Low risk	No drop-outs No missing data reported
Selective reporting (reporting bias)	Low risk	All outcomes (targeted behaviours) of interest were considered in the analysis
Other bias	Low risk	Adequate music therapy method: yes Adequate music therapy training: yes

**Buday 1995**

Methods	Allocation: randomised Blindness: assessor blinded to the nature of the hypothesis and to treatment condition Duration: 2 weeks Design: cross-over	
Participants	Diagnosis: autism N = 10 Age: range 4 to 9 years Sex: 8 males, 2 females Setting: public school	
Interventions	1. Structured receptive MT (songs used to teach signs), 5 individual sessions, n = 10 2. 'Rhythm therapy' (rhythmic speech used to teach signs), 5 individual sessions, n = 10	
Outcomes	Imitating behaviour in sessions (rating of a video recording with sound turned off to ensure blinding of raters; inter-rater agreement 98%): a) Sign imitation b) Speech imitation	
Notes		
<i>Risk of bias</i>		
Bias	Authors' judgement	Support for judgement

**Buday 1995** (Continued)

Random sequence generation (selection bias)	Unclear risk	Randomised, no further details given Additionally, counterbalancing of target signs for each treatment condition
Allocation concealment (selection bias)	Unclear risk	No details given
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Assessments were conducted by a person blinded to the nature of the hypothesis and to treatment condition
Incomplete outcome data (attrition bias) All outcomes	Low risk	No drop-outs No missing data reported
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Unclear risk	Adequate music therapy method: unclear Adequate music therapy training: unclear

**Farmer 2003**

Methods	Allocation: randomised Blindness: not known Duration: 5 days Design: parallel group
Participants	Diagnosis: autism N = 10 Age: range 2 to 5 years Sex: 9 males, 1 female Setting: homes and therapy centres
Interventions	1. Music therapy sessions (combined active and receptive: guitar playing, songs), n = 5 2. Placebo (no music) sessions, n = 5 Mostly individual sessions of 20 minutes
Outcomes	Responses within sessions (inter-rater agreement 91%): a) Verbal responses b) Gestural responses
Notes	

<i>Risk of bias</i>		
Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Randomised, no further details given
Allocation concealment (selection bias)	Unclear risk	No details given
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown
Blinding of outcome assessment (detection bias) All outcomes	Unclear risk	Unclear if assessors were masked to the randomisation result
Incomplete outcome data (attrition bias) All outcomes	Low risk	No drop-outs No missing data reported
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Low risk	Adequate music therapy method: yes Adequate music therapy training: yes

**Gattino 2011**

Methods	Allocation: balanced randomisation using a table of random numbers Blindness: assessors blinded Duration: 7 months Design: parallel group
Participants	Diagnosis: autism spectrum disorder N = 24 Age: range 7 to 12 years (mean 9.75 years) Sex: 24 males, 0 females Setting: hospital
Interventions	1. Relational music therapy (improvisation not using a structured protocol; 3 assessment sessions, 16 intervention sessions, 1 final assessment session) in addition to standard treatment, 20 thirty-minute sessions, scheduled weekly, n = 12 2. Standard treatment (clinical routine activities including medical examinations and consultations), n = 12

**Gattino 2011** (Continued)

Outcomes	a) Verbal communication (Childhood Autism Rating Scale, Brazilian version, CARS-BR) b) Nonverbal communication (CARS-BR) c) Social communication (CARS-BR)	
Notes	Funding sources: Fund of Incentive to Research of Porto Alegre Clinical Hospital (project no. 08006), Brazilian Research Council (CNPq)	
<i><b>Risk of bias</b></i>		
<b>Bias</b>	<b>Authors' judgement</b>	<b>Support for judgement</b>
Random sequence generation (selection bias)	Low risk	Randomised (computer-generated random sequence)
Allocation concealment (selection bias)	Low risk	Allocation was conducted by an external investigator
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Assessors were blinded to the randomisation result
Incomplete outcome data (attrition bias) All outcomes	Low risk	No drop-outs No missing data reported
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Low risk	Adequate music therapy method: yes Adequate music therapy training: yes

**Kim 2008**

Methods	Allocation: randomised Blindness: assessors were blinded to the treatment condition, except for parent-based measures conducted by mothers Duration: 8 months Design: cross-over
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Participants	Diagnosis: autism N = 15 at entry; N = 10 for analysis Age: range 39 to 71 months (mean 51 months) Sex: 13 males, 2 females at entry; 10 males, 0 females for analysis Setting: private practice clinic	
Interventions	1. Improvisational music therapy, 12 thirty-minute sessions, scheduled weekly, n = 10 2. Play sessions with toys, 12 thirty-minute sessions, scheduled weekly, n = 10	
Outcomes	<i>Social interaction:</i> - social approach subscale (Pervasive Developmental Disorder Behavior Inventory, PDDBI); completed by parents (not blind) and independent observers (blinded) - turn-taking duration <i>Non-verbal communicative skills:</i> - Early Social Communication Scale, ESCS, abridged version - eye contact frequency and duration <i>Initiating behaviour:</i> - initiation of engagement frequency <i>Social-emotional reciprocity:</i> - emotional synchronicity frequency and duration - musical synchronicity frequency and duration <i>Social adaptation:</i> - compliant response frequency - non-compliant response frequency - no response frequency <i>Joy:</i> - joy frequency and duration	
Notes	Funding source: Aalborg University, Denmark	
<i>Risk of bias</i>		
Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Randomised (picking the randomisation result from an opaque box)
Allocation concealment (selection bias)	Unclear risk	No details given
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown

**Kim 2008** (Continued)

Blinding of outcome assessment (detection bias) All outcomes	Unclear risk	Assessors were blinded to the randomisation result, except for non-generalised measures, ESCS, and MPIP, where a random subsample (30%) was additionally assessed by independent observers (inter-rater reliability ranging from 0.70 to 0.98)
Incomplete outcome data (attrition bias) All outcomes	High risk	High drop-out rate (5 of 15 participants dropped out) Data from drop-outs were excluded
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Low risk	Adequate music therapy method: yes Adequate music therapy training: yes

**Lim 2010**

Methods	Allocation: randomised Blindness: assessors were blind to the purpose of the study Duration: 5 days Design: parallel group
Participants	Diagnosis: autism spectrum disorder N = 50 Age: range 3 to 5 years (mean 4.8 years) Sex: 44 males, 6 females. Setting: recruiting site (schools, therapy centres, etc.)
Interventions	1. Music training ('Developmental Speech and Language Training through Music'; videotaped songs with target words), 6 individual sessions within 3 days, n = 18 2. Speech training (videotaped spoken stories with target words), 6 individual sessions within 3 days, n = 18 3. No training, n = 14
Outcomes	Behaviour observation of videotaped post-test sessions: verbal response. Inter-rater reliability 0.999
Notes	

***Risk of bias***

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Randomised, no further details given

**Lim 2010** (Continued)

Allocation concealment (selection bias)	Unclear risk	No details given
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Assessors were blind to the purpose of the study
Incomplete outcome data (attrition bias) All outcomes	Low risk	No drop-outs No missing data reported
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Unclear risk	Adequate music therapy method: unclear Adequate music therapy training: unclear

