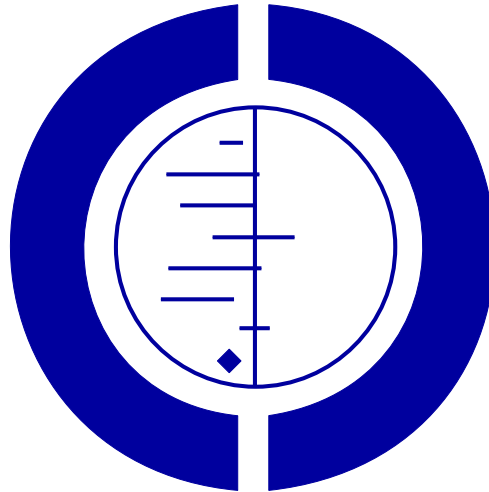


Music therapy for autistic spectrum disorder (Review)

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ABSTRACT

Background

The central impairments of people with autistic spectrum disorder (ASD) include social interaction and communication. Music therapy uses music and its elements to enable communication and expression, thus attempting to address some of the core problems of people with ASD.

Objectives

To review the effects of music therapy for individuals with autistic spectrum disorders.

Search strategy

The following databases were searched: CENTRAL, 2005, (Issue 3); Medline, (1966 to July 2004); Embase, (1980 to July 2004); LILACS, (1982 to July 2004); PsycINFO, (1872 to July 2004); CINAHL, (1982 to July 2004); ERIC, (1966 to July 2004); ASSIA, (1987 to July 2004); Sociofile, (1963 to July 2004); Dissertation Abstracts International, (late 1960's to July 2004). These searches were supplemented by searching specific sources for music therapy literature and manual searches of reference lists. Personal contacts to some investigators were made.

Selection criteria

All randomised controlled trials or controlled clinical trials comparing music therapy or music therapy added to standard care to "placebo" therapy, no treatment or standard care.

Data collection and analysis

Studies were independently selected, quality assessed and data extracted by two authors. Continuous outcomes were synthesised using a standardised mean difference (SMD) in order to enable a meta-analysis combining different scales, and to facilitate the interpretation of effect sizes. Heterogeneity was assessed using the I^2 statistic.

Main results

Three small studies were included (total $n = 24$). These examined the short-term effect of brief music therapy interventions (daily sessions over one week) for autistic children. Music therapy was superior to "placebo" therapy with respect to verbal and gestural communicative skills (verbal: 2 RCTs, $n = 20$, SMD 0.36 CI 0.15 to 0.57; gestural: 2 RCTs, $n = 20$, SMD 0.50 CI 0.22 to 0.79). Effects on behavioural problems were not significant.

Authors' conclusions

The included studies were of limited applicability to clinical practice. However, the findings indicate that music therapy may help children with autistic spectrum disorder to improve their communicative skills. More research is needed to examine whether the effects of music therapy are enduring, and to investigate the effects of music therapy in typical clinical practice.

PLAIN LANGUAGE SUMMARY

Music therapy for people with autistic spectrum disorder

People with autism spectrum disorders (ASD) have difficulties with communication, behaviour and/or social interaction. Music therapy uses music and its elements to enable people to communicate and to express their feelings. In this way music therapy addresses some of the core problems of people with ASD. This review set out to assess the evidence for the effectiveness of music therapy for individuals with ASD.

Three small studies were included which examined the short-term effect of brief music therapy interventions for autistic children. Music therapy was superior to “placebo” therapy with respect to verbal and gestural communicative skills, but it was uncertain whether there was an effect on behavioural outcomes. The included studies were encouraging, but of limited applicability to clinical practice. More research with better design, using larger samples, in more typical clinical settings is needed to strengthen the clinical applicability of the results and to examine how enduring the effects of music therapy are. 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.

BACKGROUND

Autistic spectrum disorder (ASD) is the core disorder of the pervasive developmental disorders as defined within the International Classification of Diseases and Related Health Problems, tenth edition (ICD-10) (WHO 1992) and the Diagnostic and Statistical Manual of Mental Disorders (DSM IV) (APA 1994). Individuals with autistic spectrum disorders have difficulties in all aspects of communication. They are said to lack a “theory of mind”, sometimes called lacking a “sixth sense”. They display impairments in social communication and social interaction and 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. The key construct is 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). People with autistic spectrum disorder also present with a pervasive inability to ‘mind-read’ (Baron-Cohen 1995), where a lack of perception and understanding of other people’s feelings, beliefs or emotions results in a consequential inability to respond appropriately. This has particular impact on social skills and interactions (Howlin 1998).

The clinical picture varies because individuals have variable levels of ability, from profound learning disability to a spiky cognitive profile where superior skills are present in some areas of functioning. Children and adults with autistic spectrum disorder frequently pose considerable behavioural challenges to their parents and other family members (Diggle 2002). Prevalence estimates for autistic spectrum disorder range from 5 to 63 children per 10,000, with more recent estimates showing much higher prevalence rates than those from older studies (Chakrabarti 2001, Fombonne 1999). At the high functioning end of the autistic spectrum is a disorder known as Asperger Syndrome, with the same fun-

damental core impairments as autism, but also some differences in language development, motor skills and originality of thought (Asperger 1979).

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 techniques in music therapy include free and structured improvisation, songs, and listening to music. The processes that occur within musical improvisation may help people with autism spectrum disorder to develop communicative skills and their capacity for social interaction. Musical interaction in music therapy, in particular musical improvisation, is sometimes understood and described as a kind of non-verbal and pre-verbal language which 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).

Music therapy for individuals with autistic spectrum disorders is usually provided as individual therapy. A rationale for the use of music therapy for individuals with communication disorders is based on the findings from infancy researchers such as Stern (Stern 1985, Stern 1989) and Trevarthen (Trevarthen 1999a) who describe sound dialogues between mothers and infants using “musical” terms. 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 tim-

ing (Wigram 2002). Trevarthen (Trevarthen 1999b) 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 autistic spectrum disorder. Necessary communicative behaviours, such as joint attention, eye contact and turn-taking are characteristic events in shared, active music making.

Clinical reports and pre-experimental studies suggest that music therapy may be an effective intervention for people with autism spectrum disorder. For example, Edgerton examined the development of communicative skills in eleven autistic children 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 autistic children change and develop during long-term music therapy (Schumacher 1999a, Schumacher 1999b). Two systematic reviews pertaining to the scope of this review have yielded conflicting results: One review (Whipple 2004) concluded that music therapy was effective for people with autistic spectrum disorders; however, interventions and study designs were too heterogeneous to allow clinically meaningful and methodologically strong conclusions. The other review (Ball 2004) concluded that effects were unknown, but failed to identify many possibly relevant studies. Thus, a more comprehensive systematic review of controlled studies in this area is necessitated.

OBJECTIVES

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

CRITERIA FOR CONSIDERING STUDIES FOR THIS REVIEW

Types of studies

All relevant randomised controlled trials (RCTs) and controlled clinical trials (CCTs).

Types of participants

Individuals of any age who are diagnosed with a pervasive developmental disorder, as defined in ICD-10 or DSM-IV, whether identified by a psychological assessment or a psychiatric diagnosis. 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) are not included as they do not conventionally fall within

the autistic spectrum disorders, given their significantly different clinical course.

Types of intervention

Music therapy (regular sessions of music therapy as defined above), delivered by a professional, compared with either "placebo" (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

Measures of the following including:

- Communicative and social skills, social interaction
- Quality of social interaction
- Behavioural problems (e.g. stereotypic behaviour)
- Attention and concentration
- Cognitive ability
- Hyperacusis (hypersensitivity to sound)
- Activity level
- Quality of life in both school and home environments
- Stress in the family
- Adverse events

Data sources could include 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 included only if the instrument was either a self-report or completed by an independent rater or relative (not the therapist).

SEARCH METHODS FOR IDENTIFICATION OF STUDIES

See: Developmental, Psychosocial and Learning Problems Group methods used in reviews.

The following search terms were used:

- #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)

Where necessary, 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.

Relevant trials were identified through searching the following databases:

Cochrane Central Register of Controlled Trials (CENTRAL, 2005, Issue 3)
Medline (1966 to July 2004)
Embase (1980 to July 2004)
LILACS (1982 to July 2004)
PsycINFO (1872 to July 2004)
CINAHL (1982 to July 2004)
ERIC (1966 to July 2004)
ASSIA (1987 to July 2004)
Sociofile (1963 to July 2004)
Dissertation Abstracts International (late 1960s to July 2004)

The following specific sources for music therapy literature were also searched:

musictherapyworld.net (accessed July 2004)
Music Therapy Research CD ROM (AMTA 1999)
Music Therapy World Info-CD ROM IV (Aldridge 2002)
References were also retrieved from related review articles (Ball 2004, Whipple 2004).

METHODS OF THE REVIEW

1. Selection of trials

Two reviewers independently inspected the titles and abstracts identified from the search. Potentially relevant papers were obtained. Any disagreement was resolved through discussion and/or consultation with a third reviewer.

2. Assessment of quality

The authors assessed methodological quality independently, and any disagreements were resolved by discussion. Assessment was made of all included studies, to consider the following questions: Was the assignment to treatment groups truly random?

Was allocation adequately concealed?

How complete was follow up?

How were the outcomes considered for people who withdrew?

Were those assessing outcomes blind to the treatment allocation?