**Lim 2011**

Methods	Allocation: training order randomised Blindness: assessors were blind to the purpose of the study Duration: 2 weeks Design: cross-over	
Participants	Diagnosis: autism spectrum disorder, N = 22 Age: range 3 to 5 years (mean 4.3 years) Sex: 17 males, 5 females Setting: no details given	
Interventions	1. Music training ('music incorporated Applied Behavior Analysis Verbal Behavior'; sung instructions, songs with target words), 6 individual sessions within 2 weeks, n = 22 2. Speech training (Applied Behavior Analysis Verbal Behavior; spoken instructions, sentences with target words), 6 individual sessions within 2 weeks, n = 22 3. No training, n = 22	
Outcomes	Behaviour observation of videotaped post-test sessions: verbal production	
Notes		
<i>Risk of bias</i>		
Bias	Authors' judgement	Support for judgement

**Lim 2011** (Continued)

Random sequence generation (selection bias)	Low risk	Order of sessions (including different therapeutic approaches) was randomised for each child using a random number chart
Allocation concealment (selection bias)	Unclear risk	No details given
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown
Blinding of outcome assessment (detection bias) All outcomes	Low risk	Assessors were blind to the purpose of the study
Incomplete outcome data (attrition bias) All outcomes	Low risk	No drop-outs No missing data reported
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Low risk	Adequate music therapy method: yes Adequate music therapy training: yes

**Thomas 2003**

Methods	Allocation: randomised order of treatment Blindness: no blinding Duration: 12 weeks Design: cross-over (within each session)
Participants	Diagnosis: autism, N = 6 Age: range 2 to 3 years Sex: 5 males, 1 female Setting: not known
Interventions	1. Music therapy (using songs, instruments, vocal sounds and movement to interact with the child and musically or verbally respond to the child's verbal or non-verbal behaviour), twelve 15-minute session parts, immediately following or preceding playtime session parts, n = 6 2. Playtime (attempts to interact with the child using toys and verbally responding to the child's non-verbal or verbal behaviour), twelve 15-minute session parts, immediately following or preceding music therapy session parts, n = 6

**Thomas 2003** (Continued)

Outcomes	Behaviour observation of videotaped sessions a) On-task behaviour (percentage of session time) b) Requesting behaviour (percentage of session time)	
Notes	Funding source: Mid-Atlantic Region of the American Music Therapy Association	
<b><i>Risk of bias</i></b>		
<b>Bias</b>	<b>Authors' judgement</b>	<b>Support for judgement</b>
Random sequence generation (selection bias)	Unclear risk	Randomised, no further details given
Allocation concealment (selection bias)	Unclear risk	No details given
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention was unknown
Blinding of outcome assessment (detection bias) All outcomes	Unclear risk	No details given if the assessor was blinded to the randomisation result
Incomplete outcome data (attrition bias) All outcomes	Low risk	No drop-outs No missing data reported
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Low risk	Adequate music therapy method: yes Adequate music therapy training: yes

**Thompson 2012a**

Methods	Allocation: randomised Blindness: no blinding Duration: 16 weeks Design: parallel group
Participants	Diagnosis: ASD N = 23 Age: range 3 to 6 years Sex: 19 males, 4 females Setting: participants' homes

Interventions	1. Home-based, family-centred music therapy (using songs, improvisation, structured music interactions), in addition to standard care, 16 sessions, scheduled weekly, n = 12 2. Standard care, n = 11	
Outcomes	a) Vineland Social Emotional Early Childhood Scales (Vineland SEEC) b) Social Responsiveness Scale Preschool Version (SRS-PS), rated by parents c) MacArthur-Bates Communicative Development Inventories - Words and Gestures (MBCDI-W&G), rated by parents d) Parent-Child Relationship Inventory (PCRI), rated by parents e) Music Therapy Diagnostic Assessment (MTDA): not used since rated for music therapy group only	
Notes	Funding source: Department of Education and Early Childhood Development, Victoria	
<i>Risk of bias</i>		
Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Low risk	Randomised (computer-generated random sequence)
Allocation concealment (selection bias)	Low risk	An independent statistician prepared opaque, numbered allocation envelopes
Blinding of participants and personnel (performance bias) All outcomes	Unclear risk	The fact that children with ASD participating in the study were not blinded was considered unlikely to introduce bias The possible risk of bias introduced by therapists administering the intervention is unknown
Blinding of outcome assessment (detection bias) All outcomes	Unclear risk	Parent-report based measures were used However, measures contain internal safeguards to address bias as evidenced by high correlations with non-parent rated measures or high test-retest correlations
Incomplete outcome data (attrition bias) All outcomes	Low risk	Low drop-out rate Intention-to-treat analysis
Selective reporting (reporting bias)	Low risk	All outcome measures of interest were considered in the analysis
Other bias	Low risk	Adequate music therapy method: yes Adequate music therapy training: yes

MT - music therapy; ABAC, ACAB - type of trial where interventions A, B, and C are given in this order

## Characteristics of excluded studies *[ordered by study ID]*

Study	Reason for exclusion
Applebaum 1979	Not intervention study (assessment)
Bettison 1996	Not MT (AIT - only music listening)
Blackstock 1978	Not intervention study (assessment)
Bonnel 2003	Not intervention study (assessment)
Boso 2007	Not RCT or CCT (case series)
Brown 1994	Not RCT or CCT (case series)
Brown 2003	Not intervention study (assessment)
Bruscia 1982	Not RCT or CCT (case study)
Carroll 1983	Not MT (only sung instructions)
Chilcote-Doner 1982	Not MT (rhythmic strobe and drumbeat)
Clauss 1994	Not RCT or CCT (case series, ABACA design)
Cooley 2012	Not MT (speech and language training with music)
Dawson 1998	Not intervention study (assessment)
Diez Cuervo 1989	Not intervention study (assessment)
Edelson 1999	Not MT (AIT - only music listening)
Edgerton 1994	Not RCT or CCT (case series)
Finnigan 2010	Not RCT or CCT (case study)
Frissell 2001	Not intervention study (assessment)
Goldstein 1964	Not RCT or CCT (case study)
Gore 2002	No usable data (unclear outcome measure)
Griggs 1997	Not RCT or CCT (case study) Not intervention study (assessment)

(Continued)

Hadsell 1988	Not RCT or CCT (case series) Not ASD (Rett syndrome)
Hairston 1990	Not RCT or CCT (case series)
Heaton 1999	Not intervention study (assessment)
Heaton 2003	Not intervention study (assessment)
Hillier 2012	Not RCT or CCT (case series)
Kern 2006	Not RCT or CCT (case series)
Kern 2007	Not RCT or CCT (case study)
Kolko 1980	Not intervention study (assessment)
Krauss 1982	Not RCT or CCT (case series) Not ASD (apraxia, language delay)
Laird 1997	Not RCT or CCT (uncontrolled design)
Lee 2004	Not RCT or CCT (case series)
Li 2011	Not possible to isolate MT from other interventions
Lim 2007	Not MT (speech training with music)
Litchman 1976	Not MT (listening to recorded nursery rhymes)
Lundqvist 2009	Not MT (presentation of preset vibroacoustic stimuli)
Ma 2001	Not RCT or CCT (case series)
Mahlberg 1973	Not RCT or CCT (case study)
Miller 1979	Not RCT or CCT (case study)
Mottron 2000	Not intervention study (assessment)
Mudford 2000	Not MT (AIT/only music listening)
O'Connell 1974	Not RCT or CCT (case study)
O'Dell 1998	Not MT (music listening)



(Continued)