Randomisation

Randomisation was judged as 'adequate' when computer-generated random numbers, a random numbers table, or coin-tossing were used to allocate participants to treatment conditions.

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

Allocation concealment

The trials were divided into quality categories as defined in the Cochrane Reviewers' Handbook (Alderson 2004), where:

A: adequate allocation concealment; participants and researchers were unaware of participants' future allocation to condition until after decisions about eligibility were made and informed consent was obtained;

B: unclear concealment; allocation concealment measures were not described in detail;

C: inadequate allocation concealment; 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. The rating was only used as a descriptive measure of study quality.

Blindness of assessors

It is not possible to blind either those who deliver music therapy or those who receive it to the nature of the intervention. Blindness of assessors, however, can be used and was assessed as a part of this review. Quality of blindness was determined primarily by whether those who assessed and coded outcome measures were blind to condition, and the quality of blindness was categorised where:

Adequate = assessor blind to condition;

Unclear = blinding of assessor not reported and information not available from researchers; and

Inadequate = assessor not blind to condition.

All of the above were included in the review.

3. Data management

3.1 Data extraction

This was performed independently by two reviewers (CG, TW). When necessary the reviewers contacted the authors of trials to provide missing data.

3.2 Loss to follow-up and intention to treat analysis

The included studies had complete data for all participants, and therefore an intention-to-treat analysis was straightforward. (If studies with drop-outs had been included, we would have examined the impact of drop-out rates using sensitivity analyses and imputing the negative outcome as appropriate.)

4. Data analysis

Data from washout periods in crossover studies were excluded from the analysis.

4.1 Binary data

No binary data were available from the included studies. We had planned to use risk ratio and number needed to treat statistics with 95% confidence intervals for binary outcomes.

4.2 Continuous data

For studies where outcomes were measured at several occasions during each treatment condition, we used only the last measurement at the end of treatment.

Where raw data were available, the distributions of values were visually checked in an attempt to detect 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. The alternative of using change data which tend to be less skewed was not considered because using change data may be less conservative than using endpoint data.

We used standardised mean differences (SMD) for the analysis of 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 on the same scale, either non-standardised weighted mean differences (WMDs) or SMDs could have been used. We decided to use SMDs even in this case in order to facilitate the interpretation of effect sizes as small, medium or large according to the guidelines that are commonly used in the behavioural sciences (Cohen 1988). It is noted that the choice of SMD versus WMD does not usually affect the significance level of the results, and the authors cautiously assessed whether such was the case.

All SMDs (regardless of whether the study was a parallel or a crossover 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. For crossover studies we took into account the correlations within the participants as recommended and described in the literature on meta-analysis of crossover studies (Elbourne 2002).

4.3 Meta-Analysis

A fixed effect model was used in all analyses. Fixed effect models have the advantage of being simpler and more easily interpretable. If a common effect size had not been tenable due to heterogeneity, a random effects model would have been considered. In addition to the fixed effect analyses, the authors also examined whether random effects analyses would have altered the results, and any such difference would have been reported.

5. Assessing heterogeneity

Consistency of results was assessed visually in the forest plot and by examining I^2 (Higgins 2002), a quantity which describes approximately the proportion of variation in point estimates that is due to heterogeneity rather than sampling error. We supplemented this with a test of homogeneity to determine the strength of

evidence that the heterogeneity is genuine. In case of heterogeneity, possible sources would have been investigated.

6. Assessing bias

Funnel plots were planned to be used to investigate any relationship between effect size and study precision, but were not used in this review due to the small number of included studies.

7. Sensitivity analyses

Sensitivity analysis were planned to be conducted to determine the impact of study quality on outcome if studies of different quality had been identified and included (for example studies with high attrition rates).

8. Subgroup analyses

The impact of clients' age and intensity of therapy (number and frequency of music therapy sessions) would have been examined in subgroup analyses had heterogeneity been found.

DESCRIPTION OF STUDIES

1. Excluded studies

The search yielded a total of 312 references. 52 of these were deemed possibly relevant and selected for closer inspection. Of these, 13 were excluded from the review because they concerned an assessment rather than an intervention (e.g. assessing traits of people with ASD using music therapy techniques). 24 studies were excluded because they did not have an appropriate design (5 studies with ABAB or similar designs, i.e. studies comparing different treatments that all participants received in the same order; 10 case series; 9 case studies). 7 studies employed an intervention involving only listening to music (e.g. auditory integration training), rather than music therapy. One study was excluded because the outcome measure was unclear (see table of excluded studies).

2. Awaiting assessment

Three studies (unpublished master theses) could not be retrieved and are therefore still awaiting assessment (Laird 1997, O'Dell 1998, Wood 1991).

3. Ongoing studies

One relevant longer-term study of improvisational music therapy was still ongoing when this review was written (Kim 2005).

4. Included studies

Three short-term American studies comparing music therapy to a "placebo"-type therapy met the criteria for the review (Brownell 2002, Farmer 2003, Buday 1995; see table of included studies). Two of the studies (Brownell 20002 and Buday 1995) were of crossover design; the remaining included study (Farmer 2003) was parallel group. Other characteristics of these studies are described below.

4.1 Length of trials

The length of all included studies was extremely short, varying from one (Farmer 2003) to four weeks (Brownell 2002). The duration of each treatment condition was only one week in each study. No later follow-up assessments were included in any of the studies.

4.2 Participants

The participants in the included studies were between 2 and 9 years of age, with 80-100% boys in each study. All participants had received a diagnosis of autism. In one study (Buday 1995), levels of retardation and severity of autistic traits were also specified. Participants in this study ranged from mildly to severely mentally retarded (according to DSM III-R) and from mildly to moderately autistic (according to the Childhood Autism Rating Scale, CARS).

4.3 Setting

The participants received therapy either at home, at school, or at an outpatient therapy centre.

4.4 Study size

All three studies had extremely small sample sizes, varying from four to ten participants per study. Crossover designs were used in two of the studies (Brownell 2002 and Buday 1995) to partly compensate for the small sample size.

4.5 Interventions

4.5.1 Music therapy

In all studies, music therapy was provided on a daily basis in an individual (one-to-one) setting. The duration of the music therapy condition was only one week in all studies.

All studies used a highly structured approach to music therapy, which is not unexpected given the American origin of the studies (see Wigram 2002, Gold 2005, and the discussion section of this review). All studies used receptive techniques (listening to music); one study also used active techniques (Farmer 2003).

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); 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 study (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 and in what ways the therapist improvised or otherwise played music together with the client.

4.5.2 "Placebo" therapy

All studies compared music therapy to some kind of "placebo" activity to control for the non-specific effects of therapeutic atten-

tion. Since in all 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 speech was used instead of singing (Buday 1995); or the same play activities were offered without using songs or music instruments (Farmer 2003). Therefore, the "placebo" conditions included many elements similar to the music therapy conditions, including some potentially active ingredients (but none that are specific to music therapy). The concept of a "placebo" therapy designed to control for the therapist's attention has been widely used in psychotherapy research (Kendall 2004), but has also been criticised (Lambert 2004; see the discussion section of this review).

4.5.3 Other conditions

One study (Brownell 2002) also reported the outcomes during a baseline and a washout period with no intervention. These data were not used in this review.

4.6 Outcome measures

4.6.1 Communicative skills: gestural

Non-verbal (gestural) communicative skills were examined in two studies (Buday 1995, Farmer 2003). Both studies addressed the participants' behaviour within therapy sessions. Independent observers counted the number of communicative gestures (e.g. imitating a sign or motion) in the session. In one study (Buday 1995), the outcome consisted simply of the frequency count of appropriate gestures within a session. In the other study (Farmer 2003) a completed gesture was given a score of 2 and an attempt a score of 1, and the outcome consisted of the sum of these scores for all attempted and completed gestures within a session. The exact criteria for what was seen as a communicative gesture were different between the two studies. The measures used for this outcome were not published.

4.6.2 Communicative skills: verbal

Communicative skills in verbal communication were addressed in the same two studies (Buday 1995, Farmer 2003). Independent observers rated in-session behaviour in a similar way as in the previous outcome only that it was the frequency of appropriate verbal responses that was counted for this outcome. Again, the measures used were unpublished.

4.6.3 Behavioural problems

One study (Brownell 2002) addressed individually targeted repetitive problem behaviour. Occurrence of behaviour was assessed outside therapy sessions. Independent observers (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.

METHODOLOGICAL QUALITY

1. Randomisation

Two studies (Buday 1995, Farmer 2003) stated explicitly that randomisation was used to assign participants to conditions. The remaining study (Brownell 2002) used the term “counterbalanced” to describe an assignment that was either random or quasi-random, but intended to be random. Methods of randomisation and allocation concealment were not specified in the studies.

2. Blindness and quality of outcome assessment

There was one single-blind study, with blinded assessors (Buday 1995). It was not reported whether blindness was used in the other studies. All studies used independent raters to assess outcomes. All studies reported a high inter-rater reliability for the assessment of outcomes (Brownell 2002 inter-rater reliability 0.86 to 0.94; Buday 1995: agreement rate 98%; Farmer 2003: agreement rate 91%).

3. Loss to follow-up

No dropouts were reported in any of the studies.

4. Overall

4.1 Performance bias

Medication levels were not monitored in the included studies. However, due to the short duration of the studies, it appears unlikely to assume significant differential change in medication might distort the results. All participants received the full therapy intervention as intended.