O'Loughlin 2000	Not RCT or CCT - includes three case series where all received the same treatment (no. 1, 3, 4) and one case series with an ABA design (no. 2)
Pasiali 2004	Not RCT or CCT (case series, ABAB design)
Rao 2001	Not MT (headphones with versus without music)
Sandiford 2013	Not MT (speech training with music)
Saperston 1973	Not RCT or CCT (case study)
Schmidt 1976	Not RCT or CCT (case series, AB design)
Starr 1998	Not RCT or CCT (case series)
Staum 1984	Not RCT or CCT (case study)
Stephens 2008	Not RCT or CCT (case series)
Stevens 1969	Not RCT or CCT (case series)
Thaut 1987	Not intervention study (assessment)
Thaut 1988	Not intervention study (assessment)
Toolan 1994	Not RCT or CCT (case series)
Watson 1979	Not RCT or CCT (case series, ABCA design)
Wimpory 1995	Not RCT or CCT (case study)
Wood 1991	Not MT (music listening)

MT - music therapy; AIT - auditory integration training; RCT - randomised controlled trial; CCT - controlled clinical trial; ASD - autism spectrum disorder; ABA, ABAB, AB - type of trial where interventions A and B are given in this order; ABCA, ABACA - type of trial where interventions A, B, and C are given in this order

## Characteristics of ongoing studies *[ordered by study ID]*

ISRCTN78923965

Trial name or title	Randomised controlled trial of improvisational music therapy's effectiveness for children with autism spectrum disorders (TIME-A)
Methods	Allocation: randomised Blindness: assessors of primary outcome blinded Duration: 12 months Design: parallel group
Participants	Diagnosis: autism spectrum disorder N = 300 Age: range 4 years to 6 years, 11 months
Interventions	1. Individual improvisational music therapy over a period of five months, 3 sessions per week (high-intensity) , plus standard care (see below) 2. Individual improvisational music therapy over a period of five months, 1 session per week (low-intensity), plus standard care (see below) 3. Standard care: 3 sessions of parent counselling at 0, 2, and 5 months
Outcomes	a) Autism Diagnostic Observation Schedule (ADOS) b) Social Responsiveness Scale (SRS) c) Cost-effectiveness
Starting date	01/08/2011
Contact information	christian.gold@uni.no
Notes	<a href="http://www.controlled-trials.com/ISRCTN78923965">http://www.controlled-trials.com/ISRCTN78923965</a>

## DATA AND ANALYSES

### Comparison 1. Music therapy vs. 'placebo' therapy or standard care

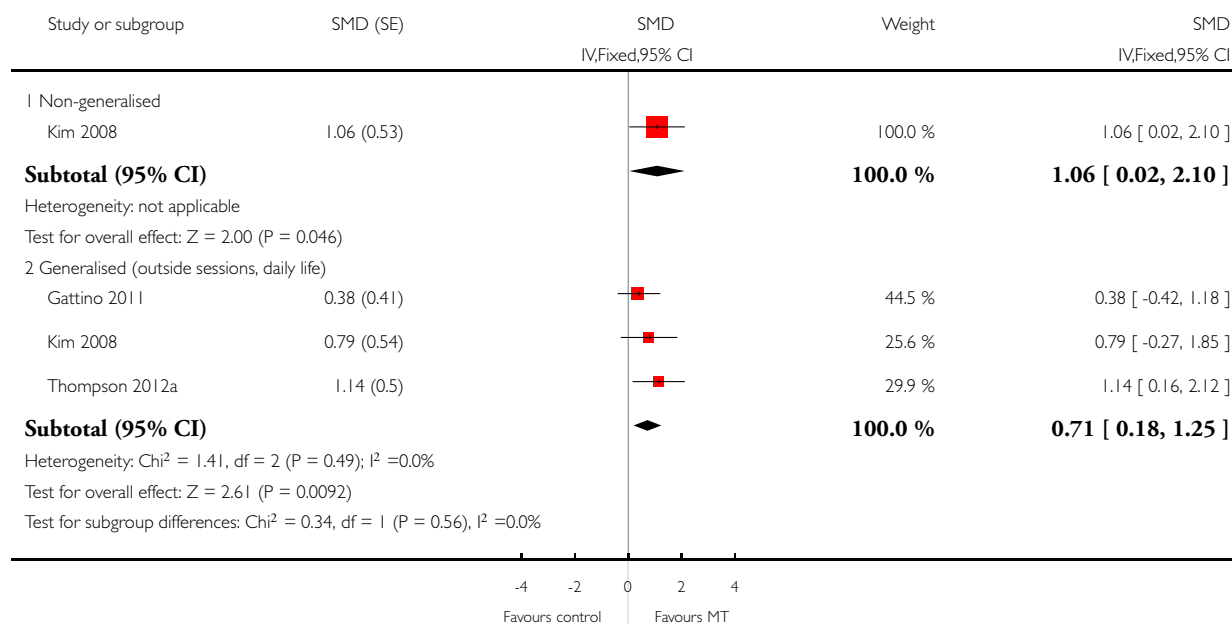
Outcome or subgroup title	No. of studies	No. of participants	Statistical method	Effect size
1 Social interaction	3		SMD (Fixed, 95% CI)	Subtotals only
1.1 Non-generalised	1		SMD (Fixed, 95% CI)	1.06 [0.02, 2.10]
1.2 Generalised (outside sessions, daily life)	3		SMD (Fixed, 95% CI)	0.71 [0.18, 1.25]
2 Communicative skills: non-verbal	5		SMD (Fixed, 95% CI)	Subtotals only
2.1 Non-generalised	3		SMD (Fixed, 95% CI)	0.57 [0.29, 0.85]
2.2 Generalised (outside sessions, daily life)	3		SMD (Fixed, 95% CI)	0.48 [-0.02, 0.98]
3 Communicative skills: verbal	6		SMD (Fixed, 95% CI)	0.33 [0.16, 0.49]
3.1 Non-generalised	4		SMD (Fixed, 95% CI)	0.33 [0.16, 0.50]
3.2 Generalised (outside sessions, daily life)	2		SMD (Fixed, 95% CI)	0.30 [-0.28, 0.89]
4 Initiating behaviour	3		SMD (Fixed, 95% CI)	Subtotals only
4.1 Non-generalised	3		SMD (Fixed, 95% CI)	0.73 [0.36, 1.11]
5 Social-emotional reciprocity	1		SMD (Fixed, 95% CI)	Subtotals only
5.1 Non-generalised	1		SMD (Fixed, 95% CI)	2.28 [0.73, 3.83]
6 Social adaptation	4		SMD (Fixed, 95% CI)	0.41 [0.21, 0.60]
6.1 Non-generalised	3		SMD (Fixed, 95% CI)	1.15 [0.69, 1.61]
6.2 Generalised (outside sessions, daily life)	1		SMD (Fixed, 95% CI)	0.24 [0.02, 0.46]
7 Joy	1		SMD (Fixed, 95% CI)	0.96 [0.04, 1.88]
8 Quality of parent-child relationship	2		SMD (Fixed, 95% CI)	0.82 [0.13, 1.52]

# **Analysis 1.1. Comparison 1 Music therapy vs. 'placebo' therapy or standard care, Outcome 1 Social interaction.**

Review: Music therapy for people with autism spectrum disorder

Comparison: 1 Music therapy vs. 'placebo' therapy or standard care

Outcome: 1 Social interaction

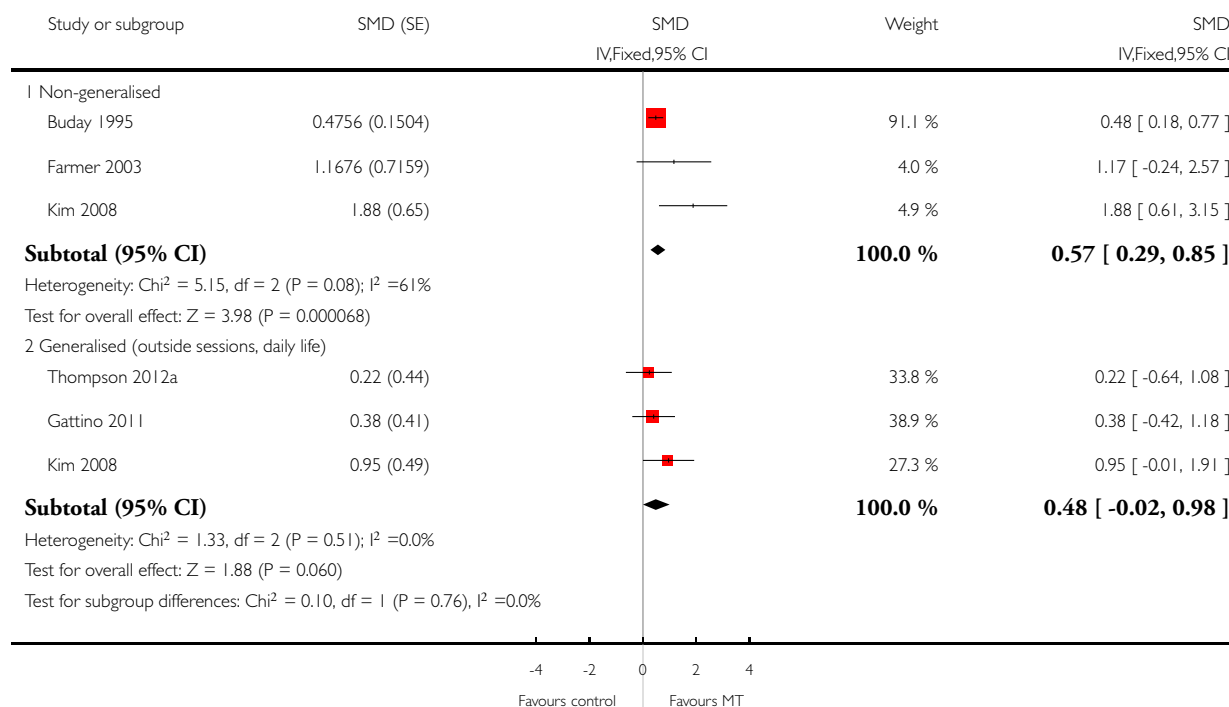


## Analysis 1.2. Comparison 1 Music therapy vs. 'placebo' therapy or standard care, Outcome 2 Communicative skills: non-verbal.

Review: Music therapy for people with autism spectrum disorder

Comparison: 1 Music therapy vs. 'placebo' therapy or standard care

Outcome: 2 Communicative skills: non-verbal

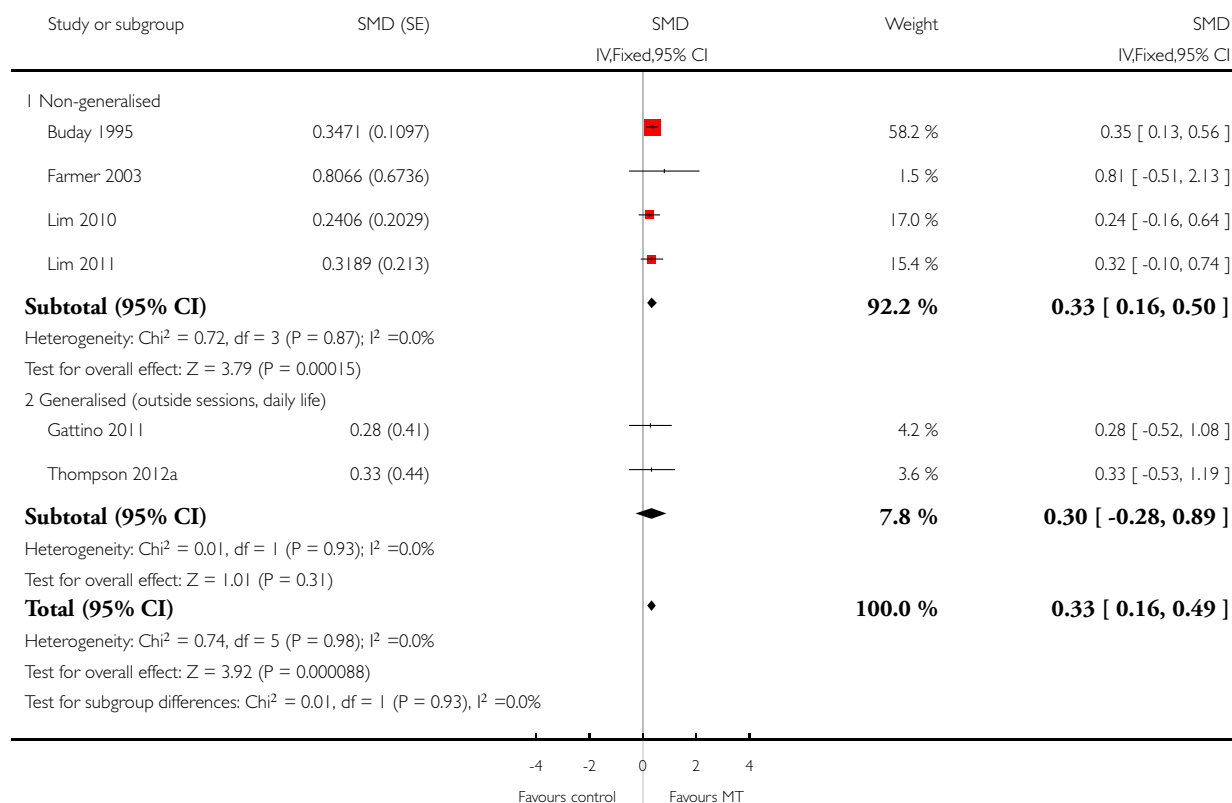


### Analysis 1.3. Comparison 1 Music therapy vs. 'placebo' therapy or standard care, Outcome 3 Communicative skills: verbal.