4.2 Data reporting and analysis

One study (Buday 1995) reported means, standard deviations, and F test results for the outcomes described above. From these statistics it was possible to calculate an SMD with a standard error as appropriate for crossover studies (see Elbourne 2002 and the section on continuous data in the method section of this review). For the other two studies, individual patient data were extracted from tables or graphs. We screened the data for skewness before data synthesis. The data of one study (Farmer 2003) 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.

RESULTS

COMPARISON 1: MUSIC THERAPY versus “PLACEBO” THERAPY

1. Communicative skills: gestural

Skills in non-verbal, gestural communication were measured on continuous scales addressing observed behaviour. The results showed a significant effect in favour of music therapy, suggesting that improvement in gestural communicative skills was more likely to occur with music therapy than with a similar therapy not

including music (2 RCTs, $n = 20$, SMD 0.50 CI 0.22 to 0.79). Results were consistent between the two studies ($I^2 = 0\%$; see figure 1.1).

2. Communicative skills: verbal

Observed skills in verbal communication were measured on continuous scales. The results showed a significant effect favouring music therapy over the “placebo” intervention, suggesting that improvement in verbal communicative skills was more likely to occur with music therapy (2 RCTs, $n = 20$, SMD 0.36 CI 0.15 to 0.57). The results were consistent between studies ($I^2 = 0\%$; see figure 1.2).

3. Behavioural problems

The frequency of observed problem behaviour was measured on a continuous scale. Data were only available from one trial with four participants, and results were not significant when examining only the last day in therapy (figure 1.3). However, when averaging participants’ behaviour over all days in therapy except the first one (figure 1.4), there was a significant effect suggesting that music therapy may be slightly more beneficial than a similar verbal therapy in reducing behaviour problems (1 RCT, $n = 4$, SMD -0.24 CI -0.45 to -0.03).

DISCUSSION

Findings

Music therapy was compared to a “placebo” therapy which attempted to control for all non-specific elements of music therapy, such as the attention of a therapist. Outcomes assessed included communicative skills and behavioural problems, and results were significant for communicative skills and reached borderline significance for behavioural problems, suggesting a beneficial effect of music therapy.

The effect sizes found in the results of this review can be interpreted in accordance with common guidelines for interventions in the behavioural sciences (Cohen 1988). The effect on non-verbal (gestural) communicative skills reached a medium effect size (SMD 0.50), which is seen as a clinically relevant magnitude when comparing an active therapy condition to a “placebo” therapy. The effects on verbal communicative skills were slightly smaller (SMD 0.36) and ranged between a small and a medium effect size. Considering that the “placebo” therapy possibly contained not only “non-active” but also some of the “active ingredients” of music therapy (see the discussion below), this can still be seen as a clinically relevant magnitude. It is however interesting to note that non-verbal communicative skills, which may be more closely related to the non-verbal communication within music therapy, seemed 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. Whether the effects of music

therapy on communicative skills are generalisable and persistent remains to be shown.

The data on behavioural problems were very limited. The outcome was only addressed in the smallest of the included studies. The effects were non-significant at the end of the short-term music therapy programme, and barely significant when averaging over the course of the therapy. Either way, the effect size was small. Clearly, more data are needed to draw any conclusions about the effects of music therapy on behaviour problems.

Applicability of findings

Music therapy conditions

The included studies were of limited generalisability to clinical practice. Only a limited subset of the music therapy techniques described in the clinical literature were used in the experimental treatment conditions. Receptive music therapy techniques with a high level of structuring predominated in the interventions; improvisational techniques were not mentioned. Approaches with a high level of structure, including behaviourally oriented and directed interventions, are more often applied in the North American context than in Europe, and the findings will therefore apply more easily to the former. However, improvisational techniques are widely used in many parts of the world (Edgerton 1994, Kim 2006, Wigram 2006).

The included studies illustrate the value of structure, which is generally an essential element for children with ASD. 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. Flexible but systematic treatment protocols for music therapy are currently developing in clinical practice and research investigations in autism (Kim 2005, Wigram 2006) as well as in other fields (Rolvsjord 2005). One small RCT is currently investigating the effects of improvisational music therapy (Kim 2005). Further such studies which are close to clinical practice will be needed to improve the applicability of findings.

Control conditions

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. Any conclusion from such comparisons will therefore address the effects of specific music therapy techniques, rather than the absolute effects of music therapy. This type of design is justified when exploring the intervention strategies in which music therapy works. 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 a specific cause-effect relationship. 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. 20-21). However, recent research on the 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. 150-152). In any case, the results of the included studies are likely to underestimate the true effects of music therapy, because the control conditions contain a number of potentially efficacious techniques which are also used in music therapy.

Duration and population

Time-limited, intensive treatments such as those examined in the included studies are relevant in acute care, and probably also in the light of limited available resources. However, ASD is pervasive developmental disorder leading to a chronic condition which requires sustained therapeutic intervention starting as early as possible. Music therapy is, in clinical reports for ASD, also described as a longer-term intervention, and given the typical emergence of entrenched and deteriorating behaviour, therapeutic intervention relies on consolidating progress over time. Conclusions about the potential benefits of longer-term music therapy cannot be drawn from the available studies. 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 adults with ASD.

Strength of the evidence

The limited information on randomisation methods and allocation concealment, the limited use of assessor blindness, the lack of using standardised scales, and the small sample size limit the methodological strength of the evidence. However, there was no performance bias (co-intervention) or attrition bias (drop-out), and inter-rater reliability was high, and these factors contribute positively to the strength of the evidence.

AUTHORS’ CONCLUSIONS

Implications for practice

The findings of this review indicate that music therapy may have positive effects on the communicative skills of children with autistic spectrum disorder. Music therapy has been shown to be superior to similar forms of therapy where music was not used, and this may be indicative of a specificity of the effect of music within music therapy. As only short-term effects have been examined, it remains unknown how enduring the effects of music therapy on verbal and non-verbal communicative skills are.

When applying the results of this review to practice, it is important to note that the application of music therapy requires an academic and clinical training in music therapy. Trained music therapists are available in many countries. 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. Academic training courses in music therapy exist in many countries, and information is usually available through the professional associations.

Implications for research

Relation of studies to clinical practice

More research is needed to examine the effects of music therapy over a longer term. Future studies should involve therapy conditions that are close to clinical practice, especially in terms of frequency, duration, and therapy techniques. This may include further efficacy studies as well as effectiveness studies. Standardised and published tools should be used to evaluate the outcome of music therapy.

Sample size, power, and the choice of research design

Future research on music therapy for people with ASD will need to pay close attention to sample size and power. All included studies had very small sample sizes and included no discussion of test power, possibly indicating that the issue has been ignored (see Gold 2004 for a discussion of the issue of test power in music therapy research). Limited sample size is common in research on ASD, and crossover designs seem an obvious choice because they help to improve test power. However, crossover designs entail other problems. First, longer-term follow-up assessments are not possible with such designs. Second, crossover designs are only adequate for interventions whose effects are thought to be short-acting (Elbourne 2002), but for music therapy it is not unreasonable to assume lasting effects if children learn new modes of behaviour as a result of therapy. In this case, crossover designs would be inappro-

priate due to carryover effects which would lead to the effect being underestimated. Large parallel group trials are necessary to resolve this dilemma. An informed discussion is needed about what designs are considered appropriate in research on music therapy for ASD.

POTENTIAL CONFLICT OF INTEREST

The authors of this review are clinically trained music therapists.

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*Indicates the major publication for the study

TABLES

Characteristics of included studies

Study	Brownell 2002
Methods	Allocation: quasi-randomised, possibly randomised ('counterbalanced') Blindness: independent assessor (teacher), blinding not reported. Duration: 4 weeks. Design: crossover.
Participants	Diagnosis: autism. N=4. Age: range 6-9. Sex: 4 M, 0 F. 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	
Allocation concealment	B
Study	Buday 1995
Methods	Allocation: randomised. Blindness: assessor blinded to the nature of the hypothesis and to treatment condition. Duration: 2 weeks. Design: crossover.
Participants	Diagnosis: autism N=10. Age: range 4-9. Sex: 8 M, 2 F. 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	
Allocation concealment	B

Characteristics of included studies (Continued)

Study	Farmer 2003
Methods	Allocation: randomised. Blindness: not known. Duration: 5 days. Design: parallel group.
Participants	Diagnosis: autism. N=10. Age: range 2-5. Sex: 9 M, 1 F. Setting: homes and therapy centres.
Interventions	1. MT 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	
Allocation concealment	B

Characteristics of excluded studies

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)
Brown 1994	not RCT/CCT (case series)
Brown 2003	not intervention study (assessment)
Bruscia 1982	not RCT/CCT (case study)
Carroll 1983	not MT (only sung instructions)
Chilcote-Doner 1982	not MT (rhythmic strobe and drumbeat)
Clauss 1994	not RCT/CCT (case series, ABACA design)
Dawson 1998	not intervention study (assessment)
Diez 1989	not intervention study (assessment)
Edelson 1999	not MT (AIT/only music listening)
Edgerton 1994	not RCT/CCT (case series)
Frissell 2001	not intervention study (assessment)
Goldstein 1964	not RCT/CCT (case study)
Gore 2002	not usable (unclear outcome measure)
Griggs 1997	not RCT/CCT (case study), not intervention study (assessment)
Hadsell 1988	not RCT/CCT (case series), not ASD (Rett syndrome)
Hairston 1990	not RCT/CCT (case series)
Heaton 1999	not intervention study (assessment)
Heaton 2003	not intervention study (assessment)