Review: Music therapy for people with autism spectrum disorder

Comparison: 1 Music therapy vs. 'placebo' therapy or standard care

Outcome: 3 Communicative skills: verbal

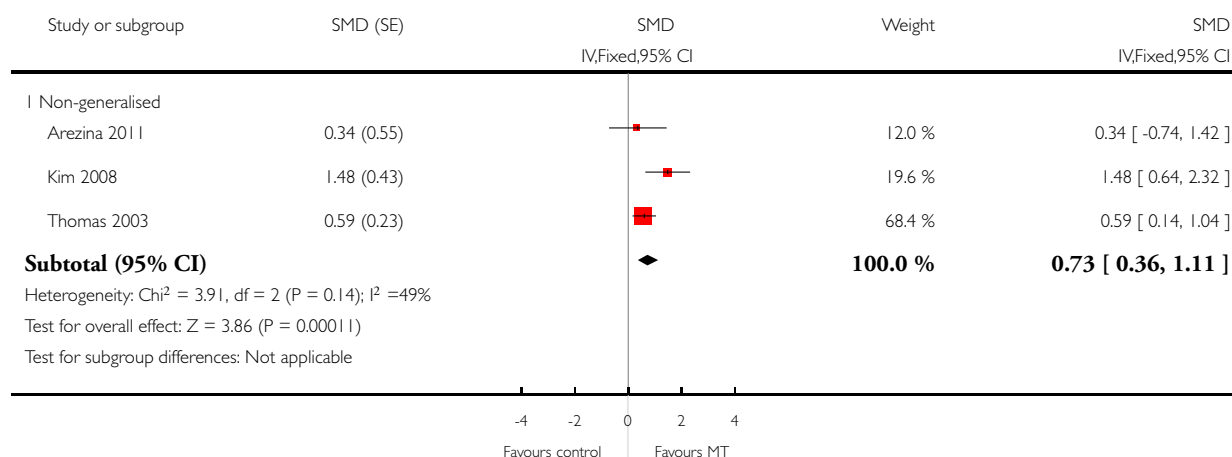


### Analysis 1.4. Comparison 1 Music therapy vs. 'placebo' therapy or standard care, Outcome 4 Initiating behaviour.

Review: Music therapy for people with autism spectrum disorder

Comparison: 1 Music therapy vs. 'placebo' therapy or standard care

Outcome: 4 Initiating behaviour

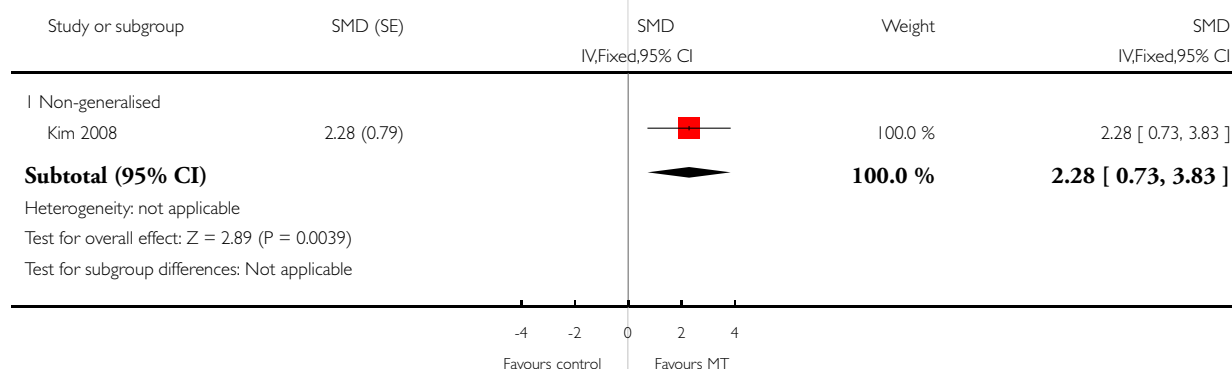


### Analysis 1.5. Comparison 1 Music therapy vs. 'placebo' therapy or standard care, Outcome 5 Social-emotional reciprocity.

Review: Music therapy for people with autism spectrum disorder

Comparison: 1 Music therapy vs. 'placebo' therapy or standard care

Outcome: 5 Social-emotional reciprocity

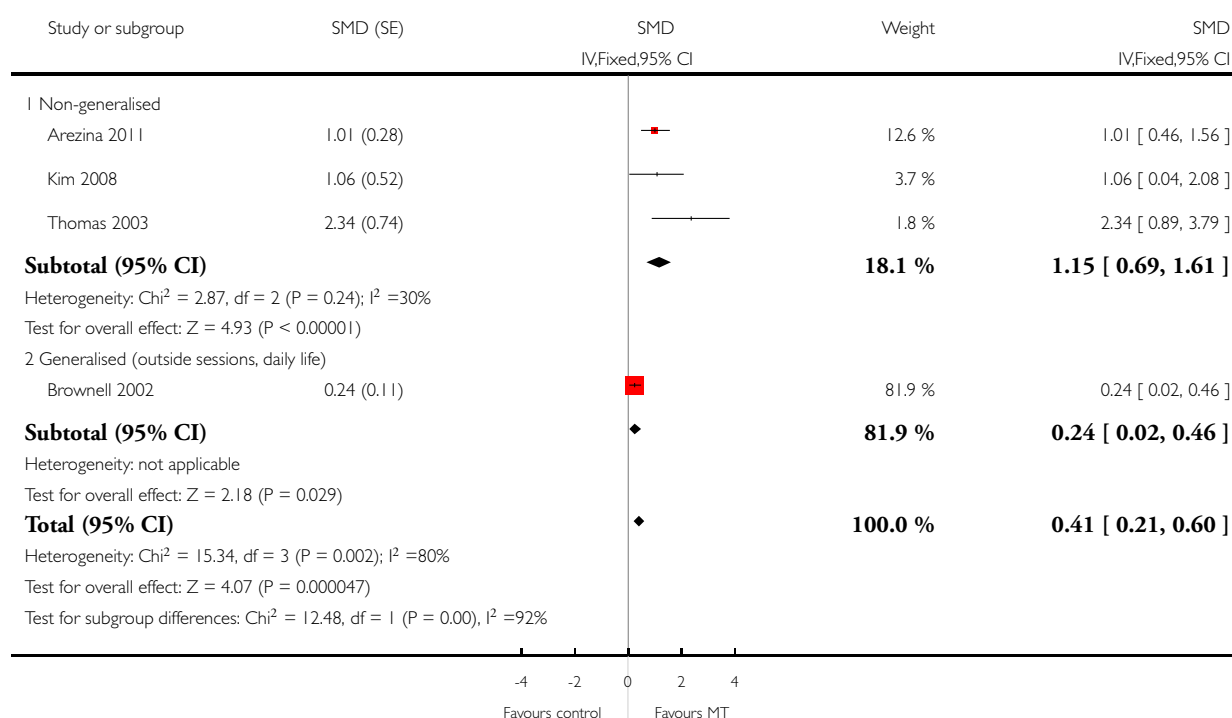


## Analysis 1.6. Comparison 1 Music therapy vs. 'placebo' therapy or standard care, Outcome 6 Social adaptation.

Review: Music therapy for people with autism spectrum disorder

Comparison: 1 Music therapy vs. 'placebo' therapy or standard care

Outcome: 6 Social adaptation



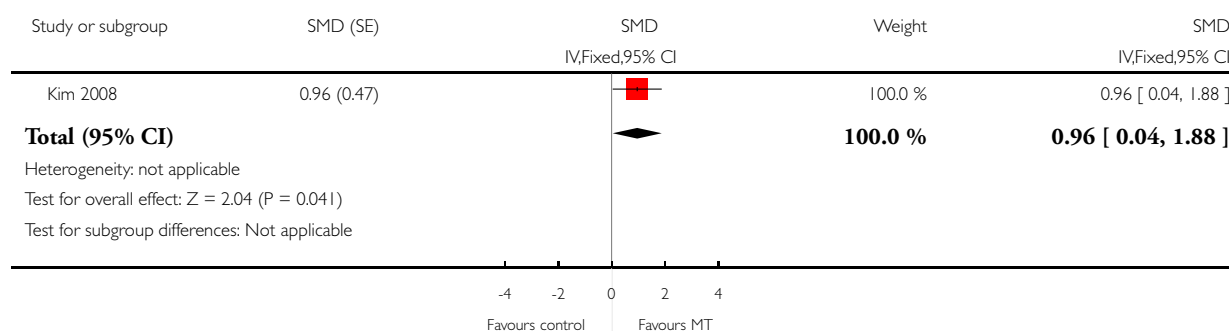


### Analysis 1.7. Comparison 1 Music therapy vs. 'placebo' therapy or standard care, Outcome 7 Joy.

Review: Music therapy for people with autism spectrum disorder

Comparison: 1 Music therapy vs. 'placebo' therapy or standard care

Outcome: 7 Joy

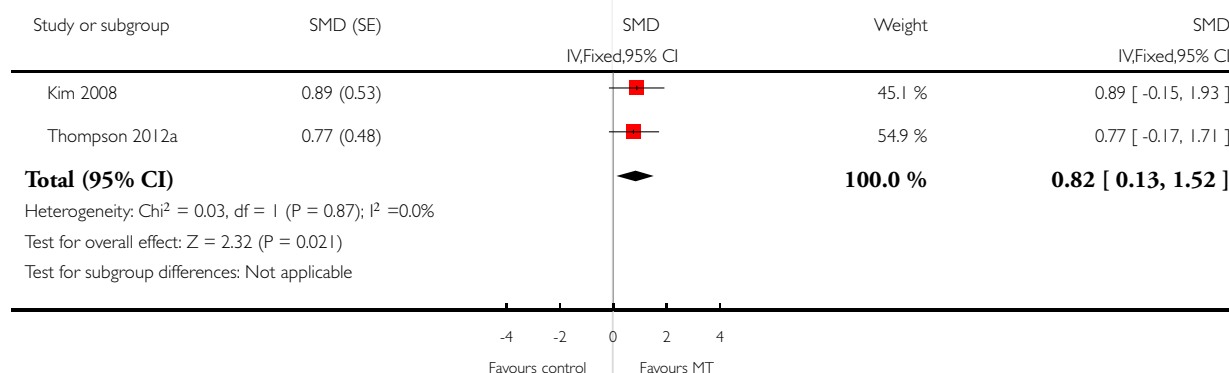


### Analysis 1.8. Comparison 1 Music therapy vs. 'placebo' therapy or standard care, Outcome 8 Quality of parent-child relationship.

Review: Music therapy for people with autism spectrum disorder

Comparison: 1 Music therapy vs. 'placebo' therapy or standard care

Outcome: 8 Quality of parent-child relationship



## APPENDICES

### Appendix I. Search strategies 2004-2013

For this update, the following search terms were added to the original strategy (reported in [Appendix 2](#)) to increase the sensitivity of the search:

(sing or singing or song\* or choral\* or choir\*)

(percussion\* or rhythm\* or tempo\*)

improvis\*

melod\*

Nordoff-Robbins

Bonny

(auditory or acoustic or sound\*) adj5 (stimulat\* or cue\*)

#### CENTRAL

2011 Issue 3 Limited by year 2004 to 2011. Searched 7 September 2011 plus new terms for all years pre-2004 [61 records]

2013 Issue 6 Limited by year 2011 to 2013. Searched 29 July 2013 [8 records]

#1MeSH descriptor: [Music] this term only

#2MeSH descriptor: [Music Therapy] this term only

#3music\*

#4((guided next imagery) near music)

#5GIM

#6vibroacoustic

#7vibro-acoustic

#8(sing or singing or song\* or choral\* or choir\*)

#9(percussion\* or rhythm\* or tempo\* or melod\*)

#10improvis\*

#11(Nordoff-Robbins\* or bonny\*)

#12((auditory or acoustic or sound\*) near/5 (stimulat\* or cue\*))

#13(#1 or #2 or #3 or #4 or #5 or #6 or #7 or #8 or #9 or #10 or #11 or #12)

#14MeSH descriptor: [Child Development Disorders, Pervasive] 1 tree(s) exploded

#15asperg\* or autis\* or kanner\* or (childhood next schizophren\*)

#16(speech near disorder\*)

#17(language near delay\*)

#18ASD or ASDs or PDD or PDDs

#19(#14 or #15 or #16 or #17 or #18)

#20(#13 and #19) in Trials

#### Ovid MEDLINE

Ovid MEDLINE 1948 to August Week 4 2011. Searched 6 September 2011. Limited by year 2004 to 2011 plus new terms for all years pre-2004 [93 records]

Ovid MEDLINE 1946 to July Week 3 2013. Searched 29 July 2013 Limited to ed=20110831 to 20130729 [24 records]

1 music therapy/

2 music\$.tw.

3 (guided imagery adj3 music).tw.

4 gim.tw.

5 (vibro-acoustic\$ or vibroacoustic\$).tw.

6 music/

7 (sing or singing or song\$ or choral\$ or choir\$).tw.

8 (percussion\$ or rhythm\$).tw.

9 melod\$.tw.  
 10 improvis\$.tw.  
 11 (Nordoff-Robbin\$ or bonny\$).tw.  
 12 ((auditory or acoustic or sound\$) adj5 (stimulat\$ or cue\$)).tw.  
 13 or/1-12  
 14 exp child development disorders, pervasive/  
 15 pervasive development\$ disorder\$.tw.  
 16 (PDD or PDDs or ASD or ASDs).tw.  
 17 autis\$.tw.  
 18 asperg\$.tw.  
 19 kanner\$.tw.  
 20 childhood schizophreni\$.tw.  
 21 (speech adj3 disorder\$).tw.  
 22 (language adj3 delay\$).tw.  
 23 or/14-22  
 24 randomized controlled trial.pt.  
 25 controlled clinical trial.pt.  
 26 randomi#ed.ab.  
 27 placebo\$.ab.  
 28 drug therapy.fs.  
 29 randomly.ab.  
 30 trial.ab.  
 31 groups.ab.  
 32 or/24-31  
 33 exp animals/ not humans.sh.  
 34 32 not 33  
 35 13 and 23 and 34

#### **Embase ( OVID)**

Embase 1980 to 2011 Week 35. Searched 7 September 2011. Limited to year=2004 to 2011 plus new terms for all years pre-2004 [133 records]

Embase 1980 to 2013 Week 30. Searched 29 July 2013. Limited to year=2011 to 2013 [ 54 records]

1 exp music/  
 2 music therapy/  
 3 music\$.tw.  
 4 (guided imagery adj3 music).tw.  
 5 GIM.tw.  
 6 (guided imagery adj3 music).tw.  
 7 (vibro-acoustic therapy or vibroacoustic therapy).tw.  
 8 (sing or singing or song\$ or choral\$ or choir\$).tw.  
 9 (percussion\$ or rhythm\$).tw.  
 10 melod\$.tw.  
 11 improvis\$.tw.  
 12 (Nordoff-Robbin\$ or bonny\$).tw.  
 13 ((auditory or acoustic or sound\$) adj5 (stimulat\$ or cue\$)).tw.  
 14 or/1-13  
 15 exp autism/  
 16 pervasive development\$ disorder\$.tw.  
 17 (PDD or PDDs or ASD or ASDs).tw.  
 18 autis\$.tw.  
 19 asperg\$.tw.