Characteristics of excluded studies (Continued)

Kolko 1980	not intervention study (assessment)
Krauss 1982	not RCT/CCT (case series), not ASD (apraxia, language delay)
Lee 2004	not RCT/CCT (case series)
Litchman 1976	not MT (listening to recorded nursery rhymes)
Ma 2001	not RCT/CCT (case series)
Mahlberg 1973	not RCT/CCT (case study)
Miller 1979	not RCT/CCT (case study)
Mottron 2000	not intervention study (assessment)
Mudford 2000	not MT (AIT/only music listening)
O'Connell 1974	not RCT/CCT (case study)
O'Loughlin 2000	not RCT/CCT - includes 3 case series where all received the same treatment (no. 1, 3, 4) and 1 case series with an ABA design (no. 2).
Pasiali 2004	not RCT/CCT (case series, ABAB design)
Rao 2001	not MT (headphones with vs. without music)
Saperston 1973	not RCT/CCT (case study)
Schmidt 1976	not RCT/CCT (case series, AB design)
Starr 1998	not RCT/CCT (case series)
Staum 1984	not RCT/CCT (case study)
Stevens 1969	not RCT/CCT (case series)
Thaut 1987	not intervention study (assessment)
Thaut 1988	not intervention study (assessment)
Toolan 1994	not RCT/CCT (case series)
Watson 1979	not RCT/CCT (case series, ABCA design)
Wimpory 1995	not RCT/CCT (case study)

Characteristics of ongoing studies

Study	Kim 2005
Trial name or title	Joint attention and attunement in improvisational music therapy with the autistic child
Participants	Diagnosis: Autism (diagnostic consents by two child psychiatrists). Exclusion criteria: diagnostic comorbidity; previous experience of music therapy or play therapy. N=15. Age: range 3-7.
Interventions	1. Music therapy: 12 individual sessions over 3 months. 2. Free play: 12 sessions over 3 months. Psychiatric medication allowed to change during trial period. Crossover RCT.
Outcomes	Pre/post (at baseline, crossover, and end of trial): 1. Mother-child free play interaction at home (MPI profile). Pre/post 2. Early Social Communication Scale (ESCS, Mundy et al, 1986, 2003), 3. Pervasive developmental disorder behavior inventory (PDDDBI-C, Cohen & Subhalter, 1999) Within sessions: 4. Treatment measurement: Every session videotaped. Treatment manual used.

Characteristics of ongoing studies (Continued)

Starting date 2003

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Notes

ANALYSES

Comparison 01. Music therapy vs. “placebo” therapy

Outcome title	No. of studies	No. of participants	Statistical method	Effect size
01 Communicative skills: gestural	2		SMD (Fixed) 95% CI	0.50 [0.22, 0.79]
02 Communicative skills: verbal	2		SMD (Fixed) 95% CI	0.36 [0.15, 0.57]
03 Behavioural problems (end of therapy)	1		SMD (Fixed) 95% CI	-0.14 [-0.42, 0.14]
04 Behavioural problems (using all measurements from 2nd day on)	1		SMD (Fixed) 95% CI	-0.24 [-0.45, -0.03]

COVER SHEET

Title	Music therapy for autistic spectrum disorder
Authors	Gold C, Wigram T, Elefant C
Contribution of author(s)	CG - designed the protocol, co-ordinated the reviewing, searched for studies, extracted and analysed data, and wrote the report. TW - extracted and analysed data, helped with writing the protocol and the report. CE - helped with data extraction and analysis and with writing the report.
Issue protocol first published	2003/3
Review first published	/
Date of most recent amendment	22 February 2006
Date of most recent SUBSTANTIVE amendment	29 January 2006
What's New	Information not supplied by author
Date new studies sought but none found	Information not supplied by author
Date new studies found but not yet included/excluded	Information not supplied by author
Date new studies found and included/excluded	Information not supplied by author
Date authors' conclusions section amended	Information not supplied by author
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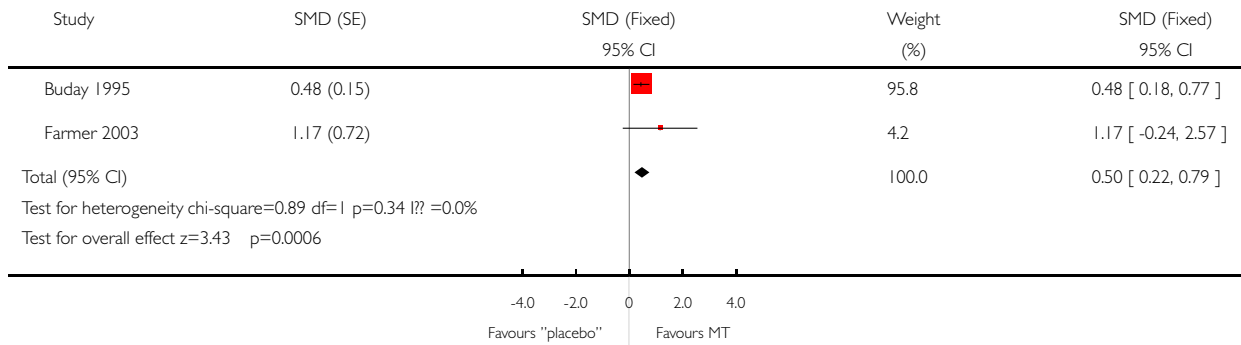
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GRAPHS AND OTHER TABLES