20 kanner\$.tw.  
 21 childhood schizophre\$.tw.  
 22 (speech adj3 disorder\$).tw.  
 23 (language adj3 delay\$).tw.  
 24 or/15-23  
 25 exp Clinical trial/  
 26 Randomized controlled trial/  
 27 Randomization/  
 28 Single blind procedure/  
 29 Double blind procedure/  
 30 Crossover procedure/  
 31 Placebo/  
 32 Randomi#ed.tw.  
 33 RCT.tw.  
 34 (random\$ adj3 (allocat\$ or assign\$)).tw.  
 35 randomly.ab.  
 36 groups.ab.  
 37 trial.ab.  
 38 ((sing\$ or doubl\$ or trebl\$ or tripl\$) adj3 (blind\$ or mask\$)).tw.  
 39 Placebo\$.tw.  
 40 Prospective study/  
 41 (crossover or cross-over).tw.  
 42 prospective.tw.  
 43 or/25-42  
 44 14 and 24 and 43

## PsycINFO (OVID)

PsycINFO 1806 to August Week 5 2011. Searched 7 September 2011. Limited to 2004 to 2011 plus new terms for all years pre-2004 [33 records]

PsycINFO 1806 to July Week 3 2013. Searched 29 July 2013. Limited to 2011 to current [14 records]

1 exp music/  
 2 music therapy/  
 3 music\$.tw.  
 4 (guided imag\$ adj3 music\*).tw.  
 5 GIM.tw.  
 6 (vibroacoustic\$ or vibro-acoustic\$).tw.  
 7 rhythm/ or tempo/  
 8 (percussion\$ or rhythm\$ or tempo).tw.  
 9 singing/  
 10 (sing or singing or song\$ or choral\$ or choir\$).tw.  
 11 melod\$.tw.  
 12 improvis\$.tw.  
 13 (Bonny or Nordoff\$).tw.  
 14 ((auditory or acoustic or sound\$) adj5 (stimulat\$ or cue\$)).tw.  
 15 or/1-14  
 16 exp pervasive developmental disorders/  
 17 pervasive development\$ disorder\$.tw.  
 18 (PDD or PDDs or ASD or ASDs).tw.  
 19 autis\$.tw.  
 20 asperg\$.tw.  
 21 kanner\$.tw.

22 childhood schizopreni\$.tw.  
 23 (speech adj3 disorder\$).tw.  
 24 (language adj3 delay\$).tw.  
 25 or/16-24  
 26 Clinical Trials/  
 27 Random Sampling/  
 28 Placebo/  
 29 treatment effectiveness evaluation/ or mental health program evaluation/  
 30 evaluation/ or program evaluation/  
 31 educational program evaluation/  
 32 ((clinical or control\$) adj5 trial\$).tw.  
 33 placebo\$.tw.  
 34 randomi#ed.tw.  
 35 (random\$ adj3 (assign\$ or allocat\$)).tw.  
 36 (singl\$ adj3 (mask\$ or blind\$)).tw.  
 37 (doubl\$ adj3 (mask\$ or blind\$)).tw.  
 38 ((tripl\$ or trebl\$) adj3 (mask\$ or blind\$)).tw.  
 39 (crossover\$ or cross-over\$).tw.  
 40 ((evaluat\$ or effectiveness\$) adj3 (study or studies or research\$)).tw.  
 41 or/26-40  
 42 15 and 25 and 41

#### **CINAHL Plus (EBSCOhost)**

CINAHL 1937 to current. Searched 7 September 2011. Limited to year=2004 to 2011 plus new terms for all years pre-2004 [50 records]

CINAHL 1937 to current. Searched 29 July 2013. Limited to year=2011 to 2013 [25 records]

S42 S21 AND S41  
 S41 S22 OR S23 OR S24 OR S25 OR S26 OR S27 OR S28 OR S29 OR S30 OR S31  
 OR S32 OR S33 OR S34 OR S35 OR S36 OR S37 OR S38 OR S39 OR S40  
 S40 placebo\*  
 S39 (MH "Placebos")  
 S38 (MH "Evaluation Research") OR (MH "Summative Evaluation Research")  
 OR (MH "Program Evaluation")  
 S37 (MH "Treatment Outcomes")  
 S36 (MH "Comparative Studies")  
 S35 TI (compar\* stud\* or compar\* research\*) or AB (compar\* stud\* or  
 compar\* research\*) or TI (evaluat\* study or evaluat\* research) or AB  
 (evaluat\* study or evaluat\* research) or TI (effectiv\* study or  
 effectiv\* research) or AB (effectiv\* study or effectiv\* research) OR TI  
 (prospectiv\* study or prospectiv\* research) or AB(prospectiv\* study or  
 prospectiv\* research) or TI (follow-up study or follow-up research) or  
 AB (follow-up study or follow-up research)  
 S34 crossover\* or "cross over"  
 S33 (MH "Crossover Design")  
 S32 (trebl\* N3 mask\*) or (trebl\* N3 blind\*)  
 S31 (tripl\* N3 mask\*) or (tripl\* N3 blind\*)  
 S30 (doubl\* N3 mask\*) or (doubl\* N3 blind\*)  
 S29 (singl\* N3 mask\*) or (singl\* N3 blind\*)  
 S28 (clinic\* N3 trial\*) or (control\* N3 trial\*)  
 S27 (random\* N3 allocat\* ) or (random\* N3 assign\*)  
 S26 randomis\* or randomiz\*

S25 (MH "Meta Analysis")  
 S24 randomis\* or randomiz\*  
 S23 (MH "Clinical Trials+")  
 S22 MH random assignment  
 S21 S14 AND S20  
 S20 S15 OR S16 OR S17 OR S18 OR S19  
 S19 speech N3 disorder\* or language N3 delay\*  
 S18 (PDD or PDDs or PDD-NOS or ASD or ASDs)  
 S17 pervasive development\* disorder\*  
 S16 autis\* or asperger\* or childhood schizophre\* or kanner\*  
 S15 (MH "Child Development Disorders, Pervasive+")  
 S14 S1 OR S2 OR S3 OR S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10 OR S11 OR  
 S12 OR S13  
 S13 (auditory N3 cue\* or auditory N3 stimul\*) OR (acoustic N3 cue\* or  
 acoustic N3 stimul\*) or (sound N3 cue\* or sound N3 stimul\*)  
 S12 Nordoff\* or Bonny\*  
 S11 improvis\*  
 S10 percussion\* or rhythm\* or melod\* or tempo  
 S9 sing or singing or song\* or choral or choir\*  
 S8 (MH "Singing")  
 S7 vibro-acoustic\* or vibroacoustic\*  
 S6 GIM  
 S5 (guided imagery) N3 (music\*)  
 S4 (MH "Guided Imagery")  
 S3 music\*  
 S2 MH music therapy  
 S1 MH music

#### ERIC (Proquest)

ERIC 1966 to current. Limited to year=2004 to 2011. Searched 9 September 2011 Limited to 2004 to 2011 plus new terms for all years pre-2004 [67 records]

ERIC 1966 to current. Limited to year=2011 to 2013. Searched 30 July 2013 [31 records]

(SU.EXACT.EXPLODE("Music") OR SU.EXACT("Music Therapy") OR SU.EXACT.EXPLODE("Music Activities") OR (music\* OR guided imag\* OR GIM OR vibro-acoustic therapy\* OR vibroacoustic therapy\* OR

Bonny\* OR Nordoff\* OR singing OR song\* OR choral\* OR choir\* OR percussion\* OR rhythm\* OR improvis\*) OR ((auditory OR acoustic OR sound\*) NEAR/5 (stimulat\* OR cue\*)) AND (SU.EXACT.EXPLODE

("Pervasive Developmental Disorders") OR (autism\* OR asperg\* OR "pervasive development\* disorder\*" OR "childhood schizophrenia\*" OR Kanner\*)) AND (SU.EXACT.EXPLODE("Pervasive Developmental

Disorders") OR (autism\* OR asperg\* OR "pervasive development\* disorder\*" OR "childhood schizophrenia\*" OR Kanner\*)) AND (SU.EXACT("Experimental Groups") OR SU.EXACT("Control Groups") OR

random\* OR control\* or group\* or placebo\* OR trial\* OR blind\*)

#### Sociological Abstracts (Proquest)

1952 to current. Limited to year=2004-2011. Searched 8 September 2011. Limited to year=2004 to 2011 plus new terms for all years pre-2004 [4 records]

1952 to current. Limited to year=2011-2013. Searched 30 July 2013. Limited to year=2011 to 2013 [0 records]

((su.EXACT("Music" ) or ((music\* or guided imag\* or GIM or vibro-acoustic therapy\* or vibroacoustic therapy\* or Bonny\* or Nordoff\* or singing or song\* or choral\* or choir\* or percussion\* or

rhythm\* or improvis\* or ((auditory or acoustic or sound\*) near/5 (stimulat\* or cue\*)))) and (su.EXACT(("autism" )) or (autism\* or asperg\* or pervasive development\* disorder\* or childhood

schizophrenia\* or Kanner\*)) AND (random\* or placebo\* or trial\* or blind\*)

## **LILACS**

All available years searched 9 September 2011 [2 records]

Searched 30 July 2013. Limited to year=2011 to 2013 [0 records]

((Pt randomized controlled trial OR Pt controlled clinical trial OR Mh randomized controlled trials OR Mh random allocation OR Mh double-blind method OR Mh single-blind method) AND NOT (Ct

animal AND NOT (Ct human and Ct animal)) OR (Pt clinical trial OR Ex E05.318.760.535\$ OR (Tw clin\$ AND (Tw trial\$ OR Tw ensa\$ OR Tw estud\$ OR Tw experim\$ OR Tw investiga\$)) OR ((Tw singl\$

OR Tw simple\$ OR Tw doubl\$ OR Tw doble\$ OR Tw duplo\$ OR Tw trebl\$ OR Tw trip\$) AND (Tw blind\$ OR Tw cego\$ OR Tw ciego\$ OR Tw mask\$ OR Tw mascar\$)) OR Mh placebos OR Tw placebo\$ OR (Tw

random\$ OR Tw randon\$ OR Tw casual\$ OR Tw acaso\$ OR Tw azar OR Tw aleator\$) OR Mh research design) AND NOT (Ct animal AND NOT (Ct human and Ct animal)) OR (Ct comparative study OR Ex

E05.337\$ OR Mh follow-up studies OR Mh prospective studies OR Tw control\$ OR Tw prospectiv\$ OR Tw volunt\$ OR Tw volunteer\$) AND NOT (Ct animal AND NOT (Ct human and Ct animal))) [Words] and

(Mh MUSIC OR Mh MUSIC therapy OR (musi\$ OR GIM OR vibro-acoustic therapy OR vibroacoustic therapy ) ) [Words] and ( Mh autistic disorder OR Mh asperger syndrome OR autis\$ OR asperg\$ or PDD

or PDDs or ASD or ASDs)

ASSIA (Proquest)

ASSIA 1987 to current. Searched 8 September 2011. Limited to year=2011-2014 plus new terms for all years pre-2004 [4 records]

ASSIA 1987 to current. Searched 29 July 2013.

((su.EXACT("Music" or "Drumming" or "Melodies" or "Singing" or "Songs") or su.EXACT(("Music therapy")) or ((music\* or guided image\* or GIM or vibro-acoustic therapy\* or vibroacoustic

therapy\* or Bonny\* or Nordoff\* or singing or song\* or choral\* or choir\* or percussion\* or rhythm\* or improvis\* or ((auditory or acoustic or sound\*) near/5 (stimulate\* or cue\*)))) AND

(EXACT(("autism" or "Infantile autism")) or EXACT(("Asperger's syndrome" or "autistic spectrum disorders" or "Pervasive developmental disorders")) or (autism\* or asperg\* or pervasive

development\* disorder\* or childhood schizophrenia\* or kanner\*)) AND ((EXACT(("Clinical randomized controlled trials" or "Cluster randomized controlled trials" or "Double blind randomized

controlled trials" or "Randomized consent design" or "Randomized controlled trials" or "Single blind randomized controlled trials" or "Urn randomization")) or randomised or randomized or

randomly or trial\*))

### **ClinicalTrials.gov**

Searched 9 September 2011 and 30 July 2013 [0 records]

Conditions: autism OR autistic or asperger or aspergers or pervasive or ASD or ASDs or PDD or PDDS

Interventions: music

### **ICTRP**

Searched 9 September 2011 and 3 July 2013 [3 records]

Condition: autism OR autistic or asperger or aspergers or pervasive or ASD or ASDs or PDD or PDDS

Intervention: music

## **Appendix 2. Search strategies up to 2004**

Searches for the original review were based on the following Ovid MEDLINE strategy:

#1 MUSIC

#2 MUSIC THERAPY

#3 musi\*

#4 gim

#5 ((guided imagery) near music)

#6 vibroacoustic

#7 vibro-acoustic

#8 (#1 or #2 or #3 or #4 or #5 or #6 or #7)

#9 (asperger next syndrome)

#10 autis\*

#11 kanner\*

#12 (childhood near schizophren\*)

#13 (speech near disorder\*)

#14 (language near delay\*)

#15 pdd

#16 CHILD DEVELOPMENT DISORDERS, PERVASIVE

#17 (#9 or #10 or #11 or #12 or #13 or #14 or #15 or #16)

#18 (#8 and #17)

The search terms were modified to suit the requirements of the other databases searched. An optimal sensitive search strategy for randomised controlled trials was also used where necessary.

## **WHAT'S NEW**

Last assessed as up-to-date: 2 December 2013.



Date	Event	Description
2 December 2013	New search has been performed	A search for new studies was conducted, resulting in the inclusion of seven new studies; based on the added studies' findings, the categories of outcome measures were revised, new meta-analyses were performed, and pre-existing results and conclusions were modified
31 March 2013	New citation required and conclusions have changed	Updated review with two new authors.

## HISTORY

Protocol first published: Issue 3, 2003

Review first published: Issue 2, 2006

Date	Event	Description
5 November 2009	Amended	Minor edit in background.
10 November 2008	Amended	Converted to new review format.
21 February 2006	Amended	Minor update
29 January 2006	New citation required and conclusions have changed	Substantive amendment

## CONTRIBUTIONS OF AUTHORS

CG designed the protocol and co-ordinated the reviewing. MG co-ordinated this review's update. CG and MG searched for studies. CE, CG, and MG screened search results. CE, CG, KM, and MG extracted data, analysed data, wrote the report, and approved the full review.

*Contribution of previous authors:* Tony Wigram, co-author of the 2006 version of this review, contributed to the development of the protocol, extracted and analysed data, and helped with writing the original report.

## DECLARATIONS OF INTEREST

The authors of this review are clinically trained music therapists.

Christian Gold is an Associate Editor of the Cochrane Developmental, Psychosocial and Learning Problems Group, and has been involved in publications from two studies included in this review (Kim 2008;Thompson 2012a), none of which supported or influenced his work on this review.

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Cochavit Elefant - none known.

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## DIFFERENCES BETWEEN PROTOCOL AND REVIEW

In compliance with the developments in systematic review methods since publication of the first version of this review (Gold 2006), a distinction was made between primary and secondary outcome measures, and 'Risk of bias' tables and a 'Summary of findings' table were included in this update.

## INDEX TERMS

### Medical Subject Headings (MeSH)

Autistic Disorder [\*rehabilitation]; Child Development Disorders, Pervasive [\*rehabilitation]; Communication; Music Therapy [\*methods]; Randomized Controlled Trials as Topic

## **MeSH check words**

Child; Humans