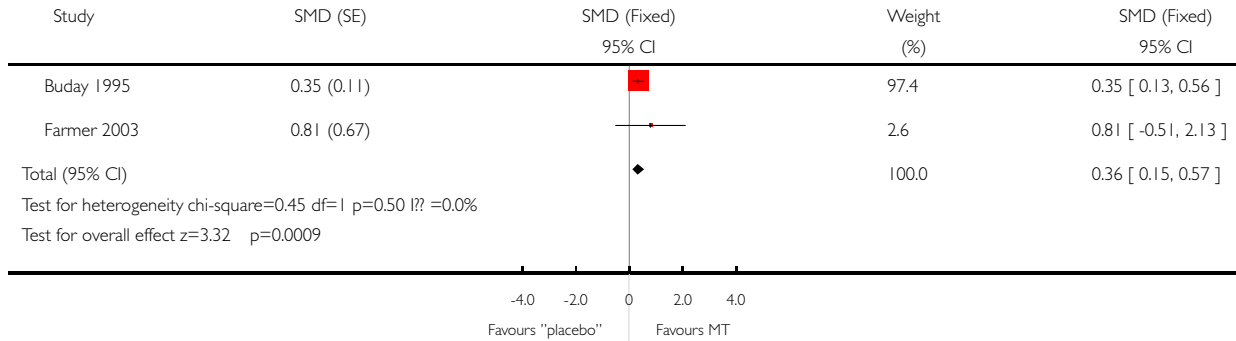
Analysis 01.01. Comparison 01 Music therapy vs. "placebo" therapy, Outcome 01 Communicative skills: gestural

Review: Music therapy for autistic spectrum disorder
 Comparison: 01 Music therapy vs. "placebo" therapy
 Outcome: 01 Communicative skills: gestural



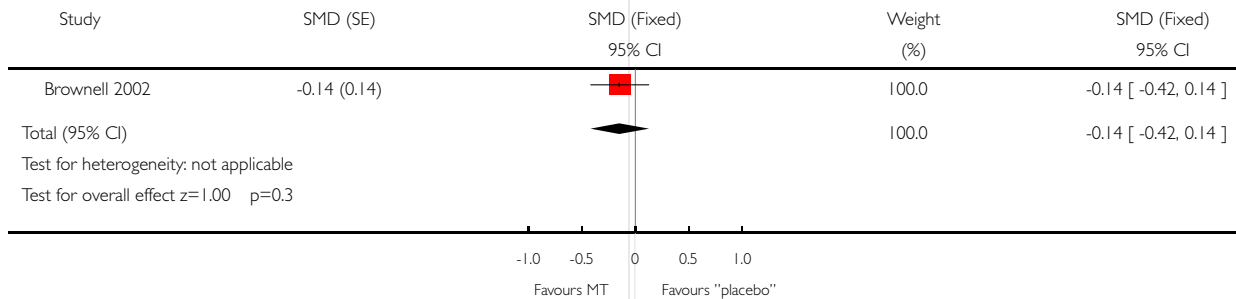
Analysis 01.02. Comparison 01 Music therapy vs. "placebo" therapy, Outcome 02 Communicative skills: verbal

Review: Music therapy for autistic spectrum disorder
 Comparison: 01 Music therapy vs. "placebo" therapy
 Outcome: 02 Communicative skills: verbal



Analysis 01.03. Comparison 01 Music therapy vs. "placebo" therapy, Outcome 03 Behavioural problems (end of therapy)

Review: Music therapy for autistic spectrum disorder
 Comparison: 01 Music therapy vs. "placebo" therapy
 Outcome: 03 Behavioural problems (end of therapy)



Analysis 01.04. Comparison 01 Music therapy vs. "placebo" therapy, Outcome 04 Behavioural problems (using all measurements from 2nd day on)

Review: Music therapy for autistic spectrum disorder

Comparison: 01 Music therapy vs. "placebo" therapy

Outcome: 04 Behavioural problems (using all measurements from 2nd day on)

